Louise Cummings *Editor*

Handbook of Pragmatic Language Disorders

Complex and Underserved Populations

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Foreword

The field of clinical pragmatics has witnessed considerable expansion in recent years. But while empirical studies of certain clinical populations have proliferated, other populations have languished in a state of almost complete neglect by clinicians and researchers. The reasons for this neglect are twofold. First, an understanding of conditions like psychiatric disorders, neurodegenerative dementias, and traumatic brain injury involves expertise from a range of disciplines, most notably psychiatry, neurology, and psychology, in addition to speech-language pathology and clinical linguistics. In clinical practice and theory, these disciplines tend to run in parallel to each other, which makes true interdisciplinary collaboration difficult to achieve. The result is that speech-language pathologists tend not to study and treat clients whose disorders are perceived to fall within the remit of specialists in fields like psychology and psychiatry, even though the expertise in language that speech-language pathologists can contribute is vital to an understanding of these disorders.

Second, not all clients with pragmatic disorders have equal access to clinical language services. Clients with substance abuse disorders and addiction, HIV infection, or who are detained in young offender institutions and prisons face social exclusion and marginalization. These clients often have social difficulties and psychiatric issues which may limit their access to, and compliance with, the very services that are best placed to address their pragmatic language difficulties. Other underserved populations include children in residential care and internationally adopted children who on account of linguistic, cultural and social factors may have pragmatic language problems that remain undetected. All these clients are underrepresented in the caseloads of speech-language pathologists notwithstanding their evident need for specialist language assessment and intervention.

This volume addresses the neglect of these children and adults by giving emphasis to complex and underserved populations of clients. In doing so, it addresses a significant gap in the clinical literature and responds to the needs of clinicians who often lack direction in the management of these clients. The chapters have been carefully crafted to ensure that they are accessible to students, researchers, and clinicians in speech-language pathology and related disciplines. The individual contributors to the volume are drawn from a wide range of disciplines, most notably speech-language pathology, but also psychology, psychiatry, neurology, paediatrics, and genetics. Collectively, they represent a vast body of clinical experience and academic learning in the conditions addressed by this volume. It is a wealth of expertise that can help establish new research priorities in clinical pragmatics.

Finally, a book can only claim to contribute to knowledge when it makes us look afresh at complex issues that we thought we understood or brings problems into focus that had previously evaded our gaze. It is hoped that in some small way, this book achieves both these outcomes for the many children and adults who must face the challenge of living with a pragmatic language disorder.

Kowloon, Hong Kong Louise Cummings

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Louise Cummings

1.1 Introduction

This is an unusual starting point for a chapter on clinical pragmatics. But I want the reader to join me in thinking about what daily life must be like for children and adults with a pragmatic disorder. The world with its millions of pieces of linguistic information and social signals must be a bewildering place to occupy. Children with pragmatic disorder must wake up in the morning unsure of whether they will be able to cope with the day's communicative challenges. They must hope that their attempts to join in games and other activities with friends in the playground will not be misunderstood and rejected. They must wonder if their teacher will not interpret their difficulties with communication as reluctance to engage or, worse still, bad behaviour and defiance. They must think about how they are going to indicate their food preferences to catering staff when they have not successfully achieved this on many previous occasions. They must worry about being read stories in class and having to answer questions about them for fear that they will not understand the narratives they have heard. And they must think about how they are going to ask the teacher or classroom assistant for permission to leave the room to attend the toilet. The difficulties for adults with pragmatic disorder are no less challenging. They must be concerned that they will appear awkward, inept, or even incompetent in front of their colleagues when they are asked to contribute to a meeting or give a presentation to others. They must think about how they are going to accept or decline an invitation to a friend's birthday party, or hold a conversation with colleagues over lunch. They must hope that they will not misunderstand an email from their line manager and make an impolite response in consequence. They must consider how to respond appropriately to a colleague who offers them a lift home.

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All these anxieties (and many more not mentioned) occupy the thoughts of children and adults with pragmatic disorders. Even those children and adults who are not fully cognizant of their pragmatic difficulties cannot escape the feeling of a lack of success in their everyday verbally mediated interactions. These difficulties limit the academic achievement of children, the employment prospects of young people and adults, and the social functioning of individuals of all ages (Cummings, 2014a; Snow & Douglas, 2017). Pragmatic disorders are also associated with psychological distress in the form of depression and anxiety and, for young males in particular, problems such as offending behaviour and engagement with the criminal justice system. These adverse consequences can be mitigated, if not wholly then partially, by effective and timely clinical language services. But what happens to those individuals who are not able to access these services, or whose pragmatic problems remain undetected or are poorly characterized? This is the central challenge for all clinicians who work with clients who have pragmatic disorders. In reflecting on how we can best address this challenge, we need to think about clinical populations which have been neglected to date by clinical language services. The individuals who constitute these populations may have complex neurocognitive and neuropsychiatric problems which are assessed and treated by professionals other than speech-language pathologists. Alternatively, they may experience social exclusion and a lack of cultural integration which may limit their access to services. It is these children and adults who are the focus of the chapters in this volume.

This chapter will unfold as follows. In Sect. 1.2, we examine some of the achievements and drawbacks of clinical pragmatic research which has been conducted to date. This research has produced an abundance of empirical findings, not all of which have facilitated our understanding of pragmatic disorders (Cummings, 2007). The reasons why this has occurred should be examined if we are to chart a productive road ahead. In Sect. 1.3, clinical populations which have traditionally not been prominent in the caseloads of speech-language pathologists are considered. The clients in these populations often have complex neurocognitive and neuropsychiatric problems which are assessed and treated by professionals other than speech-language pathologists. It will be argued that these clients must have access to the specialist services of speech-language pathology because of the interaction of these problems with language, and pragmatics in particular. These complex populations of clients have unmet pragmatic language needs. But they are not alone. In Sect. 1.4, we examine several other populations of clients who are underserved by speech-language pathology. They include children in residential care and adults in prison, both of whom may not have access to clinical language services because of factors such as social exclusion. Individuals with substance use disorders and other forms of addiction may not be able to comply with pragmatic language interventions. The pragmatic language needs of these clients are no less significant than those of many other clients with pragmatic language impairments who do receive clinical services. But they remain unaddressed for the most part because of societal prejudice and exclusion.

For clinical pragmatics to be fit for purpose in the twenty-first century, it must embrace these previously overlooked populations of clients. But it is worth asking why the pragmatic difficulties of these clients have been overlooked in the first place. Standard routes of referral between clinical services are certainly part of the explanation. Children with autism spectrum disorder access speech-language pathology services by means of referral from paediatricians and psychologists in child development clinics. However, there is not the same precedent for children with Tourette's syndrome to be referred for assessment by a speech-language pathologist even though these children can have significant pragmatic language difficulties (e.g. Eddy et al., 2010). Another part of the explanation is that speech-language pathologists have not been sufficiently proactive in making professionals like psychologists and psychiatrists aware of the relevance of clinical language services to the children and adults in their care. As a result of this lack of awareness, pragmatic language difficulties become subordinated to other behavioural problems in these clients. Also, speech-language pathologists may not have the knowledge and professional training that are required to assess and treat non-traditional clients (e.g. adults in prisons). Even if they do believe that they can offer effective clinical services to these clients, a lack of professional experience may dissuade them from this course of action. Also in Sect. 1.4, we examine these reasons in more detail, as an understanding of their true nature and complexity is vital to establishing a clinical pragmatics that can address the needs of clients in the twenty-first century.

Alongside the discovery of those factors that have led to the neglect of certain populations of clients in the past comes a responsibility to put clinical pragmatics on a firm footing for the future. This involves establishing new applications for clinical pragmatics which will sustain the continued development of the discipline. Chief among these applications is a new role for clinical pragmatics in the diagnosis of a range of disorders. This extends beyond the role that pragmatic language features currently fulfil in the diagnosis of primary pragmatic disorders such as social communication disorder. Instead, it will be argued in Sect. 1.5 that pragmatic features can also serve a role in the diagnosis of conditions such as dementia and schizophrenia (Cummings, 2012). This represents a new departure for clinical pragmatics into nosology and diagnosis. This departure is all the more significant given one of the great diagnostic challenges of our time, namely, the diagnosis of clients for whom there is a suspicion of dementia. What makes the diagnosis of dementia so challenging for clinicians is that there is a high degree of overlap in the initial presenting symptoms of several dementia syndromes. Also, there is a lack of a definitive, non-surgically invasive biomarker with which to make an *in vivo* diagnosis (Reilly et al., 2010). Against this backdrop, there are calls to develop reliable behavioural markers of the dementias. It will be argued in Sect. 1.5 that pragmatic language impairments have the potential to function as such markers.

1.2 Clinical Pragmatics: The Story So Far

Research into pragmatic disorders has proceeded apace in the last 40 years. From relatively small beginnings in investigations of speech acts (typically requests) in language impaired children (Rom & Bliss, 1983; Prinz, 1982; Prinz & Ferrier,

1983) and adults with aphasia (Wilcox & Davis, 1977; Hirst et al., 1984), the discipline has spawned an extensive array of empirical findings. There have been clinical studies into all the main pragmatic concepts including speech acts, implicatures, presupposition, deixis, context, and non-literal and figurative language (see Cummings (2009, 2014a) for an extensive review). This body of work has given clinicians and researchers considerable insight into pragmatic language function. For example, we now know that pragmatics is separable from structural aspects of language. An adult with non-fluent aphasia, for example, can have poor structural language skills (e.g. reduced grammatical structure) but still produce sufficient content words to be an effective communicator. By the same token, a child with pragmatic language impairment (or social communication disorder) can produce fluent, well-formed language. However, this same child might struggle to conduct a conversation or tell a story to a friend. We also know that improvements in structural language in adults with aphasia are not necessarily reflected in improvements in pragmatic communication (Coelho & Flewellyn, 2003), and that the pragmatic language system can selectively deteriorate in clients with early-stage dementia even as phonology and syntax remain intact (Cummings, 2021). Each of these findings has given support to the view that pragmatics is a rather unique type of competence within the wider cognitive architecture of the mind (see Cummings (2009, 2014a) for discussion).

But a separable competence is not necessarily a competence which is wholly independent of language. For it remains the case that certain linguistic structures are required in order to undertake pragmatic language functions such as producing speech acts and encoding information in the presuppositions of an utterance. An adult with agrammatic aphasia may not be able to perform the syntactic inversion that is required to produce indirect speech acts such as requests (e.g. *Can you close that window?*). This same adult may struggle to use lexical and grammatical structures that are known to generate presuppositions, including definite noun phrases (e.g. *<u>The house on the hill</u> is expensive → There is a house on the hill)*, cleft constructions (e.g. *<u>It was</u> the boy who broke the window → Someone broke the window*), and factive verbs (e.g. *Joan <u>regretted</u> leaving her job → Joan left her job*). It is an inescapable fact that several pragmatic language functions are intertwined with the ability to produce and comprehend syntactic and semantic structures. Much of the clinical pragmatic research which has been conducted to date serves to remind us that this is the case. For example, Katsos et al. (2011) found that children with specific language impairment (SLI) had difficulty comprehending statements which were quantified with expressions like ‘all’ and ‘some’. However, these children’s difficulties were comparable to those of younger, typically developing children with whom they were matched on a receptive grammar test. The finding that these children’s difficulties employing the maxim of informativeness are in keeping with their overall language difficulties is evidence, according to these authors, that pragmatic and grammatical competence are not the dissociable components that other investigators have contended.

If clinical pragmatic research has made possible an interesting line of inquiry into the pragmatics-language interface, it has permitted examination of another, equally important interface between pragmatics and cognition. In recent years, there has been prolific investigation into the relationship between pragmatics and theory of mind (Cummings, 2013, 2014b, 2015, 2017a). Theory of mind is the cognitive ability to attribute mental states to one's own mind and to the minds of others (Premack & Woodruff, 1978). Mental states include cognitive states such as knowledge and beliefs and affective states like happiness and sadness. Theory of mind allows us to predict and explain the behaviour of other people. This includes linguistic and non-linguistic behaviour during communication. It is by means of theory of mind that we are able to establish the communicative intention of the speaker who produces the utterance: *Do you know the time?* The communicative intention is the mental state that motivated the speaker to produce the utterance. In this case, the speaker does not know the time and wants his hearer to tell him the time. So the communicative intention can be described in terms of a desire to be given some information that the speaker currently lacks. A quite different communicative intention motivates the speaker who produces an ironic utterance like: *Your lack of generosity is so endearing*. In this case, the speaker entertains the belief that the hearer's lack of generosity is anything but endearing, and wishes to communicate this belief indirectly to the hearer by means of sarcasm. The same recovery process occurs in each of these instances of utterance interpretation. The hearer uses his theory of mind to recover the communicative intention that motivated the speaker to produce the utterance.

Theory of mind has proven to be a valuable explanatory concept in understanding pragmatic disorders in children and adults. We know that theory of mind in conditions like autism spectrum disorder, schizophrenia, and dementia is associated with pragmatic language impairments (Losh et al., 2012; Maki et al., 2013; Fukuhara et al., 2017). We also know why some pragmatic aspects of language pose a greater challenge to clients with pragmatic disorder than other pragmatic aspects of language. For example, the comprehension of sarcasm or irony deteriorates more rapidly for clients with Alzheimer's disease than the comprehension of metaphor (Maki et al., 2013). This is because sarcasm comprehension requires second-order theory of mind (the attribution of a mental state to the speaker about another person's mind) (Winner & Leekam, 1991), while metaphor comprehension requires first-order theory of mind (the attribution of a mental state to the speaker about the world). We also know that the relationship between theory of mind and pragmatics is unlikely to be a simple causal relationship. This is because the relationship appears to be mediated in some cases at least by executive functions such as working memory (Honan et al., 2015). Disordered pragmatic development in children can also be explained in terms of theory of mind. For example, delays in the acquisition of pragmatic language and nonliteral language in children with autism spectrum disorders have been found to reflect a delayed developmental trajectory in theory of mind abilities (Whyte & Nelson, 2015). These studies and many others not addressed here point to the versatility of the theory of mind concept in understanding the different ways in which pragmatics may be impaired in children and adults.

Theory of mind is merely one component of the cognitive substrate of pragmatic disorders (Bosco et al., 2018). Clinical pragmatic research has also investigated the relationship between pragmatic impairments and executive functions. Executive function is integral to the planning, execution, and regulation of goal-directed behaviour (Diamond, 2013). Key executive functions are inhibition, planning ability and organization, working memory, and attention. Clinicians have known for some time that executive function deficits are integral to the pragmatic communication difficulties of clients with traumatic brain injury (Douglas, 2010). But there is now a growing realisation that executive dysfunction is also associated with the communication difficulties of many other populations of clients, including adults with neurodegenerative diseases (Bambini, Arcara, Martinelli, et al., 2016b; Cummings, 2021) and right-hemisphere damage (Saldert & Ahlsén, 2007; Cummings, 2019a). (The reader is referred to Feyereisen et al. (2007) and McDonald (2000) for a different view of the relationship between executive functions and pragmatics in these populations.) It is as a result of clinical pragmatic research that we are beginning to understand the executive basis of what speech-language pathologists call 'cognitive-communication disorders' in these clients. Information management is impaired in many (or most) clients with cognitive-communication disorders. Information may be omitted, repeated, and poorly organized during discourse. Speakers may also convey incorrect and irrelevant information. We now know that these difficulties are related to executive deficits (Ash et al., 2011). We also know that problems with the use of cohesion in discourse have their basis in executive functioning (Ellis et al., 2015). With each study of this type that is conducted, more of the executive substrate of pragmatic disorders is revealed.

The reason cognitive accounts of pragmatic disorders have held such appeal is that they provide an explanatory framework for these disorders. In the absence of these frameworks, early studies in clinical pragmatics produced an abundance of empirical findings, not all of which shed light on the nature of pragmatic disorder (Cummings, 2007). Knowing that a child with pragmatic disorder cannot use cohesive devices like anaphoric reference during narrative production is certainly something very much worth knowing. But unless this aspect of a child's pragmatic function is explained in linguistic or cognitive terms (e.g. failure to retain an antecedent noun phrase in working memory), this knowledge does not progress our understanding of the child's pragmatic disorder (even less our ability to treat it). Many clinical pragmatic studies have also cast the net of pragmatics so widely that it is not clear what the term may be taken to exclude (Cummings, 2009). Not every aspect of communicative behaviour is pragmatic in nature. The ability to use facial expression to establish a speaker's communicative intention in producing an utterance is a social perceptual skill which has consequences for pragmatic language understanding. The fact that this skill contributes to pragmatic understanding does not thereby make it pragmatic – it is still a social perceptual skill. Finally, some clinical pragmatic studies have misused pragmatic concepts such as implicature, presupposition, and speech acts (Cummings, 2009). Simply recognising that a speaker has flouted a maxim is not tantamount to recovering the implicature of an utterance. Yet, this has been an assumption of several clinical studies of implicature (e.g. Surian, 1996). These drawbacks aside, this section has clearly demonstrated that clinical pragmatics can claim considerable achievements in its relatively short history to date.

1.3 Complex Clinical Populations

When a discipline first emerges, it can take some time for it to establish its scope and identity. As part of its continuing growth, a discipline may acquire new applications and areas of interest. These novel lines of inquiry are what sustain its future development and ensure that it remains relevant to all those who study it. Clinical pragmatics, I contend, is at this point in its development. It has made a substantial contribution to our knowledge of pragmatics in a wide range of clients including children and adults with autism spectrum disorder, traumatic brain injury, and social communication disorder. And that contribution will undoubtedly continue. But clinical pragmatics is now ready to address new clinical challenges and to move beyond its traditional areas of theory and practice. A significant challenge for the discipline comes in the form of clients who have pragmatic language impairments but who are not normally referred to speech-language pathology. This may be because their care is provided by medical or health professionals who do not recognise the need for referral. Alternatively, the presenting symptoms and behaviours for which these clients are receiving treatment may serve to mask their pragmatic language difficulties. A further challenge for clinical pragmatics comes from clients who are referred to speech-language pathology but for whom we lack a clear profile of their pragmatic communication difficulties. Many of these clients have complex neurocognitive and neuropsychiatric disorders which contribute to their pragmatic difficulties. However, the exact nature of that contribution is not well understood. In this section, we outline the challenge that these different clients pose for clinical pragmatics.

There is considerable heterogeneity among the children and adults who are served by speech-language pathologists. Clients of all ages, education levels, and social and cultural backgrounds are assessed and treated by speech-language pathologists. But while the clients of speech-language pathologists are heterogeneous, the conditions which they manifest are not for the most part. Certain clinical disorders have come to dominate the caseloads of speech-language pathologists. They include language disorders such as aphasia and specific language impairment and motor speech disorders like dysarthria and apraxia of speech. The language and communication problems that occur in clients with epilepsy or Tourette's syndrome are much less common or even non-existent in the caseloads of speech-language pathologists. This is not because these disorders have a low prevalence, or because there are few, if any, language and communication problems in these clients. Epilepsy is at least as prevalent as developmental stuttering in the general population[1] and its language and pragmatic impairments have been documented in clinical studies (Broeders et al., 2010; Debiais et al., 2007). We must find an alternative explanation of the lack of prominence afforded to these conditions if we are to understand why only certain clients with pragmatic disorders have been the focus of clinical pragmatics to date. That explanation should involve the following factors: (1) poor professional awareness of (pragmatic) communication disorders and the need for onward referral to speech-language pathology; (2) an understanding of how pragmatic impairments are manifested in clients with complex behavioural presentations; and (3) an understanding of how pragmatics may be compromised in neurocognitive and neuropsychiatric disorders. These factors are discussed below.

There is nothing new in the statement that many medical and health professionals have poor knowledge and understanding of communication disorders and of the work of speech-language pathologists. McCann et al. (2013) investigated awareness and knowledge of aphasia among 100 health professionals. Although health professionals had better awareness and knowledge of aphasia than members of the general public, it was still relatively low at 68% for awareness and 21% for knowledge. In a study of general practitioners, Nesbitt and Thompson (1995) reported poor awareness of the role of speech and language therapy in the management of clients with Parkinson's disease. What makes these findings so significant is that this lack of knowledge and awareness has consequences for the referral of clients to speech-language pathology. In the study conducted by Nesbitt and Thompson, referral analysis indicated that of 18 patients with Parkinson's disease referred to speech and language therapy, only one had been referred by a general practitioner. Keating et al. (1998) found that the referral rate to speech pathology services among paediatricians was associated with the quality of their training in and knowledge of communication development and disabilities. If awareness of communication disorders in general is poor, it is poorer still for pragmatic disorders. Many clients with pragmatic disorders have intelligible speech production. These clients can also often produce well-formed language. In the absence of striking communication difficulties like unintelligible speech production, it may not be immediately apparent to medical and health professionals that clients have a pragmatic disorder and should be referred to speech-language pathology. These factors explain, I believe, why many clients with pragmatic disorders have not accessed the services of speech-language pathology to date.

To address this lack of referral, speech-language pathologists need to identify the medical and health professionals who manage the care of clients with undiagnosed pragmatic disorders. For clients with conditions such as epilepsy and neurodegenerative diseases with and without dementia, the lead medical professional is usually a neurologist. For clients with genetic and other syndromes, paediatricians often lead the multidisciplinary team that provides assessment and treatment. Clinical psychologists manage the treatment of clients with obsessive-compulsive disorder

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[1] The Epilepsy Foundation of America (2020) reports that the prevalence of epilepsy in the US population is between 5–8.4/1000 persons per year or approximately 1% of the population. The point prevalence of developmental stuttering is also 1% (Bloodstein & Bernstein Ratner, 2008).

and reactive attachment disorders. Psychiatrists, psychologists, and educationalists are involved in the assessment and treatment of children with disruptive behaviour disorders. Speech-language pathologists must attempt to educate these different professionals about pragmatic communication disorders if referral of children and adults with these disorders to speech-language pathology is to occur. This educational effort will not be easy. Even experienced speech-language pathologists can struggle to identify pragmatic disorder in clients, especially when it occurs alongside other behavioural problems. There are, however, tools that professionals other than speech-language pathologists can use to help them identify clients with pragmatic disorder. One such tool is the Children's Communication Checklist (Bishop, 2003), a 70-item questionnaire which can identify pragmatic impairment in children with communication problems. The use of this checklist and other similar assessments will undoubtedly serve to improve the rate and accuracy of referral of clients with pragmatic disorder to speech-language pathology.

The accurate identification of clients with pragmatic disorder is a precondition of referral to speech-language pathology. But in clients whose pragmatic disorders have gone undiagnosed, identification is made difficult by complex behavioural problems. Children with disruptive behaviour disorders can display defiance of authority figures, angry outbursts, and other antisocial behaviours like lying and stealing. However, behaviours associated with pragmatic language impairment such as a failure to follow instructions or understand the communicative intent of a speaker who uses a speech act like '*Can you sit down?*' can easily be misinterpreted as acts of defiance. Also, it is difficult to discern if an outburst of anger is related to a disruptive behaviour disorder or is the inevitable consequence of the frustration that a young child experiences when he or she is unable to convey a message to a hearer. Disruptive behaviour disorders are not the only clinical condition where pragmatic language impairment may be effectively masked by behavioural symptoms. Children and adults with Tourette's syndrome exhibit simple and complex motor tics and vocal tics. Tics are not a feature of pragmatic language impairment. But motor and vocal tics, like pragmatic language impairment, disrupt gestural and verbal communication. If a client with Tourette syndrome had pragmatic language impairment, it is highly likely that its impact on verbal and non-verbal communication would pass undetected in the presence of motor and vocal tics. A child with reactive attachment disorder may display inhibition or hesitancy in social interactions. But so too may the child with pragmatic language impairment who has limited experience of communicative success and avoids social interaction in consequence.

Untangling the features of pragmatic disorder from the behavioural symptoms of these other conditions is complex and poses a significant diagnostic challenge for clinicians. The diagnostic specificity that is required is beyond our current knowledge of the clinical symptoms of pragmatic disorder and conditions like disruptive behaviour disorder. One way to ensure that clients with pragmatic disorder do not evade detection is for clinical evaluations of clients to be jointly conducted by speech-language pathologists and psychiatrists and/or psychologists. Joint evaluations of this type are only rarely conducted in clinical practice. But the potential that they create for discussion of the diagnostic weighting that should be attached to behavioural symptoms means that they are a productive way forward in the management of clients with complex behavioural presentations. Of course, joint clinical evaluations only work well when the professionals who are conducting them are as immersed in the terminology and frameworks of another clinician's discipline as they are in the terminology and frameworks of their own discipline. Once again, this requires a comprehensive educational effort on the part of all concerned. Speech-language pathologists must be prepared to educate colleagues in psychiatry and clinical psychology about pragmatic language impairment. For their part, psychiatrists and psychologists must make speech-language pathologists aware of the diagnostic criteria and protocols that guide their evaluations of clients with conditions like disruptive behaviour disorder and reactive attachment disorder. If conducted well, joint clinical evaluations could make a significant contribution towards reducing the lack of diagnosis and misdiagnosis of pragmatic language impairment in clients.

There is a further reason why certain clients with pragmatic disorders have not been prominent in the caseloads of speech-language pathologists. Many of these clients have pragmatic disorders against a backdrop of neurocognitive and neuropsychiatric dysfunction. Few speech-language pathologists have specialist knowledge of neurocognitive and neuropsychiatric disorders and their effect on language in general, and pragmatics in particular. All speech-language pathologists receive clinical education in the neuroanatomical and neurophysiological basis of aphasia and dysarthria. However, the same cannot be said of language disorder in neurodegenerative diseases like Parkinson's disease and in psychiatric conditions like schizophrenia and disruptive behaviour disorders. In recent years, considerable progress has been made in our understanding of the cognitive basis of language and communication disorder. Cognitive impairments in conditions like specific language impairment and developmental dyslexia have been widely investigated (Christo, 2014; Ellis Weismer, 2014). There is also considerable awareness of the role of theory of mind deficits in the communication problems of clients with autism spectrum disorder, and of the contribution of executive function deficits to communication problems in clients who sustain a traumatic brain injury (Cummings, 2009, 2013, 2014a, 2014b, 2017a). However, it still remains the case that much of this knowledge exists within the research base of speech-language pathology and is not yet part of the working knowledge of speech-language pathologists. The situation is even worse for neuropsychiatric disorders. Writing in 2001, Novak and Kapolnek describe the lack of clinical services for, and research into, clients with psychiatric disorders in speech-language pathology:

> Traditionally and in general, speech-language pathologists have not provided speech/language services for individuals with mental illness, and no articles have been found to be published on this topic in the *Journal of Speech, Language, and Hearing Research* from 1995 to date. (2001: 111)

These remarks remain as true today as they were nearly 20 years ago when they were made. Degrees in speech-language pathology rarely contain dedicated modules or courses on communication disorders in psychiatric conditions. The *International Journal of Language & Communication Disorders* published only two articles on mental health conditions in the 5-year period between January 2012 and January 2017 (one article on schizophrenia and one on emotional problems in childhood). It should not be surprising to discover that speech-language pathologists who lack formal training in certain clinical disorders, or who are unable to access research to guide their clinical management of clients, should end up not prioritising these clients or their pragmatic communication needs. It is once again the case that clients with pragmatic disorders, who could benefit from clinical language services, may remain undetected by these services.

1.4 Underserved Clinical Populations

A further aim of this volume is to highlight the pragmatic communication problems of several other groups of clients who also fail to access the specialist services of speech-language pathology. However, the reasons for this lack of access differ from the reasons we have just examined in Sect. 1.3. Clients with problems such as addiction and offending behaviour face social marginalization and exclusion. These social difficulties reduce the access of these clients to the healthcare services, including speech-language pathology, that are available to the rest of the population. Children in residential and foster care may have experienced severe physical and emotional neglect and sexual abuse at the hands of their biological parents. These events can place their social and emotional development at risk, with consequences also for language development. Residential and foster care can be fragmented, with children often experiencing multiple placements and different carers over relatively short periods of time. This lack of continuity in care may result in poor detection of language and pragmatic disorders and lead to reduced referral to speech-language pathology. It can be the case that as the number of agencies and individuals involved with the child increases, so too does the risk that a child's pragmatic language difficulties will not be undetected. As well as social barriers to clinical language services, there are also significant cultural barriers. Children who have been internationally adopted may experience pragmatic language problems. However, these problems may be dismissed as difficulty with cultural adjustment or misinterpreted as 'normal' pragmatic behaviour in a different cultural context. In this section, each of these underserved populations is examined in more detail.

There is a considerable burden of pragmatic disorder in the young offender and prison population. This burden arises in large part because pragmatic disorders are associated with several clinical conditions which have an increased prevalence in incarcerated individuals. These conditions include autism spectrum disorder, intellectual disability, schizophrenia, attention deficit hyperactivity disorder, traumatic brain injury, and conduct disorder (Cummings, 2017b). Pragmatic language impairments have particularly pernicious consequences for those juvenile offenders and prison inmates who have them. Individuals with pragmatic disorder are poorly equipped to comply with the verbally mediated rehabilitation programs which are available to inmates in prison. These programs are important in that they reduce rates of reoffending behaviour. They also help the offender achieve successful reintegration into society and secure employment on leaving prison. Rehabilitation programs address issues such as conflict resolution and encourage reflection on the factors that serve as triggers for an individual's offending behaviour. The metapragmatic and meta-cognitive demands of these programs are considerable and may exceed the pragmatic language skills of many inmates. To the extent that pragmatic disorder reduces engagement with these programs, early identification of inmates with pragmatic disorder must be a priority for clinical language services in prisons. It is unfortunately the case, however, that these services are lacking in many prisons. In written evidence in October 2016 to the UK Justice Committee inquiry into prison reform, the Royal College of Speech and Language Therapists stated that:

> There is a strong presence of speech, language, and communication needs within the prison population. There may not, however, be functional access to speech and language therapy which would allow access to rehabilitation programmes. This may be due to the following factors: a lack of identification of speech, language, and communication needs and the need for speech and language therapy as a result of a deficiency in workforce training; the availability of speech and language therapy services within prisons.

Clark et al. reported in 2012 that there was only one dedicated speech and language therapy service (21 hours per week) in Scotland's entire criminal justice system (Clark et al., 2012). Until the availability of speech and language therapy to the prison population is comparable to that of the population as a whole, it is difficult to see how prisons and other correctional facilities are going to achieve the successful rehabilitation of offenders. What is clear is that whatever clinical language services are made available to the prison population, pragmatics must be an integral part of them.

Even when individuals in prison do get access to clinical language services, they may have complex psychiatric problems which prevent them from complying fully with those services. There are high rates of substance use disorders and alcoholism in the prison population. In a systematic review of 18,388 prisoners across 24 studies, Fazel et al. (2017) reported that around a quarter of newly incarcerated male and female prisoners have an alcohol use disorder. The prevalence of a drug use disorder is at least as high in incarcerated men, and higher still in incarcerated women. There is also a high prevalence of alcohol and drug addiction in community populations (Arria et al., 2017; Krill et al., 2016). As well as reducing compliance with pragmatic language interventions, alcohol and substance use disorders are a risk factor for pragmatic language impairment. This may be on account of impaired theory of mind in individuals with alcohol and substance use disorders (Kim et al., 2011; Onuoha et al., 2016). After all, an individual who has impaired understanding of others' intentions and emotions (theory of mind) may also have impaired understanding of the communicative intentions involved in pragmatic interpretation. It appears that pragmatic language impairment can also increase liability for alcohol and substance use disorders. Najam et al. (1997) examined the language abilities of 135 children who were the offspring of men diagnosed as having a substance use disorder. These children, who were judged to be at high risk of drug abuse, were compared at baseline (10–12 years) and follow-up (16 years) to 208 children whose fathers had no psychiatric disorder or substance use disorder (low risk children).

High risk children obtained significantly lower scores than low risk children on subtests of the Test of Language Competence (Wiig & Secord, 1989) which assess pragmatic language skills. Specifically, the tests in question examined these children's ability to assign meaning to ambiguous sentences, comprehend metaphorical language, and express intents. At follow-up at age 16 years, high risk children were still significantly poorer than low risk children at comprehending ambiguous sentences and expressing intents. Najam et al. (1997: 78) concluded that '[i]mpaired linguistic ability, especially in those facets which involve the interpretation of abstract information […] appears to contribute to the liability for a substance use disorder'. Regardless of whether alcohol and substance use disorder is an independent risk factor for pragmatic language impairment, pragmatic disorder increases the risk of alcohol and substance use disorder, or both are a consequence of a third variable like theory of mind, it is clear that clients with problems of addiction do not access healthcare services to the same extent as the rest of the population (Palepu et al., 2013). This includes the clinical language services that speech-language pathology is able to offer. If community outreach programs are to be successful in tackling drug and alcohol addiction, physical and mental health needs of clients must be addressed. This includes problems with language and communication which, if left untreated, limit societal reintegration, the prospects of gaining employment, and participation in drug and alcohol recovery programs. Speech-language pathology has successfully adapted its services in the past to address the needs of clients. It must now do the same to address the language and communication needs of clients with alcohol and substance use disorder.

Incarcerated individuals and individuals with alcohol and substance use disorders are not the only marginalized clients who have undiagnosed pragmatic language impairments. Children in residential and foster care can also have pragmatic disorders which may remain unidentified, often with serious consequences for the social functioning and academic achievement of these children (Cummings, 2014a). The pragmatic impairments of these looked-after children may be missed for several reasons. A significant reason is that the number of referrals to speech-language pathology from social work departments with responsibility for these children is very low. Clark and Fitzsimons (2016) reported that one paediatric speech and language therapy service in a healthcare trust in Scotland received only 14 referrals from the local social work department in the last 5 years. This amounted to 0.13% of total referrals to the service in this period. This low referral rate may be explained by a lack of expertise and training on the part of social workers in the identification of pragmatic language impairments in the children in their care. Also, social workers have other professional priorities and responsibilities, chief amongst which is the secure placement of children with complex social and emotional needs in residential and foster homes. Language and communication difficulties may simply be overlooked against the backdrop of these other priorities.

A further reason why pragmatic language impairments may not be detected in looked-after children is that these children have often experienced chaotic home lives with their biological parents before being taken into the care of local authorities. The parents of these children may not have complied with the developmental checks that are conducted by health visitors between 0 and 5 years. Poor school attendance limits the opportunity of teachers to identify children with language problems. The language surveillance afforded by health and educational services for children in stable home environments is often not present for looked-after children. Another reason why looked-after children do not come to the attention of speech-language pathology is that multiple agencies, professionals, and foster carers are often involved in the care of these children. It might be thought that this would increase the surveillance of these children and with it the rate of detection of language problems. However, there is a significant risk that as the number of agencies and professionals involved in a child's care increases, language problems are not detected as each agency and professional focuses on a particular area of responsibility. This is even more likely to happen when communication between agencies and professionals is poor. In order for there to be improved detection of looked-after children with pragmatic impairments, it seems clear that speech-language pathologists must forge closer alliances with social workers and other professionals involved in the care of these children. Education and training in the recognition of pragmatic disorders must be an integral part of this effort.

Finally, there is another group of children with pragmatic impairments who have been underserved by speech-language pathology. However, these children do not lack access to clinical language services because of social reasons such as marginalization and exclusion. Children who have been internationally adopted are known to be at an increased risk of language impairment and pragmatic disorder (Petranovich et al., 2016; Rakhlin et al., 2015). It is not difficult to see why this is the case. Many of these children spend several years in institutions before they are accepted for adoption. During this time, they may receive less language stimulation than they might receive in a home environment. Pragmatic language skills develop early in young children as a result of the many everyday exchanges that occur between children and their parents and other adults. Children who are institutionalized in poorly staffed orphanages often receive little in the way of communicative interaction from the adults who care for them. Pragmatic language skills are particularly vulnerable to the lack of stimulation that this environment affords. If these children are eventually placed with an adoptive family, they must then embark on a process of assimilation and adjustment to the culture of a new country. This carries many hazards for these children who may already be trailing pragmatic language impairments from their time in institutions. The pragmatic language norms of a new culture may not be easily acquired, if acquired at all. To compound the difficulties of these children, pragmatic language impairments may be dismissed by the adoptive parents of these children as temporary difficulties with cultural adjustment. In recent years, there has been growing recognition among speech-language pathologists of the unique needs and challenges of internationally adopted children.

1.5 The Road Ahead for Clinical Pragmatics

Each population of clients examined in Sects. 1.3 and 1.4 will contribute to a new and more inclusive road ahead for clinical pragmatics. The relevance of clinical pragmatics in the twenty-first century can only be increased by consideration of the pragmatic difficulties of these hitherto neglected populations of clients. But there is another way in which clinical pragmatics can establish its relevance and value to clinicians and researchers in the years ahead. That way takes clinical pragmatics into the areas of nosology and medical diagnosis. These are not areas traditionally associated with pragmatics, or at least not as they are envisaged here. The proposal in this section is that clinical pragmatics is now at a point in its internal development where it can demonstrate its utility to other areas of enquiry by establishing new applications of its work and ideas. There is no more pressing application than that clinical pragmatics can play a significant role in the many diagnostic challenges that confront us in medicine and elsewhere. This new application will be examined in brief in this section, and is developed at length elsewhere (Cummings, 2012).

Speech-language pathologists have used pragmatic features of language for some time to diagnose primary pragmatic disorders[2] and to set these disorders apart from other conditions with which there appears to be some diagnostic overlap. For example, let us consider one of the long-standing issues in the nosology of child language disorder. Children who have good structural language skills but who struggle to use language in contextually appropriate ways have always presented clinicians with something of a diagnostic challenge. These children have normal non-verbal cognitive skills like children with specific language impairment (SLI). However, they lack the marked deficits in morphosyntax that typify children with SLI. At the same time, their pragmatic language impairments are similar in many respects to those of children with autism spectrum disorder (ASD). However, they lack the restricted interests, repetitive behaviours, insistence on sameness, and sensory abnormalities of children with ASD. This anomalous group of pragmatically impaired children has been variously labelled as having semantic-pragmatic disorder, pragmatic language impairment (PLI) and, most recently in the fifth edition of the *Diagnostic and Statistical Manual of Mental Disorders* (DSM-5; American Psychiatric Association, 2013), social communication disorder. For clinicians, the question is whether to characterize these pragmatically impaired children as a subgroup of children with SLI (reflecting the diagnostic overlap of PLI with SLI), or as a separate disorder which lies somewhere between SLI and ASD. Gerenser (2009) aptly captures this diagnostic quandary as follows:

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[2] Pragmatic language skills may be impaired on account of deficits in structural language (syntax and semantics) or as a result of cognitive deficits. Clients who have impaired pragmatic language skills in the presence of language and/or cognitive deficits have a *secondary* pragmatic disorder. However, in a *primary* pragmatic disorder, the pragmatic impairment does not arise on account of any structural language impairment or cognitive deficit.

> The question today involves the relationship between ASD, PLI, and SLI. There may be a closer relationship between PLI and autism than between PLI and SLI; PLI may be a subgroup of autism, typically described as high-functioning autism. An alternative to this concept is that some children with PLI may actually fall between the classifications of SLI and ASD – that is, these children demonstrate some aspects of SLI and some symptoms of autism, but they fail to reach diagnostic criteria for an autism spectrum disorder (74–75).

This is the type of diagnostic debate that clinical pragmatics has contributed to up until this point in time. And that contribution has been a significant one. This is reflected in the inclusion of social (pragmatic) communication disorder for the first time in the fifth edition of DSM. But I believe there is a more significant role still for pragmatics in nosology and diagnosis. Unlike PLI or social communication disorder, where pragmatic criteria are used to diagnose a primary pragmatic disorder, I contend that pragmatic features of language may also be used to diagnose psychiatric, cognitive, and behavioural disorders like attention deficit hyperactivity disorder (ADHD), dementia and schizophrenia (Bambini, Arcara, Bechi, et al., 2016a; Pawełczyk et al., 2018). This new diagnostic role for pragmatics is supported by several considerations, two of which are outlined here as they pertain to schizophrenia. First, the criteria that are currently used in DSM-5 to diagnose schizophrenia are essentially pragmatic in nature. Alogia or poverty of speech is a negative symptom[3] of schizophrenia. The speaker with alogia produces minimal, unelaborated turns which convey little information to the hearer. In failing to address the informational needs of his or her hearer, a speaker with alogia is in violation of the Gricean maxim of quantity – the speaker's utterances are under-informative. Disorganized speech or formal thought disorder is a positive symptom of schizophrenia. The speaker in this case produces language which lacks referential cohesion, contains irrelevant utterances, and is illogical and incoherent. Once again, the similarity of these features of disorganized speech to pragmatic language impairments is undeniable. The use of irrelevant utterances amounts to a violation of the Gricean maxim of relation. Utterances which lack cohesive links are unclear, ambiguous and difficult to follow. The Gricean maxim of manner has been compromised in this case.

Second, pragmatic language features in schizophrenia vary with the course and duration of the illness. Positive symptoms are most prominent in schizophrenia during the first psychotic episode and in the early stage of the condition. Over time, positive symptoms tend to subside and are replaced by negative symptoms. So clients with chronic schizophrenia have more negative than positive symptoms. To the extent that the symptoms of schizophrenia are pragmatic language behaviours, we might expect to see more pragmatic features like poor cohesion, irrelevance, and a lack of coherence (features of disorganised speech) in early-stage schizophrenia and verbal under-productivity and reduced information (features of alogia) in clients

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[3] Negative symptoms in schizophrenia are the absence of normal behaviours. They include alogia, avolition (lack of motivation) and a lack of affect. Positive symptoms in schizophrenia are the presence of abnormal behaviours. They include delusions (false and bizarre beliefs), hallucinations (the perception of things which do not exist), and disorganised speech. A diagnosis of schizophrenia is based on the presence of both types of symptom.

with chronic schizophrenia. This pattern of pragmatic features is supported by the findings of studies. Bearden et al. (2011) examined the speech samples of 105 adolescents, 54 of whom were considered to be at high risk of a first psychotic episode. At 1 year follow-up, adolescents who converted to psychosis used significantly less referential cohesion in their baseline speech samples than adolescents who did not convert to psychosis. Bowie et al. (2005) studied 220 geriatric patients with chronic schizophrenia. These investigators found that the verbal under-productivity of patients increased during a follow-up period of 2.3 years. However, scores for disorganized speech remained relatively stable during follow-up. Saavedra (2010) studied paranoid schizophrenic patients with duration of illness in excess of 20 years. A lack of cohesion in the narratives of a sub-group of these patients who had been long-stay residents in a care home had decreased to the point of almost disappearing.

Clearly, the psychopathology of schizophrenia lends itself to the type of analysis that must be possible if pragmatic features of language are to serve a role in the diagnosis of conditions other than primary pragmatic disorders. But for that role to be fully realized, pragmatic criteria must have greater diagnostic reach than just this one condition. Initial analysis suggests that this is indeed the case (Cummings, 2012). Symptoms of inattention and hyperactivity-impulsivity in ADHD can also undergo the type of pragmatic analysis that has just been conducted in relation to schizophrenia. An inability to wait on a speaker to complete a turn before starting the next turn and a tendency to blurt out an answer before a question is completed are both symptoms of hyperactivity-impulsivity in ADHD. But they are also pragmatic anomalies in the conversations of children and adults with ADHD. Even more exciting is the prospect that pragmatic criteria could become significant behavioural markers of the dementias. This could assist in the *in vivo* diagnosis of dementia. This is all the more important when one considers that dementia pathology can only be determined *post mortem* and is not a definitive guide to the type of dementia that a client may experience in any event. For example, as well as causing Alzheimer's dementia, Alzheimer's disease pathology accounts for around 19% of cases of primary progressive aphasia (Spinelli et al., 2017), a clinical dementia syndrome in which there is progressive deterioration of language functions alongside relative preservation of other aspects of cognition. It seems that pragmatic behavioural markers of dementia might have a diagnostic potential which exceeds that of even neuropathology itself.

The question naturally arises of what kinds of pragmatic impairments are likely to serve as diagnostic markers of different types of dementia. At this early stage, what can be said with some certainty is that a single pragmatic impairment is unlikely to distinguish one form of dementia from all other forms of dementia. It is unlikely to be the case, for example, that impaired comprehension of metaphor or irony will be able to distinguish clients with Alzheimer's dementia from those with vascular dementia or frontotemporal dementia. Pragmatic language skills operate across too many neural and cognitive levels for this to be a plausible scenario (Stemmer, 2017). But what does seem plausible is that constellations of pragmatic impairments could be used to differentiate types of dementia. In this event, a group of pragmatic impairments like poor referential cohesion, use of tangential utterances, and impaired comprehension of idioms might very well serve to distinguish different types of dementia. In two recent studies, the discourse of clients with Alzheimer's disease and primary progressive aphasia was examined (Cummings, 2019b, 2019c). Both groups of clients displayed reduced informational content in their respective discourses. This was the single most significant pragmatic anomaly for both groups of speakers with dementia – the discourse of these speakers failed to address the informational needs of listeners. However, apart from poor referential cohesion, which contributed to the informational difficulties of both groups of speakers, there was little overlap in the profiles of these clients. Lexical-semantic deficits made a large contribution to the discourse problems of adults with Alzheimer's disease, while executive planning problems were prominent in the discourse of adults with primary progressive aphasia. This work continues.

1.6 Summary

This chapter has reviewed some of the many achievements of clinical pragmatics in its relatively short history. It has been argued that if these achievements are to continue in the future, clinical pragmatics must look beyond its traditional client base and consider a range of other children and adults with pragmatic disorders. These clients have been overlooked by speech-language pathologists for a variety of reasons. Some clients have complex psychiatric, cognitive, and behavioural disorders that may mask pragmatic language impairments, making a diagnosis of these impairments difficult. Other clients experience marginalization and social exclusion on account of alcohol and substance use disorders, and fail to access the services of speech-language pathology on account of these difficulties. The clients in these complex and underserved populations deserve access to the same specialist language services that are available to the rest of the population. Ensuring that these clients achieve this access will be the next big challenge for all workers in clinical pragmatics. The chapter also addressed a new application of clinical pragmatics in the areas of nosology and diagnosis. The type of diagnostic work that pragmatic features of language might be expected to undertake was discussed in relation to schizophrenia, ADHD, and the dementias.

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Part I
Complex Populations in Childhood

Chapter 2
Social (Pragmatic) Communication Disorder

Geralyn R. Timler and Danielle Moss

2.1 Introduction

Robert is a 12-year-old who is performing within the average to above average range in most of his school subjects. Unfortunately, Robert is not as successful in his peer interactions. His peers, teachers, and parents describe Robert as someone who talks a lot, often providing more details about a topic than his conversational partners want to hear. He does not seem to be aware of, or able to interpret, the facial expressions of his peers. Therefore, Robert often misses the intended meanings of sarcastic messages delivered by a peer, such as “Oh yeah, Robert, we want to hear about that again”, when the peer’s facial expression and tone of voice clearly convey disinterest and frustration.

The diagnostic labels for individuals like Robert have varied over the decades. Rapin and Allen (1983) introduced the label “semantic-pragmatic syndrome without autism” to describe children with a primary deficit in pragmatic language despite “fluent expressive language” (p. 174). Their descriptions of the communication behaviours of these children included irrelevant responses to a partner’s questions and comments, difficulty with comprehension of abstract language, inappropriate use of language in specific pragmatic contexts, and the use of “canned phrases” (p. 179). Canned phrases, sometimes referred to as “scripted language” (for example, see Bishop, 2006), include phrases from movies or adult sentences that the child repeats without full understanding of the meaning of the phrase. As such, the child’s use of these phrases may not match the communicative context and can, therefore, be judged as incorrect, inappropriate, or unexpected by a communication partner.

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Bishop (1998, 2000) refined the diagnostic label to "pragmatic language impairment (PLI)" after finding that parent report of children's semantic skills did not always differ from typically developing peers even though report of their pragmatic language skills did. Parents reported difficulties in conversation and prosocial skills. Prosocial skills include a range of desirable behaviours such as sharing, helping, and cooperating with others to achieve a common goal, and negotiating to reach a mutually acceptable compromise when a conflict occurs. Fine-grain analyses of conversations between children with PLI and adult examiners revealed that these children were less likely than age-matched typical peers to respond to adult requests for information. Moreover, the children were less responsive verbally and nonverbally to requests for acknowledgments, such as "this is nice, isn't it?" (Bishop et al., 2000).

Further evidence that some children exhibit relative weaknesses in pragmatic language and prosocial skills when compared to their structural language skills (i.e. syntax, morphology, and semantics) was reported by Tomblin and his colleagues (Tomblin et al., 2004). They identified a subgroup of children within their epidemiological sample of 604 second graders, with and without language impairment, who had relatively poor "social communication" skills (p. 71) compared to their semantic and syntactic language skills. Teacher ratings of these children revealed difficulties in conversation management, including initiating conversation and maintaining topics, as well as difficulties in prosocial skills such as inviting others to join activities and giving compliments to peers. In short, descriptions of children like Robert have always reflected deficits in the pragmatic aspects of language defined as the "rules governing the use of language" (Bates, 1976). In turn, deficient pragmatic skills affect children's social communication success.[1] These deficits appear to have long-term impact as some adults with histories of PLI report having few, if any, close friends (Whitehouse et al., 2009).

2.2 Social (Pragmatic) Communication Disorder

The current diagnostic label for individuals like Robert is Social (Pragmatic) Communication Disorder (SCD). SCD was formally introduced in the *Diagnostic and Statistical Manual of Mental Disorders-Fifth Edition* (DSM-5; American Psychiatric Association, 2013) as a neurodevelopmental communication disorder

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[1] The distinction between pragmatic language and social communication is difficult to discern in clinical populations because a deficit in one of these developmental areas may adversely affect the other (Norbury, 2014). The terms 'pragmatic language' and 'social communication' are sometimes used synonymously. In fact, language experts are likely to label a behavior as demonstrating both a pragmatic language skill and a social communication skill if words are required to display the behavior. For example, effectively complimenting a peer is classified as reflecting both pragmatic language and social communication skills but complying with a teacher instruction is likely to be classified as a social communication skill only and not a pragmatic language skill (Izaryk & Skarakis-Doyle, 2017).

characterized by "persistent difficulties in the social use of verbal and nonverbal communication" (p. 47). Four symptoms that are required for the diagnosis include difficulties in (1) using appropriate communication for specific purposes; (2) changing communication to match the situation and the needs of the listener (e.g. speaking differently with adults than with a peer); (3) following rules for conversations and story-telling; and (4) understanding what is not explicitly stated, for example, misinterpreting sarcasm and nonliteral language such as idioms. Moreover, limitations in communication, social participation, social relationships, academic achievement, or job performance must be a consequence of specified SCD symptoms.

In addition to specifying the communication profiles of individuals with SCD, the DSM-5 provides a comprehensive summary of the onset and course of the disorder, associated co-occurring disorders, and risk factors. Parents may first note potential symptoms of SCD in the toddler years such as the child seeming to be uninterested in playing with others. Yet, a formal diagnosis of SCD should not be conveyed until children have demonstrated some advanced language milestones (e.g. storytelling/narrative discourse skills) in order to meet the four specified criteria. As such, it is expected that SCD may not be diagnosed until the late preschool years (American Psychiatric Association, 2013). Initial diagnosis may also occur in the school-age years and in adolescence, particularly for individuals with more subtle symptoms who struggle to meet the expectations of increasingly sophisticated and complex peer interactions. It is expected that symptoms of the disorder will be observed throughout the individual's lifespan although the impact of these symptoms in an individual's daily life will vary based upon the severity of the disorder and available supports from clinicians and peers (Section 2.7 provides a review of some of these supports).

Disorders that co-occur with SCD include attention-deficit/hyperactivity disorder (ADHD), behaviour disorders, and language disorder (LD).[2] A challenge for diagnosticians is that the word structure and grammar deficits observed in an individual with LD can also hinder conversation skills. As such, symptoms of LD cannot be the sole cause for the observed social communication deficits in an individual with SCD. Risk factors for SCD include a family history of autism spectrum disorder (ASD), communication disorders, or learning disabilities. Although ASD is a familial risk factor for SCD, DSM-5 stipulates that children should not receive a

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[2] The DSM-5 description of language disorder focuses on the structural aspects of language, characterized by deficits in vocabulary and grammatical knowledge and use, with subsequent impairment in discourse activities including difficulty sequencing events in a conversation so that the intended meaning is unclear to a communication partner (American Psychiatric Association, 2013). A recent international team of 57 experts has recommended the adoption of the term Developmental Language Disorder (DLD) to reference children and adults who have these linguistic deficits in the absence of accompanying intellectual disabilities, sensory deficits, or genetic syndromes (Bishop et al., 2017). Other terms for language disorder include specific language impairment (SLI) and language impairment (LI). For consistency, we use the DSM-5 term Language Disorder (LD), but also provide the specific diagnostic label used by the authors of research studies reviewed in this chapter.

dual diagnosis of ASD and SCD. Both children with ASD and children with SCD have primary difficulties in the social use of verbal and nonverbal communication, but children with ASD also demonstrate, or have a history of, restricted and repetitive behaviours, interests, or activities, usually abbreviated as RRBs. The differential diagnosis of SCD and ASD is somewhat complicated by the DSM-5 descriptors of RRBs that include stereotyped language and restricted interests (American Psychiatric Association, 2013). This is because some individuals with SCD may display these behaviours, particularly within preferred topics of interest (Norbury, 2014; Whitehouse et al., 2009).

Because SCD is a relatively new neurodevelopmental disorder, limited information is available about prevalence and incidence. In their research review of SCD, Swineford et al. (2014) noted that previous studies of children with PLI focused on small samples of children, precluding extrapolation of prior data for estimation of the percentage of children in the current population who may have SCD. Two recent studies do provide some preliminary information about prevalence. Kim et al. (2014) reported that the prevalence of SCD among 7- to 12-year olds in their population sample of 55,266 Korean children was 0.49% (95% confidence interval: 0.21–0.77). The sex ratio of males to females was 1.3:1. Mandy et al. (2017) reported on a clinically referred sample of 1081 children and adolescents, aged 4–18 years, referred to a social communication speciality clinic in England. Only 88 of these referrals, or 8.1% of the sample, met the diagnostic criteria for SCD. The majority of the children and adolescents (n = 801) received a diagnosis of ASD. The authors did not provide information about sample sex distribution.

Research efforts have been ongoing to clarify the communication and social profiles of children with SCD, ASD, and LD. Results to date suggest that the language and social differences among children with these disorders reflect a continuum of mild to severe impairment rather than discrete differences that fit neatly within separate categories. This research is reviewed in the next section. Subsequent sections provide recommendations for differential diagnosis of SCD and ASD, suggestions for a comprehensive assessment of communication skills including skills that support communication, and limitations of current assessment procedures in terms of consideration of cultural differences in social communication behaviours. The final section presents a review of intervention strategies and programs.

2.3 Review of Recent Research Differentiating SCD from Other Neurodevelopmental Disorders

Research efforts to further describe and differentiate the SCD clinical profile from the profiles of other neurodiverse populations have focused primarily on school-age children and adolescents with ASD and/or children with LD. We summarize several recent studies to illustrate research questions and methods that have been employed to examine these differences.

Gibson et al. (2013) explored similarities and differences in the social interactions of three groups of school-aged children aged 6–11 years. Children with pragmatic language impairment (the SCD group), ASD, and language impairment (the LD group) were observed in the playground interacting with a typical peer partner for 15 min. The interactions were rated using the Manchester Inventory for Playground Observation (MIPO; Gibson et al., 2011). The MIPO consists of four categories of social interaction behaviours: prosocial behaviours; conflict management; caregiving and confiding skills; and atypical stereotypic behaviours associated with ASD such as unusual sensory behaviours, preoccupation with one activity, and echolalia (Gibson et al., 2011). Higher ratings reflect fewer skills/more problematic behaviours. The results revealed that the SCD group's interactions were significantly poorer than the LD group but better than the interactions of the ASD group. Specifically, the SCD group received significantly higher (i.e. poorer) ratings in prosocial and atypical behaviours than the LD group, but these ratings were not as poor as the ASD group. As expected, the ASD group demonstrated significantly more atypical behaviours than the SCD and LD groups. The ASD group also demonstrated higher ratings (more difficulties) in caregiving and confiding skills than the other groups.

Bishop et al. (2016) examined how the social communication profile of children with ASD differs from that of children with other neurodevelopmental and psychiatric disorders. Their sample included children aged 2–12 years, with either ASD or another disorder including ADHD, language impairment (LD), intellectual disability, and mood or anxiety disorder. The Autism Diagnostic Observation Scale (ADOS; Lord et al., 2003), a standardized, semi-structured assessment of communication, social interaction, and play, was administered to all children. Exploratory factor analysis of ADOS items yielded a two-factor model of social communication: *basic social interaction* composed of items related to eye contact, facial expression, gesture use, and shared enjoyment; and *interaction quality* including items such as conversational initiations and responses, reciprocal interaction, and overall quality of rapport defined as a measure of how hard the examiner worked to maintain the social interaction. The results revealed that children with ASD performed poorly in both social communication factors. The children in the "other" disorder group also performed poorly in interaction quality, but did not generally demonstrate significantly reduced performance in the basic social interaction factor items. The authors suggest that deficits in basic social communication may be uniquely associated with the ASD profile while deficits in interaction quality are not unique to ASD. Although a SCD group was not included in this study, these results point to the need for fine-grain analyses of specific social communication behaviours in order to differentiate the social communication profiles of children with SCD from children with ASD.

Mandy et al. (2017) completed a large, retroactive case study review of 1081 children aged 2–18 years who were referred to a speciality clinic for children with social communication problems. The study utilized items from parent interview and multiple-report measures reflecting the children's ASD symptoms, communication skills, and behaviours (e.g. attention deficits, peer problems, prosocial behaviours, etc.). Differences among typical, SCD, and ASD groups were examined. The SCD and ASD groups demonstrated higher levels of structural language difficulties in speech production and syntax than the typical group. The SCD group's social relationships were rated more positively than the ASD group but were below the typical group. The SCD group's interest ratings were similar to the typical group, but the ASD group's ratings were significantly poorer than both groups, confirming that the presence of RRBs is a diagnostic marker for ASD but not SCD.

The proportion of participants within each group who received abnormal scores on the behaviour report measure was also analysed. More children in the SCD group demonstrated clinical levels of emotional and conduct problems, hyperactivity, peer problems, and compromised prosocial skills than the typical group, but the proportion of the children in the ASD group who had these difficulties was higher than the SCD group. The authors argued that these results yielded somewhat contradictory conclusions. The study provided support for the existence of SCD because over half of the SCD group demonstrated significant social communication impairments in the absence of RRBs. Yet, a number of the children identified as SCD demonstrated clinical levels of RRBs, but their social communication performance was just outside of the clinical range for ASD diagnosis. As such, the authors concluded that the SCD diagnosis represents a heterogeneous group of children, some of whom fall just below diagnostic criteria for ASD.

Adams et al. (2018) examined metapragmatic skills, defined as "explicit knowledge of pragmatics in social interactions" (p. 604), in children aged 6–11 years with SCD, developmental language disorder (DLD) and typical development. Parent report measures and direct assessment of linguistic abilities were used to classify children into one of the three groups. Although all children in the SCD group demonstrated clinical levels of social communication impairment, the authors noted that the group's language abilities varied more than the other groups, with some children demonstrating intact structural language skills while other children in this group did not. The dependent measures for this study focused on child responses to the Assessment of Metapragmatics (AMP; Collins et al., 2014). The AMP consists of 13 brief videos of social interactions between two child actors. One child violates a pragmatic rule while the other child provides a typical response to that violation. For example, the first child produces an off-topic or unexpected comment in the interaction and the other child appears confused or surprised. After the presentation of each film, the investigator asks a series of questions, "What went wrong?", "Why is that wrong?", "What could that boy (the violator) have done differently?" and finally, "What kind of boy is he?". For this final question, highest scores were given for answers that reflected psychological states such as "annoying" or "ignorant".

Group differences for responses to each question were explored after controlling for age and nonverbal IQ. Overall, the SCD group demonstrated less metapragmatic knowledge than the typical group. However, no differences were detected between the SCD and DLD groups with the exception of responses to "What went wrong?". The SCD group provided more pragmatic rules answers (e.g. "the boy said something that didn't make sense in the conversation") than the DLD group. Because both the DLD and SCD groups demonstrated similar weaknesses in awareness of pragmatic behaviours, the authors concluded that pragmatic knowledge may be an area of weakness for many children with communication disorders and not something that is unique to the SCD phenotype. Wide variations in the performance of both clinical groups suggest that more investigation is needed to further understand the factors that influence metapragmatic knowledge in both DLD and SCD groups.

Ash et al. (2017) examined if parent report of pragmatic language and social emotional behaviours yielded a unique SCD factor structure in a community sample of 125 boys and 85 girls aged 6–10 years. Some of the children in this community sample had learning disabilities or social emotional difficulties and approximately one third were receiving speech-language services. Children received a comprehensive battery of language tests and a nonverbal cognitive measure. Parents completed two report measures. The Child Behaviour Checklist (CBCL; Achenbach & Rescorla, 2001) is a measure of social emotional behaviours that yield six scales of disorder categories: affective disorder; anxiety disorder; somatic disorder; ADHD; oppositional defiant disorder; and conduct disorder. The Children's Communication Checklist-2 (CCC-2; Bishop, 2006) is the most widely used measure of children's structural and pragmatic language skills. The CCC-2 consists of 70 items and provides eight scaled scores of various language components including speech, syntax, semantics, and pragmatics plus two scales related to ASD behaviours in social relations and interests. The investigators examined a composite of the five CCC-2 pragmatic scales (i.e. PC-5) consisting of items that align with DSM-5 SCD symptoms. The scales and (sample parent-rated items) include: coherence (confuses the sequence of events when telling a story); initiation (difficulty to stop child from talking); scripted language (repeats things others have just said); context (realizes the need to be polite); and nonverbal communication (does not recognize when others are upset or angry).

Sex differences were examined for all measures. Boys and girls did not differ in the direct language measures and the CBCL report measure. Parent ratings of boys for the CCC-2 nonverbal communication and interests scales as well as the PC-5 composite score were significantly lower than girls. Sex differences in the PC-5 supported examination of separate factor analyses for boys and girls. For boys, the results revealed a three-factor solution of social emotional and behavioural competence, internalizing behaviours, and linguistic abilities. Only a two-factor solution was revealed for girls that excluded internalizing behaviours. The results of this study suggest that future research should examine potential sex differences in the SCD profile. The authors conclude that interprofessional teams of mental health professionals and speech-language pathologists/speech-language therapists will be needed to address the complex pragmatic and emotional-behavioural difficulties in children and adolescents with SCD.

The studies reviewed here reveal that children with SCD represent a heterogenous group in severity and significance of social communication impairment and linguistic abilities. Although the social performance of children with SCD is, by definition, significantly poorer than children without the diagnosis, these impairments are generally not as severe as those observed in children with ASD. The intriguing results from the Bishop et al. (2016) study, revealing that behaviours related to “basic social skills” separated children with ASD from children with other diagnoses but that “social interaction quality” did not, requires replication and further exploration. Future research may reveal specific areas of social deficits that uniquely distinguish the SCD and ASD clinical profiles. At present, the current evidence points to SCD as a milder form of ASD and the absence of RRBs in the SCD group as the primary difference from ASD. Although difficulty in pragmatic language is a hallmark characteristic of children with SCD, these children may also have structural language deficits that warrant an additional diagnosis of LD. Sex differences were also noted in pragmatic language ratings among a community sample of boys and girls with and without clinical concerns. The heterogeneity of the linguistic profiles of children with SCD point to the need for comprehensive assessment of language abilities to fully describe and then address the communication strengths and weaknesses of children with SCD.

2.4 Recommendations for Differential Diagnosis of SCD and ASD

When a child is referred to a diagnostic team for social communication concerns, the first goal of the team should be to determine whether the child has ASD or SCD. Whereas both clinical populations demonstrate significant social communication impairments, children with accompanying restricted and repetitive patterns of behaviours, interests, or activities should receive a diagnosis of ASD rather than SCD. The challenge for diagnosticians is that school-age children and older individuals with ASD may no longer display these patterns. It is, therefore, important that the diagnostic team collect a thorough history to ascertain if the individual displayed these behaviours in the past even if they are not currently observed or reported by caregivers.

Recommended diagnostic tools for documentation of RRBs include the Autism Diagnostic Observation Schedule-Second Edition (ADOS-2; Lord et al., 2012) and the Autism Diagnostic Interview-Revised (ADI-R; Rutter et al., 2003). Clinicians should note that the ADOS-2 is a direct assessment and, as such, the RRBs must be observed during the diagnostic session to confirm the presence of these behaviours. The ADI-R is a comprehensive interview assessment that elicits information about the history of RRBs even if these behaviours are no longer present (Bishop et al., 2016). Current best practice recommendations call for use of both instruments to achieve optimal diagnostic accuracy in the discrimination of ASD and SCD.

If ASD is ruled out during the diagnostic evaluation, further assessment is needed to confirm that the child meets all specified criteria for SCD. For example, neither the ADOS nor the ADI-R provide evidence for the ability to understand what is not explicitly stated such as nonliteral language, idioms, and metaphors (Foley-Nicpon et al., 2017). An inability to understand nonliteral language is the fourth symptom of persistent difficulties in the social use of verbal and nonverbal communication specified under SCD in DSM-5 (American Psychiatric Association, 2013). Moreover, the ADOS provides too few items to account for deficits in using communication for specific purposes, and for matching communication to situation and listener needs, the second and third symptom criteria of SCD in DSM-5. Details of how this further assessment should be conducted are examined in the next section.

2.5 Recommendations for Comprehensive Assessment of Social Communication

The symptom criteria for SCD in DSM-5 focus on observable verbal and nonverbal behaviours. However, these behaviours are supported by the interaction of multiple underlying cognitive abilities (Adams, 2005). Imagine a scenario whereby the examiner is making judgements about whether a child effectively met the needs of a listener when retelling a fun event that happened over the weekend (symptom criteria 2 and 3 in DSM-5). The examiner could rate the child on the following questions: Did the child produce grammatically correct sentences with appropriate word choice to convey the event? Did the child provide sufficient location and person background for the listener? Did the child attend to his or her listener's reactions and recognize when the listener was confused or wanted to hear more? The answers to these questions will reflect not only the child's syntactic, semantic and pragmatic skills, but also the child's social cognition, particularly theory of mind abilities, and executive function skills (Ketelaars et al., 2012).

Theory of mind is the ability to acknowledge and understand the mental states or thoughts, feelings, intentions, and motivations of others, particularly when these intentions and motivations differ from your own (Baron-Cohen et al., 1994). In the above scenario, the child needs to consider what the listener already knows. If the listener was present during the fun weekend event, limited background content must be conveyed. Alternatively, if the listener is a relative stranger to the child, the child will need to introduce the location and the people at the fun event. Executive functions refer to a variety of higher-order cognitive processes that support self-regulation when planning, attending to, and revising goal-directed behaviour (Pennington & Ozonoff, 1996). In this scenario, the child's self-regulation skills facilitate the child's ability to attend to and monitor listener interest, permitting the child to revise his or her comments if the listener is confused or to share more details if the listener appears especially entertained by one aspect of the retell. Theory of mind and executive function skills are particularly useful for the appropriate interpretation of sarcasm, an area of potential weakness for children with SCD. In order to interpret sarcasm correctly, a child must have some sense of the communication partner's intentions and be able to attend to and discern the mismatch between the partner's stated words, tone of voice, and facial expressions.

A comprehensive social communication assessment must document the child's strengths and weaknesses in observable verbal and nonverbal behaviours and the cognitive abilities that support or hinder the child's social communication success. Because LD, ADHD, and behaviour problems are associated with SCD (American Psychiatric Association, 2013), speech-language pathologists/speech-language therapists (SLP/SLT) and clinical psychologists must be integral members of the SCD assessment team. The SLP/SLT should conduct a screening and then, if warranted, undertake a comprehensive assessment of syntax and semantics to identify if LD is present. The clinical psychologist should collect caregiver report measures and complete observations to identify or rule out the presence of ADHD, depressive disorders, and executive function difficulties.

As is the case for most neurodevelopmental disorders, a gold standard instrument for the identification of SCD does not yet exist. Guidelines from the clinical practice literature suggest that a combination of report measures, criterion-referenced measures such as conversation and narrative language sample analyses, observation of interactions with peer and adult partners, and norm-referenced tests are needed to provide confirmatory evidence for the specified diagnostic criteria (Adams, 2002,

Table 2.1 Suggestions for comprehensive social communication assessment of school-age children and adolescents

Assessment procedure	Examples of assessment tools
Caregiver report measures	Children's Communication Checklist-2 (CCC-2; various editions available; Bishop, 2003, 2006) Theory of Mind Inventory-2 (ToMI-2; Hutchins et al., 2016) Behavior Rating Inventory of Executive Function, Second Edition (BRIEF-2; Gioia et al., 2015)
Conversation and narrative language samples (to include samples collected during peer interactions)	Semantic and syntactic analysis to assess for language disorder – see Miller et al. (2015) and Nippold (2013) for protocol and analysis details Pragmatic language analysis to identify SCD symptoms – see Adams et al. (2010, 2011), Landa et al. (1992), Landa (2011), and Prutting and Kirchner (1987) for examples of pragmatic rating scales and checklists; see Timler (2018b) for example of a language sampling elicitation protocol
Norm-referenced social language tests to assess comprehension of inexplicit language comprehension and use in oral and written language modalities; see Timler and Covey, 2021 for a psychometric review of these tests.	Comprehensive Assessment of Spoken Language-2 (CASL-2; Carrow-Woolfolk, 2017) Clinical Evaluation of Language Fundamentals-5: Metalinguistics (CELF-5: Metalinguistics; Wiig & Secord, 2014) Social Language Development Test-Adolescent: Normative Update (SLDT-A: NU; Bowers et al., 2017) Social Language Development Test-Elementary: Normative Update (SLDT-E: NU; Bowers et al., 2016) Test of Integrated Language and Literacy (TILLS; Nelson et al., 2016) Test of Pragmatic Language-Second Edition (TOPL-2; Phelps-Terasaki & Phelps-Gunn, 2007)

2015; Izaryk et al., 2015; Norbury, 2014; Swineford et al., 2014). Suggestions for assessment procedures and tests to complete a comprehensive social communication assessment are provided in Table 2.1.

Parent- and teacher-report measures and, if appropriate, self-report measures from school-age children, adolescents, and adults can provide documentation that the individual has difficulty choosing expected words and phrases in various social situations and in reading and displaying verbal and nonverbal social cues. In addition, report measures can be used to assess children's social cognitive skills, that is theory of mind, and executive function skills. Report measures consist of items that directly align with SCD verbal and nonverbal behaviours or the skills that support or hinder those behaviours. For example, items from the Children's Communication Checklist-2 (CCC-2; Bishop, 2006) include statements such as "talks repetitively about things no one is interested in", "does not recognize when people are upset or angry", and "uses terms like 'he' or 'it' without making it clear what is being talked about." The Theory of Mind Inventory-2 (ToMI-2; Hutchins et al., 2016) consists of 60 statements designed to tap a wide range of social cognitive understandings. Caregivers rate statements such as "My child understands whether someone hurts another on purpose or by accident" using a continuum of response choices anchored by 'definitely not', 'probably not', 'undecided', 'probably', and 'definitely'.

Conversation samples with multiple partners, including samples collected during peer interactions, are useful for documenting the concerns expressed by parents and teachers. For example, samples can be used to document off-topic and unexpected responses to comments or questions, and difficulties with turn-taking. It is also important to examine the individual's display of facial expressions and other forms of nonverbal communication as well as to document the individual's limitations in noticing or interpreting his or her communication partner's facial expressions and gestures.

Rating scales and checklists can be used to analyze these samples. For example, Timler (2018b) provides suggestions for collecting multiple discourse genres within a single language sample protocol. Specifically, this 10- to 12-min protocol elicits three samples: personal retell, exposition, and a narrative of a favourite book or movie retell. Clinical expertise, however, is required to provide judgement about the adequacy of these samples. Timler (2018b) suggests utilizing tools such as the Targeted Observation of Pragmatics in Children's Conversations (TOPICC; Adams et al., 2010, 2011). This rating scale utilizes 14 pragmatic categories (e.g. "giving too many details", "difficulties responding to questions"). Each category is rated on a scale from 0 (i.e. "is never observed and the behaviour is typical of mature interaction style") to 3 (i.e. "marked evidence of that behaviour across conversation; maybe very frequent or degree of abnormality tends to dominate the flavour of the conversation to the detriment of the interaction"). The clinician can also include specific examples of pragmatic behaviours for each category.

Narrative samples may reveal the individual's challenges in formulating stories organized within a traditional story grammar framework. This involves a setting, an initiating event, a character's internal response to this event, the character's plan to resolve or address the event, a well-sequenced set of actions to carry out the plan, and a resolution that satisfactorily ties all story events together (Stein & Glenn, 1979). Use of cohesive devices in the narratives, such as articles and pronouns, can also be examined to determine if the child is able to retell a story that makes sense to the listener (Adams, 2002). For example, if the narrator is retelling a story with two male protagonists, the narrator will need to clearly identify the pronoun "he" with the specific referent/male character in the story. If the narrator fails to indicate which male protagonist he/she is referring to by using specific referents, it can result in a communication breakdown, or the listener being unable to follow or understand the story. In this example, the ability to navigate the use of cohesive devices and specific referents can also be related to the narrator's theory of mind abilities or difficulty managing multiple aspects of discourse simultaneously.

Norm-referenced language tests of figurative language, idioms, multiple-meaning words, inferences, and sarcasm should also be administered to document difficulties in understanding what is not explicitly stated. Various tests from the Comprehensive Assessment of Spoken Language-Second Edition (CASL-2; Carrow-Woolfolk, 2017) assess comprehension of idioms (e.g. "all eyes on the board"), sarcasm (e.g. "Mom and dad are cleaning the house while their 16-year-old is watching TV. Dad says, "I hope *her Royal Highness* is enjoying herself." What did he mean?"), double meanings (e.g. She took the *cue*), and pragmatic rules (e.g. "Molly is offered dessert at her friend's house, but she doesn't want any. What should she say?"). Other tests listed in Table 2.1 require children and adolescents to act out a particular social situation or to state how they would resolve a peer conflict.

One caveat to use of these tests is that some children and adolescents score within the normal range on these measures even though parents and teachers report significant social communication concerns (Timler, 2018b). Real life social interactions are dynamic, requiring continuous monitoring of a partner's facial expressions, words, and gestures as well as the individual's own perceptions and motivations for the interaction. In contrast, social language tests are static, and are usually administered in quiet test rooms with limited distractions while viewing single pictures or thinking about an isolated event. As such, parent and teacher report measures should be considered the most authentic evidence of everyday performance when there is a discrepancy among report measures and test measures.

Finally, it is important to assess comprehension of figurative language, inferences, and other aspects of nonliteral language use in written language. If a child or adolescent struggles with comprehension of oral language, similar struggles will be observed in written language skills. Children and adolescents learn about the management of social situations, character motivations, and emotion vocabulary through both listening and reading as they advance in school. As such, these difficulties not only affect the ability to learn about how to manage social situations but could impact academic performance as well by reducing performance in reading tests and subjects such as language arts. Therefore, reading comprehension tests that include inference questions, or facts not explicitly written in the paragraph, as well as interpretation of character emotions and intentions should also be included in the assessment protocol.

2.6 Cultural Considerations

The specified symptoms of SCD include impairments in social behaviours such as greeting, sharing information, using eye contact, and maintaining expected distance between partners. Cultural practices modulate these behaviours. For example, within white subcultures of the United States, it is generally expected that children and adults maintain eye contact during a conversation (LaFrance & Mayo, 1978). However, it is documented in the literature that the amount and type of eye contact varies across both race and culture (Harrison et al., 2017). As such, cultural differences, such as extended periods of gaze aversion, may be interpreted as unusual, disinterest, impoliteness, or even rudeness (Harrison et al., 2017). In some non-western cultures, it may be considered disrespectful for a younger person to look an older person directly in the eye as the lowering of eye gaze is an expected display of respect (Norbury & Sparks, 2013). The methods for identifying and treating SCD include a combination of behavioural observations and parent report. When using these assessment tools, the clinician must consider if the behaviour of interest is consistent with, or deviant from, expected social communication behaviours within that child's culture.

The question that arises from this clinical decision is: what does appropriate and expected social communication look like? The answer depends on the perspective of the person who is asking and who is being asked about. The question, then, should be: what does appropriate and expected social communication look like for this particular child? Therefore, clinicians should be cognizant of the cultural variability in social norms, rules, expectations, and behaviours. Due to immigration and globalization, clinicians are likely to assess and treat individuals outside of their own culture. Moreover, individuals may be bi-cultural such that children are exposed to a diverse set of expectations from first- and second-generation immigrant relatives. Thus, awareness of potential cultural differences between the clinician, the culture of the surrounding environment, and the family of the child being served should be considered during the diagnostic and assessment process. Also, clinicians and families must work together to develop a culturally aware intervention plan that addresses the teaching of culturally expected verbal and nonverbal behaviours.

Social communication is comprised of complex and nuanced interactions that can be difficult to explain and assess outside of one's culture. Existing tests and rating scale items consist of culturally mediated verbal and nonverbal social communication behaviours. Clinicians should be aware that currently, the diagnosis and treatment of developmental disorders mostly rely on criteria constructed from Western and North American European research (Norbury & Sparks, 2013). Therefore, clinicians must be aware of and continue to work towards cultural competence, which involves acknowledging, understanding, and responding to cultural variables that derive from the child, family/caregivers, and the environment (American Speech-Language-Hearing Association, n.d.).

2.7 Social Communication Interventions

A social communication assessment must ultimately inform intervention planning. Optimal outcomes from a comprehensive assessment include a thick description of the child/adolescent's weaknesses that hinder social success as well as strengths that may support social success. As noted in Sect. 2.5, children and adolescents with SCD are likely to have one or more additional communication or behaviour disorders that will need to be addressed prior to, or simultaneously with, the implementation of a social communication intervention. Children and adolescents with co-occurring LD need direct support for the syntactic and semantic deficits that may be impacting academic and social success (see Paul et al. (2017) for review of relevant language interventions). If ADHD is present, a pharmaceutical intervention and/or behavioural treatment plan to enhance focus and motivation for participation in a social communication intervention may be needed (see Hodgson et al. (2014) for review of behavioural programs for students with ADHD).

We do not yet have sufficient research data to support the selection of one intervention over another for children and adolescents with SCD. Instead, recommendations for best practices in intervention selection focus on matching the child/adolescent's specific profile of strengths and weaknesses with an intervention (or interventions) that address the child/adolescent's needs. In this section, we review three evidence-based approaches to address social communication in school-age children and adolescents. These approaches include the most common approach, which is the development of an individualized program to target the specific social communication profile of a child or adolescent with SCD. The final two approaches are curriculum-based, social skills group interventions for teaching and practicing the skills needed for particular social situations (e.g. entering a peer group, resolving conflicts, asking someone out, etc.), and peer support interventions that focus on the peers of children and adolescents who have significant social communication impairments. We use the term 'social communication impairment' rather than SCD here, because the social skills and peer support interventions highlighted below have primarily been studied in children and adolescents with ASD. It is likely that some children with SCD would benefit from the additions of social skills and peer support interventions, but to date, no research evidence is available to back this assertion.

Individualized interventions are typically conducted within one-to-one or small group therapy sessions. These interventions directly address children/adolescents' specific linguistic, pragmatic, and social cognitive deficits. Arguably, one of the most all-inclusive individualized programs for treating the complex needs of children with SCD is the Social Communication Intervention Programme (SCIP; Adams & Gaile, 2015; Adams, Gaile, et al., 2012; Adams, Lockton, et al., 2012). The SCIP treatment manual provides comprehensive assessment instructions to help clinicians select appropriate therapy goals, implement detailed therapy lessons, and engage parent/teacher support to facilitate generalization. Therapy activities address various aspects of social understanding and social interpretation, pragmatics, and language processing skills. For example, pragmatic-based lessons include understanding information requirements, improving turn-taking, managing topics in conversation, and understanding and refining discourse style. SCIP treatment efficacy has been investigated in several case studies and one randomized controlled trial of 88 children between the ages of 5 and 10 years (Adams, Gaile, et al., 2012; Adams, Lockton, et al., 2012). Parents and teachers reported positive changes in pragmatic skills, and some children showed improvement in conversational language samples, although changes in norm-referenced language testing did not occur.

While not as comprehensive as the SCIP manual, a number of case studies and small group studies in the clinical literature provide step-by-step instructions for changing one or more social communication skills. These skills can be targeted with individualized intervention sessions. Examples include using stories and story enactment activities to teach emotion words and to interpret the emotions of others (Brinton & Fujiki, 2019), training perspective-taking skills related to theory of mind (see Peters and Thompson (2018) for a review) and facilitating responsive commenting (Fujiki et al., 2013). Other examples include using social thinking to reflect on how one's actions and words influence what others think and feel about us (Crooke et al., 2016; Winner, 2007), refining conversation skills such as adding relevant information to a conversation, and repairing conversation breakdowns (Brinton et al., 2004); also see Gerber et al. (2012) and Timler (2018a) for reviews.

The second approach is social skills interventions. These are typically group-based programs that focus on the development of skills needed to accomplish a particular social situation or task. For example, most social skills interventions have one or more curriculum units devoted to entering peer groups, making friends, and resolving conflicts. The agenda for social skills groups include a brief discussion of the social situation and the steps needed to engage in the situation, followed by role-play of all group members, and then group and leader feedback. Two examples of commercially available, evidence-based social skills curriculums for pre-schoolers, school-age children, and adolescents include Skillstreaming© (www.skillstreaming.com) and the Program for the Education and Enrichment of Relational Skills or PEERS® (www.semel.ucla.edu/peers), developed for individuals with ASD. The PEERS program also includes a manualized treatment program developed for adults with ASD focused on relationship and vocational skills (Laugeson et al., 2015).

The final approach, peer support interventions, involves the peers of children/adolescents with social communication impairments. These interventions are usually administered in school settings. Teachers and other school personnel arrange the environment so that students with and without social communication impairments work together on collaborative projects of mutual interest. The purpose of these arrangements is to encourage students of differing abilities to interact as students with communication impairments can be ignored or actively victimized by their peers (Conti-Ramsden & Botting, 2004). More intensive peer support interventions, referred to as peer-mediated intervention, include training peers in how to initiate and sustain interactions with students who have social communication impairments (Changa & Locke, 2016). In these interventions, prosocial peers who are willing to participate are selected and serve as good social role models. Peer-mediated interventions have been primarily developed for students with ASD and their peers, but it is likely that some children and adolescents with more severe profiles of SCD would benefit from these intensive interventions, especially if their social reputations are such that they are ignored or victimized by a majority of their peers.

2.8 Summary

Although children and adolescents with pragmatic language needs have been well known to clinicians and researchers for decades, the diagnostic label of social (pragmatic) communication disorder is relatively new. At first glance, it would seem that a potential advantage of this new diagnostic label is that a set of required symptoms are now specified, allowing for easier identification of this disorder. Nevertheless, the introduction of this disorder to the DSM-5 has introduced more questions than answers for clinicians and researchers who serve and study this population of children. For example, how do the social communication profiles of individuals with SCD and ASD differ? Is it by severity only or are there categorical differences that can be reliably discriminated? How does the SCD phenotype and prognosis vary in boys and girls? What is the prognosis for children and adolescents with co-occurring SCD and LD or ADHD? How do cultural practices and expectations influence the specified symptoms of SCD? Are available assessment tools culturally sensitive enough to support accurate diagnosis in culturally and linguistically diverse populations?

In addition to the need for more knowledge about diagnosis and description of the SCD phenotype, development of empirically validated assessment tools and intervention programs are needed. With the notable exception of the SCIP program described in Sect. 2.7, few interventions have been tested with children who have SCD. Some children with SCD have profiles very similar to those in ASD, but perhaps not quite severe enough to warrant a diagnosis of ASD. This may place a child at a disadvantage for receipt of intervention services. In many countries, education and health care policies mandate intensive services for children with ASD (Brukner-Wertman et al., 2016). Such mandates do not exist for children with SCD. Certainly, these children would qualify for school-based services if an educational impact can be demonstrated, but it is unlikely that they would receive the intensity of services that a child with ASD would receive.

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Chapter 3
Autism Spectrum Disorder

Soile Loukusa

3.1 Introduction

Autism spectrum disorder is characterised by persistent deficits in (1) reciprocal social interaction and social communication, and (2) a range of restricted, repetitive, and inflexible patterns of behaviour and interests in the International Classification of Diseases (ICD-11; World Health Organization, 2018) and the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association, 2013). Since social communication difficulties are core diagnostic symptoms of ASD, it is understandable that pragmatic language difficulties are also a feature of persons on the autism spectrum. The onset of ASD typically occurs in early childhood, but features may only become fully manifest later in childhood, when social demands increase. Deficits cause harm in many important areas, such as personal, family, social, educational and occupational functioning. As difficulties in social communication increase, so too does the risk of peer discrimination and difficulties with integrating into society (see also Finke, 2016).

In recent years, there have been changes in how communication and interaction deficits in ASD have been viewed. Current diagnostic criteria in ICD-11 and DSM-5 combine social interaction and communication. Thus, the triad of features of the autism spectrum (i.e. impaired in 1. communication, 2. social interaction, and 3. behavioral flexibility) is no longer used. Instead, it is referred to the dyad of symptoms. This is reasonable since social reciprocity is needed in both verbal and non-verbal communication and thus, especially in real-life situations, they are difficult to separate (see also Vaughan & Hogg, 2014; Baron-Cohen, 2009).

The prevalence of ASD has increased over time which, it is suggested, is mainly due to increased awareness. Nowadays, the population prevalence of ASD is found to be as high as 1.5% in developed countries (Lyall et al., 2017; Christensen et al.

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2016; Baxter et al., 2015). The male: female ratio for ASD prevalence is lower today than in the past and is currently about 2–5:1 (see Lai et al., 2015). The lower gender ratio may be caused by the fact that current diagnostic criteria allow more females to be categorised on the spectrum. It is probable that features of the autism spectrum vary to some extent between genders and females' features are not always as easy to detect as features in males. Accordingly, more research is needed into the female phenotype of the autism spectrum (van Wijngaarden-Cremers et al., 2014).

Concern about language development is often the first issue that parents of children that are later diagnosed with persons on the autism spectrum raise (Herlihy et al., 2015). Even if some persons on the autism spectrum have preserved or superior language abilities, most persons have structural language difficulties (Ellis Weismer & Kover, 2015; Tek et al., 2014; Boucher, 2012). According to the large data set (N = 2568) from the Autism and Developmental Disabilities Monitoring Network, a population-based public health surveillance programme in the US, around 63% of eight-year-old children on the autism spectrum also have a diagnosis of language disorder (Levy et al., 2010). About 30% of persons on the autism spectrum remain minimally verbal, which means that they do not develop phrase-level speech (see Tager-Flusberg & Kasari, 2013). Inevitably, this also affects their pragmatic functioning.

Persons on the autism spectrum also show high frequencies of comorbid developmental, psychiatric, neurological, and medical diagnoses (Levy et al., 2010). Previously, it was thought that most persons on the autism spectrum had intellectual disability, but recent studies have shown that less than half have a co-occurring intellectual disability (e.g. Postorino et al., 2016). In the Developmental Disabilities Monitoring Network Surveillance (2014), the frequency of an intellectual disability (IQ $\leq$ 70) in autism was as low as 31%. Comorbid diagnoses of persons on the autism spectrum also affect pragmatic language skills and affect the intervention of these skills. Thus, when looking at the pragmatic skills of these persons, it is important to keep possible comorbid conditions in mind.

Although the aetiology of ASD is not yet fully explained, remarkable progress has been made in the last decade. Various neurobiological and genetic risk factors exist (Lyall et al., 2017). It is known that ASD is highly heritable and complex genetic components have a role to play in most cases (Bralten et al., 2018; Lyall et al., 2017; Yoo, 2015). However, it has also been recognised that there are many environmental factors that may increase the risk of ASD. To date, there is evidence that parental age, preterm birth, prenatal exposure to air pollution and short inter-pregnancy intervals are potential risk factors for ASD (Lyall et al., 2017). In addition, there is a need for more research to examine whether certain prenatal nutrients, metabolic conditions, and exposure to endocrine-disrupting chemicals have an effect on the risk of ASD.

3.2 Pragmatic and Social Communication in Persons on the Autism Spectrum

3.2.1 Features and Background Factors

Since the diagnosis of ASD requires deficits in social communication, it follows that persons on the autism spectrum have difficulties in pragmatic language, an important component of social communication. According to DSM-5, difficulties in social communication in ASD include:

1. Deficits in social-emotional reciprocity ranging, for example, from abnormal social approach and failure of normal back-and-forth conversation to reduced sharing of interests, emotions or affect and failure to initiate or respond to social interactions.
2. Deficits in non-verbal communicative behaviours used for social interaction ranging, for example, from poorly integrated verbal and non-verbal communication to abnormalities in eye contact, body language or deficits in the understanding and use of gestures, to a total lack of facial expressions and non-verbal communication.
3. Deficits in developing, maintaining, and understanding relationships ranging, for example, from difficulties in adjusting behaviour to suit various social contexts to problems with sharing imaginative play or making friends and the absence of interest in peers.

When looking at these three criteria, it is obvious that pragmatic language deficits play a role in all of them. For example, reciprocity is central to conversation skills, non-verbal communication is important when inferring and expressing intentions, and difficulties in adopting an appropriate listener's and speaker's role in different contexts can cause difficulties in relationships. Thus, when looking at these criteria, it is important to keep in mind that pragmatic skills and other social communication skills (social cognition, social interaction, and language processing) operate together and often it is almost impossible to separate them from each other. This is also seen in methods used in autism spectrum research. For example, one of the most widely used methods to measure the ability to provide context-appropriate explanations for story characters' non-literal statements is Happé's Strange Stories Test (Happé, 1994). It has been developed to measure the skills of advanced theory of mind. However, when looking at the test scenarios and questions, it is obvious that answering contextually challenging questions demands pragmatic inference abilities and an interplay between theory of mind and pragmatic skills.

During the last twenty years, researchers have increased our knowledge of pragmatic skills in persons on the autism spectrum (e.g. Deliens et al., 2018; Dindar et al., 2021 Loukusa et al., 2018; see also Volden, 2017). Most studies have focused on specific skills of the pragmatic language domain (e.g. speech acts or contextual comprehension) and have been conducted in clinical settings. At the same time as interpreting the results of these studies, it is important keep in mind that functions of the pragmatic language domain should not only be based on studies performed in clinical settings, but research should also be done in multi-dimensional, real-life contexts (see also Volden et al., 2009; Adams, 2002). Gibbs and Colston (2012) describe pragmatic functioning as a continuously changing process in which a person adapts to the world in a communication situation. In the light of this definition, it is easy to understand that studies done in clinical settings do not give the whole picture of the phenomenon.

If we look at the pragmatic language domain from a wide-ranging viewpoint, it helps us see that pragmatic communication is not only disturbed in persons on the autism spectrum because of social communication difficulties. In real-life situations, stereotyped, restrictive, and repetitive patterns of behaviour, interests or activities and hyper- or hyporeactivity to sensory inputs also affect pragmatic communication in persons on the autism spectrum (Fig. 3.1). For example, imagine a situation where a child has hyperreactivity to auditory and/or visual stimuli and his classmates start to talk to him in a noisy corridor where there are lots of children walking and talking to each other, and at the same time there is the sound of closing doors, clattering of things, etc. The child who has hyperreactivity to sensory inputs may feel this kind of environment is overwhelming, chaotic, and even scary. This may make communication with classmates impossible. In this way, sensory hyperreactivity affects the child's pragmatic functioning, at least in certain contexts. It may also cause the child to withdraw from communication situations, which results in them not having communication experiences with their peers. This affects their pragmatic communication development.

This is an example of how sensory abnormalities may disrupt pragmatic functioning in persons on the autism spectrum more often than it is thought. This also shows that when looking at the pragmatic functioning of an autistic person, it is

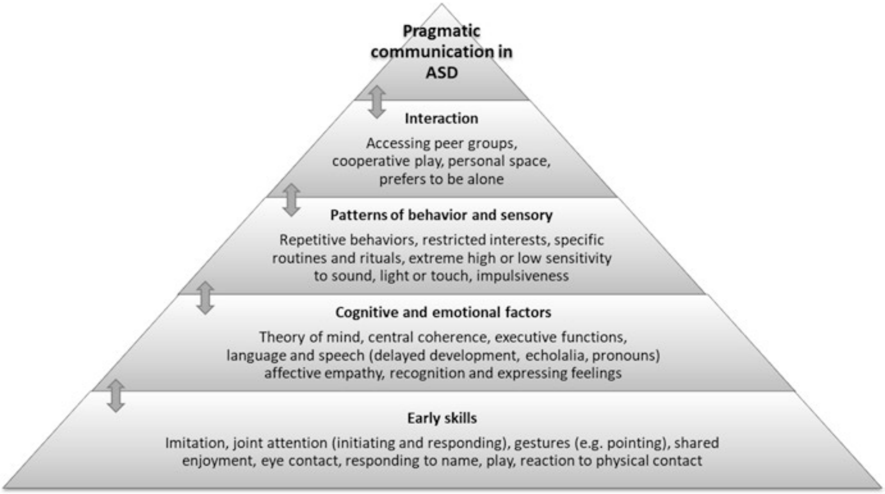

Fig. 3.1 Some factors affecting pragmatic communication in persons on the autism spectrum

important to assess their functioning as a process of adapting to changing communication situations and not only their abilities in separate pragmatic functions. One way to look at pragmatic communication disability is Perkins' (2007) emergentist model of pragmatic ability. It shows how there may be multiple cognitive, linguistic, and sensory sources behind pragmatic impairment and how pragmatics is a product of many interacting variables (see also Fig. 3.1).

It is known that pragmatic language in persons on the autism spectrum is affected by basic language abilities (Whyte & Nelson, 2015; Volden et al., 2009). However, it is good to keep in mind that the relationship is not always straightforward. For example, the study by Volden et al. (2009) showed that although pragmatic language skills in children on the autism spectrum were strongly related to structural language skills, they were not dictated by them. This suggested that pragmatic language scores on the Test of Pragmatic Language (TOPL; Phelps-Terasaki & Phelps-Gunn, 1992) measure additional language skills that are not captured by structural language competence. In earlier studies, pragmatic communication features in persons on the autism spectrum have been interpreted using cognitively-oriented pragmatic theories such as relevance theory (Happé, 1993; Loukusa, Leinonen, Jussila, et al., 2007) and Gricean maxims (Surian et al., 1996). However, the most common social and pragmatic communication difficulties in persons on the autism spectrum have been explained using theory of mind (ToM) (Martin & McDonald, 2004; Happé, 1993), weak central coherence theory (Noens & van Berckelaer-Onnes, 2005; Jolliffe & Baron-Cohen, 1999b, 2000), and executive dysfunction theory (Hill, 2004). Executive dysfunction of persons on the autism spectrum compromises planning and mental flexibility. It affects the ability to use and interpret language in a flexible way according to situation and the ability to direct one's attention to relevant factors (see also Papp, 2006). Working memory is one important component of executive function and works as a tool to integrate information from different sources, which is important in pragmatic language inference. It has been found that autistic persons have weaknesses in working memory, such as in tasks that require cognitive flexibility, planning, and greater working memory load (Kercood et al., 2014).

Empathizing-systemizing theory (Baron-Cohen, 2009, 2010) could also offer a fruitful background for interpreting pragmatic communication difficulties in persons on the autism spectrum, even if to the author's knowledge it has not yet been fully utilised in pragmatic language studies. According to empathizing-systemizing theory, autistic persons perform weakly in tasks requiring cognitive and affective empathy, but their ability to use systemising skills is average or above average (Baron-Cohen, 2009, 2010; Wakabayashi et al., 2007; Goldenfeld et al., 2005). Superior systemising skills do not just show up in understanding systems, but they widely affect the behaviour of autistic persons. According to Baron-Cohen (2009), this discrepancy between weak empathising skills (e.g. the inability to utilise social context in the comprehension of others' emotions) and intact or even superior systemising skills results in a specific processing style that may cause many kinds of strengths (e.g. easily understanding the syntax of different languages, good technical skills) but also weaknesses. Discrepancy (e.g. weak social understanding and strong technical understanding) may also make young autistic adults vulnerable to social manipulation (Al-Attar, 2016).

From a clinical point of view, different pragmatic theories used in studies should not be viewed as competing approaches. Instead, it is better to consider them as connected views and ideas to better understand the complex nature of pragmatic communication in persons on the autism spectrum. Because many kinds of neuropsychological weaknesses (e.g. Elsheikh et al., 2016), psychiatric symptoms (e.g. Mattila et al., 2010), and savant skills (e.g. Howlin et al., 2009) may persist in autistic persons in addition to the core features of the autism spectrum, every individual has a unique collection of strengths, weaknesses, and symptoms which in part affect the individual's pragmatic communication in different contexts. Currently, most pragmatic studies have been carried out with autistic children who have average or above average intelligence without comorbid disorders. It is important to keep this in mind when reading this chapter to understand that probably our current research does not provide a proper picture of the whole spectrum.

3.2.2 Pragmatic Comprehension in Persons on the Autism Spectrum

Pragmatic comprehension difficulties vary considerably in children and adults on the autism spectrum. The fact that the population with ASD is very heterogeneous and mild difficulties in pragmatic comprehension are not always easy to detect in structured test situations has sometimes caused contradictory results between studies. Researchers have achieved different kinds of results in terms of whether some types of difficulties belong to autism spectrum (e.g. Deliens et al., 2018; MacKay & Shaw, 2005) and what the reasons are behind pragmatic difficulties (see e.g. Martin & McDonald, 2004).

However, if we look at abilities across the whole spectrum, it is obvious that pragmatic comprehension difficulties vary from severe (e.g. the child understands only short literal expressions such as "take a book") to mild (e.g. difficulties understanding complex humour). It is also possible that background factors may vary. Mild difficulties are not always easy to detect in structured test situations, even if they may be present and cause harm in complex, real-life communication situations. It is also possible that qualitative analysis may show more differences than quantitative analysis can show. For example, in the study by Norbury and Bishop (2002), typically developing children performed better than children on the autism spectrum and children with language impairment in tasks involving story comprehension that required inferencing and understanding of the literal meaning. Although the scores between the clinical groups were quite similar, qualitative analysis showed that children on the autism spectrum gave most answers that were not relevant to the story context. It is also suggested that linguistically talented children on the autism spectrum are more able to use compensatory strategies in demanding tasks, which help them in their performance (Fisher et al., 2005; Happé, 1994).

Clinically, it is well known that many autistic persons tend to interpret utterances literally (see also Rapin & Dunn, 2003; Kaland et al., 2002). Children on the autism spectrum have shown pragmatic deficits in their ability to infer the implication of an utterance and to make inferences from social scripts, metaphors, and speech acts (Dennis et al., 2001). In Dennis et al.'s study, the differences between the groups increased in relation to the amount of inferencing and intentionality of the tasks. The same effect of intentionality and inferencing load for comprehension was also seen later in studies by Loukusa et al. (Loukusa, Leinonen, Kuusikko, et al., 2007; Loukusa et al., 2018) and Angeleri et al. (2016). Loukusa, Leinonen, and Kuusikko (2007) showed that children on the autism spectrum had difficulties in contextually complex processing, such as detecting implicatures, but not in comprehension of reference assignments.

In a later study (Loukusa et al., 2018), children on the autism spectrum differed from typically developing (TD) children in all question types in the Pragma test (contextual inference with ToM, contextual inference without ToM, relevant use of language, recognition of feelings, and understanding false beliefs). However, the biggest difference between the groups was in the questions that demanded contextual inference with mind-reading, showing that difficulty in understanding increased in relation to the intentionality of the tasks (see also Deliens et al., 2018; Heavey et al., 2000). In many kinds of situations, it is very common to use utterances that demand multi-level processing. In these kinds of utterances, processing load is not determined by only one factor, such as understanding mental states or interpreting verbal or physical context, but there is a need to interpret and connect multi-level information at the same time. This kind of processing demands many cognitive abilities and world and social knowledge, as well as the interplay between them, as the analysis of the following example shows. The item shown below is taken from the Pragma test (Loukusa et al., 2017, 2018).

> *Scenario (presented with paper dolls)*: There has been a race at school. Vera was the slowest runner in the whole class. Vera goes to Tina's house after school. Maddie is also there. Tina and Maddie suggest playing tag. Vera says, "I have to go home", and leaves right away.
> *Question:* Why did Vera say "I have to go home"?
> *Visually- and physically-given context:* Showing the scenario with Vera, Tina and Maddie as paper dolls.
> *Verbally-given context*: Vera was the slowest runner in the race at school. When Tina and Maddie suggest playing tag, Vera wanted to go home.
> *World knowledge*: If you are the slowest runner, you will easily be caught. You will also have difficulties catching other children who are faster than you.
> *Social knowledge:* It is not nice to be "it" all the time. If you have to be "it" for a long time, you start to feel bad about yourself.
> ↓
> *Conclusion:* She wants to go home because she does not want to play tag.
> *Example of a correct answer* (from a six-year-old boy with TD): "She doesn't want to play tag."

Example of an incorrect answer (from a six-year-old autistic boy): “Because she is in a hurry to get home.”
Example of an incorrect answer (from an eight-year-old autistic boy): “It’s her dinner time.”

To answer the question correctly, the child must use the relevant information. Using working memory, the child must connect relevant verbally-given information with his or her world knowledge and take Vera’s mental states and emotions into account by utilising social knowledge. It is probable that in the typically developing boy, the processing of this multi-level information happens automatically and in a parallel way (processing is not sequentially ordered) (see e.g. Wilson & Sperber, 2004). When looking at the incorrect responses of the autistic boys, it is obvious that the younger boy interpreted the scenario literally and had not connected the verbally-given information with his world and social knowledge. It is also possible that he was lacking world or social knowledge about this topic. A literal interpretation may mean that pragmatic inferencing (context utilisation) is missing.

When looking at the answer from the older autistic boy, it is possible to see that he had tried to infer an answer by using his world knowledge (often children must go home for dinner) (see also Loukusa, Leinonen, Jussila, et al., 2007). However, he has not connected verbally-given information with his social knowledge (or he is lacking social knowledge), and the interpretation of Vera’s mental state is missing, which lead him to provide an incorrect answer. Thus, the comparison of these two answers showed that in the younger autistic boy, there was no attempt to use contextual information or world knowledge. The older autistic boy tried to use his knowledge about the issue but failed to answer correctly since he did not interpret and connect all the relevant factors.

These two examples show how children’s incorrect answers may give us a clue about what goes wrong in the interpretation of utterances (see also Loukusa, Leinonen, Jussila, et al., 2007). The interpretation difficulties of children with ASD increase in relation to the amount of inferencing and intentionality of the tasks. Thus, it is not surprising that many studies have shown that autistic persons have difficulties in understanding irony (e.g. Deliens et al., 2018; Wang et al., 2006) and humour (e.g. Emerich et al., 2003). Interpreting irony and humour demands an ability to derive an intended meaning based on world and social knowledge and other information available in the context. Emerich et al. (2003) found that adolescents on the autism spectrum had difficulties understanding cartoons and jokes. They also had difficulties handling surprise and coherence aspects of humour simultaneously.

Kaland et al. (2002, 2006) conducted studies that contained different kinds of questions demanding the ability to infer non-literal meanings and intentions. They found that compared to physical states, inferring mental states was more problematic for children and adolescents on the autism spectrum. Compared to their control peers, they did not just have more incorrect answers, but they also needed more prompt questions and they had longer reaction times. Slower reaction times for answering have also been found in other studies (Saarinen et al., 2012; Nakakachi et al., 2008; Pijnacker et al., 2009; Bowler, 1997). In Saarinen et al.’s (2012) Finnish study, a difference between small groups of children on the autism spectrum (n = 15) and control children (n = 15) was evident, especially with regard to correct answers. This may suggest that in children on the autism spectrum, the processing of pragmatically challenging utterances does not always happen as automatically as it does in typically developing children, but to derive utterance meaning, more cognitive effort is needed. Longer processing of utterance meaning may cause problems in quickly moving between communication situations in real life.

By examining the ability to understand idioms (phrases that express an idea or sentiment that cannot be determined by what the individual words mean), homographs (words that have different meanings but share the same spelling) and scalar implicatures (e.g. all/many/some), it is also possible to increase our knowledge of the ability of children on the autism spectrum to utilise contextual information. Most studies of scalar implicatures have found that persons on the autism spectrum interpret and produce scalar implicatures well when they are explicitly required by the task to do so (Schaeken et al., 2018; Hochstein et al., 2017; Chevallier et al., 2010). However, Schaeken et al.'s measure revealed that when the option 'I agree a bit' was available in addition to "I agree" and "I disagree", the children on the autism spectrum showed a dichotomized attitude toward the speaker's meaning by tending to either fully agree or fully disagree with under-informative statements, whereas children with typical development preferred the middle option.

It has been found that at least some children on the autism spectrum show weaknesses in some kinds of homograph tasks (Hala et al., 2007; López & Leekam, 2003). In the study by López and Leekam (2003), children on the autism spectrum performed as well as controls if the context required common interpretation of a homograph. However, when the context required uncommon interpretation of a homograph, children on the autism spectrum showed weaknesses. This showed that autistic children may also have difficulties using sentence context in a homograph task. Hala et al. (2007) used an alternative approach to study whether autistic children utilise meanings of prime words when disambiguating a target homograph. In their study, children on the autism spectrum and their controls were presented with semantically related and semantically unrelated word pairs. The results showed that autistic children are usually able to draw connections between primes and targets. However, in the second presentation of the homographs, autistic children had the tendency to repeat their first pronunciation although the prime had changed. Researchers concluded that autistic children utilise meanings of related word primes, but that they have difficulties in inhibiting prior responses when a homograph presents later with different primes, which may tell us something about executive dysfunction.

Currently, it is not possible to say how and when pragmatic inference abilities develop in children on the autism spectrum. However, there are some findings that difficulties in utilising contextual information in comprehension are milder in older children on the autism spectrum (Loukusa, Leinonen, Kuusikko, et al., 2007). In addition to the number of incorrect answers, changes may be seen in the type of incorrect answers, since irrelevant answers and topic drifts diminish with increasing development (Loukusa, Leinonen, Jussila, et al., 2007). Even though pragmatic comprehension abilities progress with age in individuals on the autism spectrum, some difficulties usually persist into adulthood in even the most capable persons on the autism spectrum (Lönnqvist et al., 2017; Jolliffe & Baron-Cohen, 1999a, 1999b, 2000; see also Loukusa & Moilanen, 2009; Rapin & Dunn, 2003).

Studies using Happé's Strange Stories Test (Jolliffe & Baron-Cohen, 1999a; Happé, 1994) or part of the test (Heavey et al., 2000) have shown that adults on the autism spectrum do not differ in terms of their performance in physical control stories, but they do differ in mental state stories when they have to justify the story characters' nonliteral speech. This shows that these individuals have problems in providing contextually relevant mental state explanations. Jolliffe and Baron-Cohen (1999a) suggested that difficulties may arise in inferring a speaker's intended meaning from context or in understanding some mental states. Heavey et al. (2000) presented their Awkward Moment Test to adults on the autism spectrum with a view to measuring subtle difficulties in mental understanding. In the test, subjects had to answer mental state questions that required them to infer the film character's understanding of the social situation and the social significance of the character's actions. There were also control questions that were not related to the social content of the film. Like in Happé's Strange Stories Test, it was evident that adults on the autism spectrum had difficulties answering mental state questions and especially explaining the motives and intentions of film characters.

3.2.3 Prosody in Persons on the Autism Spectrum

Prosody is the patterns of stress and intonation in a language and includes both expressive and receptive aspects. From the framework of pragmatics, prosody can influence the meaning of a sentence by indicating a speaker's attitude to what is being said. It can indicate sympathy, irony or humour, for example. Prosody also conveys information about the speaker's emotional state. Thus, in communication prosody has an impact on linguistic, pragmatic, and emotional levels. Persons on the autism spectrum have often been reported as having unusual prosodic features (e.g. Olivati et al., 2017; Olejarczuk & Redford, 2013; Kaland et al., 2013; Diehl & Paul, 2012). However, even if unusual prosody is a feature of the autism spectrum, not all autistic persons have unusual prosody. For example, in the study by Nadig and Shaw (2012), six out of fifteen persons on the autism spectrum had typical prosody.

It has also been shown that unusual prosodic features in persons on the autism spectrum vary from the use of monotonous speech to the use of exaggerated "sing-song" intonation (DePape et al., 2012). In Olivati et al.'s (2017) study of persons on the autism spectrum, speech was louder and lower than in individuals with typical development. Interestingly, in Kaland et al.'s (2013) study, autistic persons and their typically developing controls produced functionally similar contrastive intonation, since both groups took their own and their listener's perspective into account. However, controls used a greater pitch range and were perceived as speaking more dynamically than autistic individuals, suggesting differences in the use of prosodic form. Some preserved prosodic features in the comprehension of prosodic cues have also been found (Wang et al., 2006). Children who showed weaknesses in tasks where it was central to utilise contextual knowledge of the event performed comparably with typically developing peers in tasks where prosodic cues were central. The researchers concluded that autistic children have difficulties in tasks where the interpretation of non-literal language is required and that their difficulties do not lie with prosodic cues.

3.2.4 Discourse and Narration in persons on the autism spectrum

Since discourse is essential for the expression of opinions, feelings and ideas, it is also essential for establishing relationships (Dipper & Pritchard, 2017). Thus, discourse difficulties usually cause severe harm to individuals who have these difficulties. It is generally known that persons on the autism spectrum have difficulties in discourse skills (e.g. Paul et al., 2009; Ziatas et al., 2003; Adams et al., 2002), and failure of normal back-and-forth conversation is even mentioned in the diagnostic criteria for ASD under social communication (see Sect. 3.2.1 in this chapter). Discourse skills are also included in many diagnostic or screening instruments for ASD, such as the Autism Diagnostic Observation Schedule-2 (ADOS-2; Lord et al., 2012), which shows that these are one of the core social communication features of ASD.

According to research performed by Paul et al. (2009) in adolescents on the autism spectrum, atypical conversation behaviours persist primarily in the management of topics and information, reciprocity, intonation, and eye gaze. It has been shown that in structured conversation, children on the autism spectrum predominantly refer to their desires and make less reference to their thoughts and beliefs compared to children with language impairment and typically developing children (Ziatas et al., 2003). Using applied discourse analysis, it has been found that children on the autism spectrum respond to comments and questions. However, the content of the responses is often pragmatically problematic in that the responses of children on the autism spectrum do not always fit well with the social or communicative context (Adams et al., 2002). Children on the autism spectrum gave responses that reflected problems in knowing what the other listener knew about the subject (shared information).

A systematic review of pragmatic difficulties in conversation in ASD found that persons on the autism spectrum often have difficulty staying on topic and providing novel and relevant information (Sng et al., 2018). In addition, during conversation they initiate and respond less often, and they also tend to perseverate more in conversation. However, the review also showed that persons on the autism spectrum offered a similar number of turns to partners, and that there was little difference in the way communication breakdowns were repaired or clarified. Findings on the use of eye gaze during conversation were contradictory.

Research has shown that in narration there are both typical and deviant features in children and adults on the autism spectrum (Mäkinen et al., 2014; Norbury et al., 2013; Barnes & Baron-Cohen, 2012; Rumpf et al., 2012; Colle et al., 2008). When looking at the pragmatic aspect of narration, studies have reported many weaknesses in persons on the autism spectrum, even if the results are not entirely consistent with each other. This may be due to different methodologies, age of the participants, and heterogeneity of the personson the autism spectrum. The following two examples (translated from Finnish) show how young autistic man (average IQ) and a young neurotypical man relate the content of a videoclip from the Finnish family TV series Ruusun Aika. These examples show how much it is possible to collect information about communication using narration in autistic persons. The video shows two women walking together, and one is boasting about how people are always looking at her and how hard it is because she would like to be alone. Then, at the end of the video the women meet a man who is interested in the other woman, and then this woman goes off with the man. At the end of the video the second woman says that her friend wants to be alone. The first woman leaves and stands quietly, looking at the couple in an astonished way.

24-year-old neurotypical man:

> There were two (.) female friends it seemed (.) in som- some kind of school and err (.) they were talking with each other and one of them had some kinda (.) err (.) one of them was just talking and was having a kinda identity crisis I mean she wanted to put herself forward and (.hh) wanted attention and then the other (.) friend just listened quietly and until then err (0.6) they met a man on the street who then just talked to the (.) quiet friend and she was surprised (.) the other girl that (.) this is how it went then that I'm not really so (.) electrifying and that it's not.

24-year-old autistic man:

> So (1.0) they walked err (1.4) err (0.5) towards the lift and then they err (1.6) came out of the lift .hh I think it was somehow err (1.4) mmh a stupid sce- scene because it looks like they had just walked through the (1.0) doo::r (.) the door (0.4) and hadn't (1.0) hadn't been in the lift long (1.2) and then (.) then that (.) man came to get the one wearing the woollen (.) jumper.

When examining these two examples of narratives, it is worth remembering that every individual would produce their own unique narrative after looking at the video clip. However, by comparing these two examples, it is possible to detect some core features of the autism spectrum and find some similarities and differences between narratives.

First, before examining pragmatic aspects, let us take a quick glance at the duration and other aspects of narration. The narration times in these two examples were each approximately 30 s, and both narrations contained disfluencies. However, earlier studies have shown that there might be more disfluencies in persons on the autism spectrum (de Marchena & Eigsti, 2016; Suh et al., 2014). In addition, both narrations contained pauses, but they were longer in the narration by young autistic man. His narration also contained fewer words. Some earlier studies have found that stories told by persons on the autism spectrum contain a reduced number of words (Norbury et al., 2013; Rumpf et al., 2012), but there are also studies showing that persons on the autism spectrum use a similar number of words as controls (Novogrodsky, 2013; Suh et al., 2014). If we look at the syntax of the stories, it is possible to see that simpler syntax is used by the autistic man (see also Norbury et al., 2013; Norbury & Bishop, 2003).

Both young men used reference (e.g. pronominal reference) in their stories. In the study by Mäkinen et al. (2014), children on the autism spectrum displayed similar referential accuracy to their peers. However, many earlier studies have shown that the use of reference may be weak in children on the autism spectrum and they may use more ambiguous pronouns than their controls (Suh et al., 2014; Norbury et al., 2013; Novogrodsky, 2013; Norbury & Bishop, 2003). Additionally, the use of references may be pedantic even if they have used accurate references (Arnold et al., 2009), and they may use noun phrases more often than their controls (Rumpf et al., 2012).

Barnes and Baron-Cohen (2012) detected that narratives by adults on the autism spectrum concentrated more on specific details than the overall gist of the story. This was also the case in the story in the example given above. The autistic man concentrated on the door of the lift and the time spent in the lift, which was not relevant to the story (see also Norbury et al., 2013). This may tell us something about the difficulty that persons on the autism spectrum have in processing relevant information. According to relevance theory, the story of the autistic man ran counter to the presumption of optimal relevance. Concentrating on specific details (in this case, irrelevant, visual details) and not on the gist of the story may also tell us about weak central coherence or executive dysfunction that causes difficulties with focusing on and choosing between relevant contextual factors.

To understand the content of the video clip, many kinds of social cognition skills are required, including the ability to interpret a person's mental states such as emotions, beliefs, and desires. It is generally known that persons on the autism spectrum have weaknesses in social cognition (e.g. Loukusa et al., 2014), and problems in theory of mind are even suggested to be one factor behind the symptoms of the autism spectrum (see Sect. 3.2.1 in this chapter). While narrating, the autistic man produced fewer mental state expressions than the neurotypical man (see also Rumpf et al., 2012). However, even though this example and some other studies have reported a reduced number of mental state expressions, there are also plenty of studies that have not (e.g. Mäkinen et al., 2014; Suh et al., 2014; Norbury et al., 2013). It is also possible that weak inferencing skills affect narration (see Norbury & Bishop, 2002). In this case, it could mean that the autistic man did not understand the story and thus he concentrated on an irrelevant, visual part of the video clip. It is also possible that poor working memory could affect narration in the setting of the example.

These two stories could also be interpreted from the viewpoint of empathizing–systemizing theory. Contrary to the story by the neurotypical man, the story by the autistic man may suggest difficulties both in affective and cognitive empathy. It may also suggest that this autistic man is focusing on analysing the details in the video clip and that he cannot concentrate on the gist of the story at the same time. This may show extreme visual systemising. Thus, the narration by the man with ASD may show a discrepancy between empathising and systemising skills that is characteristic of the unique cognitive processing style of persons on the autism spectrum (see Baron-Cohen, 2009).

In addition to the above analyses, the stories could be interpreted using other methods or frameworks such as story grammar. Many studies have shown, for example, that persons on the autism spectrum use fewer story elements in their narrations (Suh et al., 2014; Rumpf et al., 2012; Smith Gabig, 2008). This was also case in the narration by the autistic man. It is clear from the above analyses that narration is an effective way of collecting a wide amount of information that can be interpreted in different ways. Even if our sample narration showed many common features of the autism spectrum, it did not show all of them. It is also reported that idiosyncratic speech, such as the use of scripted or overly formal language (Suh et al., 2014), is often seen in the autism spectrum. It is also possible that the elicitation method influenced the narration. Losh and Capps (2003) reported the use of irrelevant comments by persons on the autism spectrum only in less structured personal narratives, but not in a picture-based story generation task, and Losh and Gordon (2014) found that the use of off-topic or irrelevant utterances only occurred in retelling tasks but not in story generation tasks.

3.2.5 Neural Background of Pragmatic Communication Features in Persons on the Autism Spectrum

In recent years, significant progress has been made in describing both structural and functional abnormalities associated with ASD (e.g. Pereira et al., 2018; Yamada et al., 2016). However, there is still a need for studies of neural-level processing of pragmatic communication in persons on the autism spectrum in order to better understand the background of pragmatic impairment. The processing of social cognition tasks is closely related to pragmatic communication. Understanding neural activation of social cognition tasks can, therefore, also increase our knowledge of the processing of pragmatic language. Studies have found atypical neural activation or organisation, for example, in facial affect recognition (Ciaramidaro et al., 2018; Mennella et al., 2017) and social and emotional processing during interactions (Oberwelland et al., 2017; Salmi et al., 2013). Studies focusing on inferences and the comprehension of irony have found increased activation in right hemispheric regions, which may suggest that inferencing and irony comprehension are more demanding to persons on the autism spectrum and that more cognitive effort is needed (Wang et al., 2006; Mason et al., 2008).

Using functional magnetic resonance imaging (fMRI), Hubbard et al. (2012) studied the neural processing of co-speech beat gestures in children on the autism spectrum and their control peers. *Beat gestures* are *gestures* that do not carry any specific content but accentuate the topic that is being conveyed by emphasising certain words and phrases. In their study, children with typical development showed increased activity in the right superior temporal gyrus and sulcus when listening to speech with beat gestures, whereas children with ASD did not. Compared to typically developing children, children on the autism spectrum showed greater activity within visual areas when processing co-speech beat gestures, and the severity of their social and communicative impairment was connected with increased activity in the visual region. Researchers suggested that the increased activity observed in children on the autism spectrum in visual regions may indicate a deficit in multisensory integration (auditory and visual speech integration).

Kotila et al. (2021) investigated synchrony of neural network activity in a group of neurotypical young adults and a group of autistic young adults when participants were looking at simple pragmatic non-verbal video clips containing speech acts (e.g. request, statement, and order) from the Assessment Battery of Communication (ABaCo; Sacco et al., 2008). The results showed that when looking at simple communicative-pragmatic actions, correlation of brain activity was greater within the neurotypical adults than within autistic adults in several brain areas (especially in the right dorso-central insula, the left superior frontal gyrus, the left supramarginal gyrus and the posterior insula). This may show that in neurotypical adults, brain activity has synchronised because they automatically assume and focus on similar perspectives during stimulus viewing. Atypical activation in insular regions belonging to the salience network has been linked to ASD (Odriozola et al., 2016). In the behaviour tests there were no differences between groups in these simple communicative-pragmatic items (Kotila et al., 2021). This suggests that autistic persons may use different kinds of processing styles (compensatory strategies) to interpret the speaker's speech acts.

3.3 Assessment of Pragmatic Language Skills in Persons on the Autism Spectrum

The assessment of pragmatic language skills in persons on the autism spectrum should be comprehensive and multidisciplinary in nature. Thus, it should not just consist of structural language assessment and parental reports of pragmatic aspects of language. To obtain a complete picture of a child's or adult's pragmatic communication, clinicians should connect information collected using observation, parental reports, assessment methods developed to detect features of the autism spectrum, tests for neuropsychological skills including theory of mind and affect recognition skills and, of course, tools developed for pragmatic language skills. The purpose of the assessment also affects the measures used. It is a different process to assess skills for diagnostic purposes than for educational ones.

Because of the complex nature of pragmatics, it may be challenging to capture pragmatic difficulties in a structured test situation (Volden et al., 2009; Adams, 2002). An individual may perform well in a structured test situation even if they have significant pragmatic difficulties in real-life situations. In this review, it is mostly focused on methods that are research-based and commercially or generally available (e.g. via websites). However, it is important to be aware that this review does not encompass all possible measures to assess pragmatic language in the autism spectrum. There are also other methods such as the Pragma test (see Loukusa et al., 2018) and the ABaCo (see Angeleri et al., 2016), which are used in research with good results. In future, these measures might also provide important knowledge about pragmatic functioning in the autism spectrum if they were easily available to clinicians and researchers in different languages (at present, there are only Finnish and Italian norms of the Pragma test, and the test is recently translated into English for example). Since knowledge of pragmatic communication in the autism spectrum is increasing rapidly, there is a constant need to develop sensitive, research-based measures that are directed to the study of the most central pragmatic communication difficulties in the autism spectrum.

There are many instruments available for detecting to traits of the autism spectrum for diagnostic purposes (Table 3.1). These tests also include several pragmatic tasks or questions that usually belong to the social interaction or communication part of the measure. From the viewpoint of pragmatic communication, tasks concerning routines, restricted interests, and sensory abnormalities also provide important information since they may affect a person's pragmatic functioning in real life (see Sect. 3.2.1 in this chapter). With many diagnostic instruments, such as the Autism Diagnostic Interview-Revised (ADI-R; Lord et al., 1995), the Gilliam Autism Rating Scale (GARS-3; Gilliam, 2013), the Diagnostic Interview for Social and Communication Disorder (DISCO; Wing et al., 2002) and the Developmental, Dimensional and Diagnostic Interview (3di; Skuse et al., 2004), information about a person's behaviours and social communication is collected from interviews with parents (or caretakers). The Childhood Autism Rating Scale, second edition (CARS-2; Schopler et al., 2010) combines observations of the child with interviews with parents or caretakers. In the Autism Diagnostic Observation Schedule, second edition (ADOS-2; Lord et al., 2012), a trained professional observes a child's behaviour while performing specific tasks. In Randall et al.'s (2018) review of diagnostic tests in preschool children with ASD, only ADI-R, ADOS, and CARS met the inclusion criteria for review. All three tests performed similarly for specificity. However, ADOS was the most sensitive in diagnosing ASD in preschool children.

As mentioned earlier, pragmatic performance may be difficult to assess in clinical settings. As a result, standardised checklists of pragmatics and social communication have often been used, especially for screening purposes. The Children's Communication Checklist-2 (CCC-2; Bishop, 2003) is one of the most widely used checklists in clinical practice and research. CCC-2 comprises ten scales, of which eight assess structural language and pragmatic language, and two assess the social impairments and restricted interests that are more typical of in children on the autism spectrum. CCC-2 gives a score for General Communication Composite (GCC) and a score for the Social Interaction Deviance Composite (SIDC). The SIDC identifies social communication abilities that are disproportionately impaired relative to structural language skills, as can be found in children and adolescents on the autism spectrum. Many studies have confirmed that CCC-2 accurately identifies children with social communication impairments as in the autism spectrum (Loukusa et al., 2018; Volden & Phillips, 2010; Norbury et al., 2004). There are also other standardised rating scales for the assessment of social communication skills in the autism spectrum, such as the Social Responsiveness Scale-2 (SRS-2; Constantino & Gruber, 2012) and the Social Communication Quotient (SCQ; Rutter et al., 2003). Both instruments are concerned to assess features that are typically found in ASD.

There are also tests that assess pragmatic language skills which may be suitable for assessing these skills persons on the autism spectrum. They include the Test of Pragmatic Language-2 (TOPL-2; Phelps-Terasaki & Phelps-Gunn, 2007), the Strong Narrative Assessment Procedure (SNAP; Strong, 1998) and the Expression, Reception and Recall of Narrative Instrument (ERRNI; Bishop, 2004). Young et al. (2005) investigated how TOPL and SNAP detected pragmatic impairments in children on the autism spectrum and found that TOPL scores differentiated children on the autism spectrum from matched controls. However, the researchers observed that variance within the group of children on the autism spectrum was large, resulting in some of the children on the autism spectrum performing comparably with controls. Volden and Phillips (2010) reported that TOPL identified nine of 16 children on the autism spectrum as pragmatically impaired, whereas the CCC-2 identified 13 as impaired.

In the SNAP, children on the autism spectrum performed more poorly than controls in inferential questions, but similarly in tasks assessing syntax, cohesion, story grammar, and completeness of episodes. As a result, SNAP did not clearly differentiate language abilities among children on the autism spectrum from those in typically developing children (Young et al., 2005). Volden et al. (2017) used the ERRNI in their study of 74 children aged 8–9 years on the autism spectrum. They found that among children on the autism spectrum, average performance was poorer in the ERRNI than in a language test. These authors concluded that the ERRNI revealed discourse impairments that might not be identified by tests that focus on individual words and sentences. Overall, the ERRNI provided a useful measure of communicative skill beyond sentence level in school-aged children on the autism spectrum.

Since discourse difficulties are one of the core pragmatic features of ASD, assessment should also include them. Diagnostic instruments such as ADOS-2 give some information about discourse skills but it would be useful to measure them in deeper way to get information about where exactly intervention should take place. Conversational and discourse analysis could provide a valid tool for assessing discourse skills in ASD (Reilly et al., 2016). However, they remain rather time-consuming and complex assessment methods, which may limit their clinical use. There are also some promising measurements, such as the Targeted Observation of Pragmatics in Children's Conversation observation scale (TOPICC; Adams et al., 2011) which can be useful in rating the quality of conversational exchanges (Table 3.1).

Table 3.1 Examples of methods for assessing pragmatic language skills and social communication abilities in persons suspected or diagnosed

Name of measure		Authors and year	Aspects covered	Age	Other information
General ASD screening tools and rating scales	Modified Checklist for Autism in Toddlers (M-CHAT-R/F)	Robins et al. (2014)	Early signs of ASD (e.g. pointing, responding to name, eye contact)	16–30 months	Form and instructions for free download translated in many languages can be found at: https://mchatscreen.com/mchat-rf/translations/
	Early Screening of Autistic Traits Questionnaire (ESAT)	Swinkels et al. (2006)	Early signs of ASD (e.g. joint attention, eye contact and smiling at other people)	14–15 months (0–36 months)	Link to the form for free download can be found at: http://disabilitymeasures.org/ESAT/
	Autism Spectrum Quotient AQ	Baron-Cohen et al. (2001)	ASD features in social interaction, communication and restrictive or repetitive activities and interests	From 16 years onwards	Self-assessment downloadable from: https://www.autismresearchcentre.com/arc_tests
	Autism Spectrum Screening Questionnaire (ASSQ)	Ehlers et al. (1999)	Features that are common in school-aged children on the autism spectrum (e.g. literal understanding and lack of empathy)	7–12 years	Form can be downloaded from: https://gillbergcentre.gu.se/english/research/screening-questionnaires/assq
Rating scales focusing on social communication in ASD	Social Responsiveness Scale-2 (SRS-2)	Constantino and Gruber (2012)	Discriminate autism spectrum disorder from other child psychiatric conditions by identifying presence and extent of autism spectrum social impairment	From 2.5 years onwards	

Name of measure		Authors and year	Aspects covered	Age	Other information
	Children’s Communication Checklist-2 (CCC-2)	Bishop (2003)	Focus areas of communication such as speech, vocabulary, sentence structure and social language skills of children, and screening for general language impairments. Identifying children with pragmatic language difficulties and detecting children who need deeper assessment for autism spectrum	4–16 years	Also available: Communication Checklist - Adult (CC-A), Whitehouse and Bishop (2009)
	Social Communication Quotient (SCQ)	Rutter et al. (2003)	Screen for features of autism spectrum	Mental age 2 years and above	Designed as a questionnaire version of the ADI-R. Caregivers can rate the individual’s “lifetime” features (used to support a diagnosis) or “current” features (used as an evaluation of current difficulties).
Diagnostic tests	Autism Diagnostic Observation Schedule, Second edition (ADOS-2)	Lord et al. (2012)	Accurately assesses and diagnoses ASD across age, developmental level and language skills. Focus on social interaction, communication and restrictive and repetitive interests and activities	From 12 months onwards	Standardised behaviour observation and coding. Includes Toddler Module and Modules 1–4. Cut-off scores for autism and autism spectrum classifications
	Childhood Autism Rating Scale, Second edition (CARS-2)	Schopler et al. (2010)	Identifies children on the autism spectrum and determines symptom severity	From 2 years onwards	Includes rating scales completed by the clinician based on direct observation and an unscored Parent/Caregiver Questionnaire

(continued)

Table 3.1 (continued)

Name of measure		Authors and year	Aspects covered	Age	Other information
	Developmental, Dimensional and Diagnostic Interview (3di)	Skuse et al. (2004)	Computer-based interview for the diagnosis of autism and related disorders in children	From early childhood onwards	Computes the severity of features associated with a diagnosis of autism and establishes co-morbidity across the range of child psychiatric disorders
	Autism Diagnostic Interview-Revised (ADI-R)	Lord et al. (1995)	Diagnostic instrument for assessing ASD. The instrument focuses on behaviour in three areas: 1. reciprocal social interaction, 2. communication and language, and 3. restricted and repetitive, stereotyped interests and behaviours	From 2 years onwards	ADI-R is an interview method which focuses on behaviours that are rare in unaffected individuals. It provides categorical results (not scales or norms).

Name of measure		Authors and year	Aspects covered	Age	Other information
Formal language tests and structured observations containing pragmatic language	Clinical Evaluation of Language Fundamentals®, Fifth Edition Metalinguistics (CELF®-5 Metalinguistics)	Wiig and Secord (2014)	Subtests include ambiguous sentences, listening comprehension: making inferences, oral expression: recreating speech acts, figurative language, and a supplemental memory subtest	From 9 to 21 years	A revision of the Test of Language Competence – Expanded Edition (TLC-Expanded), Wiig and Secord (1989)
	Targeted Observation of Pragmatics in Children's Conversations (TOPICC)	Adams et al. (2011)	Includes categories: reciprocity, taking account of listener knowledge, turn-taking, verbosity, topic management, discourse style, response problems	6–11 years	
	Test of Pragmatic Language, Second edition	Phelps-Terasaki and Phelps-Gunn (2007)	Subcomponents of pragmatics: physical setting, audience, topic, purpose (speech acts), visual-gestural cues, and abstraction	From 6 years to 21 years	
	Expression, Reception and Recall of Narrative Instrument (ERRNI)	Bishop (2004)	Assesses the ability to relate, comprehend, and remember a story	6 years to adult (norms available from 4 years)	
	Communication and Symbolic Behavior Scales (CSBS DP)	Wetherby and Prizant (2001)	Includes the Infant Toddler Checklist, Caregiver Questionnaire and Behavior Sample. Focus on communication and symbolic development, including gestures, facial expressions and play behaviours	Functional communication age between 6 and 24 months (chronological age from 6 months to 6 years)	The Infant Toddler Checklist is available as a free download from the publisher's website (other parts must be purchased): https://brookespublishing.com/product/csbs-dp-itc/
	Strong Narrative Assessment Procedure	Strong (1998)	Narrative generation and comprehension	7–12 years	

3.4 Pragmatic Intervention in Persons on the Autism Spectrum

Wide-ranging and careful assessments build a basis for planning an intervention in ASD. In children on the autism spectrum, early intervention is shown to be effective (Zwaigenbaum et al., 2015; Koegel et al., 2014). When intervention occurs at the point of maximal neural plasticity, it has a long-term impact on the child's development (Bradshaw et al., 2015). Since intervention should start as early as possible, it should be targeted at at-risk toddlers who do not yet have an official ASD diagnosis. Early intervention in children on the autism spectrum usually aims to increase communication and social skills (e.g. joint attention and turn-taking) that are also crucial for pragmatic communication. In addition, early intervention often aims to decrease maladaptive symptoms (e.g. stereotypes and self-injurious behaviour) and support young children's development in a comprehensive way.

Based on the research findings, best practices for providing interventions for children with suspected or diagnosed ASD have been created (e.g. Zwaigenbaum et al., 2015; Myers & Johnson, 2007). According to them, interventions should begin early, be systematically planned and intensive, involve parents and other caregivers, include both developmental and behavioural approaches, promote interaction with peers, and develop children's skills and functionality in the core and associated features of ASD. It should also include a high degree of structure and secure the generalisation of learnt skills to other situations. Finally, it should consider family circumstances, and a child's medical and other comorbid disorders.

Currently, there are many evidence-based interventions available for children on the autism spectrum that include or focus on supporting communication skills (see reviews from Will et al., 2018; Tachibana et al., 2017; Wong et al., 2015). For example, Pivotal Response Training (PRT; Koegel & Koegel, 2006) and the Picture Exchange Communication System (PECS; Bondy & Frost, 1994) are focused, evidence-based intervention practices, whereas Applied Behavior Analysis (ABA; Cooper et al., 2014) and the Early Start Denver Model (ESDM; Rogers & Dawson, 2009) can be considered comprehensive, evidence-based methods.

As intervention in children on the autism spectrum should start early, many existing methods also focus on toddlers. The ESDM is a behavioural therapy for children on the autism spectrum between the ages of 12 and 48 months (Rogers et al., 2019; Rogers & Dawson, 2009). It is based on the methods of ABA. In the ESDM parents and therapists use play to build a positive relationship with the child and through play and joint activities, they boost the child's language, social and cognitive skills. The method is based on the understanding of a normal toddler's learning and development. Parental involvement is a key part of the ESDM programme. In the intervention the therapists explain and model the strategies that they use so that families can practise them at home. As a comprehensive method, the ESDM is focused on the child's development in its entirety. The intervention style and many areas in the practice develop pragmatic skills (e.g. communicative acts) or are important building blocks for pragmatic development (e.g. joint attention). The ESDM has been shown to be an effective method for supporting children on the autism spectrum, especially in the areas of communication and language. However, it is only one of several effective methods. In their study, Rogers et al. (2019) remarked that regardless of the brand name involved, when young children on the autism spectrum receive an adequate level of high-quality, developmentally suitable intervention, their skills will develop (see also Watkins et al., 2017; Wong et al., 2015).

Providing high-quality intervention depends on consistent methods and the content of teaching, which are derived from empirical bases and delivered with acceptable treatment fidelity in terms of implementation across staff members. There is also a requirement that the treatment is altered as needed based on an ongoing evaluation of the child's progress and is delivered at adequate intensity to allow for the child's goals to be accomplished within set timelines. A review by Watkins et al. (2017) reported that it is important that evidence-based intervention strategies for social communication in children on the autism spectrum are also selected based on a variety of factors, including the child's age and the desired outcome. The increasing empirical evidence for various interventions provides both a strong knowledge base and the confidence to support persons on the autism spectrum in achieving intervention goals.

Because the area of pragmatics is understood and defined in different ways, the focus of intervention studies varies considerably. Parsons et al. (2017) undertook a systematic review of pragmatic language interventions for children on the autism spectrum and found 20 different intervention programmes that were reported across 21 studies, of which four were modifications of the Joint Attention, Symbolic Play and Engagement Regulation model (JASPER; see Kasari et al., 2006). JASPER is an evidence-based method targeting social communication (joint attention, imitation, play). It uses naturalistic strategies to develop social communication. Research has shown that JASPER develops children's joint engagement, social communication and emotion regulation and increases parental co-regulation strategies (e.g. Kasari et al., 2012).

In their review, Parsons et al. approached pragmatics from a broad perspective and included methods such as therapeutic horse-riding (Gabriels et al., 2015) and emotion recognition training (Ryan & Charragain, 2010). These methods also support many aspects of pragmatics, even if the focus is more on interaction and/or social cognition than pragmatics. Since pragmatic development interacts strongly with development of social cognition, interaction and language, supporting these skills may develop pragmatic skills also. Because in persons on the autism spectrum there are often weaknesses in all areas of social communication, intervention methods often target these areas in general and do not focus simply on the area of pragmatics. However, since in this chapter we are concentrating on pragmatic communication, it may be worth highlighting one promising intervention programme where pragmatics is an important focus area of intervention.

The Social Communication Intervention Project (SCIP; Adams et al., 2012) is a manualised intervention framework developed for 6- to 11-year-old children who have pragmatic language impairments. It is also suitable for verbal children on the autism spectrum. In the SCIP, intervention consists of three components: 1. social understanding and social interpretation (e.g. understanding social context cues, thoughts, and intentions); 2. pragmatics (e.g. understanding and managing topics in conversation and understanding information requirements); and 3. language processing (e.g. narrative construction and understanding and using non-literal language). Adams et al. (2012) reported that a significant treatment effect was found in children with pragmatic impairment in terms of perceptions of conversational competence, both in parent-reported measures of pragmatic functioning and social communication, and teacher-reported ratings of classroom learning skills.

Since emphasis is placed on interventions with children, it is important to highlight that social communication intervention can also be effective during adolescence and adulthood. It has been shown that methods such as PEERS (Laugeson & Frankel, 2010) and LEGO®-based therapy (Legoff et al., 2014) can support social communication skills, such as discourse abilities and responsiveness. PEERS is a social skills training intervention for social challenges and it has shown to be effective for use with adolescents and young adults on the autism spectrum (Laugeson et al., 2012). LEGO®-based therapy is a social skills programme for children and adolescents with social communication difficulties such as in autism spectrum. Key to this approach is building LEGOs collaboratively and at the same time developing social and communication skills. Therapy utilizes strong systemizing skills of persons on the autism spectrum and is effective and fun for participants (Owens et al., 2008; Legoff & Sherman, 2006).

Although there are many evidence-based interventions available, with increasing knowledge of autism spectrum features there is a constant need to develop new intervention strategies. For example, it is known that persons on the autism spectrum show significant deficits in relation to recognizing and processing human stimuli whereas many of them show a heightened interest in non-social stimuli (see Atherton & Cross, 2018). In their review, Atherton and Cross discussed how persons on the autism spectrum can even show preserved theory of mind when they are dealing with animals, robots, or human cartoons. The review also stated that reduced oxytocin neurohormonal release during human interaction in persons on the autism spectrum may make eye contact too sensitizing, as one of the purposes of oxytocin is to reduce anxiety during social interaction. Thus, in the future it would be interesting to explore more, for example, the use of animals to compensate for reduced oxytocin release when practicing eye gaze, gestures, and other social communication skills with persons on the autism spectrum and to investigate if animals could function as a natural bridge for persons on the autism spectrum to interact and communicate with humans.

3.5 Summary

Knowledge of pragmatic and other features of ASD has increased substantially over the last twenty years. As a result, diagnosis is more reliable and children on the autism spectrum are recognised more effectively. It is known that social communication, including pragmatic skills, belong to the core features of ASD and there are various theories that try to explain pragmatic difficulties. It is also known that children on the autism spectrum may have many kinds of developmental and neuropsychological difficulties, such as memory, attention and linguistic difficulties, which do not match their intelligence level. There is also some knowledge of atypical neural functioning during pragmatic tasks in persons on the autism spectrum. With this increasing knowledge, intervention methods are also being developed and the prognosis in ASD is better than it has been in the past. It is now known that intensive intervention should start as early as possible and the focus should be on social communication (including pragmatics), not forgetting other features of ASD and the child's possible comorbid disorders.

Current research has focused mainly on weaknesses in persons on the autism spectrum. However, in the future it would be good to focus more on the strengths of persons on the autism spectrum as well. Because of their uniquely good systemising skills (Baron-Cohen, 2009), many persons on the autism spectrum could make an important contribution to our society. To help them use this capacity, it is also important to gain more knowledge about the processing mechanisms of persons on the autism spectrum. For example, currently we do not know enough about how possible atypical neural activation affects their learning of pragmatic communication skills. Today's technologically-oriented world offers persons on the autism spectrum a good opportunity to communicate via the internet without the need for direct social interaction. This offers persons on the autism spectrum the opportunity to have contact with other people and to build new relationships. While this is a good thing, we must also be aware that the internet has made it easy to contact and manipulate socially vulnerable people in new ways. Therefore, intervention or support for social and pragmatic inferencing is needed throughout a person's development, and not just during early childhood.

ASD is a complex disorder and, in the future, more research into pragmatic communication is required in a wide-ranging multidisciplinary framework. To date, there are not enough studies that connect multi-level information such as self-assessment, behaviour tests, and neural measures. Currently, pragmatic communication research is mostly focused on persons on the autism spectrum with average or above average intelligence. We need more studies to be undertaken at the other end of the spectrum (severe cases) to get a better understanding of the whole spectrum. If research is centred on the mildest cases, it distorts the clinical picture. Accordingly, despite impressive development in our understanding of the nature of pragmatic communication in the persons on the autism spectrum, there is still a lot to do to better understand individuals' features and their developmental pathways across this complex spectrum. Also the terminology of autism research is changing. In this chapter, when referring individuals diagnosed with ASD, it is used identity-first term "persons on the autism spectrum" or "autistic persons" as peferrad by persons on the autism spectrum (Bottema-Beutel et al., 2021; Bury et al., 2020; Kenny et al., 2016). When talking about diagnosis or diagnostic criteria, official diagnostic term (ICD-11, DSM-5) autism spectrum disorder, ASD, is used.

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Chapter 4
Fragile X syndrome

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4.1 Introduction

Fragile X syndrome (FXS) is the most common known hereditary form of intellectual disability (ID). Additionally, FXS is the most common known single-gene condition associated with autism spectrum disorder (ASD). In this chapter, we review the literature on pragmatic language in FXS. We begin by discussing the general phenotypic characteristics of FXS, including cognition, global language abilities, and social-communicative functioning. We then describe findings from studies of pragmatic language in males and females with FXS, focusing on group comparison studies where individuals with FXS were compared to younger children with typical development (TD) or those with Down syndrome (DS) in efforts to control for mental and/or language age to capture pragmatic differences existing beyond general cognitive and language delays. The impact of ASD status on pragmatic impairments in FXS is examined. We evaluate evidence for the influence of additional related domains on pragmatic language. The chapter concludes with a discussion of critical directions for future research and clinical implications.

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4.2 The Cognitive and Language Phenotype of Fragile X Syndrome

FXS is the most common known hereditary form of ID, affecting about 1/2500–1/5000 persons (Coffee et al., 2009; Hagerman, 2008; Pesso et al., 2000). FXS is caused when there are greater than 200 trinucleotide cytosine-guanine-guanine (CGG) repeats on the fragile X mental retardation 1 gene (*FMR1*) on the X chromosome. This in turn reduces or eliminates production of the Fragile X Mental Retardation Protein (FMRP), which is important for synaptic maturation in early brain development (Weiler et al., 1997). Females with FXS are usually less impaired than males, given that they have a second "protective" X chromosome which contains a normal copy of *FMR1* able to produce FMRP (Hagerman & Hagerman, 2002; Loesch et al., 2002). The majority of males with FXS have moderate or severe ID, whereas females typically have mild intellectual disability or normal intellectual functioning (Hagerman & Hagerman, 2002; Loesch et al., 2003; Reiss & Dant, 2003).

In addition to general cognitive delays, structural language skills (e.g. vocabulary and syntax) are impacted in FXS as well. As such, it is important to consider pragmatic impairments in the context of broader language and cognitive delays, to understand those pragmatic language skills that are impacted in FXS above and beyond developmental delays. Deficits in expressive vocabulary (Martin, Losh, et al., 2013b; Roberts, Hennon, et al., 2007a; Roberts, Martin, et al., 2007b; Sudhalter et al., 1991) and syntax (Estigarribia et al., 2011; Levy et al., 2006; Martin, Losh, et al., 2013b; Price et al., 2008; Roberts, Hennon, et al., 2007a; Roberts, Martin, et al., 2007b) have been reported for boys with FXS. Although much less research has been conducted with girls, one study of school-age girls with FXS found a relative strength in vocabulary compared to non-verbal cognition (Sterling & Abbeduto, 2012). Speech intelligibility, or understandability, is also reduced in boys with FXS (Barnes et al., 2009; Madison et al., 1986; Paul et al., 1987) and can clearly impact overall communicative effectiveness by making it difficult for a communication partner to understand an individual's communicative message.

An important additional factor that can influence pragmatic language profiles in FXS is the presence of ASD symptoms. As noted, FXS is the most common single-gene condition linked with ASD, a condition defined in part by pragmatic language impairments (Hagerman & Hagerman, 2002). About half to three quarters (43–74%) of males meet criteria for ASD (Clifford et al., 2007; Hall et al., 2008; Kaufmann et al., 2004; Philofsky et al., 2004; Rogers et al., 2001). Boys with comorbid FXS and ASD (FXS-ASD) typically display more severe language impairments than males with FXS only (FXS-O) (Bailey et al., 2001; Estigarribia et al., 2011; Roberts, Martin, et al., 2007b). ASD is less common in females with FXS, although available evidence suggests that 13–45% of females may also have ASD (Clifford et al., 2007; Hagerman et al., 1992; Hall et al., 2010; Klusek et al., 2014a; Lee et al., 2016).

In addition to language difficulties and ASD symptoms, the FXS phenotype is characterized by deficits in theory of mind (ToM), or the ability to understand that others may have different knowledge and thoughts than oneself. ToM deficits may also be related to ASD in FXS (Garner et al., 1999; Grant et al., 2007; Lewis et al., 2006; Losh et al., 2012). For example, Losh et al. (2012) reported that boys with FXS-ASD performed more poorly on a comprehensive battery of ToM tasks compared to boys with TD, whereas boys with FXS-O did not differ from TD controls. Males and females with FXS also demonstrate signs of social anxiety, such as gaze aversion and social withdrawal, and hyperarousal (Freund et al., 1993; Hessl et al., 2001; Klusek et al., 2013; Lesniak-Karpiak et al., 2003; Mazzocco et al., 1998; Roberts et al., 2001). For example, Klusek et al. (2013) found that boys with FXS (in a group composed of those with and without comorbid ASD) showed higher rates of hyperarousal than boys with TD. How these phenotypic features may underlie pragmatic deficits in FXS is discussed in Sect. 4.6.

4.3 Pragmatic Language in Males with Fragile X Syndrome

Because males with FXS are typically more severely affected than females, the vast majority of studies of pragmatic language in FXS have focused on males. In this section, we focus on studies of boys with FXS-O (without comorbid ASD) or studies where ASD status was not specified. The impact of ASD status on pragmatics in FXS is discussed in Sect. 4.5. Some older investigations reported pragmatic deficits in males but did not include comparison samples. Two studies reported non-contingent language (i.e. language that is off-topic or tangential) and perseveration (i.e. inappropriate self-repetition of words, sentences, and topics) in males with FXS (Hanson et al., 1986; Madison et al., 1986), based on small samples of 5–10 males where ASD status of participants was not reported. Similarly, Sudhalter et al. (1991), in a larger sample of 19 males without ASD, observed perseverative language.

More recent studies have included comparison groups of younger children with TD and children with Down syndrome (DS), to determine whether pragmatic features in males with FXS may be explained by developmental level or ID in general. Using a standardized measure of pragmatic ability and controlling for structural language skills in addition to mental age in order to isolate pragmatic ability in particular, Martin, Losh, et al. (2013b) found that boys with FXS-O scored lower than TD boys overall and also developed pragmatic skills more slowly over time but did not differ from boys with DS. Similarly, using a comprehensive rating scale of pragmatic ability, Klusek et al. (2014b) reported that boys with FXS-O and DS performed similarly and showed more impairment overall than boys with TD.

Investigations have also focused on particular pragmatic features, including noncontingent language, perseveration, strategies for managing communication breakdowns, narrative, and prosody. These findings are described below.

4.3.1 Non-contingent Language

Males with FXS have been found to produce more non-contingent language than boys with TD and DS (Klusek et al., 2014b; Sudhalter & Belser, 2001; Wolf-Schein et al., 1987), although two other studies (Martin et al., 2018; Roberts, Martin, et al., 2007b) reported no such differences. Note, however, that Sudhalter and Belser (2001) and Wolf-Schein et al. (1987) did not report the ASD status of participants, and the finding of Klusek et al. (2014b) is based on one item (inappropriate topic shifting) from a rating scale. On the other hand, the two studies reporting no differences (Martin et al., 2018; Roberts, Martin, et al., 2007b) relied on turn-by-turn coding of transcribed language samples. Thus, while findings are mixed, it is possible that non-contingent language is not a core feature of the pragmatic phenotype of FXS (but see Sect. 4.5 for the impact of ASD status on non-contingent language).

4.3.2 Perseveration

Similarly, boys with FXS have been found to produce more perseverative language than boys with TD and DS (Levy et al., 2006; Roberts, Martin, et al., 2007b; Sudhalter et al., 1990; Wolf-Schein et al., 1987), although some other studies did not find these differences (Martin et al., 2012, 2018). This discrepancy is harder to explain. Again, Wolf-Schein et al. (1987) did not report the ASD status of participants, and the “deviant repetitive language” variable used by Sudhalter et al. (1990) included perseveration but also other features such as echolalia. Additional studies of perseveration in males with FXS-O are needed to draw firm conclusions regarding whether or not it is a core feature of the pragmatic profile in this group.

4.3.3 Strategies for Contending with Communication Breakdown

Individuals with speech and language difficulties are likely to experience communication breakdowns (i.e. misunderstandings between communication partners) during interactions. Barstein et al. (2018) examined communication repair skills, or the ability to respond to requests for clarification, and found that boys with FXS-O did not differ from TD controls. A related skill that is necessary for managing communication breakdowns is non-comprehension signaling, or the ability to *initiate* a repair in the face of confusing messages. Abbeduto et al. (2008) found that young males with FXS-O (along with five females) signaled non-comprehension of confusing messages less often than TD controls. However, Martin et al. (2017) did not find differences between boys with FXS-O and TD for non-comprehension signaling. The discrepancy in findings between these two studies may be explained by differences in exclusionary criteria based on ASD status. Whereas Martin et al. (2017) excluded from the FXS-O group all those who met criteria for ASD, Abbeduto et al. (2008) excluded only those who met the stricter criteria for "autistic disorder" and thus likely included some in the FXS-O group who would meet broader ASD criteria.

4.3.4 Narrative

Studies of narrative, or story-telling, skills in FXS-O have yielded some mixed results, but overall suggest that this aspect of pragmatics represents a relative strength. Keller-Bell et al. (2007) reported that males with FXS-O (along with females) performed similarly to TD controls, with both groups employing less evaluation (e.g. mental state verbs and character dialogue) than young individuals with DS. Similarly, Hogan-Brown et al. (2013) found no group differences in thematic maintenance and other macrostructural narrative skills between FXS-O and TD groups, although in this study boys with DS also did not differ from any other group. In another study, young males with FXS-O again performed similarly to young individuals with DS, with both of these groups telling stories with *more* sophisticated introductions than TD controls (Finestack et al., 2012). Only one study has found a disadvantage for boys with FXS-O regarding narrative ability. Estigarribia et al. (2011) found that boys with FXS-O referenced goal-driven activities of the protagonist less often than boys with TD. A possible explanation for the discrepancy between this study and the other studies of narrative in FXS is that Estigarribia et al. (2011) administered the Bus Story Language Test (Cowley & Glasgow, 1994). This story includes an anthropomorphized bus as the main character, which may have made it more difficult to take the perspective of the protagonist.

The following example is used to illustrate aspects of the narrative profile documented in FXS. This narrative was produced by a 12.6-year-old boy with FXS-O (non-verbal mental age of 5.6 years, non-verbal IQ of 48), who was asked to narrate the children's book, *A Bed Full of Cats* (Keller, 2003). This book was modified for use as a wordless picture book. This story depicts the tale of a young boy whose pet cat goes missing during the night. The boy and his family search for the cat, and the story concludes with the cat returning with her litter of kittens.

> One day David, he was David was David was he, was in his room. He had a cat named, uh um, the cat's name was, um uhh, what's the cat's name? Why don't you ask her what's the cat's name? Ask her. Come on. [Examiner: you have to think of it.] How about, uh, Zoe? Zoe and David went in, they were in, uh, Zoe's room. Zoe and David went to bed. David's up. He looks surprised. They look under his bed. But he played rough. Then he lied in his room. He started crying. Family. David wanted to look out the window. Then he looked in the trash. And then looked at the tree. But there was a spider. He started crying with his cat Zoe. One day he fell asleep. When he woke up he looked surprised. He turned his light on. And he, she got pregnant. With four kittens. He seems so happy said Zoe. So David loved Zoe's cats. The end.

Similar to findings reported by Estigarribia et al. (2011), this child's narrative contains several instances of labeling as opposed to inferring goal-driven activities of characters (e.g. "family", as opposed to inferring that the boy's family is assisting with the search for the boy's cat), and includes instances of perseveration (e.g. "he was David, David was he…"). However, the story contains a clear introduction, complicating action (e.g. surprise that his cat was pregnant), and resolution (i.e. the boy loving the kittens), as well as some descriptions of thoughts and emotions that help to drive the story.

4.3.5 Prosody

Prosody refers to the "rhythm" of speech, including variations in rate and intonation. Although prosody is not considered a purely pragmatic skill, it can serve critical pragmatic functions. For instance, rising or falling intonation can indicate a question or comment. Intonation contours and rhythmic qualities of speech are also important tools for conveying emotions and attitudes. When these skills are impaired, they may seriously undercut pragmatic competence. Atypical prosody may also impact social communication by creating an impression of "oddness" to peers, as has been demonstrated in studies of idiopathic ASD (Mesibov, 1992; Van Bourgondien & Woods, 1992).

The perception of a fast speaking rate has been reported for boys with FXS (Borghgraef et al., 1987; Hanson et al., 1986). Note that ASD was not an exclusionary criterion in Borghgraef et al. (1987) and ASD status was not reported in the study by Hanson et al. (1986). However, Zajac et al. (2006), using objective acoustic analyses, did not find differences for speaking rate between boys with FXS (in a group composed of those with and without ASD) and boys with TD of similar chronological age, suggesting to the authors that other aspects of prosody (e.g. pausing, intonation) may underlie perceptions of fast rate in FXS. In fact, boys with FXS-O showed more atypical intonation than boys with DS and TD in the study by Klusek et al. (2014b) discussed at the beginning of this section.

4.3.6 Summary of Pragmatic Features in Boys with FXS

Studies of pragmatic skills in boys with FXS-O reveal a heterogeneous portrait. In large part, this is likely due to inconsistent characterization of ASD status across studies. Nonetheless, they indicate that pragmatic language deficits exist beyond general language or cognitive delays that characterize FXS, differentiating boys with FXS from younger, typically developing males of similar developmental level and males with DS. Although inconsistent across studies, reported deficits include non-contingent language, perseveration, and non-comprehension signaling. However, responding to requests for clarification and some aspects of narrative may represent relative strengths in boys with FXS-O. Studies of prosody are too few in number to draw clear conclusions. Particular attention is needed to understand others' perceptions of a fast speaking rate and to better understand how prosodic skills may impact social communication in boys with FXS.

4.4 Pragmatic Language in Females with Fragile X Syndrome

Compared with the literature on males with FXS, considerably less empirical attention has focused on females with FXS, which is true of research in neurodevelopmental disabilities more generally (Messinger et al., 2015; Rinehart et al., 2011; Thompson et al., 2003). Because females with FXS tend to have less cognitive impairment than males (see Sect. 4.2), studies of pragmatic impairment in this group can help to isolate core pragmatic features impacted in FXS that are not as conflated by general cognitive or structural language deficits. This section generally follows the structure of Sect. 4.3 on pragmatic features in males with FXS. We describe distinct pragmatic features documented in studies of females with FXS-O (without ASD) or studies where ASD status was not reported. However, given the relatively sparse literature on girls, we describe findings on different pragmatic features together, rather than in subsections. We discuss the impact of ASD status on pragmatics in females with FXS in Sect. 4.5 below.

One early case study described selective mutism (i.e. the inability to use language in certain social contexts) along with shyness and social anxiety in a 12-year-old girl with FXS (ASD status not described) (Hagerman et al., 1999). Similarly, two group studies found that females with FXS (ASD status also not described) were less likely to initiate communication than controls (Lesniak-Karpiak et al., 2003; Mazzocco et al., 2006), although another study (Martin et al., 2018) found no differences between girls with FXS-O (or FXS-ASD) and controls in initiations. This discrepancy may be explained in part by differences in sampling context, as the two group studies reporting differences used a role-play scenario where an unfamiliar adult acted as a stranger, was fairly non-responsive, and showed little affect. However, the Martin et al. (2018) study examined conversations with a trained examiner who was responsive to communicative bids. Together, results suggest that girls with FXS may initiate less depending on context and the communication style of their conversational partner.

Although few studies have been conducted overall, girls with FXS-O do not appear to differ from younger girls with TD of similar mental age in non-contingent language, perseveration, communication repair, or non-comprehension signaling (Barstein et al., 2018; Martin et al., 2017, 2018). Only a single study has examined narrative ability in females with FXS specifically (Simon et al., 2001). In this study, adult females with FXS (ASD status not described) exhibited greater difficulty than TD controls and women with the *FMR1* premutation (i.e. those with a CGG repeat length between 55 and 200) in selecting appropriate conclusions to jokes compared to stories. Understanding of humor, and the discursive structure of jokes, may place greater demands on metalinguistic and metacognitive skills than the simple narratives that were examined in this study. Indeed, whereas the simple narrative condition only required subjects to select a correct story ending in a logical sequence of events, the joke condition required participants to infer a humorous, or unexpected conclusion, drawing on the more complex ability to understand anticipated and alternative outcomes, as well as cultural expectations and conventions of humor. Further work, using more complex narrative stimuli, is therefore necessary to understand how narrative ability may be impacted in females with FXS.

A few studies have examined sex differences in FXS-O. No differences between boys and girls with FXS-O have been detected for non-contingent language, perseveration, or repair skills (Barstein et al., 2018; Martin et al., 2017, 2018), although Murphy and Abbeduto (2007) reported that males with FXS more frequently repeated rote phrases than females. In addition, a marginally significant difference (with a medium effect size) reported by Martin et al. (2018) suggests that males with FXS-O may initiate more than females. Sex difference findings for non-comprehension signaling are more mixed. Thurman et al. (2017) found better non-comprehension signaling in females with FXS than males, whereas another study found no sex differences between males and females with FXS (Martin et al., 2017). This inconsistency may be due to ASD status, as Thurman et al. (2017) did not use ASD as an exclusionary criterion for their FXS group as Martin et al. (2017) did. ASD severity was higher for boys than girls in the Thurman et al. (2017) study, although this was not a significant predictor of non-comprehension signaling in either group.

In sum, although relatively few studies have examined pragmatic language in females with FXS, the available evidence suggests similar or stronger pragmatic skills in girls versus boys. One possible exception is initiations, where girls may show a particular weakness which may be related to social context.

4.5 The Impact of Autism Spectrum Disorder on Pragmatic Language in Fragile X Syndrome

Given that pragmatic deficits are a hallmark of ASD and are included as a diagnostic criterion for ASD in the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association, 2013), it is unsurprising that the presence of ASD in FXS appears to result in distinct pragmatic language profiles. In this section, we discuss the literature on pragmatic language in males and females with FXS-ASD. We examine how children with FXS-O compare to children with FXS-ASD and, in the case of males, how pragmatic profiles in FXS-ASD compare to that of boys with idiopathic ASD (ASD only, or ASD-O).

In the same longitudinal study referenced in Sect. 4.3, Martin, Losh, et al. (2013b) found that boys with FXS-ASD performed more poorly overall on a standardized test of pragmatics than boys with FXS-O. Using the same standardized measure, Losh et al. (2012) reported that FXS-ASD and ASD-O male groups performed similarly, displaying more deficits than those with FXS-O and TD. In the study by Klusek et al. (2014b), also referenced in Sect. 4.3, young males with FXS-ASD and ASD-O demonstrated comparable deficits and the same types of pragmatic violations on a rating scale based on semi-structured conversation, with both groups showing more impairment overall than boys with FXS-O, DS, and TD.

Of those studies examining specific pragmatic features in FXS-ASD, a few studies have reported that boys with FXS-ASD produced more non-contingent language (Martin et al., 2018; Roberts, Martin, et al., 2007b) and perseveration (Martin et al., 2012, 2018) than boys with FXS-O. Martin et al. (2018) additionally included a group of boys with ASD-O and found them to show highly similar non-contingent and perseverative language impairments as the FXS-ASD group. To illustrate, below is an example of perseveration produced by an 11.4-year-old boy with FXS-ASD during a semi-structured conversation with an examiner. This boy has a non-verbal mental age of 5.6 years and a non-verbal IQ of 50:

Examiner: It's like a what?
Child: Just like a coo coo.
Examiner: A coo coo? It doesn't look like a coo coo bird to me.
Child: It's a coo coo.
Examiner: I see.
Child: It's a coo coo.
…
Examiner: Is that heavy?
Child: It's a coo coo bird, see?
Examiner: mhm.
Child: It's a coo coo bird. It's a coo coo bird.
…
Child: See coo coo?
Examiner: I know, it's cool.
Child: Two coo coo birds.
...
Examiner: Time to put these things away.
Child: It's a coo coo bird.

In addition to the striking overlap in non-contingent language and perseveration observed in FXS-ASD and idiopathic ASD, Martin et al. (2018) also reported some important pragmatic differences between these groups. Young males with ASD-O initiated turns less often than those with FXS-ASD. Similarly, whereas boys with ASD-O were more non-responsive to questions than those with FXS-O and DS, young males with FXS-ASD did not differ from the FXS-O and DS groups on this variable. Together, these differences suggested to the authors that boys with FXS-ASD may be more socially motivated to interact with a communication partner than boys with ASD-O.

With regard to communication breakdowns, Barstein et al. (2018) reported that boys with FXS-ASD and ASD-O both showed difficulty responding to requests for clarification but in different ways. Specifically, boys with FXS-ASD responded more inappropriately than boys with ASD-O and DS, consistent with research on non-contingent language in boys with FXS-ASD (Martin et al., 2018; Roberts, Martin, et al., 2007b). However, males with ASD-O responded less to clarification requests than males with FXS-ASD and FXS-O, also consistent with findings of non-responsiveness and reduced initiations in ASD-O (Capps et al., 1998; Jackson et al., 2003; Loveland et al., 1988; Martin et al., 2018). Another study found that boys with FXS-ASD were less likely to signal non-comprehension compared to boys with ASD-O, FXS-O, and TD (Martin et al., 2017), suggesting another area where the pragmatic profile of FXS-ASD and ASD-O may diverge.

In the study of narrative skills by Estigarribia et al. (2011) reported in Sect. 4.3, boys with FXS-ASD (like boys with FXS-O) remembered fewer goal-directed behaviors than TD controls. In addition, the FXS-ASD group scored more poorly in story grammar overall than controls (whereas the FXS-O group did not), providing further evidence that ASD status in FXS negatively affects pragmatic ability. However, in the study by Hogan-Brown et al. (2013), also described in Sect. 4.3, no group differences in narrative skills were detected for boys with FXS-ASD, FXS-O, ASD-O, DS, and TD. More research on the impact of ASD status on narrative ability is needed.

Another area where limited research on the impact of ASD symptoms on FXS exists is prosody. As discussed in Sect. 4.3, the perception of a fast speaking rate has been reported for boys with FXS (Borghgraef et al., 1987; Hanson et al., 1986). Zajac et al. (2009) reported that boys with FXS-ASD (but not FXS-O) were perceived to speak more quickly than boys with TD, even when overall actual articulation rate was purposefully equated across groups. Articulation rate with the final word of the sentence excluded, along with sentence-final drop in fundamental frequency (pitch), appeared to explain perceptions of rate. In the study by Klusek et al. (2014b) discussed previously, ratings on an item tapping atypical intonation (i.e. attenuated or exaggerated variation in pitch) were comparable for FXS-ASD, FXS-O, and ASD-O male groups, which differentiated all three groups from boys with DS and TD.

Even less research has been conducted with girls with FXS-ASD. Similar to boys, Martin et al. (2018) found that girls with FXS-ASD produced more non-contingent language than girls with FXS-O, DS, and TD, and more perseveration than the DS group. In the same study, however, girls with FXS-ASD responded less often to questions compared to female FXS-O and DS groups, and also less often than boys with FXS-ASD. This suggests an important area of divergence in male and female FXS-ASD groups and that intervention efforts may need to be different depending on sex in this population. Girls with FXS-ASD also exhibited less inappropriate responses, but more gestures, than boys with FXS-ASD on a communication repair task (Barstein et al., 2018). Although Martin et al. (2017) found no sex differences in FXS-ASD for non-comprehension signaling, girls with FXS-ASD in this study (like boys) performed more poorly than TD controls.

In summary, boys with FXS-ASD show more pragmatic impairment than boys with FXS-O, and in many cases show similar types of pragmatic language impairments as are observed in ASD-O. These findings have important implications for understanding the roots of variable phenotypic expression in FXS, as well as clinical implications for the subgroup of individuals with comorbid FXS and ASD. It is equally important to recognize, however, that there are differences in the pragmatic profiles of boys with FXS-ASD and ASD-O, which should also inform tailored interventions. More research needs to be conducted with girls with FXS-ASD, including studies that compare them to girls with ASD-O, to more fully understand the overlap of FXS-associated and idiopathic ASD.

4.6 Related Abilities Associated with Pragmatic Difficulties

A critical question regarding pragmatic deficits in FXS is what underlying abilities may account for such difficulties. This is especially important given that comparisons with TD and DS groups of similar mental age, along with studies that have also controlled for structural language differences, suggest that general cognitive and language deficits alone cannot explain pragmatic language difficulties in FXS.

In Sect. 4.2, we reviewed some key characteristics of the phenotype of FXS that may be related to pragmatic language. Very few studies have directly examined the links between features such as theory of mind (ToM) and hyperarousal, and pragmatics. Examining a single false-belief task, Abbeduto et al. (2008) did not find a relationship between ToM and non-comprehension signaling in FXS-O (or DS). However, Losh et al. (2012), using a composite ToM score that comprised false belief as well as other tasks tapping desires and intentionality, reported that impaired ToM skills were related to poorer performance on a standardized assessment of pragmatics for all groups studied (i.e. boys with FXS-ASD, FXS-O, ASD-O, DS, and TD). Utilizing the same composite score and overlapping samples, Barstein et al. (2018) found few associations between ToM and repair skills for children with FXS. In young males and females with FXS-O, better ToM was related to fewer inappropriate responses. Of course, boys and girls with FXS-O did not show a weakness in repair skills in this study. But boys with FXS-ASD did show difficulties with repair, and this was not associated with ToM. Future studies should examine the relationships between particular ToM skills *and* particular pragmatic skills to more fully understand this relationship.

Hyperarousal and anxiety are commonly observed in FXS and may also importantly contribute to the pragmatic deficits in FXS (e.g. Belser & Sudhalter, 1995; Cohen, 1995). Elevated arousal can be a biological sign of stress, and hyperarousal has been hypothesized to manifest in anxious behavior (e.g. social withdrawal). For instance, in a preliminary study of two males with FXS, heightened arousal was related to increased perseveration and non-contingent language (Belser & Sudhalter, 1995). However, Klusek et al. (2013), using larger samples, found that arousal was not predictive of pragmatic language in boys with FXS (with and without ASD), as measured by a standardized assessment and comprehensive rating scale, despite the fact that this relationship was observed in ASD-O (for the rating scale). However, arousal dysregulation was marginally associated with weaker pragmatic language for boys with FXS in this study. More research is needed into the role of hyperarousal in pragmatic difficulties in FXS, including the examination of particular pragmatic features like perseveration and non-contingent language with larger samples and also extending this line of investigation to females.

Finally, problems with executive function in FXS have been reported (Garner et al., 1999; Hooper et al., 2008; Munir et al., 2000; Wilding et al., 2002). Although underlying executive dysfunction has been *hypothesized* to cause pragmatic impairment in FXS (Abbeduto & Hagerman, 1997; Martin et al., 2012; Murphy & Abbeduto, 2007), direct investigation of this relationship is lacking.

4.7 Research Directions

The literature examining pragmatic skills in FXS has produced important findings, highlighting non-contingent language, perseveration, and non-comprehension signaling as key pragmatic skills impacted, and illustrating the importance of considering ASD when evaluating pragmatic skills of individuals with FXS. However, as is clear from this review of the existing literature, several important areas for continued investigation remain.

First, more research needs to be conducted with girls with FXS with and without ASD. This should compare them both to boys with FXS and to girls with idiopathic ASD, to determine whether a unique profile exists that would suggest tailored approaches to assessment and intervention as well as enhanced insights into the overlap of FXS and ASD. Given previously described sex differences in the pragmatic profiles of boys and girls (Martin et al., 2018), as well as known differences in the cognitive and behavioral phenotype of males and females with FXS, it is expected that important differences likely exist across pragmatic domains, and thus intervention aimed at treating these impairments in males may not directly translate to similar deficits in females.

Second, we have noted previously in this chapter how inconsistent characterization (or no characterization) of ASD status could help to explain some discrepancies between different study findings. Most studies reviewed here, when they *did* include subgroups of children with FXS-O and FXS-ASD, based these subgroupings on results of the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2001). Although the ADOS is certainly a gold-standard ASD diagnostic instrument, future studies should carefully define subgroups using additional information from other gold-standard assessments of ASD. These include the Autism Diagnostic Interview-Revised (an in-depth parent interview measure) (Lord et al., 1994) and the ASD criteria set forth in DSM-5 (American Psychiatric Association, 2013). Consistent characterization of ASD status across future studies will make findings more comparable.

Third, further research aimed at delineating the role of related abilities, such as ToM, executive function, and anxiety are critical to a better understanding of the impact of cognitive and behavioral skills on pragmatic language in FXS. The role of these related abilities may also differ based on sex and ASD status. Together, such work is needed to inform the roots and causes of pragmatic impairment in FXS, as well as to reveal effective targets for intervention. Finally, the vast majority of existing studies on conversational abilities in FXS involved the individual with FXS interacting with a trained and unfamiliar examiner. Future research should examine pragmatic performance in more ecologically valid contexts, such as interactions with parents and peers, that are more representative of daily interactions.

4.8 Clinical Implications

In general, language interventions for children with ID should focus on improved communication for social, academic, and vocational needs (American Speech-Language-Hearing Association, 2005). Regarding assessment in FXS in particular, Klusek et al. (2014b) found that evaluation during a more naturalistic context was more sensitive for detecting group differences in pragmatic language than a standardized assessment. Therefore, assessment should utilize a variety of methods, including examination of pragmatic performance during conversation – in differing contexts (e.g. school, home) and with different communication partners (e.g. family members, educators) – in addition to standardized testing.

Certainly, pragmatic language should be assessed in all children with FXS. Given considerable evidence that ASD status has a substantial impact on pragmatic language in FXS, whether a child with FXS has comorbid ASD should also be considered during assessment. In a survey study of males and females with FXS living in the United States, Martin, Ausderau, et al. (2013a) reported that speech-language therapy was the most common therapy type, with both males and females with FXS-ASD more likely to receive speech-language intervention than those with FXS-O. For clinicians familiar with ASD-O but not FXS, it is also important to note that pragmatic profiles in ASD-O and FXS-ASD may be similar but not identical. For example, although boys with ASD-O and FXS-ASD both exhibited non-contingent language and perseveration in the study by Martin et al. (2018), only boys with ASD-O demonstrated reduced initiations and responsiveness to questions. This suggests potentially more social motivation in boys with FXS-ASD, a potentially positive prognostic factor for intervention success. It also suggests that targets of intervention may need to differ to some extent. Of course, in the same study, girls with FXS-ASD were less responsive than boys, suggesting that clinicians should also be aware of potential sex differences in FXS.

Roberts, Martin, et al. (2007b) and Roberts et al. (2008) provide a number of suggestions for children with FXS who produce non-contingent language or perseveration, including using familiar routines, materials and topics that the child is interested in, and simple redirection away from a perseverative topic. If, as suggested in Sect. 4.6, anxiety does indeed underlie the expression of these behaviors, it may also be the case that better anxiety management could help reduce non-contingent and perseverative language. That said, Klusek et al. (2013) interpreted their finding that social interaction did not atypically influence arousal levels in boys with FXS to suggest that clinicians should not be overly cautious in socially interacting with a child with FXS. For management of communication breakdowns, Dollaghan et al. (1990) have described an intervention program where children are taught to identify and respond to confusing messages. This treatment proved to be successful in increasing requests for clarification in a small sample of children with language impairment without FXS (Dollaghan & Kaston, 1986). In the area of prosody, Zajac et al. (2009) argued that prosodic intervention focusing on rate and intonation might lessen the social stigma of peculiar speech patterns.

Unfortunately, intervention studies targeting pragmatic language in FXS are lacking. Given known overlap in the pragmatic profiles of individuals with FXS-ASD and ASD-O, it may be that evidence-based intervention techniques used in ASD-O can be applied among children with FXS. Although further research would be needed to determine efficacy in this population, especially given the complex evidence discussed previously of both overlap and difference in pragmatic profiles in FXS-ASD and ASD-O, these techniques could include approaches such as parent-child interventions (e.g. Beaumont & Sofronoff, 2008; Casenhiser et al., 2013; Kasari et al., 2014; Warren et al., 2011) or the use of peer role models or mentors (e.g. Llaneza et al., 2010; Orsmond et al., 2004). Further determination of the underlying mechanisms of pragmatic difficulty in FXS in particular will help to guide intervention, as intervention focused on domains like ToM and hyperarousal/anxiety may have reverberating effects on pragmatic language. Of course, well-designed intervention studies focused specifically on individuals with FXS will be critical.

4.9 Summary

Pragmatics is a highly complex domain that is impacted in individuals with FXS. Most research has focused on boys with FXS, and found that pragmatic deficits may be greater in, or in some cases specific to, boys with FXS-ASD versus boys with FXS-O. Patterns of overlap and divergence in pragmatic profiles of FXS-ASD and ASD-O may have important implications for assessment and intervention efforts. Future studies should continue to use a cross-syndrome comparison design, to better understand pragmatic patterns across clinical groups and other aspects of the social-behavioral phenotype (e.g. anxiety, ToM) as well as their impacts on pragmatics. Importantly, future research should also focus more on females with FXS, as pragmatic language in this group has been understudied thus far, as is the case for research in developmental disabilities more generally. Research in FXS should use ASD-O research as a model, as female-specific phenotypic expression went unrecognized for decades in idiopathic ASD research. Only more recently is literature emerging suggesting critical differences between males and females with ASD-O. These findings have serious implications for understanding etiology and informing targeted and maximally effective interventions (e.g. Cauvet et al., 2019; Halladay et al., 2015; Lai et al., 2015). Further research that continues to delineate the pragmatic profile of males and females with FXS, and address this in relation to key underlying mechanisms throughout development, is warranted. Of course, intervention research is critically needed as well.

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Chapter 5
Down Syndrome

Angela John Thurman and Laura del Hoyo Soriano

5.1 Introduction

Much of the behavioral research focused on intellectual and developmental disabilities, such as Down syndrome (DS), has been on language development. Importantly, as discussed by Jackendoff and Pinker (2005), language is "not a monolith but a combination of components" (p. 223). At the heart of children's mastery of the meaning of words, and the ways in which sounds and words can be modified and combined, lies mastery of the function of communication itself, or pragmatics (Airenti, 2017). Learning how to use communication for different reasons, change communicative strategies in response to a listener or situation, and to follow the rules governing communicative exchanges, is a complex process that interfaces with both language and several other cognitive domains (Airenti, 2017). Therefore, it is not surprising that individuals with DS, who experience overall delays in cognitive functioning as well as clinical features that likely influence their social experiences, should also experience pragmatic problems.

In this chapter, we summarize what is known about the DS phenotype, including epidemiology, clinical features, and cognitive profile. In addition, we review what is known and unknown about the nature and extent of pragmatic communication difficulties in individuals with DS, as well as the associations between pragmatic communication skills and other domains of functioning in individuals with DS. In doing so, we focus largely on the pragmatic features associated with the DS phenotype – while considering within-syndrome variability- from the prelinguistic period into

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adulthood. Finally, we also briefly consider general issues in the treatment of pragmatic difficulties in individuals with DS.

5.2 Epidemiology

In 1959, due to the advent of karyotyping, it was determined that DS results from the presence of an extra copy of the long arm of chromosome 21. It is currently understood that ~95% of cases of DS are due to the presence of three full copies of chromosome 21. The remaining ~5% of cases of DS are due to either an unbalanced translocation, in which part of a third copy of chromosome 21 attaches to another chromosome, or due to mosaicism, in which there is a mixture of cells with three copies of chromosome 21 and cells with two copies of chromosome 21 (1–2%; Mutton et al., 1996). Although the prognosis for those individuals with mosaicism is not always better than for those with other forms of DS (Carr, 2002), on average people with mosaicism have higher IQs when compared to those with three full copies of chromosome 21 or a translocation (Fishler & Koch, 1991).

It is currently estimated that the prevalence rate of DS is approximately 1 in 691 live births (Parker et al., 2010). As such, DS remains the most common genetic cause of intellectual disability (ID). There is some data from the US indicating some differences in the maternal age-adjusted prevalence rates of DS across three major maternal racial/ethnic groups, with a prevalence ratio of 0.77 for non-Hispanic black mothers and 1.12 for Hispanic mothers when compared to non-Hispanic white mothers (Canfield et al., 2006). Multiple reasons for these differences are possible, including methodological differences across the different surveillance systems considered, differences in factors relating to access to health care (e.g. socioeconomic status or education levels), and/or racial/ethnic differences in the use of health care access or practices (e.g. Canfield et al., 2006; Sherman et al., 2007).

The most common risk factor for DS is advanced maternal age, with the likelihood of having a child with DS rising from less than 1 in 1000 in mothers under 30 years of age to 1 in 12 in mothers 40 years of age (Roizen, 1997). In addition to advanced maternal age, altered recombination patterns have been shown to be an additional risk factor for DS (e.g. Ghosh et al., 2009; Sherman et al., 2006). Other potential risk factors have also been implicated in epidemiological studies of DS, including maternal weight (BMI > 30) during pregnancy, socioeconomic conditions, cigarette smoking, and radiation exposure (Yang et al., 1999; Ghosh et al., 2011; Sperling et al., 2012; Hunter et al., 2013; Hildebrand et al., 2014). However, large, population-based studies are required to confirm whether or not these factors confer risk beyond the factors already identified (e.g. Sherman et al., 2007).

5.3 Clinical Features

Because of the genetic mechanisms underlying the phenotype, nearly all individuals with DS demonstrate a characteristic set of craniofacial features, including brachycephaly, depressed nasal root, upward-slanting palpebral fissures, and epicanthal folds. Most individuals with DS ($\geq 50\%$) present with congenital heart disease (e.g. ventricular or atrioventricular septal defects), hearing loss, frequent ear infections, vision difficulties, hypotonia and ligament laxity (e.g. Bull & Committee on Genetics, 2011; Sherman et al., 2007). In addition, although less frequent in their occurrence, other medical conditions such as thyroid issues, seizures, hematological problems (such as anemia, iron deficiency, and leukemia), and celiac diseases are commonly observed in individuals with DS (e.g. Bull & Committee on Genetics, 2011). Finally, it is important to recognize that the presence of these medical comorbidities can negatively impact the level of functioning demonstrated by individuals with DS. For example, comorbid sleep difficulties (e.g. Breslin et al., 2014), thyroid dysfunction (Fernandez & Reeves, 2015; Lott, 2012), heart difficulties, and seizures (e.g. Arya et al., 2011; Eisermann et al., 2003) have also been shown to be negatively associated with cognitive development in individuals with DS. Thus, medical comorbidities in individuals with DS remain important factors to consider, even when they are not the focal area of investigation. Because individuals with DS are at increased risk of presenting with numerous other medical conditions, medical providers are recommended to monitor their patients with DS for the presence and progression of a variety of medical issues from birth (e.g. Bull & Committee on Genetics, 2011).

There are other behavioral features that are also considered to be characteristic of the DS phenotype. For example, one of the recurrent themes that emerges when considering the DS behavioral phenotype includes descriptions of increased sociability (e.g. Carr, 1995; Wishart & Johnston, 2008). These behavioral observations have also been supported by empirical research considering the social development of children with DS (Iarocci et al., 2008). Even though the social abilities and interests demonstrated by individuals with DS are thought to reflect areas of strength, these strengths do not preclude the presence of social difficulties. Toddlers and older children with DS are often described as demonstrating sudden changes in mood, being stubborn, or withdrawing from situations when frustrated, which can lead to task refusal (Carr, 1995; Fidler, 2005; Jahromi et al., 2008; Pueschel et al., 2008). Moreover, when placed under demanding/challenging conditions, children may rely on their social strengths in an attempt to avoid the demands placed on them (Pitcairn & Wishart, 1994). Although this strategy may not cause much concern early in development, as children grow, and the demands placed upon them increase, the use of this strategy can become increasingly problematic, and potentially can negatively impact later achievement (Gilmore & Cuskelly, 2009).

It is frequently noted that, as a group, individuals with DS demonstrate a lower risk of psychiatric symptomatology and comorbidities relative to their peers with non-specific ID or other genetic neurodevelopmental disorders (e.g. Dykens, 2007;

Tassé et al., 2016). These findings often overshadow the fact that the rates of psychiatric symptomatology and comorbidities observed in individuals with DS are indeed higher than what is reported for the general population (e.g. Dykens, 2007; Tassé et al., 2016). Although our understanding of the nature and consequences of psychiatric symptomatology in individuals with DS remains limited, and findings have been somewhat inconsistent across studies, likely due to variations in methodological procedures, it is clear that this is an area of growing consideration.

In general, there is evidence that the presence and severity of psychiatric symptomatology and comorbidities change across development (Dykens, 2007; Tassé et al., 2016; Urv et al., 2010). During the early childhood period, externalizing behaviors such as oppositionality, attentional difficulties and impulsivity have been noted. Ekstein et al. (2011), when using the DSM-IV-TR (American Psychiatric Association, 2000) to evaluate children with DS from 5 to 16 years of age, found that ~44% of their sample met DSM criteria for Attention-Deficit/Hyperactivity Disorder (ADHD). With regard to oppositionality, it is estimated that 10–15% of children with DS are diagnosed with disorders relating to conduct/oppositionality, primarily manifested by behaviors such as noncompliance, disobedience, and low-level aggression (e.g. Dykens, 2007).

In adulthood, depression may be an area of particular concern (Tassé et al., 2016). Moreover, changes in mood/behavior in the form of more depressive behaviors may forebear the later presentation of dementia (Grieco et al., 2015; Urv et al., 2010). However, it remains unclear the extent to which increases in depressive symptomatology is related to developmental changes in the DS psychiatric phenotype or is associated with the development of dementia (Esbensen et al., 2016). Additionally, although it is generally believed that rates of conditions such as psychosis, schizophrenia, and personality disorders are relatively rare in individuals with DS (Tassé et al., 2016), there have been multiple reports documenting the presence of psychotic symptomatology in individuals with DS. When comparing individuals with DS to those with other ID conditions (chronological age (CA) range: 13–29 years), Dykens et al. (2015) reported higher rates of Psychosis, Not Otherwise Specified in individuals with DS (35% versus 13%) and an additional 8% of individuals with DS presenting with depression associated with psychotic features. Behavior features associated with this presentation included hallucinations and/or delusions, withdrawal, and increased agitation. Similar features have also been reported in other investigations considering psychiatric symptomatology in individuals with DS (e.g. Jap & Ghaziuddin, 2011; Myers & Pueschel, 1991; Urv et al., 2010).

Finally, individuals with DS also appear to be at greater risk of presenting with symptoms of autism spectrum disorder (ASD). To date, although estimates of the prevalence of ASD have ranged from 5% to 39%, nearly all have studies have reported a prevalence of ASD greater than the reported 1% prevalence rate observed for the general population (e.g. Baio et al., 2018; Capone et al., 2005; DiGuiseppi et al., 2010; Lowenthal et al., 2007). That said, very few studies have utilized "gold-standard" diagnostic measures when considering the prevalence of ASD in individuals with DS. Despite the methodological limitations, our understanding of the presence of ASD symptomatology in individuals with DS has indeed improved. Evidence suggests that even when excluding individuals with DS who are described as having co-morbid ASD, increased rates of ASD symptoms are observed among individuals with DS (Channell, Phillips, et al., 2015). Moreover, individuals with DS who present with more limited cognitive or linguistic abilities appear to be more likely of presenting with increased severity of ASD symptomatology (Channell, Phillips, et al., 2015; Reilly, 2009). The implications of these findings remain unclear, with some disagreement as to the nature of ASD symptoms in individuals with DS. Nonetheless, methodologically-rigorous investigations focused on understanding the causes and consequences of the presence of ASD symptomatology in individuals with DS are needed.

5.4 Cognitive Profile

The DS phenotype is characterized by significant cognitive limitations. That said, it is important to recognize that at every stage of development, considerable heterogeneity is observed across individuals (Karmiloff-Smith et al., 2016). During the first year of life, differences relative to their peers with typical development (TD) are relatively subtle. As children move beyond the first year of life, nearly all demonstrate developmental delay (DD). Moreover, as children grow into the school-age years, children with DS increasingly lag behind their peers with TD, with most individuals demonstrating ID (see Carr (2005) and Couzens et al. (2012) for review). Because of the slower rate of learning demonstrated by individuals with DS relative to their peers, studies using standard scores from standardized measures of intelligence often report that the standard scores of individuals with DS decline as they age (see Carr (2005) and Couzens et al. (2012) for review). Unlike standard scores, both raw scores and growth scores provide metrics that reflect absolute levels of ability. Longitudinal investigations considering these types of metrics clarify that, albeit slowly, cognitive skills are indeed progressing across childhood and into early adulthood years (Channell et al., 2014; Couzens et al., 2012).

In adulthood, considerations of cognitive functioning for individuals with DS shift to considerations of cognitive loss. Because of multiple risk factors associated with chromosome 21, individuals with DS are at particular risk for not only an increased incidence of Alzheimer ‘s disease (AD) but also an earlier onset of the disorder as well (Hartley et al., 2017; Horvath et al., 2015; Patterson & Cabelof, 2012). Indeed, the neuropathology associated with AD is observed in nearly all individuals by the fourth decade of life and by their 60s more than half of adults with DS are demonstrating clinical symptoms of the disorder (Coppus et al., 2012; McCarron et al., 2017).

When considering performance across the different domains of cognitive functioning, at the group level, individuals with DS often demonstrate a specific pattern of strength and challenge. More specifically, although it is important to point out that these domains are still delayed relative to CA-expectations, individuals with DS are often described as demonstrating relative strength in the areas of nonverbal communication, gross motor skills, visual motor integration, and visual imitation (e.g. Brock & Jarrold, 2005; Fidler, 2005; Klein & Mervis, 1999). In contrast, areas that are considered particularly challenging for individuals with DS include auditory short-term memory, episodic memory, aspects of executive function, and expressive language (Abbeduto et al., 2016; Daunhauer & Fidler, 2011; Edgin et al., 2010; Fidler et al., 2005; Jarrold et al., 2009). In addition, difficulties in some aspects of visuospatial construction have been observed (Cornish et al., 1999).

Studies considering the language and communication skills of individuals with DS clearly exceed in number studies focused on other cognitive domains. Similar to findings about the DS cognitive profile, findings from these studies have demonstrated that a profile of areas of relative strength and challenge can be observed within the language domain specifically. As a group, individuals with DS often have better receptive language skills than expressive language skills (e.g. Chapman, 1997). However, even when considering only receptive language skills, areas of strength and challenge may occur. Individuals with DS may demonstrate a weakness in the comprehension of conceptual/relational vocabulary as compared to concrete vocabulary. In addition, syntactic skills are more significantly impaired than vocabulary skills (e.g. Finestack et al., 2013; Phillips et al., 2014).

Expressive language skills appear to be particularly affected in individuals with DS, with difficulties apparent even relative to individuals with other neurodevelopmental disorders (Abbeduto et al., 2016). Difficulties in the prelinguistic precursors to spoken language are apparent by late infancy for children with DS (e.g. Adamson et al., 2009; Mervis & Robinson, 2000; Roberts, Price, & Malkin, 2007) and difficulties with spoken language typically present by the toddler years and persist into adulthood (Abbeduto et al., 2007). For example, Chapman et al. (1998) found in their sample of 5- to 20-year-olds with DS that the average sentence length was only approximately three morphemes long. Finally, difficulties with speech intelligibility often remain a lifelong challenge and can impede their communicative success (e.g. Chapman et al., 1998; Kumin, 2006).

Importantly, data contributing to our understanding of the DS cognitive profile have been based on group-level findings. It remains unclear the proportion of individuals with DS who demonstrate this specific profile since the DS phenotype is also associated with considerable heterogeneity at every level and system of development (Karmiloff-Smith et al., 2016). Furthermore, several factors have been observed to account for within-syndrome variation. For example, variations in genetics, neurobiology, severity of affectedness, medical comorbidities, psychiatric comorbidities, and developmental period have all be shown to contribute to the presentation and development of the DS phenotype (e.g. Chapman et al., 1998; Fernandez & Reeves, 2015; Karmiloff-Smith et al., 2016).

5.5 Pragmatic and Social Communication Skills

5.5.1 Pragmatic Communication: Prelinguistic Period

In early interactions with their infants, caregivers often interpret their infant's gaze, affective, vocal, and/or physical productions as the child taking a turn in the "conversation" (Stephens & Matthews, 2014). By doing so, caregivers are modeling a conversational-like interaction, thereby setting the stage for the development of more complex interactions in which infants can follow the attention of another, share and direct their communicative partner's attention to objects/events of interests, as well as elicit aid from their communicative partner (Stephens & Matthews, 2014). As children grow, they begin to take a more active role in these interactions. It is through these bi-directional experiences, which occur well before a child is able to utter a word, that children with TD master the foundation upon which much communicative success as adults is built. Thus, these early skills (e.g. gaze use, affective expressions, and gestures) are often considered to serve as precursors to pragmatic language development (Airenti, 2017).

5.5.1.1 Gaze Behavior

Children's use of gaze is central to the development of early communicative exchanges observed between caregivers and children and is believed to contribute to the later development of intentionality (Tomasello & Carpenter, 2007). Thus, alterations or changes in the way children use gaze can potentially influence their social interactions, thereby leading to downstream negative consequences in the development of social cognitive and pragmatic skills (Tomasello, 1995).

Discussions involving gaze use in infants and young children often note the striking similarities observed between young children with DS and their peers with TD, particularly when considering children of similar developmental levels (Carvajal & Iglesias, 2000; Kasari et al., 1995; Moore et al., 2008; Slonims & McConachie, 2006). At the same time, some differences have been noted. Gaze use seems to emerge more slowly in children with DS relative to their CA-matched peers (Berger & Cunningham, 1981; Carvajal & Iglesias, 2000). Similarly, developmental changes in gaze use that are observed for children with TD seem to occur later for children with DS as well. In this regard, around 6 months of age, children with TD typically begin to reduce the amount of time they spend attending to caregivers and increase their attention toward their surrounding environment (Mundy & Jarrold, 2010). In contrast, studies considering gaze use during this developmental period have not observed children with DS to make this shift at the same time as their peers with TD.

This lack of shift in attention likely contributes to the between-group differences that have been observed, demonstrating that children with DS spend significantly more time attending to their caregivers than do their peers with TD (Berger & Cunningham, 1981; Carvajal & Iglesias, 2000; Legerstee & Bowman, 1989). There is also evidence demonstrating increased rates in the amount of time spent attending to people by children with DS into the toddler and preschool years (Berger & Cunningham, 1981, 1983; Kasari et al., 1990; Lewy & Dawson, 1992). Although more research is needed to elucidate the mechanisms underlying developmental differences observed in the area of early gaze use by children with DS, researchers have posited cognitive and motor limitations as potential influencing factors (Krakow & Kopp, 1983; Virji-Babul et al., 2006).

Findings like these indicating that children with DS spend more time attending to the people around them are often interpreted as early evidence of children's preference for social interaction. Interestingly, even though children may be looking more at their partner, there is some evidence suggesting that the looks produced by children with DS may be more ambiguous than those of their peers. Walden et al. (1997) had unfamiliar raters view video clips of children with TD, children with DD, and children with DS interacting with their caregivers and asked raters to (1) determine whether or not the child looked at the caregiver and (2) rate their confidence in their judgment using a 5-point Likert scale. Gaze use in all samples had been previously coded by trained researchers. Results indicated that unfamiliar raters were significantly less accurate when rating looks to the caregiver's face made by children with DS than they were at rating the looks made by either the children with TD or children with DD. Moreover, even when considering only those ratings that were accurately classified, untrained raters were less confident in their assessment of the looking behavior demonstrated by the children with DS in comparison to the other two groups (Walden et al., 1997). Because prompt and appropriate responses to a child's overtures, such as gaze use, allows the child to more ably learn about how their behavior influences the surrounding social world, increased ambiguity in the looks of children with DS could potentially change the back and forth exchange happening between caregivers and children and, in turn, negatively impact the trajectory of children's pragmatic development (Fidler et al., 2011).

When considering triadic attention, although there have been some discrepancies across findings, it is generally believed that joint attention, or the ability to share an experience, object, or event with another person, is an area of relative strength in children with DS, with performance comparable to that of mental age (MA)-matched TD peers (Hahn et al., 2018). Interestingly, Kasari et al. (1995) examined attention regulation by children with DS in two different situations designed to press for triadic interactions: a semi-structured play session designed to press for joint attention and requesting, and a social referencing paradigm designed to assess triadic attention in an unambiguous context. Results indicated that even though children with DS (mean CA = 24.60 months) did not differ from the children with TD matched on verbal MA (mean CA = 14.22 months) on rates of joint attention during the semi-structured play session, children with DS were less likely to demonstrate joint attention during the social referencing task, regardless of the emotional display (joy or fear). Further analyses indicated that children with DS spent approximately the same amount of time looking at the stimulus and looking at the adult, whereas the children with TD spent considerably more time attending to the stimulus than to the adult, resulting in less time looking at the adult relative to the DS group. The authors interpreted their findings to suggest that children with DS were less able to appraise the situation in the social referencing paradigm, theorizing that they may have failed to make the connection between the intended emotional message and the stimulus (Kasari et al., 1995). Similar findings were reported by Knieps et al. (1994) who also considered children's gaze use during a social referencing task.

5.5.1.2 Facial Expressions

Although facial expressions of emotion are generally viewed as behavioral responses indicative of underlying mental states, they also serve to regulate the behavior of other individuals (Sorce et al., 1985). It is this interpersonal regulatory function that makes affective expression an important component of the communicative process (Holodynski & Friedlmeier, 2005). From a young age, TD children are very attentive to the emotional expressions of those around them. Beginning around 6 months of age, infants with TD are observed to respond differentially to their mother's positive and negative expressions (Cohn et al., 1990; Termine & Izard, 1988) and around 1 year of age, infants are able to use the emotional information communicated by another person to help appraise an ambiguous situation (Feinman, 1992; Sorce et al., 1985).

To date, our understanding of the development and use of affective expressions by children with DS is more limited than our understanding of other developmental domains. That said, research conducted thus far does indeed offer some insights. Subtle differences are thought to be present both in children's use of affect and in the way they respond to the emotional expressions of those around them. More specifically, in infancy, children with DS have been described as demonstrating more muted affective displays in comparison to their peers with TD, at least in some circumstances (Cicchetti & Sroufe, 1976). During the preschool years, Kasari et al. (1990) observed that although affective displays by children with DS were similar to those of MA-matched TD children, the children with DS were observed to change the length of their affective displays more frequently than did their peers with TD. In addition, similar to the findings on gaze use, there is some evidence that it may be more difficult to interpret the affective states of infants with DS as compared to their peers with TD (Carvajal & Iglesias, 2006). As children grow, however, these patterns may change (Hyche et al., 1992).

Moreover, when considering the emotion recognition skills of children with DS relative to both MA-matched children with TD or MA-matched children with other forms of DD, Kasari et al. (2001) found that although in both groups, children were more accurate at recognizing, identifying, and labeling positive affective expressions as compared to fearful expressions, overall, children with DS performed significantly worse than their peers with TD. Furthermore, in a follow-up study, the authors found that when children with DS incorrectly identified the emotion "fear", the responses of the children with DS were significantly more likely to be that of positively valenced emotions than were the responses of the MA-matched children with TD or MA-matched children with other forms of DD.

Finally, a key socio-communicative development that typically occurs within the first year of life for children with TD, involves the regulatory function of social referencing. In general, the social referencing process involves an interactive situation in which one considers whether a child uses an adult's appraisal of an ambiguous situation, communicated primarily through affective displays, to guide his or her own behavioral response. Knieps et al. (1994) found that, in addition to spending more time looking at the adult in comparison to the MA-matched children with TD (as was previously discussed), the toddlers with DS were more likely to demonstrate positive affect in response to the adults' negative emotional displays. More recently, Thurman and Mervis (2013) compared the social referencing skills of preschoolers with DS and preschoolers with Williams syndrome (WS), a neurodevelopmental disorder associated with significant pragmatic difficulties. Results indicated that the majority of children in both groups did not regulate their own behavior in accordance with an adult's expression of fear toward an ambiguous stimulus. In fact, the authors found that although children with DS demonstrated strengths relative to the children with WS with regard to initiating eye contact with the examiner and in following the examiner's gaze in triadic situations, neither the DS children nor the WS children were observed to regulate their behavior in response to expressions of fear. In contrast, the majority of children in both groups demonstrated positive responses regarding the ambiguous stimulus in situations in which the examiner demonstrated a joyful expression.

5.5.1.3 Gestures

Gestural production provides children with yet another tool for learning from and about the social world around them. Overall, young children with DS demonstrate a relative strength in the use of gestures, albeit gesture production still lags relative to CA-expectations. Results from investigations of early gestural development in children with DS suggests that, in many ways, the frequency and diversity of gestural productions is akin to those observed in younger children with TD with similar communicative skills. For example, for both children with DS and children with TD, deictic gestures (e.g. points, shows, reaches), which are intended to draw attention to an object or event, typically emerge prior to the onset of a child's first word (Capone & McGregor, 2004; Chan & Iacono, 2001). Interestingly, multiple studies have observed increased gesture use by children with DS when compared to younger peers with TD matched on MA (Caselli et al., 1998; Singer Harris et al., 1997). These findings have led some researchers to posit that children with DS may use gestures to compensate for the spoken language challenges they experience and improve their communicative success.

In support of this argument, investigations of the development of representational gestures (i.e. gestures that symbolize a referent) in children with TD over the course of the first 2 years of life have observed a gestural trajectory in which production steadily increases, appears to reach a peak, and then steadily decreases until it levels off. This pattern of performance has been described as an inverted U-shaped trajectory (Camaioni et al., 2003). The observed decrease in representational gestural productions occurs as children transition into the first words stage of spoken language development (Iverson et al., 1994). Comparisons of representational gesture production between young children with DS and MA-matched peers with TD seems to show a profile in which the children with DS produce fewer spoken words but more gestures, when compared to their MA-matched peers, such that when communicative productions take into account both of these domains, no differences are observed between the groups (Caselli et al., 1998; Galeote et al., 2008; Stefanini et al., 2007). That said, not all investigations of gestural productions have yielded findings in support of a "gestural advantage in DS" (see Iverson et al., 2003).

Despite these indications of relative strength in gestural use, there is some evidence suggesting that there may be subtle differences in the way in which children with DS use gestures according to their pragmatic function. Although there have been some discrepancies across findings, it is generally believed that children with DS are similarly able, when compared to their MA-matched peers with TD, to use gestures when producing joint attention acts (e.g. Hahn et al., 2018). In contrast, children with DS produce fewer nonverbal communication acts intended to request help than MA-matched peers with TD (Fidler et al., 2005; Mundy et al., 1995; Sigman et al., 1999). For example, Fidler et al. (2005) found that toddlers with DS were less likely to produce requests for help, but not joint attention behaviors, when compared to MA-matched children with TD. Nonverbal requesting performance was comparable between the children with DS and the MA-matched sample of children with mixed etiology DD. However, unlike the children with DS, children in the mixed etiology DD group did not show any discrepancies between their requesting and joint attention skills. Interestingly, when considering requests to elicit a social response, no difference was found between the DS and MA-matched TD group. Moreover, results from this study demonstrated that although joint attention was significantly associated with language across all three groups, nonverbal requesting was significantly associated with expressive language skills only for the children with DS (Fidler et al., 2005). The ability to request help appears to serve as a precursor for the later development of language and executive functioning (Fidler, 2005).

Finally, although gesture use is known to decline as children transition into the first words stage of spoken language development, gestures remain an important part of children's communicative repertoire. The production of gesture-word combinations play a pivotal role in children's transition from the first words to word combinations phase of spoken language development (Capirci et al., 1996; Iverson & Goldin-Meadow, 2005). Research on children with TD indicated that gesture-word combinations that convey two elements (e.g. pointing to a snack container while saying "more") predicts the onset of two-word combinations (Iverson & Goldin-Meadow, 2005). It is believed that these types of gestures encourage caregivers/communicative partners to provide more advanced verbal input to the child. By "translating" the children's gestures (e.g. "Yes, more cookie"), the caregiver models the two-word combination, facilitating the child's learning. Although the general ability to pair gestures with words has been associated with the onset of two-word combinations, children with DS seem to demonstrate delays in the production of these gesture-word combinations that convey two elements when compared to MA-matched peers with TD. Thus, once again, these findings support a tight link between gesture use and spoken language skills in children with DS.

5.5.2 Pragmatic Communication: The Linguistic Period

5.5.2.1 Overall Pragmatic Language Profile

When considering the pragmatic skills of individuals with DS during the school-age period and beyond, there is clear evidence indicating that individuals with DS do indeed demonstrate overall pragmatic difficulties, even relative to MA-level expectations. However, it is important to note that not all facets of pragmatic skills are equally impacted. That is, findings from research in this area suggest that individuals with DS are likely to demonstrate areas of both relative strength and weakness within the pragmatic domain itself.

To date, multiple studies have used informant-report measures, particularly the Children's Communication Checklist-2 (CCC-2; Bishop, 2003), or the prior version of this measure, to characterize the pragmatic profile associated with the DS phenotype. By 6 years of age, although children with DS demonstrate strengths in pragmatics relative to their structural language skills, impairments in all areas of pragmatics (i.e. inappropriate initiations, stereotyped language, use of context, and nonverbal communication) are apparent relative to their MA-matched peers with TD based on caregiver-report (Smith et al., 2017). More specifically, caregivers reported their children with DS to have particular difficulty using context to interpret what was said to them, with performance significantly weaker in this domain relative to the other aspects of pragmatics considered (Smith et al., 2017). Furthermore, difficulties in the areas of initiating conversation and using stereotyped language were reported by caregivers to be more significant than were children's difficulties in the area of nonverbal communication.

These early weaknesses in pragmatic skills continue to be an area of challenge beyond the early school years. Laws and Bishop (2004) observed that when using the Children's Communication Checklist (CCC; Bishop, 1998), teachers rate individuals with DS (mean CA = 15;11 years; CA range: 10–22 years) as being more pragmatically impaired in all areas considered (i.e. inappropriate initiations, coherence, stereotyped conversations, use of context, and rapport) than younger children with TD (mean CA = 6;4 years; CA range = 4;11–6;8 years). In fact, a significant number of participants with DS (40%) were classified by the measure as demonstrating a clinically significant pragmatic impairment. In addition, when compared to younger children with Specific Language Impairment (mean CA = 6;0 years; CA range = 4;05–7;02 years), teachers' ratings for the individuals with DS reported more difficulties in the areas of inappropriate initiation, stereotyped conversation, and use of context. No differences were observed between the groups in the areas of coherence and rapport. Overall, although these data are consistent with the findings reported by Smith et al. (2017) for 6-year-olds with DS, it is important to note that the subscales comprising the pragmatic domain of the CCC versus the CCC-2 differ. Moreover, Laws and Bishop (2004) did not attempt to match the groups considered in their study on CA or MA, but the authors reported that pragmatic scores were not associated with CA in any of the groups. These methodological differences complicate comparisons across these studies.

Although the data from these studies certainly provide insight into the pragmatic performance of individuals with DS, it is important to recognize that informant-report measures are one of several types of tools available to assess pragmatic skills. Utilization of multiple methodological assessment procedures is likely to expand the scope of our understanding of the pragmatic skills of individuals with DS. Indeed, Lee et al. (2017) took just this approach in their characterization of the pragmatic communication profile of individuals with DS, by including not only informant reports, but also standardized and naturalistic measures within their methodological design. The authors found that their sample of school-aged children with DS demonstrated weaknesses relative to the younger, sex-matched controls with TD of similar MA when considering performance on nearly all metrics stemming from the informant-report assessment and on the standardized assessment included in the project. In contrast, a more complex profile of performance was observed when utilizing naturalistic observation methods. More specifically, when considering the total number of pragmatic violations, females with DS, but not males, were observed to commit significantly greater pragmatic violations when compared to sex-matched peers of similar MA (Lee et al., 2017).

In general, these data differ from a prior cross-sectional study showing significant differences between boys with DS and boys with TD (Klusek et al., 2014), in which the same standardized test and semi-naturalistic methods were used. Incongruences across both studies are likely due to differences in participant characteristics. Mean verbal MA of the participants in the Klusek et al. (2014) study was higher than in the Lee et al. (2017) study. This could suggest that advancing language skills may be linked with greater opportunities for committing pragmatic language violations. Indeed, in the Lee et al. (2017) study several higher-order pragmatic skills worsened for boys with DS over time, such as taking too lengthy conversational turns or being redundant in topics introduced.

5.5.2.2 Referential Communication Skills

In their attempts to provide a more in-depth understanding of the pragmatic difficulties experienced by individuals with DS, a number of investigators have focused specifically on investigating referential communication skills, or the ability to manage and respond to misunderstandings and communicative breakdown (e.g. Abbeduto et al., 2006). For example, Abbeduto et al. (2006) used a non-face-to-face referential task in which participants were to describe a novel shape so that the listener could identify the referent from the other novel shapes in the array. Some between-group differences were apparent when comparing the performance of adolescents and young adults with DS to that of MA-matched peers with TD. More specifically, although the overall strategy used by the participants in both groups was similar, the adolescents and young adults with DS were more likely to use a non-discriminating description of the intended referents than were the MA-matched children with TD. Moreover, results from the Abbeduto et al. (2006) study also suggested that the adolescents and young adults with DS were significantly less likely to use scaffolding to help support the listener's understanding of the description (by saying, for example, "it looks sort of like a boat" rather than simply "it's a boat."). Taken together, the authors posited that individuals with DS may be less able to consider the informational needs of their communicative partner which, in turn, can increase the likelihood of miscommunication. These types of difficulties have the potential to negatively impact the ability of individuals with DS to establish and maintain social relationships.

When serving in the role of the listener in a communicative exchange, Abbeduto et al. (2008) found in school-age children and adolescents with DS a weakness, relative to MA-level expectations, in the ability signal non-comprehension to a speaker, with individuals with DS verbalizing that a problem was encountered only 30% of the time as compared to 70% of the time for the MA-matched participants with TD. Moreover, the authors found that performance varied as a function of the nature of the problem encountered, with the overall profile of non-comprehension signaling similar between the two groups (Abbeduto et al., 2008). That is, when directions were "incompatible" (i.e. the referent identified was not available), individuals with DS were most successful signaling to the speaker that a problem was encountered. Developmentally speaking, this type of signaling is the most salient and easiest to resolve. In contrast, individuals with DS had more difficulty signaling message inadequacy in situations in which the directions were ambiguous (i.e. directions for which multiple, plausible referents were available) or directions that contained an unfamiliar label.

Finally, as mentioned previously, not all facets of pragmatic skills are equally impacted. In a recent study, Barstein et al. (2018) considered the communication repair strategies of youth with DS (CA range: 6–14 years) using a picture description task designed to probe for different types of communicative repair strategies (e.g. statement repetition, elaboration, nonverbal and paralinguistic supplementation) and to mimic the types of circumstances participants encounter in their daily lives. Overall, participants with DS demonstrated proficiency repairing communication breakdowns with no significant differences in their performance relative to the comparison group with TD, after controlling for differences in nonverbal cognition and language ability. The DS group responded appropriately to requests for clarification and varied their strategies in response to different prompts (Barstein et al., 2018).

5.5.2.3 Narrative Skills

Narrative language competence, or the ability to generate or retell a story, is an important skill that contributes to the ability to maintain cohesive conversational interactions in social situations (McCabe & Bliss, 2003). Furthermore, narration, much like conversation, requires perspective taking and inferences about the mental states (e.g. emotions, plans, and goals) of story characters as well as predictions about character actions and reactions. Thus, consideration of narrative language can provide a window into children's development across the pragmatic domain.

Multiple investigations of narrative language competence have been conducted to date. Findings from these studies clearly indicate the importance of considering both nonverbal cognition and expressive syntax when considering participant performance. For example, a study exploring narrative language competence in adolescents with DS (e.g. Channell, McDuffie, et al., 2015), found that individuals with DS expressed fewer of the elements of episodic structure than did peers with TD matched on nonverbal cognitive level. However, once mean length of utterance was accounted for, the between-group differences in the expression of episodic elements were eliminated. A similar pattern of findings has been reported when considering the use of inferential language by individuals with DS during a narrative task (Ashby et al., 2017). Indeed, there does appear to be a pattern emerging in the literature, with studies that match participants based on nonverbal ability more likely to observe group differences in narrative performance (e.g. Chapman et al., 1998) than do studies that match participants based on expressive language performance (e.g. Thordardottir et al., 2002).

Most research conducted to date considering the nature and consequences of pragmatic skill development in individuals with DS has matched participant groups on overall cognitive ability or MA. Given the pattern of findings emerging from the narrative language literature, it will be important for research on pragmatic skill development to also consider the impact of matching nonverbal versus verbal cognitive ability on study findings. Importantly, although narrative language can provide a window into children's development across the pragmatic domain, it is important to recognize that the rules and principles governing narrative indeed differ from those used in everyday conversation. Moreover, because narrative can require a greater syntactic demand of the speaker than is required by day-to-day conversation, different patterns may be observed in the areas of challenge experienced by the individual as well as in the correlates and consequences of any experienced difficulties. Nonetheless, insights from the narrative language literature will likely support pragmatic research development and may provide a useful tool for elucidating the mechanisms underlying both narrative and pragmatic competence.

5.5.2.4 Sex-Related Differences

It is important to note that there are many other factors to consider when interpreting patterns of strength and weakness within the pragmatic domain. For example, sex-related differences in the pragmatic skills of individuals with TD have been documented (e.g. Kothari et al., 2013). Although much more work is needed in this area, some researchers have considered the potential presence of sex differences in the pragmatic language skills of individuals with DS. In this regard, Lee et al. (2017) included as part of their investigation, in addition to comparisons to sex-matched samples of peers with TD, direct comparisons between the male and female participants with DS in their study. Results from these analyses indicated that, although sex differences were not observed on their informant report and standardized assessment measures, differences were apparent when considering their naturalistic observation measure.

More specifically, when considering the total number of pragmatic violations, females with DS were found to commit significantly more errors relative to the TD-female participants, but this finding was not observed when comparing males with DS to the male participants with TD (Lee et al., 2017). Closer consideration of the data demonstrated sex-related differences in the nature of pragmatic difficulties experienced by those with DS relative to the samples with TD. Coding of the naturalistic observation measure indicated that females with DS were more likely than their male counterparts to use excessive detail, produce inappropriate topic shifts, and demonstrate limited eye contact. In contrast, males with DS demonstrated greater difficulty than did their female peers in modulating their rate of speech. Interestingly, these sex differences differed from the sex differences observed in the sample with TD.

del Hoyo Soriano et al. (2018) did not observe differences in parental reports of pragmatic language performance on the CCC-2 between their male and female participants with DS (mean CA = 12.7 years) matched on CA and nonverbal cognition. However, this study did not include naturalistic measures of pragmatics, nor did it include a sample with TD for additional comparisons. Interestingly, however, the authors did observe significant, sex-related differences in favor of girls in expressive vocabulary and expressive syntactic skills when assessed through both standardized and naturalistic methods. Moreover, males with DS were also observed to be more unintelligible than were females with DS (del Hoyo Soriano et al., 2018). Other studies focused on the adolescent period have also failed to observe sex differences with regard to pragmatics when using direct assessment methods (Martin et al., 2017). In sum, more research is needed to determine if males and females with DS do indeed demonstrate pragmatic differences relative to each other or their sex-matched peers with TD and to understand if any such sex-related differences in pragmatic abilities vary as a function of CA, overall cognition, language ability, and methods of assessment (e.g. semi-naturalistic conversational context vs. standardized methods).

5.5.2.5 Comparisons to Other Neurodevelopmental Disorders

When interpreting the pragmatic profile associated with the DS phenotype, another factor to consider is the comparison group used within the methodological design. Even though individuals with DS demonstrate pragmatic weaknesses relative to their peers with TD, areas of pragmatic strength for individuals with DS are often noted when they are compared to individuals with other neurodevelopmental disorders.

Consider, for example, the research comparing pragmatic performance between individuals with DS and individuals with WS. Although initial comparisons between these two phenotypes were focused on elucidating why different structural language profiles were observed across these neurodevelopmental disorders when samples were seemingly comparable on overall cognitive ability, it became clear that there are notable differences in cognitive and socio-communicative profiles across these conditions. For example, overall IQ scores for individuals with DS are generally observed to be significantly lower than those observed in individuals with WS, particularly beyond the early school age years (e.g. Klein & Mervis, 1999). In addition, although expressive language impairment is a key feature of the DS phenotype, expressive language skills are generally considered to be an area of relative strength for those with WS (Abbeduto et al., 2016).

Interestingly, despite this pattern of performance, research comparing pragmatic performance between individuals with DS and individuals with WS frequently find that individuals with DS demonstrate pragmatic strengths, at least in some skill types, relative to individuals with WS and that areas of pragmatic strength can be observed even in early childhood (e.g. Thurman & Fisher, 2015). For example, when comparing participants with DS (mean CA = 15;11 years; range = 10;02–22;09 years) to children with WS (mean CA = 14;10 years; range = 6;05–25;02 years), although teacher ratings of overall performance seemed comparable between the groups, approximately 80% of the participants with WS were classified as having a pragmatic impairment, compared to 50% of the participants with DS. In addition, the participants with DS were reported to demonstrate better performance in the areas of “use of context”, “inappropriate initiation”, and “stereotyped conversation” than did the participants with WS (Laws & Bishop, 2004). No data was available on the cognitive performance levels of the participants within this study.

Similarly, a number of studies have compared the pragmatic skills of participants with DS to participants with fragile X syndrome (FXS), a single-gene X linked neurodevelopmental disorder that is also the most common single gene cause of ASD (Betancur, 2011). Results of these comparisons have also identified multiple areas of pragmatic strength in individuals with DS relative to those with FXS. For example, del Hoyo Soriano et al. (2018) found that caregivers of adolescents with DS reported fewer difficulties in the areas of “inappropriate initiation”, “stereotyped language”, and “nonverbal communication” difficulties than did caregivers of males with FXS. In addition, others have found use of perseverative language to be lower in males with DS than in males with FXS during the school-age years, after controlling for nonverbal cognitive ability and regardless of ASD status. When comparing participants with DS to participants with FXS who also have a comorbid ASD diagnosis (FXS + ASD), there is evidence of lower rates of noncontingent discourse in males with DS than in males with FXS + ASD (Roberts, Martin, et al., 2007). Moreover, Barstein et al. (2018) found that males were DS evidenced a significantly higher rate of suprasegmental cue use than did males with FXS + ASD. The authors theorized that the higher rates of suprasegmental cue use demonstrated by the participants with DS may be an adaptive strategy used by the participants with DS to compensate for their intelligibility difficulties (e.g. Kent & Vorperian, 2013).

It is important to recognize that, when interpreting findings like the ones demonstrated in the comparisons above, both the WS and FXS phenotypes are associated with a relative weakness in pragmatics. Thus, although individuals with DS may demonstrate strengths relative to these neurodevelopmental disorders, this does not mean that individuals with DS are not in need of support in these areas as well. Moreover, phenotypic differences across neurodevelopmental disorders, outside of the area of pragmatics, are likely to influence the nature and consequences of pragmatic development itself. Therefore, even in instances in which pragmatic performance appears to be at similar levels across neurodevelopmental disorders, the factors influencing, and being influenced by, pragmatic development may differ. More research is this area is likely to significantly improve our understanding of pragmatic language development in DS and elucidate treatment approaches.

5.5.2.6 Social Cognitive Considerations

Language development, and therefore the development of pragmatics, is closely linked to developments in social cognition. Social cognitive advancements, such as developments in early social learning and theory of mind skills, provide the necessary foundational skills to decipher word meanings and establish pragmatic competence (Macnamara, 1972; Ninio & Snow, 1996). Moreover, advancements in language provide a necessary foundation and support for social cognitive development; thus, language and social cognitive development are intricately intertwined (Fitch et al., 2010). Consistent with the findings from the literature on pragmatics in DS, the degree to which individuals with DS show impairment or strength in social cognitive skills also varies over the course of their development.

Early development of the socio-cognitive skills of children with DS are considered to be relatively similar to patterns observed in children with TD, although some subtle differences are noted. In fact, many of the same prelinguistic pragmatic skills reviewed earlier (e.g. gaze use, social referencing skills, pragmatic function of gestures) are also considered foundational skills for the development of social cognition. Indeed, these early skills are believed to serve as the roots for the development of later, more complex, socio-cognitive abilities such as theory of mind (ToM) (Cebula et al., 2010). ToM refers to the ability to attribute cognitive and affective mental states (e.g. beliefs and emotional states) both to one's own mind and to the minds of others. It allows us to explain and predict the behavior of others and to recognize how mental states can differ from our own (Carpendale et al., 2007).

Individuals with DS demonstrate significant delays in the development of ToM relative to their peers with TD (see Cebula et al., 2010 for review). Research comparing ToM development in individuals with DS relative to individuals with other intellectual and developmental disabilities is more limited. That said, findings from this area demonstrate the importance of looking beyond overall levels of performance to consider the different aspects of ToM performance profiles as well as the potential mechanisms underlying social cognitive development (e.g. Cornish et al., 2005; Giaouri et al., 2010). For example, Cornish et al. (2005) found that even though participants with DS and participants with FXS demonstrated similar overall performance levels on the ToM task considered, the types of errors underlying this performance differed between the groups. More specifically, the authors administered an appearance-reality task in which an orange filter was placed over a white piece of paper, thereby changing the apparent color. On this task, participants with DS were more likely to answer the question "what color is it really?" with the answer orange, suggesting that participants had difficulty ignoring the perceptual information provided even when it contradicted his/her knowledge of the object. This type of data highlights the importance of digging deeper in order to understand the factors influencing, and being influenced by, ToM development. Thus, future studies should consider including assessment tools that allow consideration of different aspects of false belief understanding and consider conducting more in-depth, qualitative analyses of error patterns in order to better describe the DS phenotype.

Social cognitive profiles and their developmental trajectories play an important role in the development of pragmatics. ToM, for example, is believed to provide the foundation for a variety of important pragmatic functions, such as monitoring and addressing the informational needs of one's communicative partner (Frith, 1996). Indeed, Barstein et al. (2018) found that increases in ToM performance were related to lower rates of off-topic responses, higher rates of adding information to responses, and higher rates of revisions during conversation in children and adolescents with DS. Moreover, Lee et al. (2017) reported that associations between ToM and pragmatic competence diminished for children and adolescents with DS and for peers with TD when controlling for MA and structural language abilities (i.e. mean length of utterance in morphemes), suggesting a strong bidirectional relationship between ToM and syntactic development (de Villiers, 2007). Thus, more research is needed to disentangle the relations between ToM, pragmatic competence, and structural language abilities.

Importantly, ToM is one aspect of social cognition. There are many other sociocognitive skills/domains that should be considered in relation to pragmatic language performance. For example, more research is needed to clarify the nature of impairments in emotion processing and ToM as well as to determine their influence on interpersonal challenges commonly reported among individuals with DS (e.g. overly friendly approaches toward strangers; "hypersocial" or excessive displays of affection such as hugging that are not appropriate to the situation) (Capone et al., 2006; Iarocci et al., 2008; Steingass et al., 2011). As discussed earlier in this chapter, although individuals with DS are often described as being friendly and "hypersocial", behavioral difficulties are not uncommon and may leave them vulnerable to exploitation (e.g. Jahromi et al., 2008). Empirical research is needed to explore the relation between this domain of social cognition, as well as others, and pragmatic language development.

5.6 Intervention

As we have reviewed in this chapter, despite their social strengths, individuals with DS do indeed have pragmatic difficulties. Findings from the studies reviewed in this chapter shed light on potential intervention targets. Given the vital role pragmatic language competence plays in daily living skills, interventions that are tailored to address the specific needs of individuals with DS across a variety of contexts are likely to support the development of positive peer relationships, intimate relationships, community integration, and employment success. The benefit of such improvements is likely to significantly improve long-term outcomes for individuals with DS. Importantly, the implementation and development of such interventions are likely to be most successful if treatment planning not only considers the pragmatic needs of those with DS, but also the potential contributions of other phenotypic factors on pragmatic success (e.g. hearing and speech-motor impairments, medical and behavioral comorbidities).

Despite extensive documentation showing pragmatic communication impairments in individuals with DS, there are very few empirically validated interventions with this population. In addition, most of the interventions that have been developed are designed to target the general communicative needs of individuals with DS, rather than specifically targeting pragmatic skills. Neil and Jones (2016) conducted a systematic review to examine the characteristics of effective interventions (e.g. strategies and intensity), whether interventions are tailored to the DS behavior phenotype, and the effectiveness (i.e. percentage non-overlapping data and effect size) of interventions. The systematic search identified 37 studies, and results were largely positive, including behavior analytic strategies (prompting and reinforcement) for increasing communication. Based on findings from this meta-analysis, Neil and Jones reported benefits of using behavior analytic techniques to improve communication outcomes among individuals with DS.

In addition, parent-implemented language approaches, in which clinicians, using a collaborative model, train and coach parents to implement evidence-based practices with their children in the context of daily routines and activities, have also been commonly used to target language skills. In a recent Cochrane systematic review, O'Toole et al. (2018) aimed to examine the effects of parent-implemented interventions for improving verbal and non-verbal communication and language skills, socialization, and behavior in young children with DS, between 29 months and 6 years of age. The study included randomized and quasi-randomized controlled trials that compared parent-implemented interventions designed to improve communication and language versus teaching/treatment as usual, or no treatment, or delayed (wait-listed) treatment, in children with DS. Results from the meta-analysis provided insufficient evidence to determine the effects of parent-implemented interventions for improving communication of children with DS. This review highlights the need for well-designed studies, including randomized controlled trials, to evaluate the effectiveness of parent-implemented interventions compared to placebo or to other types of intervention.

Clearly, limited research is available that considers the effectiveness of various intervention approaches for use in individuals with DS. Indeed, the development of evidence-based interventions for DS have lagged behind those for other neurodevelopmental disorders. However, renewed calls for action are fueling efforts to address this critical need (National Institute of Child Health and Human Development, 2014). Research on other populations has demonstrated the utility of behavioral interventions, peer-mediated interventions, and social communication treatments (e.g. social skills groups, social stories) (Cummings, 2009). There is a clear need for evidence-based practices that utilize these types of approaches, targeting pragmatic skills across development in individuals with DS.

5.7 Summary

Results of the research reviewed in this chapter clearly indicate that individuals with DS demonstrate pragmatic language impairments that begin to emerge early in development. In addition, however, some skills are impacted more than others. Overall, children with DS are often described as demonstrating a relative strength in gaze use, use of affect, and use of gestures. That said, some differences have been noted when comparing children with DS to their peers with TD of similar CA or MA. Moreover, there may be functional differences in the ways in which children with DS rely on these tools, depending on the context and/or cognitive demands placed on them. Early differences in the use or saliency of these communicative cues have the potential to alter the ways in which children's social partners interact with them, thereby shaping the development of social communication and pragmatics more generally (Fidler et al., 2011).

Once individuals reach a point at which they rely on speech as their primary mode of communication, this presentation of difficulties across all aspects of pragmatics continues. Although there is a need for more investigation, particularly using longitudinal designs at different linguistic stages and age levels, the literature so far describes a complex profile of strengths and weaknesses. Challenges may include the initiation and elaboration of topics, initiation of communicative repairs, and some linguistic aspects of narratives. Strengths tend to include use of a variety of communicative functions, ability to stay on topic, responses to requests for clarification, and storytelling with sufficient content when visual supports are used. This pattern of relative strengths and weaknesses remains during young adulthood.

However, later on, dementia in older adults with DS may compromise several of these strengths related to verbal communication.

To date, evidence-based interventions that have been specifically validated to be effective for individuals with DS is limited. However, the growing literature on the pragmatic needs of individuals with DS clearly demonstrates the need for treatment studies targeting pragmatics. Moreover, research should consider the characteristics of the responders and non-responders, monitor adherence to treatment protocols, as well as maintenance and generalization of outcomes. A wide adoption of these standards may establish a clearer picture of the effects of the intervention and may constitute the basis for decision-making in public health and social policies related to DS.

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Chapter 6
Williams Syndrome

Daniela Plesa Skwerer

6.1 Introduction

When encountering a person with Williams syndrome (WS) for the first time, one can be overwhelmed by an impression of instant and unbounded friendliness. This experience is often hard to reconcile with the understanding that the same person is likely to have intellectual disability, and has serious difficulties making sense of the social world. This is one of the many paradoxes that have made WS a fascinating target of investigations across several research fields – from genetics to the philosophy of language – over the last decades. The chapter opens with a few historical notes about how WS – a relatively rare neurodevelopmental disorder of genetic aetiology – came to be at the center of heated theoretical debates concerning the relations between language, cognition, and social behavior, and ultimately about the structure of the human mind. The brief excursion into the history of research on this syndrome provides a context for understanding the current state of knowledge about pragmatics in WS, another area of seemingly paradoxical capacities demonstrated by people with this intriguing neurodevelopmental disorder.

In the next section of the chapter, the neuropsychological and behavioral profile of individuals with WS is outlined, with a particular focus on two domains that play a critical role in the development of pragmatic skills: language and social-cognition. These domains had initially been considered to be 'intact' or 'spared' in people with WS, despite their 'severe' intellectual disability' (Von Arnim & Engel, 1964; Bellugi et al., 1988). Later research findings, however, indicated that this interpretation was premature, and that the linguistic, cognitive and social-behavioral phenotypes associated with WS consist of a complex mixture of strengths and deficits within these domains (for reviews see Järvinen-Pasley et al., 2008; Martens et al., 2008; Mervis, 2006; Plesa Skwerer, 2017).

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The rest of the chapter presents an overview of the main assessment methodologies that have been used so far in research investigating pragmatic functioning in WS. These range from parent-report questionnaires to qualitative conversational analyses. Results of these varied methodological approaches have converged toward demonstrating that pragmatics represents an area of particular difficulty for people with WS across the lifespan. What may account for these pragmatic difficulties is then discussed in light of developmental findings about the early socio-communicative behaviors shown by infants and young children with WS. The final sections of the chapter address the clinical implications of the research findings reviewed and suggest potential directions for future research.

6.2 Williams Syndrome – A Brief Research History

The history of research into WS provides clues about why this rare neurodevelopmental disorder with a relatively well-understood genetic basis gained such prominence in theoretical debates about fundamental human capacities in cognition, language, communication and social behavior more generally. The syndrome was first described in the early 1960s by physicians in New Zealand (Williams et al., 1961) and Germany (Beuren et al., 1962), who independently noted the remarkably similar medical and cranio-facial characteristics of a group of patients with a common heart condition – supravalvular aortic stenosis – and with developmental delay/mental retardation. Several psychological commonalities were also noted in the clinical description of these cases and soon the condition was recognized as a particular developmental disorder, which eventually came to be known as Williams syndrome, or Williams-Beuren syndrome.

In their original clinical observations of six children with infantile hypercalcemia[1] described in 1964, von Arnim and Engel highlighted “an unusual command of language” as a salient feature of the children’s “psychological structure”, which appeared to be in sharp contrast to “the severe mental retardation that is invariably present” (Von Arnim & Engel, 1964, p. 367). Considering the “mental similarities” between these children, the authors stated: “Their IQ is about 40–50 but they show outstanding loquacity and a great ability to establish interpersonal contacts” (p. 376). At face value, such statements resonate strongly with the view of independence of language or social cognition from other cognitive domains, a theoretical perspective that became very influential in the 1980s after the publication of Fodor’s (1983) seminal work, *Modularity of Mind*. According to this view, language and other core knowledge domains constitute innately specified modules that operate independently and, therefore, may be selectively ‘spared’ or ‘impaired’.

Early studies of the cognitive phenotype associated with WS emphasized dissociations between domain-specific abilities (e.g. “language” and “cognition”; see

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[1] A condition involving excessive blood calcium levels.

Bellugi et al., 1988, 1992, 1994), and the syndrome started to be cited as providing evidence in support of the modularity of mind hypothesis (Clahsen & Temple, 2003; Jackendoff, 1994; Piattelli-Palmarini, 2001; Pinker, 1999). This interpretation, with its strong theoretical implications for understanding the organization of cognitive systems more generally (Anderson, 1998; Pinker, 1994, 1999), led to a surge in research on the structural aspects of language (morphology, phonology, syntax, semantics) in WS, and to assertions of 'intact language' in the face of severe intellectual disability/cognitive impairment (Bellugi et al., 1988, 1990, 1994; Rossen et al., 1996; Pinker, 1994).

Over the past two decades, however, initial claims of preserved language, independent from other domain-specific abilities (e.g. non-verbal cognition), have been replaced by a more nuanced view of uneven linguistic and cognitive profiles developing interdependently, with relative strengths and deficits both across and within domains, in the mature WS phenotype (Brock, 2007; Mervis, 1999, 2004, 2006; Mervis & Bertrand, 1997). Nevertheless, descriptions of people with WS as having 'striking language' and strong social skills alongside severe cognitive deficits persisted for decades in the literature and are still prevalent to this day in the popular media (Finn, 1991; Dobbs, 2007; Mervis & John, 2010). One consequence of this prevailing view of the WS behavioral phenotype has been a relative lack of clinical interest in targeting pragmatic language in interventions for people with WS. Moreover, in early studies, the pragmatic skills of individuals with WS were described as a particular strength, often by comparison to the significant impairments that are found almost universally among people with autism, regardless of their structural language abilities (Karmiloff-Smith et al., 1995).

Research on language in WS underwent a period of intense focus on the morphosyntactic skills of individuals with WS (Bellugi et al., 1988, 1997; Clahsen & Almazan, 1998; Levy & Bechar, 2003; Perovic & Wexler, 2007; Ring & Clahsen, 2005; Zukowski, 2004) in the hope of documenting the functional independence and modular organization of such capacities. However, initial claims of 'intact' language or 'preserved' components of language, presumed to be under genetic specification, developing independently from other non-linguistic domains, failed to be validated by empirical findings. A similar mixed picture of abilities and clear deficits, more in line with individuals' overall level of cognitive functioning than with notions of 'selective sparing', eventually emerged from research on social cognition and social perception in WS, as will be described later in the chapter.

Inconsistencies between initial and later findings about the language and social cognitive abilities of people with WS can be explained by methodological shortcomings of earlier empirical research on WS, such as small samples, with participants from a wide age range, and problematic choices of control groups or group-matching procedures, given that the majority of investigations relied on group-comparison designs (for critiques see Brock et al., 2009; Martens et al., 2008; Mervis, 2004; Mervis & Robinson, 2003). For instance, individuals with Down syndrome (DS) were often selected as a contrast group for participants with WS in studies of language abilities (Bellugi et al., 1988, 1994, 1999; Vicari et al., 2002) even though many aspects of language are particular weaknesses in people with DS, relative to their overall cognitive abilities. Many studies compared participants with WS with individuals developing typically, matching groups on mental age, which inevitably required the inclusion of much younger typically developing (TD) children as a contrast group for older participants with WS. Finally, the view of a fractionated cognitive profile with syndrome-specific 'peaks' and 'valleys' in abilities was almost entirely missing any developmental or cultural dimensions (Karmiloff-Smith et al., 1997; Paterson et al., 1999; Thomas, 2005).

Later research studies, using appropriate comparison groups, larger samples, and taking developmental processes into account, have shown that neither language (or any structural aspect of language, e.g. grammar, morphosyntax) nor social cognition or social perception proved to be 'intact' domains in WS (Karmiloff-Smith et al., 2003). In light of these findings, research interest gradually shifted toward investigating how people with WS used language for social purposes, given their distinctive profile of social engagement. Thus, the study of pragmatic language in WS had a late start, but it is growing rapidly, encompassing a variety of assessment approaches and settings. So far, studies on WS over the last almost three decades have yielded a complex cognitive and behavioral profile that shows significant heterogeneity across many domains of abilities, including pragmatics (Laws & Bishop, 2004; Porter & Coltheart, 2005). However, despite substantial individual variability, the majority of people with WS can be characterized by a distinctive profile of neurogenetic, psychological and behavioral phenotypes, outlined next.

6.3 Phenotypic Characteristics of Williams Syndrome

Williams syndrome is among several neurodevelopmental disorders of known genetic aetiology associated with a unique cognitive and social-behavioral phenotype, in which language and social communication appear to be distinctive features. While initially considered to be a very rare disorder, current estimates indicate that it occurs in approximately 1 in 7500 live births and in about 6% of individuals with intellectual disability of genetic aetiology (StrØmme et al., 2002). The syndrome's genetic basis is a hemizygous contiguous deletion of approximately 28 genes in chromosome band 7q11.23[2] (Hillier et al., 2003; Osborne, 2006). The deletion region includes one copy of the ELASTIN gene and elastin deficiency is considered to account for some of the vascular and connective tissue abnormalities associated with WS. The condition can be diagnosed at or shortly after birth by using a fluorescence *in situ* hybridization probe for the missing ELASTIN gene, a technique which has been available since the 1990s.

A characteristic set of physical, medical and behavioral attributes commonly seen in people with WS include connective tissue and cardiovascular abnormalities

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[2] The deletion refers to loss of one of the two copies of a segment of the long arm of chromosome 7, which includes a number of adjacent missing genes.

(Ewart et al., 1993), distinctive craniofacial features, infantile hypercalcemia, sensory modulation problems such as hypersensitivity to sound, failure to thrive or growth deficiencies, and premature aging (Morris, 2006, 2010). The majority of individuals with WS have intellectual disability in the mild to moderate range (mean full-scale IQ between 50–60), although there is wide variability in intellectual functioning within the population (Mervis & John, 2010; Porter & Coltheart, 2005). On standardized assessments of cognitive functioning, verbal ability scores tend to be better than non-verbal (performance) IQ scores, although the discrepancy is not universal, and it depends in part on the type of test administered (Jarrold et al., 1998; Martens et al., 2008; Mervis & John, 2010). However, global assessments of cognitive functioning may mask a checkered pattern of markedly uneven abilities within particular domains. Initial research on WS highlighted severe deficits in spatial cognition, number processing and problem-solving alongside apparent strengths in language, face processing and social cognition (Donnai & Karmiloff-Smith, 2000; Mervis et al., 2000).

A distinctive personality and social-behavioral profile, which has been consistently described as ‘hypersocial’ (Jones et al., 2000; Järvinen et al., 2013; Porter et al., 2007) with an ‘undercurrent of anxiety’ (Mervis & Klein-Tasman, 2000), is demonstrated by both children and adults with WS. At all ages people with WS show an exaggerated ‘affiliative drive’ manifested in unusually friendly, affectionate, outgoing, gregarious, and empathic behavior (Doyle et al., 2004; Fidler et al., 2007; Gosch & Pankau, 1997; Järvinen-Pasley et al., 2010; Klein-Tasman & Mervis, 2003; Klein-Tasman et al., 2011; Plesa Skwerer & Tager-Flusberg, 2016). Despite these characteristics, individuals with WS have significant difficulties in social functioning, in forming and maintaining friendships, and often develop high levels of anxiety and social isolation as they age (Davies et al., 1998; Einfeld et al., 2001; Elison et al., 2010).

6.3.1 *Language Profile in Williams Syndrome*

Pragmatic language abilities emerge at the intersection of linguistic and social-cognitive skills. Both language and social cognition have been extensively studied in WS (see Brock, 2007; Brock et al., 2009; Järvinen et al., 2013; Mervis & Becerra, 2007; Plesa Skwerer & Tager-Flusberg, 2006, 2011 for reviews). The history of research in these domains presents a similar evolution from initial assumptions of either ‘intact’ or ‘sparing’ of abilities, to accumulating evidence of significant difficulties. Over time, the theoretical controversies that fueled continued interest in examining the language abilities of individuals with WS have shifted from debates about modularity and the independence of language from other aspects of cognition, to discussions of developmental trajectories and sources of heterogeneity in linguistic and non-linguistic communicative abilities in this disorder (Thomas & Karmiloff-Smith, 2003; Stojanovik et al., 2006; Van Herwegen et al., 2011).

To date, comprehensive characterizations of the language phenotype associated with WS have been based on analyses of performance on a variety of standardized language tests or psycholinguistic tasks, administered in the context of cross-sectional research designs (see Brock, 2007; Mervis & Becerra, 2007 for reviews). A growing body of evidence from research across several languages (see Bartke & Siegmüller, 2004) suggests that overall language skills in individuals with WS are commensurate with their mental-age levels, with certain areas of specific abilities showing better performance than expected based on non-verbal cognitive functioning (e.g. receptive vocabulary, particularly knowledge of concrete words). Within the language domain, according to Mervis and Velleman (2011), “concrete vocabulary and phonological skills are relative strengths, grammatical abilities are at the level expected for overall intellectual abilities and relational language and pragmatics are clear weaknesses” (p. 99).

From a developmental perspective, the linguistic profile of individuals with WS is characterized by considerable delay in language onset and slow development over infancy and toddlerhood (Mervis & John, 2012), yet “relatively verbose, intelligible and fluent speech in late childhood and adulthood” (Krishan et al., 2015, p. 82). By the time children with WS reach school age, their expressive language, including vocabulary, syntactic forms and fluency appear to be relative strengths (Mervis & Becerra, 2007). However, these advances in structural language do not lead to adequate conversational and discourse abilities, despite the appearance of speech proficiency demonstrated in various contexts by most school-age children, adolescents and adults with WS. Using language efficiently in social contexts involves not just the availability of a well-developed linguistic system (e.g. varied lexicon, ability to comprehend and produce complex syntactic forms), but more importantly, it requires the ability to attribute mental states to the people involved in the communicative interaction, an aspect of social understanding to which I turn next.

6.3.2 Social Cognition in Williams Syndrome

For communicative exchanges to be successful, both the speaker and the hearer need to rely on a set of socio-cognitive skills commonly referred to as ‘mentalizing abilities’ or ‘theory of mind’ (ToM). These include the ability to infer the communicative intent of a speaker, based on attributing mental states to the communication partners, the ability to monitor the knowledge state and informational needs of the speaker and hearer, and the ability to perceive and interpret non-verbal cues such as facial expressions, eye gaze, and gesture to aid in the interpretation of verbal messages and of other communicative exchanges.[3] Impairments in any aspect

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[3] Having knowledge about the social and conversational rules that apply to verbal interactions in one’s culture, as well as other skills related to efficient use of contextual information, attention monitoring, planning, and the abilities usually subsumed under the label ‘executive function’ are

of these competencies may compromise the pragmatic processes that underlie the effective use of language in social contexts. Strong relations between ToM and social communication skills have been reported both for TD children (Astington, 1990; Tomasello, 1995) and for a range of clinical populations (Abbeduto et al., 2004; Hale & Tager-Flusberg, 2005; Losh et al., 2012; see Cummings, 2013 for a review).

Initially, based on their heightened motivation for social engagement and relatively good language skills, it was hypothesized that people with WS would have a good understanding of the social world, showing domain-specific sparing in social cognition or ToM (Karmiloff-Smith et al., 1995; Tager-Flusberg et al., 1998). One early study (Karmiloff-Smith et al., 1995) that included older individuals with WS (ranging from 9 to 23 years) reported that the majority of them passed standard first-order false belief and higher-order, ToM-related tasks, which are often failed by individuals with autism (based on prior studies). The authors concluded that ToM might be an "islet of preserved ability" in WS (p. 202). However, given the age of the individuals with WS tested and the lack of a matched, non-autistic comparison group (Brock et al., 2009; John et al., 2009), this interpretation remains problematic.

In fact, later studies, based on age-appropriate ToM tasks, and including appropriate control groups, found that the performance of children and adolescents with WS on ToM tasks was similar to that of participants with intellectual disability (ID) matched on age and IQ. In a series of studies probing systematically different types of mentalizing abilities, Tager-Flusberg and Sullivan (2000) found that participants with WS did not perform any better than a group of individuals with Prader-Willi syndrome (PWS) and a group of participants with non-specific aetiology of ID matched on age, IQ, and standardized language measures to the WS group. This was demonstrated on three different first-order false belief tasks, on second-order belief reasoning (Sullivan & Tager-Flusberg, 1999), on distinguishing between lies and jokes (Sullivan et al., 2003), and on using trait information to attribute intentionality (Plesa Skwerer et al., 2006). Similar findings have been reported when non-verbal ToM tasks, using a picture sequencing method, were used to assess understanding of pretence, intention or false belief (Porter et al., 2007; Santos & Deruelle, 2009). Thus, across a variety of studies and task formats (language-based or non-verbal ToM tasks), the performance of individuals with WS in social reasoning was no better than predicted by mental age.

While findings for tasks of social reasoning have been generally consistent across studies, there is less agreement about the social perception abilities of people with WS (Hepburn et al., 2011; Plesa Skwerer, 2017). Once more, researchers hypothesized that WS would be a paradigmatic case illustrating a dissociation between 'social cognitive and social perceptual components of theory of mind' (Tager-Flusberg & Sullivan, 2000). Based on the apparent emotional sensitivity of people with WS, it was assumed that, while having difficulties with inferential

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also critical in communication processes. However, given the paucity of research on WS addressing these areas, the discussion of findings presented in this chapter remains focused on the contributions of language and theory of mind-related abilities to pragmatic functioning in WS.

aspects of social information processing, individuals with WS would nevertheless show ‘sparing’ of social perception abilities, such as being proficient at recognizing facial and vocal expressions of emotion, skills important for social communication. Experimental studies, however, revealed that on explicit measures of emotion recognition from faces or voices, children and adults with WS performed no better than comparison groups matched on mental age on tasks of discriminating, matching, or labeling expressions of emotion (Gagliardi et al., 2003; Plesa Skwerer et al., 2006; Porter et al., 2007).

Overall, evidence from a growing number of studies involving different methodological approaches indicates that the ability to decode mental state information at the perceptual or at the cognitive, inferential level of mentalizing is impaired in WS. Such impairments, together with the distinctive language and personality profile characteristic of people with WS, likely shape their use of language in context as a social communicative tool. The following sections of the chapter provide a description of the features of pragmatic language that have been examined so far in individuals with WS, organized by the type of assessment or methodological approach used to evaluate pragmatic language in WS.

6.4 Assessing Pragmatic Language Abilities in Williams Syndrome

Pragmatic skills in children, adolescents and adults with WS have been assessed mostly in the context of cross-sectional research designs, involving groups of TD participants and several different clinical populations, including Down syndrome (DS), autism spectrum disorders (ASD) and specific language impairment (SLI). More recently, researchers have started to use longitudinal and developmental trajectory approaches to investigate syndrome-specific features of pragmatic development (John et al., 2012; Thomas et al., 2010) but these types of studies are still rare in the literature on WS. Relevant findings based on each type of methodological approach used in research so far are described next.

6.4.1 Caregiver Report-Based Measures

One of the assessment instruments commonly used to evaluate children’s pragmatic abilities based on caregiver report is the Children’s Communication Checklist (CCC; Bishop, 1998 and CCC-2; Bishop, 2003). The checklist includes 10 subscales, of which four comprise items directly related to pragmatic language behaviors (i.e. Inappropriate Initiation, Stereotyped Language, Use of Context, and Nonverbal Communication). The rest of the subscales comprise items related to structural language (four subscales) and autism symptomatology (two scales – Social Relations and Interests). Based on this parent-rated assessment, Laws and Bishop (2004) found evidence of significant communication impairment in a sample of 19 children and young adults (aged 6–25 years) with WS relative to TD, DS and SLI control groups. In their study, 79% of the participants with WS scored in the range considered to be indicative of pragmatic impairment, compared to 50% of the participants with DS and 41% of the children with SLI. The WS group differed from the TD controls in all areas of pragmatic competence covered by the subscales – inappropriate initiation of conversation, coherence, stereotyped conversation, use of context, and development of conversational rapport – and scored worse than the two clinical groups in two domains: the use of stereotyped language and inappropriate initiation of conversation.

Using the revised version of the same instrument (CCC-2; Bishop, 2003), Philofsky et al. (2007) investigated the pragmatic language profiles of school-age children with WS and children with ASD, developmental disorders that are often considered to exemplify contrasting social phenotypes. When compared to parent ratings of the abilities of TD school-age children in the control group, both clinical groups showed communication impairment. Overall, the WS and ASD groups did not differ from each other on the General Communication Composite summary score (designed to identify communication impairment that could be based on either structural or pragmatic language deficits, or both). However, when considering the purely pragmatic subscales of the CCC-2, the children with WS appeared less impaired than the ASD group on Coherence, Stereotyped Language, Nonverbal Communication and Social Relations subscales, although no significant group differences were found on Inappropriate Initiation, Use of Context, and Interests subscales.

An examination of the item-level ratings on the CCC-2 showed that the WS children were considered by their caregivers as more skilled at use and understanding of affective expressions, prosody, learned phrases, social responsiveness, empathy and social relatedness with others, suggesting relative strengths in some areas of pragmatics compared to the children with ASD. These findings provide a complex picture of similarities and subtle differences in various aspects of pragmatic functioning across developmental disorders, pointing to types of communication impairments and strengths that may be syndrome-specific. Further research, using a combination of assessment measures and contexts, is needed to be able to determine whether particular features of pragmatic language differentiate individuals with WS from other neurodevelopmental disorders, while accounting for general cognition as well as for structural language level (Martin et al., 2017).

6.4.2 Standardized Direct Assessments of Pragmatic Skills

Few studies have tested directly the pragmatic abilities of people with WS using individually-administered standardized tests of pragmatic language. These types of instruments usually require the responder to make judgments about social situations based on brief stories accompanied by pictures, or pictures alone. One standardized test that has been given to children and adolescents with WS is the Test of Pragmatic Language-2 (TOPL-2; Phelps-Terasaki & Phelps-Gunn, 1992, 2007), which probes pragmatic reasoning in several domains relevant to successful communication (e.g. abilities to appraise and monitor physical context, audience, topic, visual-gestural cues, purpose/speech acts and abstraction). Hoffmann et al. (2013) compared directly the performance of school-age children and adolescents with WS (aged 8–16 years) on the TOPL-2 with parent ratings of the same participants' communication abilities on the CCC-2 (Bishop, 2003). While there were no significant differences in terms of overall scores between the TOPL-2 and the CCC-2, the two types of assessment yielded different results with respect to the number of participants each classified as having pragmatic language impairment (PLI). In particular, the TOPL-2 identified significantly more participants with WS as meeting cut-off for PLI than did the CCC-2 instrument (70% vs. 30%, respectively).

This pattern of results was in direct contrast to findings reported by Volden and Phillips (2010) for a group of children with ASD who had age-appropriate structural language skills. Using the same instruments, they found that the CCC-2 classified significantly more of the children with ASD as having PLI than did the original TOPL (Phelps-Terasaki & Phelps-Gunn, 1992) assessment (81% versus 56%, respectively). Whether the reason for the discrepant findings in the two studies is related to syndrome-specific aspects of pragmatic impairment or to other factors remains unclear. The TOPL-2, a test of a participant's ability to "view a social situation as an objective bystander" (Hoffmann et al., 2013, p. 200), draws on meta-pragmatic skills, and may rely more heavily on social cognitive rather than linguistic abilities, which may have rendered the test more challenging for the cognitively impaired children and adolescents with WS.

Given the differences in the rate of PLI detected among the WS participants by the two assessment instruments, it is critical that future studies of pragmatic skills involve multiple methods and measures in order to provide a comprehensive evaluation of pragmatic functioning in this population. Evaluations that include more ecologically valid contexts of assessment, based on quantitative and qualitative analyses of real-life interactions with different types of communication partners, both familiar and unfamiliar, should complement standardized and caregiver-report measures. Semi-structured conversations with researchers and narrative elicitation tasks have already provided a wealth of information about the distinctive communication style exhibited by people with WS, while revealing a checkered picture of strengths and weaknesses in their socio-communicative skills.

6.4.3 Elicited Narratives and Conversation

Early reports of unusual language proficiency in WS were based on comparing narratives produced by a small group of adolescents with WS to those of two comparison groups: an age-and IQ-matched group of adolescents with DS and a group of younger, mental age-matched TD children. In the first study to use a wordless picture book – *Frog, Where Are You?* (Mayer, 1969) – to elicit narratives from four adolescents (aged 10–18 years) with WS, Reilly et al. (1990) reported that the WS participants showed more prosodic features of speech (e.g. instances of pitch changes, vocalic lengthening, modifications in volume) than did either comparison group, and produced more evaluative language (including references to the emotional and mental states of story characters, character speech or sound effects, exclamatory phrases, and emphatic markers meant to capture the attention of the listener) than did the DS group.

Follow-up studies using the same picture book (Losh et al., 2000; Reilly et al., 2004) compared larger groups of school-age children with WS to age-matched groups of TD children and to children with specific language impairment (SLI). These studies found a similar abundant use of evaluative language in the narratives produced by the children with WS relative to either the SLI or the TD groups. However, the children with WS made significantly more grammatical errors than the TD group, and their grammatical performance was no better than that of the children with SLI. An analysis of the type of evaluative devices used by the children with WS indicated a higher prevalence of ‘social engagement device’ (e.g. sound effects, character speech, and “audience hookers”), but fewer cognitive inferences than in the narratives of the comparison groups.

Several researchers have reported a lower use of linguistic devices that contribute to narrative coherence and cohesion (cohesive ties, grammatical markers, and complex syntactic structures) in stories told by children with WS, relative to both TD and SLI comparison groups, even when individuals with WS produced longer narratives overall. On narrative measures that tap cognitive inferencing skills, such as story structure and integrating themes, children with WS usually scored significantly lower than comparison groups across narrative elicitation studies in several languages (Diez-Itza et al., 2018; Lacroix et al., 2007; Lorusso et al., 2007; Losh et al., 2000; Stojanovik et al., 2004). These findings suggest that the excessive use of social engagement devices in narratives, which has been observed across different languages and cultures, including English, French, Spanish, Italian, Dutch and Portuguese (Reilly et al., 2005; Gonçalves et al., 2010; Jones et al., 2000; Lacroix et al., 2007; Van Den Heuvel et al., 2016), may mask difficulties with both macrostructural and cognitive aspects of narrative performance in WS, while reflecting the most enduring characteristic of the children, adolescents and adults with this syndrome – their heightened sociability (Losh et al., 2000; Järvinen et al., 2013). The contrast between the strong desire to engage an audience and the ability to do so competently is also reflected in the tendency of individuals with WS to use an abundance of affective prosody and social evaluation even in the second story-telling instance to the same listener, or when not being paid attention to, which suggests a lack of pragmatic sensitivity, because the speaker with WS does not take into account the state of the listener (Järvinen-Pasley et al., 2008).

The strong motivation to keep a social interaction going and to capture the attention of a conversational partner seems to shape the speech style exhibited by people with WS across a variety of situations and often irrespective of their familiarity with the interlocutor. People with WS tend to engage conversationally with strangers just as readily as with people they know very well, and rarely adjust their style of speech based on what they know about the person they are speaking with (Järvinen-Pasley et al., 2010; Jawaid et al., 2012; Jones et al., 2000). For instance, several researchers provided anecdotal reports of participants with WS 'turning the tables' on the experimenter during interviews and asking personal questions, often accompanied by poor turn-taking and topic maintenance (Jones et al., 2000; Harrison et al., 1995; Semel & Rosner, 2003; Stojanovik et al., 2001). Udwin and Yule (1990) remarked on the conversational inadequacies demonstrated by a significant proportion of school-age children with WS in naturalistic interactions. Based on 30 min of conversation with a researcher, they found that 37% of the children with WS in their study met criteria for hyperverbal speech (also referred to as 'cocktail party speech'). This involves fluent speech with an excessive use of stereotyped phrases, an overfamiliar manner, introduction of irrelevant personal experiences and perseverative responding.

Several other studies included qualitative analyses of dyadic conversational interactions conducted mainly with researchers as the conversational partner. It should be noted that these studies were either case-reports or included a small number (4–12) of participants with WS. Stojanovik (2006) used semi-structured conversations around photographs depicting everyday situations to probe the abilities of five school-age children with WS to talk about their own experiences related to the topic, and to respond to the researcher's questions. Compared to a group of children with SLI matched on receptive vocabulary and grammatical ability, and to a group of TD peers, the children with WS were less likely to give adequate responses to the interlocutor's request for information or clarification, or to produce responses that would advance the conversation, even though they were likely to produce extended responses (e.g. more than yes/no replies). However, significantly more of their utterances were inadequately informative, providing too little information or misinterpreting what the speaker meant, and tending to overrely on the conversational partner's lead.

Similar findings have been reported for French-speaking children with WS observed in parent-child interactions or in conversation with an examiner. Lacroix et al. (2007) found that, compared to TD controls, their participants with WS showed less turn-taking in conversations, more non-contingent responding, and provided less information in response to an adult's request for clarification. While these studies provide generally consistent descriptions of the deficits shown by people with WS in conversation across several languages, the predictors and correlates of the pragmatic language difficulties reported remain to be systematically investigated.

Recently, Rossi and Giacheti (2017) examined associations between speech-language, general cognitive functioning (IQ) and behavior problems in a sample of Brazilian children and adolescents with WS (aged 8–18 years). Besides administering tests of receptive vocabulary, syntactic comprehension, IQ, and a parent questionnaire involving ratings of behavioral problems (Child Behavior Checklist-CBCL; Bordin et al., 2001, 2013), the authors analyzed 30-minute conversations with a researcher, which were coded for five speech-language characteristics considered to be indicative of pragmatic difficulties: clichés; echolalia; perseverative speech; exaggerated prosody; and monotone speech. They reported that 61.5% of the participants with WS showed perseverative speech in conversation, while 50% used exaggerated prosody, 42.3% used clichés, and 19.2% used echolalia during the conversation task. On the CBCL questionnaire, 73.8% of the participants were rated by their parents as 'talking too much'. The two types of repetitive verbal behavior coded during conversation – perseverative speech and echolalia – were negatively correlated with performance on standardized assessments of language, while exaggerated prosody use in conversation was associated with higher ratings of problem behaviors on the CBCL (Total problem scale).

These associations between lower language performance, increased reliance on prosody and paralinguistic devices in conversation, and behavioral dysregulation in individuals with WS may provide insight into why so many people with WS experience serious difficulties in forming and maintaining social relationships, especially with peers, despite their strong need for social contact and relatively proficient expressive language. Hargrove et al. (2013) analyzed spontaneous conversations between adolescents with WS and age-matched TD peers with respect to their use of paralinguistic features such as laughter, sound effects, fillers and repetitions/reformulations. While the TD peers produced significantly more fillers and reformulations than the adolescents with WS did, the WS group used an abundance of sound effects and laughter, which may be used to mask difficulties with the content of their conversations: while superficially engaging, their contributions to topic maintenance were largely irrelevant. As these authors commented, 'What may be engaging or "cute" in younger children could be viewed as awkward or immature by adolescent communicative partners', and may end up 'isolating the adolescents with WS from peers rather than engaging them' (p. 157).

In summary, at all ages, people with WS tend to engage in conversation readily and enthusiastically, but often lack the tools to initiate and maintain a meaningful conversational exchange. In a variety of discourse contexts, such as storytelling, picture descriptions, and biographical interviews involving questions about participants' interests, family, or everyday activities, children and adolescents with WS were more likely than control groups to use more adult vocabulary, social phrases and an overly-familiar conversational style. Their exaggerated use of prosodic and paralinguistic features of speech, instead of reflecting expressive communication competence, may serve mainly a social function: this speech style appears to be an attempt to compensate for the inability to contribute meaningfully to the substance of a conversation, while trying to hold the audience engaged in the interaction. Over time, this characteristic conversational style may have the opposite effect of that intended by the speaker with WS, and might, in fact, contribute to the serious difficulties that people with WS have with developing friendships and meaningful social relationships, especially as they age.

6.4.4 Experimental and Laboratory-Based Tasks

Recently, researchers have started to use experimental tasks to deconstruct the complex pattern of pragmatic deficits demonstrated by people with WS across naturalistic discourse contexts. Such tasks enable investigators to focus on discrete pragmatic skills (e.g. signaling non-comprehension of a message, providing information to clarify a message or conversational repair skills) and may lead to identifying particular sources of pragmatic deficits and strengths. So far, two areas of pragmatic competence have been probed experimentally in individuals with WS: referential communication and interpretation of non-literal language.

In the first study to examine the referential communication abilities of children with WS, John et al. (2009) focused on the listener's role. They investigated whether 6- to 12-year-old children with WS were able to recognize and verbalize the referential problem when a communication partner's messages were inadequate. In the experimental setting used (modeled after Abbeduto et al., 2008), the child and the experimenter were separated by an opaque barrier, and the child was required to identify a referent (a picture) from an array of items based on the information conveyed by the speaker. The researchers examined children's verbal responses to messages that were ambiguous, or included a word the child did not understand, or conveyed an impossible action because the requested item was not available.

The children with WS indicated that there was a problem with the speaker's message less than half the time, and they had difficulty verbalizing the nature of the referential problems encountered. The type of message inadequacy influenced their responding. They performed better in the 'impossible condition', reporting the problem 55% of the time, whereas they communicated a problem with an ambiguous message or with a message containing an unknown word only about 20% of the time. Children's performance on false-belief tasks and age were significant predictors of the likelihood and effectiveness of their verbalizing that a speaker's message was inadequate in the more difficult conditions (ambiguous and unknown word). The strong relationship found in this study between theory of mind and listener-role referential communication skills in children with WS is consistent with findings from research on TD children and on individuals with other neurodevelopmental disorders (Abbeduto et al., 2004; Losh et al., 2012; see Matthews et al., 2018 for a review), adding to the evidence of close connections between pragmatic skills and social cognitive skills across typical and atypical development.

Asada et al. (2010a) focused on the 'speaker role' in a study involving an object-choice situation used to elicit verbal requests and corrections/reformulations from the children engaged in the task. These researchers examined the communication repair skills of 4- to 11- year-old children with WS relative to those of MA-matched TD children. After the children indicated their choice of a preferred object, the experimenter proceeded to give them verbal feedback (i.e. whether the child's choice was understood or misunderstood) and one of the objects (i.e. the desired or undesired object). Thus, four conditions ensued from the combination of the experimenter's verbal feedback and object-giving action. While the overall frequency of verbalizations across the four conditions was similar across groups, children with WS produced fewer requests and rejections of the wrong object than did the TD group, particularly when they were verbally misunderstood. In contrast to the TD children who increased their corrections in the verbal misunderstanding condition compared to the correct understanding condition, the children with WS did not vary their verbalizations for clarification based on condition. Their behavior suggested that they often failed to take into account the perspective of the interaction partner, even when it was explicitly mentioned verbally. These results indicate that children with WS have difficulty in using communication repair skills, such as providing verbal corrections in order to share what they meant with others, to establish mutual understanding.

In another study based on a modified referential communication paradigm, Plesa Skwerer et al. (2013) probed experimentally the ability of children with WS to monitor the informational adequacy of both the communication partner's message and of the children's own verbal messages. The researchers used a collaborative game format, in which a child and a researcher took turns in placing small objects on a large mat illustrated with their pictures to 'build' a toy-size 'farm' or 'wildlife park'. The game partners had to indicate to each other the objects they needed help with (e.g. for those that were outside their reaching space), and to collaborate in the selection of the items. Some items were of the same identity but distinguishable by a relevant attribute (e.g. white bear and brown bear) while others were unique. When in the 'builder'/speaker role, the researcher sometimes expressed insufficiently informative indirect requests for items of the same type (e.g. "I think I need the bear now") directed toward the 'helper'/hearer.

The children with WS (ranging in age from 5 to 13 years) performed significantly worse than a group of TD children matched for chronological age (CA), but similarly to a group of TD controls matched for verbal mental age (VMA) on quantitative measures of pragmatic comprehension (i.e. number of clarification requests) when in the 'helper'/hearer role. However, they were less likely than both comparison groups to take into account the available visual information about the objects (i.e. to look both in the partner's and in their own space) before interpreting the intended referent of the partner's request. When they *did* realize that the speaker provided insufficient information to help them distinguish between objects of the same type, the children with WS were more likely to use pointing or picking up and showing one of the objects than to verbalize their request for clarification. In contrast to this reliance on gesture and guessing, the TD children showed a developmental trend toward using increasingly definite verbal questions to obtain the needed information. Another tactic used by the children with WS when confused about the partner's message was to comment or ask personal questions unrelated to the collaborative game. This exchange illustrates such a common situation. The experimenter makes an indirect and ambiguous request for a non-unique item (there are two toy dogs similar in shape and color but different in size):

> Experimenter: I need the dog now…
> Child with WS: (after a brief pause, looking at E's hand) Oh, I know what your ring means. You are married, right?

When the participant played the role of the 'builder'/speaker, the children with WS provided fewer adequately informative referents (i.e. mentioning the distinguishing feature of a non-unique item) than both the CA and the VMA controls did, and significantly fewer of them provided adequately informative referents on all the relevant trials (Ammerman, 2013). However, as the game progressed, learning effects were observed for the WS children, who did not differ from either comparison group in conversational repair attempts (offering appropriate information when notified that the listener needs more information). As the game continued, the children with WS were just as likely as the control groups to improve their responding to the feedback provided by the game partner, and to repair communication breakdowns, especially after the partner modeled a clarification response. This finding suggests that scaffolding could have beneficial effects for children with WS, who appear to be able to learn from specific feedback and improve their communication strategies during social interaction.

Longitudinal studies are needed to determine whether such gains in communication adequacy remain temporary and are circumscribed to the context of the particular interaction or may endure and contribute to lasting improvements in pragmatic functioning. So far, only one study has examined longitudinal relations between aspects of pragmatic behavior demonstrated by 4-year-old children with WS during a play session with their mother, and their conversational ability about 6 years later. John et al. (2012) found that the ability to verbally contribute new information within a social interaction at age 4 years showed stability from preschool to primary school age in children with WS. Differences in this pragmatic skill at school age were predicted by children's ability to pair verbalizations with eye contact in triadic interactions (secondary intersubjectivity) as preschoolers. When taken together with the previously mentioned findings about the potential role of scaffolding for improving communicative strategies in WS, these longitudinal findings underscore the importance of establishing pragmatic language outcomes as intervention targets for young children with WS, to maximize their beneficial effect for acquiring and consolidating pragmatic skills in this population.

For effective communication speakers are also required to consider the attentional focus of their interlocutor, to ensure that their message is processed as intended. Typically developing children by age two are able to track speakers' intentions and to take into account what they attend to, or what they know in a particular situation (O'Neill, 1996; Moll & Tomasello, 2006). Asada et al. (2010b) investigated the ability of children with WS (mean CA = 10; 2 years) to evaluate the attentional focus of a partner in communication and to modify their verbal message accordingly with a view to sharing information. The children in their study were asked to complete simple actions with toys while the experimenter either attended to them or did not pay attention to them during the time they accomplished the task.

While a comparison group of TD children who were individually matched on vocabulary age to the participants with WS verbalized more about their accomplishment when they were not attended to than when they were attended to, the children with WS showed the opposite pattern, meaning that their verbalizations did not take into account the attentional state of the listener. The authors interpreted the verbal behavior of the children with WS as a violation of the pragmatic rule "to speak the appropriate amount according to other's state" (Asada et al., 2010b, p. 456). They suggested several possible explanations for the atypical verbal communication pattern demonstrated by the children with WS, including possible impairment in the ability to direct or share attention to something (i.e. declarative function deficit), or impaired understanding of the 'seeing-leads-to-knowing' principle (socio-cognitive deficit), or higher motivation to interact verbally when they are attended to, regardless of the goal of the communication. More research is needed to disentangle the potential contribution of each of these factors to pragmatic deficits, or to clarify how their combination may impact the quality of communication efforts in people with WS.

In summary, across different types of referential communication tasks, it appears that children with WS have difficulties with a set of processes involved in efficient communication, including evaluating the informational adequacy of messages, requesting and providing verbal corrections or clarifications when needed, understanding the attentional focus and state of the hearer and, more generally, evaluating and taking into consideration the perspective of another during a social-communicative interaction. This promising line of research should be continued with investigations of whether and how each of these discrete pragmatic behaviors and processes may be turned into targets for interventions aimed at improving pragmatic functioning in people with WS.

6.4.5 Non-literal Language Comprehension and Production

Comprehending and using figurative language are important aspects of pragmatic competence because they involve the ability to distinguish between intended meaning and the 'surface' expression (literal meaning) in the process of utterance interpretation. This process can be challenging as it usually relies on background knowledge and the ability to draw links and find similarities between often conceptually disparate domains (Keil, 1986). Not surprisingly, comprehension and production of figurative language represent late achievements in typical development, continuing to progress throughout childhood (see Falkum, 2019 for a review). Several studies have examined comprehension of non-literal language in WS, including metaphors and metonyms (Annaz et al., 2009; Van Herwegen et al., 2013), perceptual simile (Thomas et al., 2010), idiomatic expressions (Mervis et al., 2003; Lacroix et al., 2010), irony (Sullivan et al., 2003), jokes (Krishan et al., 2017;

Sullivan et al., 2003) and sarcasm (Karmiloff-Smith et al., 1995; Godbee & Porter, 2013).

The majority of evidence from these studies points to significant delays shown by individuals with WS in interpreting language in context when the intended meaning differs from the literal meaning. Some researchers have suggested that comprehension of different forms of non-literal language by individuals with WS follows an atypical developmental trajectory. Across several studies, it was found that comprehension of metaphor and novel metonymy was not only delayed but did not increase with increasing chronological age in WS, in contrast to the pattern of performance of the TD participants (Annaz et al., 2009; Thomas et al., 2010; Van Herwegen et al., 2013). This seems at odds with reports of increasingly rich expressive language used by individuals with WS as they age. As will be discussed later, many individuals with WS do *produce* figurative language, especially as they increase their vocabulary knowledge, but it is possible that they do so without understanding its meaning (Bertrand et al., 1994; Thomas et al., 2010).

Being able to interpret appropriately the communicative intent implied in various forms of non-literal language such as jokes, ironic statements, lies and sarcastic remarks is important for relating to peers in everyday discourse. Sullivan et al. (2003) tested the ability of adolescents with WS to distinguish between lies and ironic jokes, using a series of short stories in which a character's final statement was false, but the communicative intent was either deception/lying or irony/joking. Adolescents with WS and two comparison groups, one of adolescents with PWS, the other of adolescents with non-specific aetiology of ID, matched on age, IQ and verbal abilities to the WS group, were asked to classify the characters' statements as lies or jokes, and to justify their responses. While almost all of the participants in the three groups were unable to identify which intentionally false utterances were intended as ironic jokes, classifying them as lies instead, the WS group differed from controls in their justification responses. They usually referred back to the facts of the story, instead of using mental state-based explanations, as did the control groups, which suggests that social-cognitive impairments may be a major source of these adolescents' pragmatic deficits in interpreting non-literal language.

Godbee and Porter (2013) presented stories in which characters made non-literal comments that were either sarcastic in intent, or voicing a metaphor or a simile, to participants with WS (ranging in age from 5;4 years to 43;8 years) and to TD controls who were individually matched either for mental age (MA) or for chronological age (CA) with the WS participants. In this study, the individuals with WS performed worse than their CA-matched controls in explaining 'what did the story character mean' on all forms of non-literal language comprehension, but their performance was not significantly different from that of the MA-matched controls, although sarcasm comprehension was particularly poor (at floor) in the WS group.

Only one study to date has focused on humor comprehension, in relation to mental state language use, in adolescents with WS and those with DS. These adolescents were compared to two groups of TD participants, one matched on MA and another matched on CA to the clinical groups. When asked to explain 'what was funny?' about 23 humorous cartoons, both groups with intellectual disabilities obtained scores indicating lower levels of humor comprehension than the CA controls, but were not different from each other, or from the MA controls, either in humor comprehension scores or in total mental state language use. These results also indicate that the relatively better expressive language abilities of the older participants with WS compared to other individuals with intellectual disability such as DS, PWS or much younger TD participants, did not play a compensatory role in their performance on tasks of non-literal language comprehension.

Using a developmental trajectory approach, Naylor and Van Herwegen (2012) found that, during a fictional narrative task, 7–18 year-olds with WS *produced* a similar amount of figurative expressions as did a TD comparison group, an apparent contrast to the poor performance usually demonstrated on tasks of figurative language comprehension. Semel and Rosner (2003) also noted the use of idioms and figurative language by individuals with WS during conversations. What could explain this discrepancy between understanding and production of figurative language by individuals WS? Anecdotal reports as well as qualitative analyses of conversations (Udwin & Yule, 1990; Jones et al., 2000) have suggested that their use of idioms, social phrases and various forms of figurative language often appears to be somewhat inappropriate to the social context, raising doubts about their conceptual understanding of the expressions produced (Bertrand et al., 1994). Parents of individuals with WS have also reported instances of using language they clearly do not understand, which may explain the impression of unusual vocabulary (e.g. low-frequency word choices) that has been anecdotally reported to be a distinctive feature of the speech produced by adolescents and adults with WS (Bellugi et al., 1992; Rossen et al., 1996).

Thomas et al. (2010) suggested that using low-frequency words, or peppering their speech with clichés, idioms and figurative language they have previously heard and memorized, may be ways in which individuals with WS attempt to capture the attention of an ‘audience’ and to keep the social interaction going. In this case, the production of figurative language may be a pragmatic device serving social ends for individuals with WS and may not reflect the conceptual understanding required by a meaningful use of these forms of speech. In short, the findings reviewed so far strongly suggest that people with WS tend to enroll all the linguistic tools available to them in the service of social engagement, sometimes at the expense of a meaningful and socially appropriate use of verbal content in their communicative attempts.

6.5 Developmental Precursors and Correlates of Pragmatic Skills

Why is pragmatics an area of language functioning that is particularly problematic for children, adolescents and adults with WS? High interest in social engagement and relatively good structural language achieved by school age, despite a delayed onset, would be expected to sustain and bolster the development of pragmatic skills. To gain insight into the possible origins of pragmatic deficits in WS, we need to understand the developmental relations among a complex set of abilities that interact across developmental time (Karmiloff-Smith, 1998), resulting in the profile of pragmatic language functioning described in this chapter. This set of relevant skills and processes involve early socio-communicative behaviors emerging in infancy and their relationship with later developments in the structural aspects of language, as well as in social perception and social cognition, attention monitoring processes and executive functioning, behavior regulation abilities and learning processes underlying the acquisition of cultural knowledge. There has been little or no research involving individuals with WS in some of these areas, and studies of infants or young children with WS are still scarce. However, a number of key findings that have emerged from recent research could shed light on the developmental origins of the pragmatic language profile associated with this intriguing neurodevelopmental disorder.

Pragmatic language shows a protracted development in TD individuals and many skills continue to emerge through adolescence and beyond. However, a variety of pragmatic skills emerge early in life, starting with pre-verbal turn-taking around 8–9 months (Ninio & Snow, 1996) followed by a sequence of processes closely related to social interaction. In describing this sequence, Adams (2002) notes that "early social exchanges revolve around objects which are the focus of joint attention followed by rapid development of communicative acts between 14 and 32 months" (p. 975). By contrast, from early in life, the attention of infants with WS revolves almost exclusively around the people they interact with, as reflected in their atypical eye contact and limited gaze following away from the partner's face during social exchanges. In one of the first studies to focus on social interactive behaviors in WS, Mervis et al. (2003) observed that a 10-month-old girl with WS displayed unusually prolonged and intense-looking behavior toward her play partner (mother or unfamiliar adult) compared to both developmental-age and chronological-age matched TD female infants.

Similar unusual eye contact was reported in other contexts where infants, toddlers and young children with WS directed their attention almost exclusively to the people present, at the expense of sharing and coordinating attention between their social partners and surrounding objects and events (Thurman & Fisher, 2015; Mervis et al., 2003; Laws & Bishop, 2004). During semi-structured interactive assessments of early socio-communicative abilities, such as the Early Social Communication Scales (ESCS; Mundy & Hogan, 1996), toddlers with WS showed less object-related behaviors (declarative and instrumental pointing, reaching, requesting toys and response to joint attention bids) than a group of TD toddlers matched on developmental age (Laing et al., 2002). While they engaged readily in dyadic interactions and used more social interactive behaviors (requests for tickling, turn-taking behaviors, eye contact not related to objects) than the control group, the toddlers with WS showed significant impairments in triadic interactions relative to MA-matched controls. Eye contact was used more in dyadic interaction and less for social referencing or in combination with requesting or reaching behaviors by the toddlers with WS compared to controls.

This finding was corroborated by a later study involving preschoolers with WS (Thurman & Mervis, 2013), who were compared to age- and gender-matched children with DS in their social-referencing behaviors and its associated component abilities – initiating eye contact, gaze following and emotional responsivity. More specifically, Thurman and Mervis (2013) found that children with WS were less likely to initiate eye contact (unsolicited) and to follow another person's gaze in triadic situations than were children with DS, although both groups showed difficulty utilizing the communicative significance of facial expressions of fear in social-referencing processes.

Similar difficulties with both initiation of, and response to, joint attention were demonstrated by children with WS during the administration of the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 1999) – a semi-structured standardized assessment designed to elicit behaviors that are directly relevant to the diagnosis of Autism Spectrum Disorder (ASD). The ADOS involves a series of interactive activities, appropriate for a child's developmental level/language and age that create opportunities for observing and evaluating joint referencing, social relatedness, communication skills, and restricted and repetitive behaviors. Several studies in which individuals with WS were administered the ADOS (Klein-Tasman et al., 2007; Lincoln et al., 2007; Tordjman et al., 2012) indicated some overlap in their socio-communicative functioning with that of children with ASD. While the proportion of children with WS who met the cut-off for a classification of ASD differed across studies (ranging from 10% to 50%), reports of abnormalities demonstrated by children with WS in the use of gestures, declarative pointing, initiating joint attention and showing objects were consistent across studies. At the same time, compared to the children with autism, those with WS showed relative strengths in their quality of social overtures, social smiling and directing facial expressions and vocalizations to another.

Besides the prelinguistic, socio-communicative difficulties described above, children with WS have been reported to begin to talk before they begin to either point or to show objects in triadic interactions, which is an atypical developmental sequence. The use of referential language prior to the onset of communicative gesture use has been described both in longitudinal and in cross-sectional studies of infants with WS, and has been observed both by parent report and in structured laboratory settings (Laing et al., 2002; Mervis & Bertrand, 1997; Singer-Harris et al., 1997). Even preschoolers with WS demonstrate a lack of ability to use gaze-shift to infer a partner's communicative intent (John & Mervis, 2010) or to evaluate the attentional focus of a partner (Asada et al., 2010b). Researchers have pointed out that this pattern of relations between prelinguistic and linguistic developments in WS appears to be atypical not just relative to the normative trajectory mapped for TD children, but also relative to the sequence of communicative developments found in other neurodevelopmental disorders, including DS and ASD (Mervis & John, 2010). The impact of this atypical course of communicative development on the acquisition of pragmatic skills in WS remains to be investigated systematically.

In summary, the pragmatic difficulties demonstrated by individuals with WS are likely to be rooted in the delays shown by young children with WS in establishing joint attention and secondary intersubjectivity during social interactions. Such difficulties and delays in the use of eye gaze, gesture and directing and sharing attention around objects and events, in combination with the failure to use communicative gestures to express intentions prior to the onset of language, are likely to trigger cascading effects on later socio-communicative developments and, in particular, on the trajectory of pragmatic skill acquisition in WS.

6.6 Clinical Implications

As noted at the beginning of this chapter, early research reports on individuals with WS emphasized their 'striking language proficiency' despite their cognitive deficits, so it is not surprising that the need for interventions targeting the use of language in social contexts by people with WS was not fully recognized until fairly recently. Even though teacher- and parent-report descriptions of 'incessant chatter', 'an old fashioned and formal style of speech, including the use of stock phrases' (Udwin et al., 1987, p. 306), 'poor turn-taking and topic maintenance, inappropriate responses, repetitive phrases and hyperverbalization' (Meyerson & Frank, 1987, p. 260) have been noted in some of the earliest published studies about WS, clinicians and speech therapists rarely prioritized addressing these speech peculiarities until recently, when their impact on the social-adaptive functioning of individuals with WS has started to be recognized and documented (Howlin et al., 2010).

Pragmatic abnormalities often have negative consequences on social skills and interpersonal relationships, interfering with the ability of individuals with WS to engage with peers and to participate in age-appropriate social activities, and may even put them at risk for social victimization (Elison et al., 2010; Jawaid et al., 2012). Understanding and taking into account how pragmatic deficits impact the adaptive skills of individuals with WS is a necessary step toward establishing targeted goals for intervention. Because recent research has uncovered significant heterogeneity in abilities among individuals with WS, despite the many common strengths and challenges discussed so far, it is possible that different factors, learning and social experiences may differentially contribute to progress in pragmatic language development for different children with WS. As Mervis and Velleman (2011) suggest, to determine the intervention needs of a child with WS, the child needs to be carefully observed in interaction with caregivers, teachers and other children in a variety of settings, in addition to conducting formal assessments of the child's language level.

This is important for several reasons. From an intervention-planning standpoint, it is critical to take into account the atypical developmental sequence between the onset of referential communicative gestures and referential expressive language in WS. It may not be the case that once a child with WS is already talking, "basic referential gestures have been mastered” (Mervis & Velleman, 2011, p. 100), as is the case in typical development. If this misleading assumption is made, early intervention planning may miss opportunities to address some of the non-verbal skills (e.g. ability to establish triadic joint attention, comprehension and production of communicative gestures) that are essential for the development of pragmatic language competence. Also misleading may be the fluency and good articulation that many children with WS demonstrate once they have acquired basic expressive vocabulary, because these verbal characteristics are sometimes taken to signal that speech/language therapy is no longer needed. Such verbal strengths may in fact mask serious difficulties with the pragmatic aspects of communication, which can persist into school years and later in life, and are likely to hinder successful functioning in the social world.

In one of the first comprehensive books published about WS, Semel and Rosner (2003) suggested several practical goals for interventions aimed at improving language pragmatics. They indicate that mediational strategies could be effective in modifying behavior such as, for instance, inappropriate greeting behaviors, which are displayed by individuals with WS “with almost every new person they meet” (p. 89), inappropriate requests for attention, persistent questioning, topic perseveration or poor turn-taking/turn-yielding. These strategies involve pointing out the social role differences of various types of people (e.g. strangers, acquaintances, family members, service people, professionals, etc.) when encountered in various contexts, and modeling appropriate behaviors (e.g. role-playing alternative ways of communicating, videotaping mock situations and having the instructor provide specific feedback, training to use “self-talk” to help restrain from compulsive greeting, etc.). Semel and Rosner (2003) suggest that interventions using “modeling, role playing, puppetry, playacting, or improvisational dramatization” (p. 94) may be especially effective, given the personality characteristics of people with WS, who tend to be dramatic in their emotional expressions and to crave social praise.

One of the most salient characteristics of the behavioral phenotype of people with WS is an openly declared, strong love for music (Thakur et al., 2018). Their affinity for music and rhythm may be used toward therapeutic goals, such as improving the quality and structure of conversational exchanges (e.g. teaching turn taking/turn yielding and the appropriate use of prosodic features of speech). The use of music in interventions targeting pragmatic language outcomes may be a particularly beneficial approach for individuals with WS. Conducting studies to evaluate systematically the efficacy of various intervention protocols aimed at improving pragmatic functioning in people with WS should be a priority for future research.

6.7 Directions for Future Research

This review of what is currently known about the pragmatic language profile of people with WS suggests several potentially interesting avenues for future research. First, identifying the predictors of progress in pragmatic language development in WS should be an important research goal because this knowledge could contribute directly to informing the design of pragmatic language intervention protocols for this population. This effort implies a systematic investigation of the developmental relationships between particular types of pragmatic processes and the trajectories of structural language, sociability, and non-verbal cognition as they interact across developmental time in individuals with WS. So far, in addition to extensive investigations of linguistic abilities in WS, research has focused mainly on socio-cognitive (ToM) and socio-perceptual processes that may directly influence the acquisition of pragmatic skills. But very little is known about how executive functions and inferencing abilities, memory processes, behavior regulation skills or the acquisition of cultural knowledge (e.g. learning about, and internalizing socially appropriate norms and rules of communication) contribute to particular aspects of pragmatic functioning in people with this intriguing syndrome.

Second, it should be noted that there are still major gaps both in the phenotypic characterization of the pragmatic profile of people with WS, and in understanding the course of development of pragmatic skills in WS. As described in this chapter, interest in pragmatic language in WS has surged in the last decade, yet many aspects of pragmatic functioning, including different types of speech acts, conversational implicatures and discourse processes remain to be investigated systematically in this population. Research on the trajectory of pragmatic skill acquisition and its complex relations with other features of cognitive and behavioral functioning in WS is extremely limited. Therefore, longitudinal studies with larger samples or studies based on cross-syndrome comparisons of developmental trajectories of particular abilities are much needed in the field of neurodevelopmental disorders research. Such studies are critical for identifying syndrome-specific phenomena, including potential specificity in the social uses of language, and may open avenues for eventually linking genetic abnormalities to brain development and to behavioral outcomes.

Although recently researchers have acknowledged the substantial individual variability within domains of skills found in the WS population, little is known about possible gender differences in pragmatic language in WS. A similar discussion of potential cultural differences in the pragmatic profiles of people with WS is timely. Previous cross-cultural research on perceived sociability and on narrative production in children with WS has revealed a rich set of similarities and differences in phenotypic profiles in WS across languages and cultures (Zitzer-Comfort et al., 2007; Reilly et al., 2005). Conducting cross-cultural research using a multitude of assessment instruments, as well as through naturalistic observations in a variety of different contexts, could be a particularly useful avenue for distinguishing between syndrome-specific and cultural/linguistic influences on pragmatic functioning in people with WS. Using multiple methods and conducting research across different settings and time points could provide valuable insights not only for understanding the development of pragmatic language and social communication in WS, but also for designing and timing appropriately the implementation of interventions, in order to maximize their beneficial effects for the adaptive, social and even emotional functioning of people with WS.

6.8 Summary

Despite its relatively short history, research on pragmatic language skills in WS has addressed a number of important pragmatic concepts, including communicative intentions, reference resolution, informational adequacy of messages and clarification requests, conversational repair strategies, non-literal language, and a range of discourse functions examined in narrative tasks and in dyadic conversations. This chapter reviewed some of the key findings and methodological approaches used to investigate a number of discrete pragmatic skills and behaviors demonstrated by children, adolescents and adults with WS in several settings, including lab-based experimental tasks, narrative elicitations, conversations and dyadic interactions with an adult, standardized tests and parent-report questionnaires. Probably one of the most interesting findings of this research is the consistency with which people with WS seem to use their language abilities primarily for social engagement, while the content of their communication appears to be secondary to their dyadic interaction goals. This social use of language may be a syndrome-specific feature of the WS behavioral phenotype. Although much remains to be learned about the complex interplay of linguistic, cognitive, social, and cultural factors shaping pragmatic functioning in WS, research conducted so far has made significant strides towards characterizing the profile and the precursors of pragmatic language in people with a neurodevelopmental disorder that continues to challenge our understanding of the organization and development of the human mind.

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Chapter 7
22q11.2 Deletion Syndrome

Ellen Van Den Heuvel, Jeroen Breckpot, Elfi Vergaelen, and Ann Swillen

7.1 Introduction

The 22q11.2 deletion syndrome (22q11DS) is the most common genomic disorder, with an incidence of 1/4000 live births (Devriendt et al., 1998; Goodship et al., 1998) and 1/1000 fetuses (Grati et al., 2015). Its high prevalence is related to the presence of eight low copy repeats (LCRs), repetitive DNA stretches with high similarity, on the long arm of chromosome 22, which are referred to as LCR22-A to LCR22-H. Classical 22q11DS is also known as Velo-Cardio-Facial syndrome (VCFS), which refers to its cardinal features: velopharyngeal insufficiency (i.e. failure of the soft palate to close against the posterior pharyngeal wall), conotruncal heart defects, and facial dysmorphic characteristics, including auricular abnormalities, hooding of the eyelids and tubular nose. Other medical concerns of this syndrome include immunodeficiency, hypocalcemia (often related to hypoparathyroidism), gastrointestinal anomalies, feeding difficulties, renal anomalies, and sensorineural and/or conductive hearing loss (McDonald-McGinn et al., 2015) (see Table 7.1). Most patients with 22q11DS have speech delay and learning difficulties and/or intellectual disability. In addition, there is a high prevalence of behavioural problems (e.g. Bassett & Chow, 1999; Biswas & Furniss, 2016).

22q11DS is characterized by phenotypic heterogeneity. Interestingly, none of the 'typical' 22q11DS-related features are fully penetrant, implying that the 22q11.2 deletion is the major driver for these features, but genetic background or environmental factors are contributive. The variable expression of anatomical defects

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Table 7.1 Common medical issues in pediatric patients with 22q11DS (adapted from McDonald-McGinn et al., 2015)

Medical issue	Type of defect	Incidence
Heart defects	Conotruncal heart defects (tetralogy of Fallot, truncus arteriosus, interrupted aortic arch) or septal defects (ventricular septal defect)	50–75%
Palatal defects	Velopharyngeal insufficiency, occult submucosal cleft palate bifid uvula; overt cleft palate (11%) and cleft lip (1-2%) less common	65%
Genitourinary defects	Renal agenesis, duplicated collecting systems, hydronephrosis, hypospadias, cryptorchidism	30%
Hypocalcemia	Hypoparathyroidism	50%
Immunodeficiency	Impaired T cell production, impaired humoral immune response, thymic hypoplasia, autoimmune disorders	60–70%
Feeding difficulties	Secondary to palatal, gastro-intestinal, endocrine, neural and/or cardiovascular problems	30%
Short stature	Intra-uterine growth delay in 4%	15%
Scoliosis	Idiopathic, vertebral defects	30–40%
Hearing loss	Sensorineural or conductive (e.g. chronic otitis media)	6–60%

(e.g. palatal defects), hearing problems, deficits in cognitive abilities and behavioural issues in 22q11DS, has shaped the unique and complex profile of language disorders in this syndrome.

7.2 Neurodevelopmental Outcome in 22q11.2 Deletion Syndrome

The diagnosis of 22q11DS in a child or adolescent raises a lot of questions for parents and caregivers. Not only medical but also developmental/educational and behavioral/psychiatric aspects of 22q11DS are major concerns for most families. One of the first and most important questions parents ask is what the impact of the 22q11.2 deletion will be on the global cognitive development of their affected child (Swillen & McDonald-McGinn, 2015). Indeed, the syndrome places individuals at an increased risk for neurodevelopmental disorders. Deficits common in 22q11DS include poor cognitive functioning and intellectual disabilities, motor deficits, speech-language disorders, attention and executive functioning deficits, learning disorders (in particular mathematical deficits), emotional dysregulation and impairments in social processing (Swillen et al., 2018). Early detection of emerging problems is important since it will allow for early intervention, individualized educational plans, and environmental adaptations.

In the case of 22q11DS, brain development (and neurodevelopmental outcome) is compromised because of the underlying genetic error, namely a 22q11.2 microdeletion. Over the last 25 years, neuroanatomic alterations in 22q11DS have been investigated in several single-site studies. Early magnetic resonance imaging studies reported whole-brain volumetric reductions in 22q11DS, particularly in midline cortical regions (e.g. Kates et al., 2001; Campbell et al., 2006). A rostrocaudal gradient of volumetric reduction was also reported, with greatest reduction in occipital lobes, while frontal regions were relatively preserved (Tan et al., 2009). More recent studies have mapped the cerebral cortex in detail, investigating measures of cortical thickness and surface area (Schmitt et al., 2015). Increases in cortical thickness in 22q11DS relative to healthy, typically developing controls, with focal thinning in the superior temporal gyrus and cingulate cortex, along with global reductions in surface area are observed (e.g. Schmitt et al., 2015; Jalbrzikowski et al., 2013; Sun et al., 2019). Due to altered brain structure and connectivity, neurodevelopmental outcome in 22q11DS is different/atypical but also variable, because multiple factors play a role: person-specific (risk and protective) factors (genes/nature), family and environmental (risk and protective) factors (nurture), and time/development itself.

Known *person-specific risk factors* with implications for brain development and neurodevelopmental outcome in 22q11DS include: having the 22q11DS (by definition), origin of the deletion (inherited deletions may result in a more severe cognitive phenotype, related to a combination of socioeconomic factors and heritable components contributed by the unaffected parent) (De Smedt et al., 2007; Swillen et al., 1997), size of the deletion (Sun et al., 2019; Zhao et al., 2018), genetic variation within the 22q11.2 region (Gothelf et al., 2005; Raux et al., 2007), variants in genes on the intact 22q11.2 (McDonald-McGinn et al., 2013), prematurity (Van et al., 2016), untreated neonatal hypocalcemia (Cheung et al., 2014), peri-operative seizures (McDonald-McGinn et al., 2015), and undetected and untreated thyroid disease in children with 22q11DS (Shugar et al., 2015). However, much work is still to be done in identifying other possible risk factors contributing to the variable neurodevelopmental outcome in 22q11DS such as genes within the region (*COMT*, *PRODH*, *TBX1*, *CRKL1*, etc.), the remainder of the genome/genetic background, impact of medical problems (e.g. type and severity of congenital heart defects, number of hospitalizations, stress related to medical events, thyroid function), psychological features such as personality and temperament, the level of stress sensitivity and anxiety, and the presence of developmental disorders such as intellectual disability, attention deficit hyperactivity disorder, autism spectrum disorder, and psychosis.

Family and environmental factors contributing to the neurodevelopmental outcome in 22q11DS include socio-economic status and parental and sibling IQ (De Smedt et al., 2007; Olszewski et al., 2014; Shashi, Keshavan, Kaczorowski, Schoch, et al., 2010). As of yet, many other important environmental factors are still understudied such as early parental behaviour, parent-child interaction (attachment), parental stress/resilience, and parenting style and coping. For many (young) parents, the diagnosis of 22q11DS in their child and the variable presentation of the condition bring uncertainty and can cause significant stress (Briegel et al., 2007; Mercer-Rosa et al., 2015). Also life-threating events in the perinatal period (e.g. heart surgery) and the immature and/or altered development and behaviour of their child with 22q11DS may increase overall parental psychological distress and may alter the interaction between child and parents. Furthermore, the impact of therapy/ remediation/anticipatory guidance, quality of life, and availability of social network support and resources on neurodevelopmental outcome in 22q11 DS is unclear and needs to be studied. Finally*, time/development* itself has its impact, and contributes to the changing and variable neurodevelopmental outcome in individuals with 22q11DS (Swillen et al., 2018).

7.2.1 Cognitive Development in 22q11.2 Deletion Syndrome

Knowledge of the cognitive capacities of a child are of great clinical relevance since it plays a key role when planning intervention and re-evaluating an individualized educational plan. In this respect, it is important to keep in mind that both genes and environmental factors play essential roles in shaping brain development and growth and neurodevelopmental outcome throughout life. In the end, each infant/ child / adolescent/ adult and his/her context with 22q11DS are unique (Swillen et al., 2018; Swillen, 2016).

Infancy and early childhood (0–4 years): Central nervous system involvement is common in 22q11DS: neonatal seizures (Hopkins et al., 2018), developmental delays and neuromotor deficits especially in the domains of balance and coordination occur early (Swillen et al., 2005; Sobin et al., 2006; Van Aken et al., 2007). Few studies have been published on the neurodevelopmental outcome in (very) young children with 22q11DS. During infancy and toddlerhood, gross/fine and neuromotor difficulties, expressive language delays, and speech problems dominate (Gerdes et al., 1999; Solot et al., 2001). Roizen et al. (2007) reported retrospective data about developmental milestones from 88 parents with a child with 22q11DS. Compared to sibling and community control participants, expressive language and gross motor milestones were more delayed than other areas of development.

From preschool to adolescence (4–18 years): From preschool age onwards, learning difficulties and/or intellectual disability become apparent. The level of intelligence in children and adolescents with 22q11DS is highly variable and follows a normal distribution (similar to the intelligence quotient (IQ) distribution in the general population), but is shifted about 30 IQ points to the left. The average mean full scale IQ (FSIQ) is in the mid-seventies (70–75), with about 55% having a borderline to normal intelligence (FSIQ >70), about 45% having a mild (to moderate) intellectual disability (FSIQ 55-70), and a minority experiencing moderate to severe intellectual disability (Swillen et al., 1997; De Smedt et al., 2007). Although the intelligence profile is highly variable, a subgroup of children with 22q11DS show, during early primary school age, a discrepancy between verbal abilities and perceptual reasoning abilities, favoring the verbal domain (Antshel et al., 2005; Moss et al., 1999; Niklasson et al., 2002; Shashi et al., 2006). However, this VIQ > PIQ cognitive profile seems to change with age. By the end of primary school age (age 10 years and older) this VIQ > PIQ profile is less commonly observed in children with 22q11DS due to the increasing problems they have within the domain of verbal and language comprehension, and abstract verbal reasoning (Campbell & Swillen, 2005).

In a recent cross-sectional study the cognitive functions of a large sample of 137 subjects with 22q11DS (ages 8–21) were compared with the performance of youth with a developmental delay and medical comorbidities and with typically developing controls (Gur et al., 2014). Cognitive functions were measured by a neuropsychological test battery. Complex cognition, specifically language and nonverbal reasoning, was most impaired in the 22q11DS group. Thus, learning difficulties are very common from primary school age onwards, especially within the domains of mathematics (e.g. Brankaer et al., 2017; De Smedt, Reynvoet, et al., 2009; De Smedt, Swillen, et al., 2009; Tobia et al., 2018) and language comprehension (Glaser et al., 2002; Van Den Heuvel, Manders, et al., 2018).

Besides a global intellectual delay and slow maturing, many children and adolescents with the 22q11DS show an academic and neuropsychological profile of strengths and weaknesses. Typically, areas of relative strengths are reading (decoding), spelling, and (auditory/verbal) rote memory (Antshel et al., 2008; Campbell & Swillen, 2005; Moss et al., 1999). Areas of relative weaknesses are reading and language comprehension, arithmetics, visual-spatial perception and memory, working memory, and executive skills (planning, problem-solving, cognitive flexibility, monitoring) (e.g. De Smedt, Reynvoet, et al., 2009; Gur et al., 2014; Simon et al., 2005; Stoddard et al., 2011; Wong et al., 2014). Coordination problems are increasingly recognized as a feature of 22q11DS (Swillen et al., 2005; Sobin et al., 2006; Van Aken et al., 2007). It is important to screen for these problems during primary school since they may have a major impact on school performance (writing, speed, fine-motor skills) and on daily life functioning (self-reliance). These cognitive/learning and motor impairments continue into adulthood, with challenges in the transition to adulthood including the provision of educational and vocational supports (Fung et al., 2015; Butcher et al., 2012).

7.2.2 Divergent Cognitive Trajectories in 22q11.2 Deletion Syndrome

An optimal design for studying developmental trajectories is to combine cross-sectional designs with longitudinal follow-up (Thomas et al., 2009). Studies of cognitive development in 22q11DS children have suffered from several methodological limitations: cross-sectional studies; small sample sizes; wide age range; and no use of a control group. However, in order to define and understand the phenotypic effect of a microdeletion, it is crucial to use a control group. In the case of 22q11DS, siblings, sex- and IQ- matched controls, and controls with a developmental delay and medical comorbidities could be adequate control groups. Only recently, longitudinal studies have started to include a control group: typically developing controls (Antshel et al., 2010; Gothelf et al., 2005; Hooper et al., 2013), siblings (Chawner et al., 2017), and IQ-matched controls (Van Den Heuvel, Jonkers, et al., 2018).

Longitudinal studies in 22q11DS using typically developing children as controls have found a negative correlation between age and IQ scores, particularly a decline in verbal IQ, suggesting that at least a subgroup of individuals with 22q11DS show a gradual decline in cognitive development as they grow into adulthood (Green et al., 2009; Duijff et al., 2012; Vorstman et al., 2015). In a recent, case-control, longitudinal study of the cognitive development of children with 22q11DS and their unaffected siblings, children with 22q11DS exhibited deficits in all cognitive domains. When individual cognitive trajectories were examined, some participants showed significant decline over time, but the prevalence was similar for 22q11DS and control siblings (Chawner et al., 2017). So, the findings of cognitive decline are more likely to reflect normal developmental fluctuations than a 22q11DS-specific abnormality.

In another recent longitudinal study (2-year follow-up) in primary school-aged children with 22q11DS and IQ-matched peers, several types of cognitive trajectories were observed: a relatively stable IQ trajectory over time (trajectory of delay), a 'growing into deficit' trajectory, and a trajectory of an absolute decline in IQ (Van Den Heuvel, Jonkers, et al., 2018). However, these different trajectories were very similar in prevalence between both groups (22q11DS subjects and IQ-matched controls). In conclusion, there seem to be different subgroups of cognitive development within the 22q11DS population, with some children's IQ decreasing over time (Duijff et al., 2012; Vorstman et al., 2015), while others seem to make progress (Chawner et al., 2017; Van Den Heuvel, Jonkers, et al., 2018).

7.3 Psychiatric Manifestations in 22q11.2 Deletion Syndrome

The psychiatric burden in children and adults with 22q11DS is high and diverse. A majority of children, adolescents, and young adults (82–85%) will fulfill DSM-5 criteria for at least one psychiatric diagnosis (Tang, Yi, Calkins, et al., 2014; Serur et al., 2019). The presence of comorbid psychiatric disorders is also frequent, with almost 1 in 3 children and 1 in 2 adolescents having two or more psychiatric diagnoses at the same time.

During childhood, attention deficit hyperactivity disorder (ADHD) is most prevalent (37–60%) (Tang, Yi, Calkins, et al., 2014; Serur et al., 2019; Schneider, Debbané, et al., 2014; Schneider, Van der Linden, et al., 2014; Niarchou et al., 2015). In contrast with ADHD in the general population, the majority of children with 22q11DS will have ADHD of the inattentive subtype (±60%) (Niarchou et al., 2015; Schneider, Debbané, et al., 2014; Schneider, Van der Linden, et al., 2014). In contrast, the combined subtype of ADHD appears to be present in 1 out of 2 diagnoses in 22q11DS, while a minority (<10%) has a diagnosis of the hyperactive-impulsive subtype (Schneider, Debbané, et al., 2014; Schneider, Van der Linden, et al., 2014; Niarchou et al., 2015). As in the general population the prevalence of ADHD appears to decline with age towards a diagnosis of about 1 in 5 during adulthood (Tang, Yi, Calkins, et al., 2014; Schneider, Debbané, et al., 2014; Schneider, Van der Linden, et al., 2014). In line with other psychiatric disorders in 22q11DS, ADHD appears to be undertreated in children with 22q11DS. Methylphenidate appears to be an effective treatment for ADHD in this population (Gothelf et al., 2003). Treatment does seem warranted, as a diagnosis of ADHD during childhood has been associated with the development of psychotic symptoms later in adolescence as well as a more pronounced executive dysfunction and decreased overall functioning in early adulthood (Taylor et al., 2018; Niarchou et al., 2018).

Autism spectrum disorder (ASD) is also a prevalent psychiatric disorder in children with 22q11DS (12–16%) (Schneider, Debbané, et al., 2014; Schneider, Van der Linden, et al., 2014; Serur et al., 2019). Prevalence peaks during adolescence with a diagnosis of ASD in about 1 in 4 adolescents with 22q11DS (Schneider, Debbané, et al., 2014; Schneider, Van der Linden, et al., 2014). In adulthood the prevalence declines again to 16% (Schneider, Debbané, et al., 2014; Schneider, Van der Linden, et al., 2014). In addition, more than half of children and adolescents with 22q11DS appear to score above the cut-off in at least one of the three domains of the Autism Diagnostic Interview-Revised (Antshel et al., 2007; Ousley et al., 2017; Serur et al., 2019). This included around 40% for the reciprocal social interaction score, 32% for the communication score, and around 47% for the restricted, repetitive, and stereotyped behaviour score (Ousley et al., 2017; Serur et al., 2019).

Another highly prevalent category of psychiatric disorders in children with 22q11DS is anxiety disorders. A large, international, multicenter study found the prevalence to be around 35% in children and adolescents (Schneider, Debbané, et al., 2014; Schneider, Van der Linden, et al., 2014). Specific phobias, social phobias, and generalized anxiety disorders are most prevalent. Prevalence remains high during adulthood, with about 1 in 4 adults fulfilling the criteria for at least one anxiety disorder. Anxiety has been related to the development of psychotic symptoms, worse social competency, and worse working memory performance in adolescents with 22q11DS (Shashi et al., 2012; Sanders et al., 2017; Gothelf et al., 2013). Anxiety has also been associated with adaptive functioning in children but not in adults with 22q11DS (Angkustsiri et al., 2012; Fabbro et al., 2012; Butcher et al., 2012).The prevalence of major depressive disorders is also elevated and it increases with age, going from around 2% in primary school children to 15% in adults (Schneider, Debbané, et al., 2014; Schneider, Van der Linden, et al., 2014). Comorbidity between anxiety disorders, mood disorders, and psychotic disorders in 22q11DS is high. The diagnosis of a mood disorder significantly increases the likelihood of the presence of both an anxiety disorder and a schizophrenia spectrum disorder (Schneider, Debbané, et al., 2014; Schneider, Van der Linden, et al., 2014).

Prodromal psychotic symptoms often arise during adolescence. They are present in more than half of adolescents with 22q11DS (Tang, Yi, Moore, et al., 2014; Weisman et al., 2017; Stoddard et al., 2010; Mekori-Domachevsky et al., 2017). An ultra high risk (UHR) state for psychosis as defined by the internationally accepted criteria seems to be present in 20–30% (Weisman et al., 2017; Mekori-Domachevsky et al., 2017; Schneider et al., 2016). Prodromal symptoms are subdivided into positive symptoms such as delusion-like or hallucination-like experiences, negative symptoms, and disorganized symptoms. Within 22q11DS adolescents with an UHR state, negative symptoms appear to be more prominent compared to individuals with an UHR state in the general population (Armando et al., 2012). As in the general population an UHR state in 22q11DS is associated with a high chance of transition. About 1 in 3 will make a transition from prodromal psychotic symptoms to a psychotic disorder (Schneider et al., 2016).

The prevalence of psychotic disorders in adults with 22q11DS is 30 times higher than in the general population and 10 times higher than in individuals with an intellectual disability. Most psychotic disorders in 22q11DS are diagnosed as schizophrenia and are associated with cognitive deficits and negative symptoms including reductions in speech, avolition and decreased expression of emotions. Moreover, a large longitudinal study of 411 individuals with 22q11DS showed that a decline in IQ, most pronounced in verbal IQ, was associated with an increased risk of developing a psychotic disorder later in life. This cognitive decline was already distinguishable at age 11 (Vorstman et al., 2015). Hence, clinically significant psychotic symptoms should be screened throughout the lifetime of people with 22q11DS (Swillen & McDonald-McGinn, 2015).

Finally, it appears that negative symptoms in 22q11DS are associated with worse cognitive performance and impairments in daily functioning. This includes a negative association with processing speed and visual memory as well as a negative association with occupational domains and socialization (Schneider et al., 2012; Schneider, Debbané, et al., 2014; Schneider, Van der Linden, et al., 2014). Therefore, monitoring the emergence of social communication deficits in children with 22q11DS from school-age onwards should be part of the interdisciplinary management of children and adolescents with 22q11DS (Bassett et al., 2011). Social communication issues can be a consequence of different neurodevelopmental and psychiatric disorders that are prevalent in 22q11DS and can lead to severe disability during adolescence (Swillen & McDonald-McGinn, 2015).

7.4 Language and Social Communication in 22q11.2 Deletion Syndrome

Language is a key predictor of developmental outcomes and a warning sign for families to note that their child might be at risk of a neurodevelopmental disorder. Evaluation of language ability in children with genetic syndromes supports the interactional exploration of the behavioural phenotype, which Dykens (1995) referred to as “the heightened probability or likelihood that people with a given syndrome will exhibit certain behavioural or developmental sequelae relative to those without the syndrome" (p. 523). This definition implies that when describing symptoms of syndromic conditions, different domains of development and within-syndrome variability need to be considered. The "linguistic/communicative component" of the behavioural phenotype, including form, content, and use aspects of language, should be linked to other developmental domains, such as cognitive and social skills and adaptive functioning (see Fig. 7.1).

7.4.1 Language Form and Content

Although considerable research has been devoted to speech disorders and velopharyngeal insufficiency in 22q11DS, rather less attention has been paid to the language abilities of children with this condition. Delayed onset of expressive language and language levels below what is expected for their developmental age have been demonstrated. Around the age of 2 about 90% of children with 22q11DS are non-verbal and only use gestures to communicate. The majority of children with 22q11DS (80%) are still non-verbal or use only single words or two-word phrases at the age of 3. From 4 years old onwards, most children are able to express themselves in short sentences. However, 30% of 4-year-olds remain non-verbal or use telegraphic fragments (Persson et al., 2003; Rommel et al., 1999; Scherer et al., 1999; Solot et al., 2000, 2001). Positive trajectories of improvement in language skills in preschool children are expected (Gerdes et al., 1999). However, nearly 90%

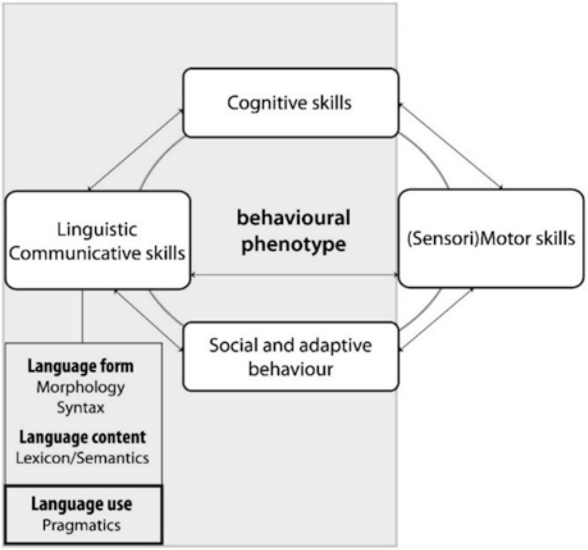

Fig. 7.1 Behavioural phenotype components and aspects of language

of primary school-aged children with 22q11DS have persistent language problems (Nayak & Sell, 1998; Scherer et al., 1999).

The first language research in children with 22q11DS (Golding-Kushner et al., 1985) reported on receptive vocabulary using the Peabody Picture Vocabulary Test-Revised (Dunn & Dunn, 1981). Three groups, preschool children (under 6 years), primary school-aged children (6–10 years) and a group older than 11 years, were assessed. The youngest group had a near-normal score, in contrast to primary school children who obtained well below average to (extremely) low scores. From the age of 11, scores were slightly better.

Subsequent investigations showed typical and atypical discrepancies between receptive and expressive language skills in children with 22q11DS. Young children with 22q11DS (<6 years of age) scored higher on receptive language than on expressive language (Gerdes et al., 1999, 2001; Solot et al., 2001). Mean receptive language scores exceeded mean expressive language scores when using the standardized language scales of The Preschool Language Scales-3 (Zimmerman et al., 1991). Cognitive abilities and language results were closely related in preschoolers. About one fourth of them scored ten or more points lower on the language assessment than on the intelligence assessment (Solot et al., 2001). In primary-school age children with 22q11DS both expressive and receptive language deficits were found. Based on results from the Clinical Evaluation of Language Fundamentals-Revised (CELF-R; Semel et al., 1987), the mean receptive and expressive language scores did not differ significantly from each other in a cognitively variable group (Moss et al., 1999). Solot et al. (2001) reported on 26 children aged 5.9 to 16.7 years using the total language score of the CELF-R. The mean receptive language score (64.8 ± 11.5) was found to be in the same range as the expressive language score (68.2 ± 13.8).

Some other studies have suggested an atypical and syndrome-specific limitation of the receptive over expressive language advantage in children with 22q11DS. Scherer et al. (2001) compared the language proficiency of four children with 22q11DS to that of four children with Down syndrome (chronological age 30–54 months). The receptive and expressive language skills of children with 22q11DS were both equal to or below what would be expected based on their mental age. In contrast, children with Down syndrome had receptive language skills equal to or above what would be expected based on their mental age, but showed more severe expressive language impairments. Glaser et al. (2002) used the Clinical Evaluation of Language Fundamentals Third Edition (Semel et al., 1995) to compare the language skills of 27 children and adolescents (6–19 years) with 22q11DS to an idiopathic developmentally delayed control group matched for gender, age, and IQ. Children with 22q11DS had lower results on receptive language subtests compared to expressive language subtests. Lowest scores were reported on the Semantic Relationships and Sentence Structure subtests, both measuring receptive language skills. Highest scores were reported on Word Structure and Recalling Sentences, both expressive subtests (Glaser et al., 2002). The opposite pattern was demonstrated in the developmentally delayed control group.

The inconsistent findings in receptive versus expressive language skills in children with 22q11DS are likely to be attributed to the following confounding factors: (1) developmental trajectories, and (2) level of cognitive functioning. Hence, the language profile of children with 22q11DS was re-evaluated after 18–24 months and compared to children with idiopathic intellectual disability (IID) and children with IID and comorbid ASD (Van Den Heuvel, Manders, et al., 2018). When comparing language profiles across groups, the characteristics of language abilities in the IID + ASD group, which often included relatively well-preserved expressive language ability but impaired receptive language ability, were also noted in some children with 22q11DS. Receptive language skills are not likely to exceed expressive language skills in children with 22q11DS, making them prone to be overestimated in their language and learning competence.

The standardized language tests used in the abovementioned language studies primarily evaluate semantics, syntax, and morphology (i.e. language form and language content). However, pragmatic language impairments may significantly interfere with the outcomes, especially with sentence comprehension and production (Botting & Conti-Ramsden, 1999). Pragmatic language issues, and more specifically their possible influence on standardized language tests, have received little attention in children with 22q11DS. Difficulties with interpreting and using contextual cues might influence expressive language responses at sentence level. Analyzing responses to the Formulating Sentences subtest of the CELF-4-NL (Kort et al., 2010) showed that semantic-pragmatic challenges often induce low scores on expressive syntax subtests in school-aged children with 22q11DS (Van Den Heuvel, Manders, et al., 2018). The latter finding was considered to be important to promote in-depth assessment of language use in children with 22q11DS.

7.4.2 Language Use

Using language effectively for social and functional purposes entails identifying what to say in a given situation (Bishop & Adams, 1991; Nilsen et al., 2012). In earlier studies (Golding-Kushner et al., 1985; Scherer et al., 1999) reduced responsiveness to simple questions, lack of initiating utterances, failure to recognise and appropriately react to verbal and nonverbal social cues were observed in children with 22q11DS and were suggested to be related to limited insight into the listener's needs (i.e. poor presupposition skills). In the last decade, there has been increasing interest in pragmatic and socio-communicative skills in children with 22q11DS and in their relationship to neuropsychological and psychiatric manifestations.

A parental report is a good starting point for highlighting specific socio-communicative challenges. The concerns of parents will corroborate the need for a comprehensive evaluation of individual socio-communicative competences and challenges. Some communicative characteristics reported by parents of children with 22q11DS resemble the behavioural pattern of children with ASD. Therefore, questionnaires used to diagnose ASD are often used to explore social and communicative impairments in children with 22q11DS.

By means of the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 1999) and the Social Communication Questionnaire (SCQ; Rutter, Bailey, & Lord, 2003), weaknesses in abstract thinking and lack of imagination were found to be characteristic of all children with 22q11DS (Angkustsiri et al., 2014). Parents of children with 22q11DS and comorbid ASD more often noticed atypical behaviours and social difficulties than parents of children with 22q11DS only. Antshel et al. (2007) suggested that these behavioural findings were related to the presence of a larger right amygdala, a structure that is likely to play a role in flexible processing of emotive and communicative information. Nonetheless, the authors acknowledged the inevitable overlap between the groups in their study, as the ASD diagnoses were solely based on parent reports using the Autism Diagnostic Interview-Revised (Rutter, Le Couteur, & Lord, 2003). Using this standardized interview, gestural communication, initiating conversation, and sharing attention seemed present in all children with 22q11DS. Difficulties with nonverbal social interactions, make-believe play, and peer relationships only occurred in children with 22q11DS when comorbid for ASD. Restricted interests are common in all children with 22q11DS, but rituals, motor stereotypies or repetitive use of objects are only reported when ASD has been diagnosed (Kates et al., 2007).

Van Den Heuvel, Manders, et al. (2017) used the Children's Communication Checklist-2, Dutch Edition (CCC-2-NL; Geurts, 2007) to explore socio-communicative similarities and differences in school-aged children with 22q11DS and two groups of children with IID, one with and one without comorbid ASD. When composite scores – summations or subtractions of subscale scores – were considered, not many significant differences were found across groups. However, children with 22q11DS and children with IID + ASD both met the cut-off score for the presence of general socio-communicative difficulties and specific pragmatic language problems. Children with IID displayed scores just below these boundaries, indicating less evidence for socio-communicative impairments. Major concerns of parents of children with 22q11DS in this study included the inability to use contextual information to understand, organise, and express language in an adequate way. Children with 22q11DS had difficulties grasping the implicit meaning of words and phrases and abstract information processing. Parents frequently reported on inappropriate information transfer, including vague word choice, perseverations, and focus on side issues or details. These pragmatic language impairments prevented them from adapting their language appropriately in changing environments and resulted in communicative breakdowns. Conversational initiation problems were found to be extremely variable in the school-aged 22q11DS group. Some parents indicated these issues but they could not be considered to be statistically significantly different from those of children with IID or from those of children with IID + ASD. The broad behavioural spectrum described by Swillen et al. (1997) with children with 22q11DS being impulsive and disinhibited on the one hand and more introverted or shy on the other hand, seemed to be reflected in the results of the initiation subscale of the CCC–2–NL (Van Den Heuvel, Manders, et al., 2017).

Direct pragmatic language assessments can confirm the (in)abilities indicated by parents of children with 22q11DS. Study of language use targeting evaluation of (a) narrative abilities, (b) referential communication, and (c) conversation skills allows us to identify whether a child with 22q11DS is able to judge the listener's informational and social needs and is able to adapt one's message content and form to consistently changing contexts (Ketelaars et al., 2009). In children with 22q11DS assessment of language use is challenging. Motivation, intellectual disability, attention span, and concentration are possible factors that can influence the results of a language test besides the actual language ability. Moreover, pragmatic language impairments can be very subtle and hard to detect.

(a) *Narrative abilities:* A retelling task, the Bus Story Test, was used by Persson et al. (2006) to investigate narrative abilities in 19 children with 22q11DS between 5 and 8 years old. Impoverished information transfer with scores significantly below age-related norms was demonstrated. A negative correlation between the scores and chronological age provided some evidence for a growing gap between children with 22q11DS and peers. The absence of an IQ-matched comparison group prevented any conclusion that the identified challenges are syndrome-specific or are part of a more general delay.

(b) *Referential communication:* Failure to concisely convey and organise information seems to be related to limitations in referential communication skills (Cummings, 2009). Since referential communication relies on both linguistic and socio-cognitive abilities, all children with intellectual disability have an increased risk of challenges in this domain (Abbeduto et al., 2004). Limitations in referential communication skills, including perspective- and role-taking abilities, have a direct impact on daily communication and may lead to social-emotional issues when a child cannot express his intended feelings or thoughts (Hatton, 1998; Rondal, 2001).

Van Den Heuvel, Reuterskiöld, et al. (2017) examined perspective-taking and role-taking abilities in children with 22q11DS. Compared to both chronological age and younger receptive vocabulary age equivalent (RVAE) matched typically developing children, children with 22q11DS produced less complete sentences containing core message components. Compared to younger RVAE matched controls, they added significantly more information regarding visual details. Frequent use of these irrelevant details made the contributions of children with 22q11DS confusing. Children with 22q11DS transferred less essential information and used shorter, less grammatically complex sentences than both control groups. Children with 22q11DS often talked in a chain of unconnected utterances and seldom used cohesive devices. This resulted in a significantly higher number of utterances in comparison to typically developing peers.

Several explanations for these perspective-taking challenges are hypothesized. First, poor structural language skills, and particularly limitations in language comprehension, may lead to poor referential communication skills. Second, some studies demonstrated abnormalities in visual scanpath strategies for both socio-facial stimuli as well as complex pictures. Children with 22q11DS demonstrated shorter fixation time, leading to inaccurate information processing (Campbell et al., 2010; Glaser et al., 2010; McCabe et al., 2011). This may explain the focus on visual details and the increased amount of illogical utterances triggered by deficient visual interpretation. Third, judging what is appropriate or inappropriate to share requires the ability to inhibit use of one's own knowledge as the common ground perspective (Epley et al., 2004; Keysar et al., 1998). It is suggested that children with 22q11DS have difficulties differentiating between their own perspective and the perspective of others (Van Den Heuvel, Reuterskiöld, et al., 2017).

(c) *Conversation skills:* Children use conservations to establish cooperative relationships. Poor conversation skills will negatively influence social acceptance and may lead to social rejection or isolation from peers (Hemphill & Siperstein, 1990). Children who have difficulties with interacting in a naturalistic context such as a conversation, are known to experience difficulties with peer relationships and have a higher risk of being bullied than typically developing children (Conti-Ramsden & Botting, 2004).

Several subskills are related to conversation success, including the following: (1) the ability to take into account a listener's knowledge; (2) discourse topic management; and (3) responding contingently and extending a topic by providing relevant information on a conversational partner's turn (Nadig et al., 2010; Schegloff, 2000). Children with intellectual disability may encounter challenges developing these subskills. Van Den Heuvel, Botting, et al. (2017) assessed these three subskills in eight children with 22q11DS aged 7–13 years. These children were re-evaluated after 18–24 months, and compared to peers with idiopathic intellectual disability (IID) and IID + ASD. Fragmentary and disorganized conversations were observed in children with 22q11DS. These were caused by impaired turn-taking and a passive discourse style. Children with 22q11DS used significantly more follow-up statements, i.e. additional optional contributions that did not elicit or provide information, than children with IID. This finding suggests that children with 22q11DS contribute less to topic maintenance.

The first language study of young children with 22q11DS also reported withdrawn and less responsive behaviour in these children (Golding-Kushner et al., 1985). A follow-up assessment after 18–24 months showed that children with 22q11DS took less initiative to start and continue a conversation. There was an increase in responses due to a more distant conversation style over time. The opposite profile (i.e. more active conversational involvement over time) was revealed in children with IID. Children with IID + ASD did less take account of listener knowledge than children with 22q11DS. In general, conversation abilities of children with IID+ASD were rated as being more severely impaired than those of children with 22q11DS and children with IID (Van Den Heuvel, Botting, et al., 2017; Van Den Heuvel, Manders, et al., 2017; Van Den Heuvel, Reuterskiöld, et al., 2017).

7.4.3 Approaching Pragmatic Language Challenges in Children with 22q11.2 Deletion Syndrome

A point to consider is the existence of subgroups of language impairments in children with 22q11DS similar to those demonstrated in children with specific language impairment (SLI) or developmental language delay (DLD). Children with SLI/DLD show impairments in spoken language development despite average nonverbal intelligence, adequate hearing and vision, absence of neurological, physical, emotional or social problems and a good language-learning environment (Norbury et al., 2008). One subgroup delineated within the heterogeneous group of children with SLI/DLD are children for whom pragmatic challenges are felt to be more at the foreground than one would expect from linguistic impairments alone (Botting, 2004). These children are diagnosed with pragmatic language impairment (PLI; Bishop, 1998).

When reviewing the description and core features of children with PLI, the overlap with the linguistic features described in school-aged children with 22q11DS is notable. Children with PLI appear to be verbal and sociable at first sight despite their language limitations (Botting, 2004). Some children with 22q11DS have language form and language content results in line with their (nonverbal) mental age, while others have language impairments beyond cognitive level expectations. Children with 22q11DS do show willingness to interact with others. The following language and socio-communicative characteristics of children with 22q11DS are similar to the profile of children with PLI (Botting & Conti-Ramsden, 2003): (1) poor understanding of functional communication including turn-taking difficulties; (2) poor insight into the listener's needs; (3) limited conversational topics; and (4) a tendency to provide too much or too little information. Hence, it seems reasonable to state that, despite their cognitive limitations, a subgroup of children with 22q11DS is eligible for a diagnosis of PLI. Currently, children with social (pragmatic) communication difficulties without repetitive or rigid behaviours are considered a separate diagnostic category in the DSM-5 (American Psychiatric Association, 2013). Several characteristics of this diagnostic entity mirror features of children with 22q11DS. Therefore, when specific pragmatic language problems are indicated in a child with 22q11DS, it seems reasonable to consider comprehensive neuropsychological assessments to investigate the social communication.

Results of several studies suggest that children with 22q11DS may perform better or similar to children with ASD in some pragmatic areas. Given the fact that 20 to 30% of children with 22q11DS receive a comorbid diagnosis of ASD (Niklasson et al., 2001, 2009), clinicians should be aware of the overlap in pragmatic language challenges across these groups. Particularly, deficits in nonverbal communication, profound problems with initiating conversation, and extremely restricted behavioural patterns should be considered as “red flags” to further investigate the presence of a comorbid ASD in children with 22q11DS. Since modifications of genes on chromosome 22 have been indicated in children with “idiopathic” ASD (Vorstman et al., 2006), it seems reasonable to consider the potential for a shared aetiological background. However, differences across children with 22q11DS and children with ASD highlight the phenotypic variability. Epigenetic factors and exposure to environmental modifiers contribute to variable expression of autism-related behaviours (Muhle et al., 2004). This might also be a reason why some pragmatic language characteristics such as nonverbal communication, use of stereotyped language and coherence measures were demonstrated to be less severely impaired in children with 22q11DS in comparison to children with ASD (Van Den Heuvel, Manders, et al., 2017).

7.4.4 Factors Influencing Pragmatic Language Challenges in Children with 22q11.2 Deletion Syndrome

Some cognitive factors that are likely to contribute to uneven patterns of language outcomes in children with 22q11DS should be considered when interpreting results of assessments of language use. Difficulties with executive functions and maintaining attention consistently have been reported in children with 22q11DS. Limitations in selecting information and filtering distractions permanently influence language processing. Furthermore, executive dysfunction reduces habituation and impacts transfer of learned language skills to various contexts (Harding, 2011). Although the relationship between theory of mind (ToM), a social-cognitive ability, and pragmatic language skills is not straightforward, insight into characters in stories and taking the perspective of a listener in conversation play a pivotal role in the ability to narrate a story and converse with others (Abbeduto et al., 2004; Cummings, 2014). Children with 22q11DS are known to perform poorly on ToM tasks (Charman & Campbell, 2002).

Other social-cognitive challenges including difficulties with (1) the recognition and understanding of emotions, (2) the interpretation of social interactions and (3) social information processing have been demonstrated in children with mild to borderline intellectual disability (Leffert et al., 2010; Van Nieuwenhuijzen et al., 2011; Van Niewenhuijzen & Vriens, 2012). These challenges have the potential to influence pragmatic language assessments in children with 22q11DS. However, more research is needed to delineate how cognitive limitations and specific socio-cognitive shortcomings will influence pragmatic language skills.

7.5 Implications for Clinical Practice

In this section, we will discuss the implications for clinical practice of both neurodevelopmental issues and language impairment in individuals with 22q11DS. Given the increased risk of impaired neurodevelopmental outcome in several neurodevelopmental domains (such as motor skills, (pragmatic) language skills, attention, executive functions, visual-spatial abilities, emotion recognition), early neurodevelopmental follow-up and multidisciplinary intervention is warranted for individuals with 22q11DS. It will be important to find a balance between follow-up and intervention, to monitor in a flexible way changing and increasing needs with age, and to adapt the environmental demands with age (Swillen et al., 2018).

Because of delays in many developmental domains, a holistic and multidisciplinary approach is required in follow-up and intervention programs. This means that different caregivers (parents, teachers) and therapists from different disciplines (developmental pediatrician, infant mental health specialist, speech-language pathologist, physiotherapist and/or occupational therapist, and clinical educational psychologist) should work together to integrate findings. Management of 22q11DS requires an individualized, multidisciplinary, and coordinated care plan that takes into account the associated medical, developmental, and psychological features of the individual. Because of the complexity of 22q11DS in many cases, this comprehensive care should be provided in multidisciplinary 22q11DS clinics.

7.5.1 Recommendations for Assessment

Awareness of the neurodevelopmental, cognitive, and pragmatic language features of children with 22q11DS is crucial to avoid situations in which environmental expectations exceed the abilities of the child. Given the changing cognitive phenotype with age (and the possible cognitive decline in a subgroup of patients), the cognitive abilities of children and adolescents should be followed-up and re-evaluated on a regular basis. As part of anticipatory care, individuals with 22q11DS should be screened for social processing deficits and anxiety and mood disorders throughout their lifetime (Swillen & McDonald-McGinn, 2015).

Many recent studies highlight the importance of comprehensive assessment and follow-up of pragmatic language and social skills in children with 22q11DS (e.g. Persson et al., 2006; Van Den Heuvel, Botting, et al., 2017; Swillen et al., 2018). The combination of both direct and indirect assessments should be considered when evaluating language proficiency in general and pragmatic aspects more specifically. The added value of parental and caregivers' reports in children with 22q11DS is twofold: (1) they are the starting point for comprehensive assessment and provide insight into the type of communicative breakdowns that weigh heavily on parent-child or child-caregiver interactions; and (2) they are an additional source of information to validate observations and outcomes of direct assessments.

Speech-language pathologists should be aware that due to the disharmonic language profile of strengths and challenges, composite scores of standardized language assessments may conceal syndrome-specific challenges. At first glance, children with 22q11DS can give a strong verbal impression due to strengths in morphology and repetition tasks. These children also show willingness to interact. Receptive language difficulties and their failure to appropriately adapt language in changing contexts are often not recognized when using general standardized assessment tools. We recommend that multiple measures should be used in children with 22q11DS to establish a child's language level, including standardized tests, parent report, and natural language samples. This is consistent with the viewpoint of Norbury (2014) who mentioned that evaluation of social communication disorder should include: (a) formal assessments of pragmatic language skills; (b) structured observation of conversation; and (c) parent-teacher reports to obtain detailed insight into the everyday communicative challenges of the child. Regular multidisciplinary psychological and speech and language evaluations are crucial to examine the level of development in order to adjust home, school and remediation conditions accordingly.

7.5.2 Recommendations for Intervention

Alongside assessment of the child's development in different areas, it is also very important to pay attention to the parent-child interaction/relation. Early attachment between the parent and a child with a 22q11DS can be complicated or disturbed by the major medical and developmental problems these children experience during their first months and years of life. Acknowledging the high burden for parents and/or children with 22q11DS and supporting parents to talk about this is crucial. In our experience, parents often benefit from psychological guidance, interventions aimed at the parent/child interaction, and contact with other parents (Swillen et al., 2018).

It is important to provide tools and recommendations for parents on how to support effective communication in different contexts. In children with 22q11DS the microdeletion not only influences behavioural outcome, it also modifies the environment in which the child with a genetic syndrome develops. The dyadic interaction will be adjusted to the parent's expectations (Karmiloff-Smith et al., 2012). Therefore, it is essential that parents (a) are informed about common language and socio-communicative problems characteristic of the syndrome, (b) are trained to recognise these features and react to them appropriately, and (c) are advised on how to anticipate these features and adjust their interaction style to limit communicative breakdowns. Children with 22q11DS seem to benefit from a "sensitive though directive interactional approach" (Swillen, 2001, p.136). Parents should encourage their child to talk about recent activities and feelings associated with past events. The use of a reward system with clearly defined socio-communicative goals can be very useful to create a positive relationship.

Parents and speech-language pathologists can use a 'recycling' technique (i.e. a repetition of the child's response in a different form). This will allow confirmation that the transferred message was well understood. This seems particularly useful in conversation where limited information transfer leads to poor understanding. Furthermore, it can be very helpful when the message is too confusing due to a large amount of irrelevant details. This technique allows a child to observe appropriate turn-taking and to repair the conversation when needed.

Given within-group variability in 22q11DS, it is difficult to achieve specific recommendations applicable for all preschool and primary school-aged children with the condition. The type of education and support should be chosen depending on the overall cognitive capacities (borderline intelligence vs. intellectual disability) of primary school-aged children and adolescents with 22q11DS. For some children with 22q11DS a regular school program with an individualized educational plan (IEP) will suffice. In other situations, children are better off following special education with IEPs that are adapted to the individual needs of the child/adolescent. Implementation of the IEP, and tight control over the quality of service delivery are critical. Also, the changing pragmatic language strengths and challenges of children with 22q11DS support the need for an IEP. We summarize and propose some general educational guidelines and some ideas to target language use in children with 22q11DS. This should not be considered as an exhaustive list and some children with 22q11DS will only benefit to a limited extent from them.

Educational guidelines: During school years, many children and adolescents with 22q11DS are helped with a highly structured learning environment, the utilization of concrete (visual) materials and experiences, and a step-by-step approach with much repetition and rehearsal. Children with 22q11DS will benefit from an encouraging and reinforcing learning environment with clear learning goals and frequent feedback, from instructions on how to learn and how to memorize (with visual aids and schemes), and from pre-teaching (for learning new material) (Swillen et al., 2018). Particularly, teachers should take into account possible underlying physical problems (ear infections, leg pains, and fatigue, for example) that might affect their learning process and their overall functioning in school. In case of a changing developmental/cognitive trajectory, realistic expectations and an adapted learning environment will be necessary to provide a good balance between the individual's capacities and the environmental demands. In this way, anticipatory guidance can be implemented in school thereby averting unnecessary stress.

Recommendations to improve pragmatic language skills: One must be careful not to overestimate receptive language abilities in children with 22q11DS. In the 22q11DS group, expressive skills of an individual child are a good reflection of his/her language proficiency, whereas more abstract and deductive language reasoning skills may be very challenging. By enhancing the understanding of (complex) instructions, children with 22q11DS will be able to respond in a more adequate way. School-aged children with 22q11DS will have a greater chance of understanding what is being said, when their communicative partners adjust their utterances to the child's expressive language level. Precise, short, clear and single directions should always be preferred over complex directions. If possible, parents, teachers, and care givers should avoid the use of idioms, metaphors, or abstract language.

The understanding of directions should be verified before moving to the next item of a task or to a new topic. We suggest ensuring the comprehension of instructions by means of indirect repetition (e.g. Could you explain to your friend, your brother/sister what you need to do?). Asking the child with 22q11DS to summarise the core features of the directions in some key words, symbols, or drawings can be useful as well. Complex verbal constructions, which require high-level language processing, could be combined with a form of alternative and augmentative communication (e.g. gestures, symbols, pictures, written language). Children with 22q11DS should be provided with reminders that will help them to easily recall the directions.

By enhancing meta-linguistic and specific pragmatic language skills, a child with 22q11DS will become better in conveying information and in structuring his or her thoughts. Children with 22q11DS should be explicitly stimulated for reliance on internal language (i.e. verbalisation of the thought process). This will support the organisation of discourse and adjustments to contextual cues. When some additional time is given, children with 22q11DS can better structure their contributions. Speech-language pathologists can give guidance on how to do so. Sources on "think-aloud methods and tasks" can provide useful ideas (e.g. Laing & Kamhi, 2002; Rosenzweig et al., 2011; Veenman et al., 2004). A child can practice to structure a story by explicitly stating when it happened, where it happened, and who was involved. After this clear statement of the context, the listener will be focused and prepared (i.e. "warmed-up") to follow the story. An overview of narrative-based intervention is described by Petersen (2011).

Although these ideas have the potential to be useful, no evidence is currently available that these children will benefit from pragmatic language intervention. To the best of our knowledge, outcomes of socio-cognitive intervention have been investigated in adolescents and young adults with 22q11DS only (Shashi et al., 2015; Glaser et al., 2018). The emergence of social deficits can already during primary school age represent a major source of disability in children with 22q11DS. Awareness of and psychoeducation about these issues for parents, therapists, and school teams is recommended. Interventions should focus on appropriate adaptation of social demands by use of socio-cognitive remediation programs and/or cognitive/behavioural therapy to improve social skills. Even at a young, preschool age it seems important to enhance pragmatic language in children with 22q11DS through the use of role-play activities, barrier-games and interactive story telling.

7.6 Future Research Directions

Given the divergent cognitive and language courses and persisting pragmatic shortcomings in school-aged children with 22q11DS, further research and follow-up are both desirable. When profiling language trajectories, it is important to start in the early stages of life. Pragmatic language studies in preschool children and infants with 22q11DS will allow us to gain insight into the full spectrum of precursors of the challenges that have been identified in school-aged children. In young children (ages 0–3 years) with 22q11DS it seems very interesting to investigate the development of joint attention and attention skills during social interaction. These processes are found to be important for social competence, for language learning, and especially for the development of receptive language skills (Striano et al., 2006). In primary school-aged children, it seems worthwhile to investigate further how oral language skills influence academic achievement.

A remaining question is whether pragmatic language problems are also found in more cognitively advanced school-aged children with the 22q11.2 deletion (FSIQ ≥85). This issue could be addressed in future research to complement the current findings. Research through adolescence and adulthood should at the same time focus on social cognition, self-image and self-perception. Abstract thinking, problem-solving, and planning skills have been identified as challenging in adolescents and adults with 22q11DS (Henry et al., 2002) and require regular re-evaluations. This will support our understanding of the interactional difficulties that individuals with 22q11DS encounter in daily activities. On the same note, with increasing age it becomes more important to investigate and report on the impact of social communication disorders on quality of life (Botting & Hilari, 2011). Efficacy of socio-cognitive, cognitive-behavioral and pragmatic language remediation programs, long-term effects of these interventions, and their impact on socio-communicative behaviour of children with 22q11DS should be further explored. Therapy outcome measures and research are needed to justify (early) functional communication intervention and/or socio-cognitive remediation (Glaser et al., 2012; Mariano et al., 2015).

Several studies have indicated that (pragmatic) language problems correlate highly with behavioural problems (Ketelaars et al., 2010; Law et al., 2014; Mackie & Law, 2010; Van Agt et al., 2011). Feelings of frustration, stress, and failure in communicative interactions have repercussions on the emotional well-being of the child. The relationship between pragmatic language challenges and challenging behaviour should be explored in a bidirectional way. Challenging behaviour may hinder interactions and communicative breakdowns may evoke and encourage problematic behaviour (Bunning & Buell, 2011). Finally, the interplay between auditory processing, visual processing, and language use should be further explored, and could lead to a better understanding of the complex behavioural phenotype of children with 22q11DS.

7.7 Summary

Central to the study of pragmatic language skills is the important role of ‘context’. A wide range of contextual factors influences the interpretation of words and utterances, and how language is used in varying social contexts (Cummings, 2014). Children with 22q11DS exhibit varying (pragmatic) language abilities with age-appropriate behaviour in some contexts, while showing immature and deviant behaviour in others. Swillen et al. (2001) described this phenomenon as “person- and situation dependency”. Parents of children with 22q11DS are highly concerned about the inability of their child to use contextual information to organise, understand, and express language effectively. This problem distinguishes children with 22q11DS from children with IID.

Parents of children with 22q11DS more frequently report specific socio-communicative challenges concerning initiation of conversation, including (a) perseveration of questions, (b) talking excessively about a favourite topic, and (c) leaving limited room for interests of others during conversations. Reduced number of core information elements, short sentence length, and limited use of subordinate clauses were indicated as pivotal narrative challenges (Persson et al., 2006). From a language use perspective, utterances of children with 22q11.2DS can be defined as being verbose, ambiguous, and irrelevant given the pictured scenes. Elaborations on visual details and off-topic information transfer force a listener to consistently infer the intended message (Van Den Heuvel, Reuterskiöld, et al., 2017).

Conversational analysis demonstrated that children with 22q11DS take a passive role during conversation and become less 'active' conversationalists over time. Therefore, children with 22q11DS might be prone to being less involved and skilled in social interaction and activities. Poor management of reference for the listener and poor interpretation of contextual cues are likely to be closely related to the weaker social competence of individuals with 22q11DS (e.g. Campbell et al., 2015; Norkett et al., 2017; Swillen et al., 1997). Consequently, they might have difficulties bonding and creating new relationships as they grow older. Given the reports of a high risk (10–20%) of developing a psychiatric disorder including depression, anxiety, and even schizophrenia from adolescence onwards in individuals with 22q11DS (Angkustsiri et al., 2012; Bassett & Chow, 2008; Stephenson et al., 2015), it seems highly worthwhile to follow-up developmental transitions in language and socio-communicative development in children with 22q11DS. Given the overlap of features between children with 22q11DS and children with ASD and the notable within-group variability in 22q11DS (e.g. Swillen et al., 2001; Swillen et al., 1997), care needs to be taken not to overdiagnose ASD in children with 22q11DS.

Comprehensive pragmatic language assessments will provide further insight into the ability of children with 22q11DS to integrate and adapt cognitive and linguistic competences in changing environments. Although there are some guidelines and recommendations relevant for all children, support and treatment must be targeted to best suit the individual needs of a child with 22q11DS, incorporating age or developmental stage, and the specific constellation of associated medical features, severity, and need for treatment.

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Chapter 8
Tourette Syndrome

Clare M. Eddy

8.1 Introduction

First described by George Gilles de la Tourette (1885, 1889), the disorder typically referred to as Tourette syndrome (TS) is thought to affect approximately 1% of school age children, with an onset around the age of 5 years (Leckman et al., 2006). More children than this, perhaps 15–25% (Khalifa & von Knorring, 2003; Scahill et al., 2005; Robertson, 2008), will experience a short period of 'transient tics' in childhood. However, a diagnosis of TS relies on tics being present for at least 12 months (American Psychiatric Association, 2013). Tics are rapid, rhythmic and stereotyped movements or vocalisations. They can be simple (e.g. a knee jerk, grimace, or sniff) or more complex (e.g. bending over to touch the ground, changing the tone of voice) and can also occur in combinations or sequences. Tic severity and frequency characteristically waxes and wanes over time (Coffey et al., 1994), on a scale of both weeks and years (Leckman et al., 1998). Many young people with tics will experience remission during early adulthood, although some people continue to experience more severe symptoms throughout the lifespan. Tics are more common in young males than females, with a ratio of approximately 3–4:1, although the reason for this is currently unknown (Robertson, 2015).

Most people with TS describe feeling a sensory or psychological urge (e.g. like wanting to sneeze) before they perform a tic, and can sometimes hold back the tic for a certain period of time. Hence, tics have been referred to as being semi-voluntary or unvoluntary (Jankovic, 1997). After a tic has occurred, this is usually followed by a feeling of relief, although sometimes clusters or severe bouts of tics can occur, often in association with stressful environments (e.g. Caurín et al., 2014). Indeed, although some tics appear quite consistent across environments, people with TS can

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experience marked contextual influences on both the nature and severity of their symptoms (Conelea & Woods, 2008; Eddy & Cavanna, 2013a). For example, tics or 'tic-like' behaviours can develop in association with certain people (e.g. Robertson, 2000; Kurlan et al., 1996), or certain objects e.g. compulsive touching (Eddy & Cavanna, 2014). A review into the contextual factors affecting tic occurrence found an exacerbating effect of anxiety, fatigue and social events, although tic reducing events included social interactions with familiar people, situations in which the individual was a passive observer and during sport or leisure activities. Interestingly, tics have been found to be more frequent when watching television versus other situations, including talking with a stranger or focusing on a comprehension task (Barnea et al., 2016).

People with tics often experience a spectrum of behavioural or psychiatric problems including obsessive-compulsive type intrusive thoughts, urges and behaviours; attention difficulties; problems with emotional reactivity and controlling impulses; and mood disorders. More specific behavioural features associated with TS include paliphenomena (repeating one's own speech or actions); echophenomena (copying others' speech or actions); and coprophenomena (obscene remarks or actions). These latter features were described by Gilles de la Tourette (1885, 1889) and will be discussed in greater detail in the following section. In comparison with some other neurodevelopmental disorders, TS is not generally associated with intellectual disability or significant cognitive dysfunction (Eddy et al., 2009).

It is believed that TS (or a propensity to tics) is likely to be inherited (Pauls & Leckman, 1986; Deng et al., 2012), but no clear genetic marker has yet been identified. The neural basis for TS implicates motor areas such as the basal ganglia, and some patients experience symptom amelioration after surgical treatments of the globus pallidus or thalamus (e.g. Cavanna et al., 2011). A number of brain imaging studies have supported the possibility that TS is a neurodevelopmental disorder (e.g. Church et al., 2009), possibly associated with delayed maturation in terms of synaptic pruning (Frick & Pittenger, 2016), although findings are not conclusive. Typically, TS is treated with dopamine blocking agents, and these tend to be most effective in the more moderate to severe cases of TS (Eddy, Mitchell, et al., 2011). Accompanying behavioural problems or mood disorders may be addressed using methylphenidate, tricyclic or serotonergic anti-depressants, although behavioural therapies are also used with some success (see McGuire et al., 2014). More rarely, patients may be referred for deep brain stimulation (Cavanna et al., 2011; Baldermann et al., 2016; Schrock et al., 2015).

This chapter will review in Sect. 8.2 the clinical evidence relevant to the study of pragmatics in TS, followed by the existing empirical literature in Sect. 8.3. In Sect. 8.4, findings are drawn together to address three key questions, i.e. whether both phonic and motor tics constitute fragments of communication; if TS could be conceptualised as a right-hemisphere communication disorder; and finally, whether atypicalities of social cognition underlie the apparent link between TS and pragmatics. This is followed by a final summary with suggested directions for future study.

8.2 Clinical Evidence: Key Symptoms of Tourette Syndrome Relevant to Pragmatics

Sacks (1981: 3) describes TS as "an excess of nervous energy, and a great production and extravagance of strange motions and notions: tics, jerks, mannerisms, grimaces, noises, curses, involuntary imitations and compulsions of all sorts…," highlighting a range of symptoms with social, affective and communicative functions. Tics are worsened by stress and frequently contain emotionally loaded content (Freeman et al., 2009). Vocal tics can include animal sounds, prosodic changes, stuttering, talking to oneself with multiple characters, inappropriate pathological laughter (Cavanna et al., 2010) or 'mutational falsetto' (Kerbeshian & Burd, 1988). This section will explore descriptions of the clinical features of TS most likely to encapsulate aspects of communication and which can therefore offer insight into the potential relationships between tics and pragmatics.

It has been suggested that tics may be more common in children who have speech and communication problems than children who do not have such impairments. For example, when considering a population characterised by dysfluency, one study found a history of tics was present in 50% of children (Abwender et al., 1998). However, while typical dysfluencies may be more common in association with TS, this is perhaps not the case in relation to stutter dysfluency (De Nil et al., 2005). Of course, the presence of vocal tics could incidentally contribute to speech dysfluency or appear to disrupt specific aspects of pragmatic communication (e.g. turn taking). While motor tics may occur throughout speech, vocal tics most frequently occur when listening to others' speech, and particularly just after a speaking partner's clause and just before one's own clause, thereby exaggerating the normal pauses and gesture patterns during conversation (Frank, 1978).

A review by Burd (2014) discusses these issues in detail. Burd describes how tics are typically inserted into ongoing speech at pauses and points of change in inflection. Vocal and motor tics rarely interrupt words and often occur when pausing for punctuation, between large units of speech and when speech is modulated. Contextually non-specific tics occur without meaning. Complex vocal tics tend to adhere to phonological and syntactic rules of speech formulation but violate semantic and pragmatic rules as they are not related to the overall message. Burd concludes that tics can be easily identified given their stereotypical nature and because they do not derail ongoing speech. In sum, many tics appear irrelevant to intended communication and can interfere with effective pragmatic communication.

Although some tics appear contextually out of place, in some cases this may be because they are ironic (Eddy, 2018), and meaning does appear to be central to the urge to perform at least some kinds of tics. One interesting source of evidence for this arises from the study of deaf patients with TS. For example, Morris et al. (2000) describe sign language tics in a deaf man, amongst other pragmatic deficits (e.g. not indicating when he did not understand questions). Phonic and obscene signing tics, including palilalia (repeating one's own speech) and echolalia (copying another's speech), have also been described, in the deaf population (Dalsgaard et al., 2001;

Robertson, 2000). The occurrence of obscene and socially inappropriate tics (e.g. miming vomit when cooking) in deaf patients highlight the importance of the meaning of the communication rather than the sound. In a similar vein, coprophenomena, which primarily consist of scatological words and racial or social epithets, appears dependent on regional cultural knowledge and context such that bilinguals expressing coprolalia may use English in public but revert to their mother tongue when at home amongst family (Burd, 2014). The prevalence of coprolalia is still debated, with estimates ranging from 10% to perhaps 40% of patients attending specialist clinics (Eddy & Cavanna, 2013b).

Other features such as echophenomena, or urges to copy other people's speech and actions, are directly related to the social context. These symptoms may constitute the pragmatics of non-verbal mirroring within conversation, the over-expression of an automatic and primal communicative behaviour shared with many animal species, but which when experienced as a tic may not exert such a facilitative or desired social effect. Indeed, echophenomena may imply an increased sensitivity to environmental social cues, a characteristic that perhaps underlies many symptoms of TS (Eddy et al., 2017). This is supported by the fact that some patients with TS report that they can 'pick up' new tics from a person with a particular tic, or develop a tic specifically related to a certain situation, e.g. prosodic mannerisms that are only elicited around that person (e.g. Burd et al., 2008; Freeman et al., 2009).

Some of the most contextually relevant tic-like behaviours include non-obscene socially inappropriate remarks and actions (NOSIS; Kurlan et al., 1996; Eddy & Cavanna, 2013c). NOSIS are reported by 25–50% of patients seeking treatment, with urges more common than actions (Eddy & Cavanna, 2013c). As socially inappropriate remarks seem to occur on the basis of their likely emotive effect, they are pragmatic by nature. However, many individuals who experience these urges find them to be ironic and most troublesome because there is no actual desire to offend. They frequently struggle to hold back from acting out these urges, mindful of their potential antisocial element. NOSIS are clearly different to coprophenomena given that they are much easier to link to specific environmental cues. The socially inappropriate remarks used by patients frequently contain colourful, complex and witty symbolic associations (e.g. saying 'bacon' when seeing a police officer or shouting 'pizza' when introduced to an Italian). Early reports suggested that NOSIS could be explained by poor inhibition of impulses (Kurlan et al., 1996). But one difficulty with this is that there is an implicit assumption that these urges must therefore apply naturally as a default in all persons (Eddy & Cavanna, 2013a). Therefore, a combination of increased sensitivity to environmental cues (further evidence for this possibility is explored in the next section) and knowledge of appropriate pragmatics, combined with poor impulse control, provides a more parsimonious explanation for the acting out of NOSIS.

Socially inappropriate behaviours could clearly have a detrimental impact on social interaction for those people with tics who experience them. Additionally, they could raise the question of whether there is a relationship between TS and Autistic Spectrum Disorder (ASD). Indeed, the parents of young people with TS may report increased evidence of social problems on scales frequently used in ASD, including aspects of social interaction, and obsessional and inflexible behaviours (McGuire et al., 2013; Güler et al., 2015). In addition, one recent study found ASD was comorbid in 12.6% of children with tic disorders (Pringsheim & Hammer, 2013). However, the non-verbal difficulties that can be typical of an autistic population (e.g. reduced eye contact) are rarely apparent in TS. Further work is needed to understand the exact relationship, given that subtle atypicalities in social cognition have been reported in adult patients with TS (e.g. Eddy, 2018) and the pattern of performance differs to that typically associated with ASD, such that people with TS do not tend to make less use of reasoning about people's mental states. On the contrary, tics may be associated with 'over-thinking' social interactions, or 'hyper-mentalizing' (Eddy & Cavanna, 2015). Moreover, it is possible that there is an ironic link between over-thinking about people's mental states and socially inappropriate urges (Eddy, 2016, 2018). Social cognition will be further explored in the following section that discusses empirical research relevant to pragmatic communication in TS.

8.3 Empirical Evidence: What Can Be Drawn from Existing Studies?

This section will discuss the available literature relating to studies that aim to explore a range of skills relevant to pragmatics, communication and social interaction in TS. These skills include reading and vocabulary, verbal fluency, interpretation of social exchanges including humour and non-literal language, and non-verbal skills related to imitation and prosody.

In relation to formal studies of language, there is limited evidence that young people with TS experience difficulties. However, one study did find that parents of offspring with TS plus a comorbid diagnosis of attention deficit hyperactivity disorder had vocabulary limitations in comparison to parents of children with TS alone or typically developing children (Casey et al., 2000). In addition, there can be decreased scores in young people with TS on tests of oral word reading and reading comprehension in comparison to controls (Ludlow et al., 1982). De Nil et al. (2005) found TS was associated with increased difficulty in speech and language formulation in expressive, receptive and written language. Language deficits in TS were also reported by Brookshire et al. (1994), including specific impairments on expressive language measures. In contrast, other studies have reported no evidence of language difficulties on tasks such as stem-completion (Channon et al., 2003), naming tests (Schuerholz et al., 1996) and sensitive language batteries, although more severe tics and comorbid disorders could potentially interfere with task performance (Legg et al., 2005) as with school performance in general (e.g. Singer et al., 1995). In sum, the evidence that TS is associated with language difficulties *per se* is inconclusive, and it can be difficult in any case to know whether noted language and communication difficulties may result incidentally due to having tics or are integral to the disorder.

A few studies have produced contrasting evidence that TS may actually be associated with relative strengths during certain kinds of language tasks. For example, Mostofsky and Ullman (2007) found that children with TS were faster at producing rule-governed past tenses of words, but worse if responding for irregular or unpredictable past tenses. More recently, Dye et al. (2016) found that children with TS performed faster than healthy controls on a non-word repetition task, providing further support for better or faster access to the procedural memory system for rule-based language processing. Perhaps most interestingly, a study by Walenski et al. found that children with tics were also faster than typically developing controls at naming objects you can use (e.g. scissors, hammer) versus those you could not use (i.e. animals), with these findings highlighting potentially stronger links between language, semantics and the motor system in TS. Such findings are in line with the possibility that environmental cues (in this case, objects) may elicit more efficient automatic responses in TS, perhaps leading to social and pragmatic effects within certain contexts. This could lead to an advantage in some respects (e.g. semantic associations may be more quickly accessed) but may be potentially detrimental when combined with ironic effects and poor impulse control (e.g. compulsive urges).

In adult studies, people with tics perform similarly to healthy controls on verbal fluency tasks, i.e. when asked to generate as many words as possible in response to a phonemic or semantic verbal cue (e.g. Eddy et al., 2012). However, the Hayling sentence completion task (Shallice & Burgess, 1996) has been associated with impairment. This task requires participants to hear sentences that strongly prime a particular word, but not to name this word, and instead produce an alternative word. Numerous studies have reported mild deficits on this task, particularly in relation to longer reaction times than controls (e.g. Channon et al., 2004; Eddy et al., 2010a). This task differs from the fluency task in that the fluency task requires generation of many related responses, whereas the Hayling task demands suppression of a contextually relevant response. The Hayling task may, therefore, tap into a skill of prime importance in TS, that of responding in a contextually relevant way (or based on learned associations) to an environmental cue. Indeed, many of the symptoms of TS suggest that stimulus response relationships may be over-learned, leading to obsessional thoughts and environmentally driven compulsions.

Other relevant studies have explored inferences about communicative intent during tests of social cognition in TS. One study reported no impairments in understanding lies, persuasion, and double-bluff in adults with TS compared to typically developing controls (Channon et al., 2004), while another that included people with TS as a clinical control group for ASD, did note that some patients with TS performed below the standard of controls (Rajendran et al., 2005). Eddy et al. (2010b) explicitly assessed the ability of adults with TS to understand hints, sarcasm, and metaphors, distinguishing between these kinds of remarks and literally correct or nonsense remarks during a verbal task. No differences to healthy controls were found for the Hinting Task (Corcoran et al., 1995). However, patients exhibited impairment in understanding sarcastic and metaphorical remarks (on the Pragmatic Story Comprehension Task: Langdon & Coltheart, 2004), despite being able to correctly determine that literal remarks were contextually appropriate and that nonsense statements did not make sense. These rather specific difficulties with non-literal language could reflect atypical theory of mind (i.e. unusual interpretations of mental states), as appreciation of the speaker's mental state (e.g. belief plus intention to produce a conflicting meaning) is needed to understand a nonliteral remark. Many errors reflected a judgment that a non-literal remark was intended to be literally true.

Few studies have explored the understanding of non-literal language in children with TS. However, one such study has proved to be insightful. Drury et al. (2018) reported that children/adolescents with TS (some also with ADHD) were worse than controls at understanding indirect sarcasm on a written measure, but they performed normally for direct sarcasm. Literal interpretations were apparent as found by Eddy et al. (2010b) in adults with TS.

It is interesting to note difficulties understanding non-literal language in a clinical population where symbolic language can frequently occur, such as in the form of socially inappropriate remarks. Perhaps because people with TS can use symbolic language in an inappropriate way (i.e. against social norms), this means they experience more ambiguity when interpreting the communicative intent of others. In relation to understanding hints, for which there was no deficit in TS, it is interesting to note that these were indirect requests. Therefore, correct interpretation did not require the person with TS to infer the opposing meaning or intention, which is required to understand direct sarcasm and also faux pas. Numerous studies have indicated that adults with TS can fail to interpret faux pas correctly, either believing these accidental remarks to be literally intended, failing to notice them, or identifying faux pas when it is not present (Eddy et al., 2010a; Eddy, Mitchell, et al., 2011; Channon et al., 2012). In sum, both the expression and interpretation of pragmatic communication can be atypical in adults with TS, but this may be more likely when opposing meanings are in question.

In addition to remarks, non-verbal communication, which also contributes to the understanding of pragmatics, may be misinterpreted by people with TS. One study showed that when viewing animated shapes moving randomly (e.g. drifting), adults with TS were more likely than typically developing adults to attribute mental states to the triangles, suggesting a baseline tendency to interpret ambiguous stimuli in a meaningful, socially salient way (Eddy & Cavanna, 2015). When asked to interpret the meanings of humorous cartoons (Eddy, Mitchell, et al., 2011) involving irony, sarcasm, and slapstick style humour (i.e. interpret the communicative intent of the cartoonist), adults with TS again made more 'literal interpretation' errors than controls, especially for sarcasm. Therefore, it seemed that the humorous context did not influence the interpretations of patients in the same way as it did for typically developing adults. This study also revealed difficulties on the 'Harry/Yoni' task in TS (see Shamay-Tsoory & Aharon-Peretz, 2007), which involved recognition of social emotions (gloating and envy) based on arrangements of faces with different expressions. More recent studies have extended the evidence that sometimes complex facial expressions are misinterpreted in TS (e.g. Eddy, Mitchell, et al., 2011) and facial expressions may be processed atypically on a neural level (e.g. Eddy et al., 2017; Neuner et al., 2010; Mermillod et al., 2013). Non-verbal cues that are relevant to pragmatics may, therefore, be interpreted in an atypical way in people who have TS.

It is important to consider whether the affective cues contained in prosody, frequently missing from lab tasks of understanding non-literal language, influence the interpretation of people with TS. Drury et al. (2012) found that for a complex prosody task where participants had to recognise angry tones with conflicting semantic content, children with TS plus accompanying attention deficit hyperactivity disorder were impaired, and adults with TS only also exhibited a deficit. While containing affective cues, the task eliciting a deficit still involves conflicting content in relation to semantic and emotional meaning, which may pose a problem in TS. Only a few studies have looked at other aspects of non-verbal communication in TS, including Devinsky et al. (1993), who found that a small group of adults with TS actually scored above norms on the Profile of Nonverbal Sensitivity (Rosenthal et al., 1979). Using an imitation inhibition task, Jonas et al. (2010) showed that adults with TS exhibit more interference with their own movements from the movements they are observing that other people are making. Similarly, a study conducted by Finis et al. (2012) found that adults with TS echoed the actions of people they observed, i.e. imitated actions to a greater extent than controls when this was not requested. The findings of these studies accord with symptoms such as echophenomena and that patients with TS are more ‘wired to imitate.’ Indeed, there can be abnormal activation within parts of the mirror neuron system in TS when patients appraise emotional facial expressions (e.g. Eddy et al., 2017).

Finally, a study carried out by Channon et al. (2003) may offer further insight. Adults with TS were required to generate possible solutions in response to a series of video-clips showing awkward situations and predicaments, e.g. having a neighbour with noisy barking dogs, and were scored in relation to factors such as the number of solutions generated and the appropriateness of these solutions. The TS group’s responses indicated fewer solutions and poorer selection of the best solution in comparison to typically developing controls, although some other ratings such as effectiveness and appropriateness were not statistically significantly different across groups. In general, the TS group appeared to be less able to fully appreciate the social and practical nuances of the predicaments, and the authors suggested that emotional processes could underlie these differences rather than higher-level cognitive difficulties. Problems responding to the pragmatics of a situation could, therefore, have contributed to patients’ limitations in this study.

8.4 Discussion: Tourette Syndrome as a Disorder of Pragmatics

8.4.1 Are all Tics Fragments of Communication?

The suggestion that TS is an intrinsically social disorder, featuring automatic responding to environmental cues (especially those with social and affective salience) raises the possibility that many tics could reflect fragments of communication. Although there has been no formal study of this, some preliminary observations may be drawn in relation to factors such as the form that tics take, the environment in which they occur, and their typical content.

Firstly, a diagnosis of TS necessitates the presence of at least one phonic tic. If language is essentially the medium of communication, then all tics involving language could constitute a form of communication. This will include the signing tics used by deaf people with TS, which can take the form of both symbols and spelled out words (Lang et al., 1993; Morris et al., 2000; Dalsgaard et al., 2001; Robertson et al., 2015). In addition, it may be argued that all forms of vocalisation are potentially communicative, and in the least occur through a system biologically intended for such. Alternatively, one may argue that a communicative message is not so clear in relation to phonic tics in the form of coughs or sniffs, or even tic words that are semantically irrelevant and used across contexts (although in the author's experience, the latter are rare in the case of traditional presentations of TS). However, even simple noises can communicate affect. For example, people may clear their throat when anxious. Certainly, in the case of non-human animals, many simple noises possess a clear communicative element, such as seeking and alarm calls, although others may appear more limited to expression, such as a yelp of pain. It is interesting, therefore, to note that animal noises can indeed sometimes form part of a tic repertoire and are included in standardised assessment (Robertson & Eapen, 1996). Stimulation of the cingulate cortex can result in affective vocalisations in animals (Devinsky et al., 1995), and many studies report activation of the cingulate in individuals with tics, including in association with vocal tics such as coprolalia (e.g. Gates et al., 2004).

Secondly, the environment in which tics occur may offer insight. Patients may say their vocal tics can occur in private, when there is the lack of a presence of others to communicate with. However, self-directed communication no doubt occurs (i.e. we often think in words, even 'out loud'), so this finding does not seem to challenge the possibility that all tics occur as part of a broader spectrum of intrinsically communicative elements. Indeed, some tics and tic-like behaviours seem to be harder to resist in contextually relevant social situations, and the presence of others can worsen symptoms. This is most clearly seen in the case of socially inappropriate remarks that are specifically linked to the physical characteristics of others present in the immediate environment (Kurlan et al., 1996; Eddy & Cavanna, 2013c). In such cases, taking into account social context is central to the interpretation of the tic. The importance of the social environment on tic expression may also be seen when patients attempt to suppress their tics or restrict activities. For example, Conelea et al. (2014) found tic suppression may occur in association with concerns over their embarrassing or potentially disruptive nature. It has also been shown that related social anxiety can impact quality of life in TS (Eddy, Mitchell, et al., 2011). It is notable that few studies have formally explored the relationship between a patient's individual tics and the contexts and environments they occur in, and such an approach could prove most informative.

Thirdly, the finding that speakers of multiple languages with TS may alter the language used during tics according to environment (Burd, 2014) also seems to suggest that at least for some tics, communication of meaning is important. Other tics or tic-like symptoms (coprolalia, echolalia, other socially inappropriate remarks) are most clearly communicative in terms of meaning whether they take the form of words or symbols. Many motor tics take the form of social gestures (e.g. nodding, smiling, eye contact, facial expressions, kissing, hitting, clapping, spitting etc.) which may be interpreted as affiliative or hostile signs. Echoing and mirroring behaviours also seem to reflect a primitive form of affiliative communication, which are shared with non-human animals, and have been described previously as the 'chameleon effect' in humans (Chartrand & Bargh, 1999). Besides these more complex tic-like behaviours, other types of motor tic (e.g. touching an object, reaching, pointing) may communicate an intention or a desire, which can be detected and/or interpreted through the Mirror Neuron System (MNS; e.g. Iacoboni et al., 2005; Rizzolatti & Fabbri-Destro, 2008; Rizzolatti et al., 2014). The relevance of the nonverbal communicative aspect of tics is further supported by studies revealing abnormal activity within brain regions that are thought to underpin the MNS in individuals with TS (e.g. Eddy et al., 2017). However, there is an important distinction between tics that appear more like goal-directed actions versus seemingly purposeless repetitive movements such as stereotypies. The former seem more frequent in TS, whereas the latter may be more characteristic of ASD (Goldman et al., 2009).

In summary, many tics can be viewed within a social context and are therein potentially communicative by nature. Whether the form of communication is less typical of normal hemispheric dominance will be explored in the next part of this section.

8.4.2 Could TS Involve Right Hemisphere Language Disorder?

When surveying the form and content of tics, one cannot help but note the propensity for "strange, often witty associations…restless reacting to the environment…sounds and rhythms… rhymes and repetitions…" as noted by Oliver Sacks (Schleifer, 2001). Language activated by virtue of its semantic meaning and association with context rather than its phonological or linguistic form is the realm of the right hemisphere, which is thought to be the dominant processor of prosody (Ross, 1981), affective language (Borod et al., 1992), figurative language (Myers & Linebaugh, 1981) and pragmatics (Hough, 1990). Indeed, it is suggested that both language and intended motor acts originate from shared intentions within the common semantic system of the right hemisphere (Richards & Chiarello, 1997). One possibility is, therefore, that atypical language processing in the form of right hemisphere dominance could be related to a range of the symptoms seen in TS. For example, words are thought to activate a broader range of meanings when processed in the right hemisphere, but without selection (Richards & Chiarello, 1997). Poorer specificity could help to explain some of the difficulties shown by people with TS when completing language tasks, particularly those involving ambiguous or non-literal language (e.g. Eddy et al., 2010b). However, it is also possible that fast access to alternative meanings of words may have some advantages and could help explain some of the more creative uses of language seen in TS.

The presence of coprolalia could also be interpreted as support for greater right hemisphere processing in TS versus typically developing individuals, given that studies imply that the right hemisphere is critical for non-propositional language such as swearing (e.g. Van Lancker & Cummings, 1999). Lees (1990: 99) describes tics as "forms of non-verbal communication which are difficult to interpret because they occur as the expression of some inner emotional conflict or turmoil." He notes the pragmatically inappropriate taboo sexual and aggressive displays that many tics allude to, which are thought to be associated with the primitive emotional speech centre (i.e. the cingulate and limbic system). Indeed, swearing may be considered a form of limbic language, along with other affectively expressive vocalisations (e.g. Van Lancker & Cummings, 1999). Interestingly, right side basal ganglia lesion has been shown to lead to loss of the ability to swear and produce other forms of automatic speech (Speedie et al., 1993). The basal ganglia have been suggested to play a fundamental role in the neural dysfunction underlying tics (e.g. Mink, 2006), and a range of studies have revealed structural abnormalities including volumetric differences in comparison to healthy controls (e.g. Singer et al., 1993; Petersen et al., 1993).

The picture is further complicated by the results of some studies, which may run counter to the argument that the right hemisphere is dominant for language processing in TS. For example, a few studies have shown that literal interpretations may be more likely in TS than in typical subjects during non-literal language tasks (Eddy et al., 2010b; Eddy, Mitchell, et al., 2011; Drury et al., 2018), and it has been suggested that a lack of right hemisphere input could result in reliance on literal meanings (Winner & Gardner, 1977). Indeed, it is thought that right hemisphere operations are essential to understand a speaker's overall communicative intent based on global context, leading to accurate interpretation of, for example, hints, humour and sarcasm (Mitchell & Crow, 2005). Therefore, a lack of right hemisphere processing could be an alternative explanation for TS patients' difficulties with interpreting sarcastic and ironic utterances.

Perhaps the most parsimonious account for study findings may be atypical inter-hemispheric communication or reduced hemispheric specialisation or dominance in TS. Outside of the language domain, a handful of studies do raise the possibility that cerebral dominance, or perhaps more subtle alterations in inter-hemispheric connectivity, may underlie TS. These include studies of motor dexterity (e.g. Avanzino et al., 2011; Georgiou et al., 1997; Bradshaw & Sheppard, 2000), auditory attention (Plessen et al., 2007), and structural connectivity involving the corpus callosum (e.g. Peterson et al., 1994). There is a reduction in the leftward bias for line bisection with more bisection on the right side in TS (Sheppard et al., 2002; Yazgan et al., 1995), and evidence for greater attention to the right side affecting movement preparation time (Mei Yoke Goh et al., 2002). There is also a loss of the typical left greater than right asymmetry in TS striatum (Petersen et al., 1993) and reduced blood flow in left side basal ganglia structures (Moriarty et al., 1995). However, children with TS can also show reduced attention to the right side of space as assessed by motor preparation (Bradshaw & Sheppard, 2000), and there can be reduced right caudate volume and left lateral ventricle volume (Hyde et al., 1995). In addition to inconsistencies across studies, most relevant existing studies only assessed children with TS. It is difficult to know if the reported effects may change with age, or how they track with symptom severity.

In sum, tendencies to communicate non-verbally, and to use expressive and emotive language and communicative displays implicate right hemisphere brain regions, perhaps the basal ganglia and limbic system in particular. However, neuroimaging of language processes is an under-studied area in TS. Therefore, future studies should explore activation in key language areas (e.g. Wernicke, Broca, limbic language centres) in both hemispheres, using a range of affective and non-affective expressive and receptive language tasks.

8.4.3 Is Social Cognition the Link Between TS and Pragmatics?

Overall, the results from social cognitive research could suggest that adults with TS have specific problems interpreting communications where intention and action (or speech), or intention and literal meaning, conflict directly with each other. Many errors appear to reflect an erroneous assumption that intention necessarily underlies action, and that people generally intend to do what they appear to do, or to communicate the precise words they say. This pattern may reflect the use of more basic or automatic systems for the interpretation of intentions from actions, e.g. the MNS, particularly because this system is rather inflexible providing only a simple backward inference of intention that action/outcome directly reflects intention (see Eddy, 2018). In other words, people with TS may be more likely than typically developing individuals to rely on select motor-related strategies when interpreting social information. This may lead them to be less flexible when making inferences about people's abstract mental states, and to assume that meaningless movements are goal directed, that accidents are intended, and that non-literal utterances are literal.

Indeed, it can be difficult to separate TS from social context as many of the symptoms that quintessentially define this disorder arise only through comparison with accepted cultural and social norms. This includes all socially inappropriate symptoms, echo/pali/coprophenomena, and frequently even simple tics given their interference with social silences, conversation turn-taking, and lack of motion during certain practices. Moreover, neuroimaging studies have found that activity within brain areas that are particularly important for social cognition, including areas of the human MNS, can be correlated with symptoms such as echophenomena (Eddy et al., 2016, 2017). Basal ganglia dysfunction in TS may also contribute to an emphasis on interpreting non-verbal cues during social interaction as opposed to more abstract mentalizing based on semantics or prior knowledge.

While it is apparent that people with TS can automatically react to the pragmatics of a situation, and do not demonstrate the more marked impairments of individuals with ASD, for example, there are difficulties in making conscious judgments about subtle social cues. Such difficulties may arise as a result of emotional reactivity (Eddy et al., 2017) or the weight given to non-verbal aspects of communication as explained previously. However, the potential impact of the lived experience of TS should not be ignored, and having involuntary tics and socially inappropriate urges could lead people with tics to develop an altered notion of pragmatic norms. That is, experiencing ironic urges which lead one to unintentionally break pragmatic norms could simply make reasoning about the intentions and motives behind social behaviour more complex or ambiguous for someone with TS. If individuals with TS are more influenced by action cues (or MNS processes) than typically developing individuals, then removing these visual cues could disproportionately hamper the ability to unravel the meaning of social communications. Alternatively, visual tasks may avoid the potential for interference from irrelevant non-verbal cues. Future studies are needed to explore these possibilities.

From a neurodevelopmental perspective, the right temporoparietal junction is implicated in understanding the mental states of others (see Eddy, 2016) and in interpreting speech acts such as requests (e.g. Egorova et al., 2013). In addition, various brain areas involved in motor function, including areas associated with the MNS (i.e. inferior parietal and sensorimotor cortex), are thought to be involved in interpreting language implying actions rather than, for example, naming (Egorova et al., 2013). Studies in adults with TS have shown atypical activity within these aforementioned areas (Eddy et al., 2016, 2017) when processing facial expressions and inferring the beliefs and intentions of others. In some cases, activity was correlated with symptoms including the urge to tic. It is, therefore, possible that the same brain areas involved in tic symptoms are critical for aspects of social cognition and the interpretation of pragmatic language.

8.5 Summary

There is little evidence that language processes are typically impaired in children or adults with TS. Indeed, many fundamental pragmatic skills appear normal, such as use of deixis, asking questions, matching communication to speaker awareness, and appropriate detail. However, the occurrence of tics could affect some aspects of pragmatic communication. For example, tics can lead to inappropriate body language during communication (e.g. excessive mirroring of others' actions or speech, or tics affecting eye contact). Sometimes tics may contradict the semantic meaning of spoken utterances (e.g. head nodding when stating 'no') or alter the perceived emotional meaning of a message (e.g. changes in intonation or pitch as part of a phonic tic). Furthermore, irrelevant repetition may occur in the form of palilalia. Tics can also interrupt communication and disrupt turn-taking, as well as challenge social norms.

In summary, although people with TS may occasionally breach pragmatic rules, this does not appear to be due to a lack of understanding, as may be seen in other disorders such as ASD. Indeed, a tendency to be more aware of pragmatic factors could combine with behavioural impulsivity to produce tics, especially those that challenge social norms. This is best demonstrated in the case of those tics closely linked to contextual cues including the appearance or attributes of the speaker (including socially inappropriate remarks or gestures), and which can contain complex implicature in the form of witty outbursts (e.g. shouting "bacon" on seeing a policeman). Such characteristics prompted the investigation of social cognition in TS, leading to the discovery that people with TS can respond atypically to tests of social cognition, including those involving non-literal language.

The available data raise the possibility that aspects of language processing are unusual in TS, but much work remains to be done. A good starting point would be to explore more about the types of tics expressed in accordance with different contexts and environmental cues. More specifically, future research should investigate the possibility of atypical hemispheric dominance or inter-hemispheric processing, in addition to further evaluating the relationship with social cognition and behavioural flexibility in general. In particular, studies should explore the developmental trajectory of both language and social skills, and determine whether an emphasis on actions or non-verbal cues could make individuals with TS more likely to misinterpret the overall nature of a social interaction or the intended meaning of a communication. People with TS appear to be 'wired differently,' and it should be emphasised that in some cases and contexts this may be a help rather than a hindrance. TS certainly has much to teach us about creative language including wit and humour, as well as the silent side of communication.

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Chapter 9
Sensory Loss

Kathryn Crowe and Jesper Dammeyer

9.1 Introduction

The disruption to the social context in which children with sensory loss develop can lead to differences in language and pragmatic development. Different kinds of sensory loss impact on pragmatic language development in different ways. Vision loss impacts on early developing foundation skills for social communication, such as joint attention, and limits access to nonverbal communication, but has less impact on the structural aspects of communication. Hearing loss can be disruptive both to the linguistic basis of pragmatics and the opportunities to successfully engage in social communication. Deafblindness impacts on both, and often significantly affects pragmatic skills and outcomes as these children cannot compensate by use of vision or hearing.

Of all the human senses, hearing and vision are the two primarily used for communication. Through hearing and vision humans are able to perceive information from a distance, making these senses dominant for communication and social interaction. Vision and/or hearing loss are, therefore, the two senses of interest in this chapter. However, having vision and/or hearing loss is a very broad classification that incorporates a multitude of subgroups of children with diverse characteristics. The first important distinction is whether the vision and/or hearing loss is congenital or acquired before the child's development of language, or acquired later in life after the development of language (pre-lingual versus post-lingual sensory loss).

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A sensory loss will most often have more severe consequences for development, including pragmatic language skills, if it has restricted acquisition of language and perception of the social context during early development.

The second important distinction is the severity of the sensory loss. Whether a loss is total or profound (total blindness and/or profound deafness) or whether there is usable residual hearing and/or vision has a major impact on communication and social interaction, as well as development more broadly. Whereas a child with total blindness and profound deafness has to rely entirely on tactile and bodily senses for communication, a child with some functional residual hearing and/or vision will have many different opportunities to interact socially and linguistically. The third important distinction concerns whether the sensory loss is accompanied with other impairments that create additional barriers to communication and interaction with the social context. Often, but not always, the aetiology of the sensory loss may be associated with other disabilities such as intellectual impairment, cerebral palsy, or specific cognitive impairments. Thus, individuals with sensory loss are a heterogeneous group, as illustrated in Fig. 9.1.

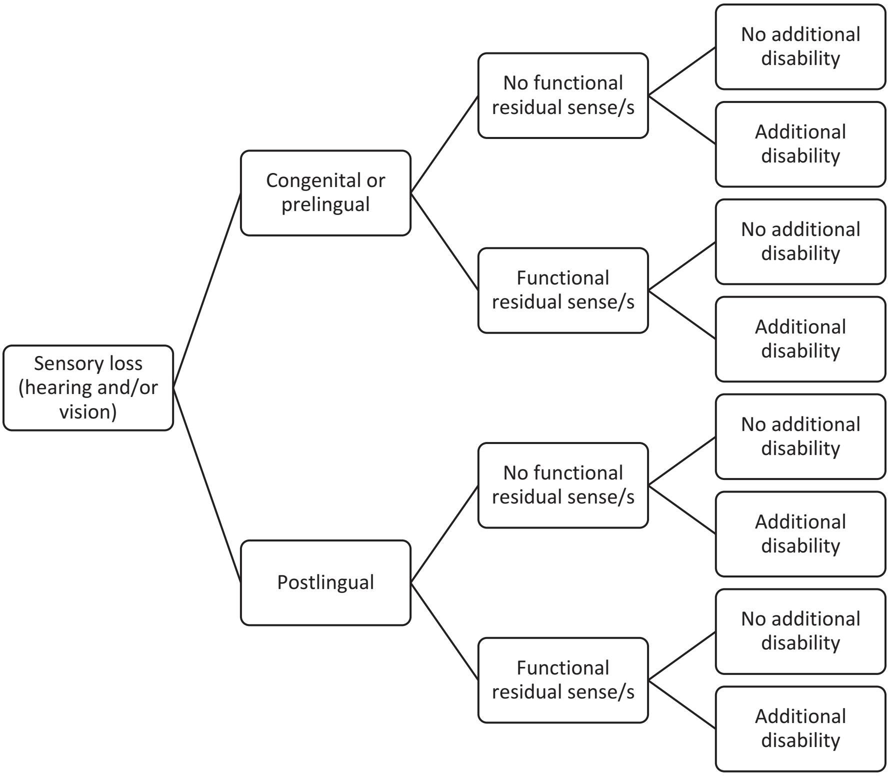

Fig. 9.1 Heterogeneity in individuals with sensory loss

A few cases can illustrate how different the subgroups can be. Where a child with congenital and total blindness and additional severe intellectual development is non-verbal and does not use any intentional communication there is a need for intensive and specialized care. Conversely, a child with congenital vision loss, but functional residual vision and no additional disabilities, is able to use low vision aids and technologies (e.g. using low vision optical devices to enlarge the print in books) and may be able to participate in all activities in the local community school without any further support. A child with profound congenital hearing loss and no additional disabilities could use a sign language and participate in mainstream education and the local Deaf community with few communication barriers. By contrast, a child with a moderate hearing loss acquired at the age of 13 years may be able to communicate effectively using spoken language through the use of hearing aids.

Sensory loss in childhood—whether congenital or acquired, mild or total, and accompanied by an additional disability or not—poses a risk for participation in the social context. As such, the presence of a sensory loss potentially affects all aspects of a child's pragmatic language development. Therefore, in this chapter we will review all aspects of pragmatics related to hearing, vision, and dual sensory loss, and use a broad definition of pragmatic language.

9.2 Children with Hearing Loss

9.2.1 Introduction to Hearing Loss

Hearing loss is a term used to describe an impairment in the ability to detect, discriminate, and localise sound, as well as the ability to perceive and discriminate the speech signal used in spoken language. Hearing loss differs across individuals in terms of the regions of the peripheral and central auditory system impaired, the volumes at which sounds of different pitches can be detected, how auditory stimuli can be interpreted, and by the onset and progression of the hearing loss. Hearing loss can be categorised as conductive (involving the outer and/or middle ear), sensorineural (involving the inner ear), mixed (conductive and sensorineural), or retrocochlear (involving the auditory nerve, brainstem, and/or central auditory system). In other words, a hearing loss can be caused by disorders from the outer ear to the auditory cortex.

The degree of hearing loss is a description of the average volume at which sounds of different pitches can be heard and encompasses mild (26–40 dB), moderate (41–60 dB), severe (61–80 dB), and profound (81 + dB) (Olusanya, Davis, & Hoffman, 2019). The World Health Organization (2018a) estimates that approximately 466 million people worldwide, or 5% of the global population, have a hearing loss that is mild or greater and that 34 million of these people are children. The incidence of permanent, bilateral childhood hearing loss in countries of the Global North is estimated to around 1:1000 live births, and up to 24:1000 in the Global

South (Mathers et al., 2000; Korver et al., 2017). Many children with hearing loss also present with additional disabilities, with the estimated incidence being over 25% (Cupples et al., 2018; Gallaudet Research Institute, 2011).

There are many aetiologies of hearing loss and the predominant causes vary widely around the world, particularly between the Global North and South. Congenital causes of sensorineural hearing loss include maternal infection (e.g. rubella, cytomegalovirus, syphilis), maternal drug use (e.g. cytotoxic drugs, anti-malaria drugs), prematurity, jaundice, and genetic causes (e.g. Connexin 26, CHARGE syndrome) (World Health Organization, 2018a; Korver et al., 2017). Causes of acquired hearing loss in early childhood can include infection (e.g. meningitis, measles), medication (e.g. ototoxic drugs), and genetic causes (e.g. some types of Usher syndrome) (World Health Organization, 2018a). The impact of hearing loss that is sensorineural and either congenital or acquired when a child is prelingual (i.e. before they have acquired language) has the most significant impact on the development of communication and pragmatic language, and such children shall be the focus here.

9.2.2 Communication of Children with Hearing Loss

Research has shown that just a mild and temporary hearing loss in early childhood due to infection in the middle ear (otitis media) increases the risk of long-developmental difficulties (Niclasen et al., 2016). The reason is that hearing loss impacts on a child's ability to acquire spoken language. An estimated 90% of children born with hearing loss have parents who are hearing and spoken language users, with few children with hearing loss born into families where a sign language is used. Whether the child will develop spoken and/or signed language, this situation often causes barriers for language acquisition due to the inability or limitations in the ability of the child to access language models. Specifically, these difficulties relate to receiving either no auditory input or a degraded auditory signal, or to not being in contact with users of a sign language. Early access to language has been found to lead to better speech and language outcomes for many children with hearing loss. Therefore, factors such as the age of onset of the hearing loss and how this relates to age of hearing aid fitting, age of cochlear implantation, and age first enrolled in early intervention are significant predictors of language and educational outcomes for deaf and hard of hearing (DHH) learners (Moeller & Tomblin, 2015; Ching et al., 2017, 2018).

As a population, children with hearing loss are extremely heterogeneous. There are many factors which can impact on children's speech and language outcomes that vary across this population. The presence and type of disabilities in addition to hearing loss can impact on outcomes, especially where the disability is autism, cerebral palsy and/or developmental delay (Cupples et al., 2018, 2018). Children's cognitive ability is also important, with higher cognitive abilities not surprisingly associated with better outcomes (Cupples et al., 2018; Cejas et al., 2018). Family factors such as socioeconomic status (Marschark et al., 2015; Suskind et al., 2016), parental education level (Cupples et al., 2018), and cultural and linguistic background (Crowe, 2018a) may also impact on outcomes. While there are many factors that have been identified as impacting on the outcomes of children with hearing loss, the impact of each is both small and variable, and contributes little to our ability to predict which children with hearing loss will achieve age-appropriate outcomes and which will not (Marschark & Knoors, 2018).

Despite advances in technology (including better hearing aids and cochlear implants) and education practices, the majority of children with hearing loss will experience delays and differences in their speech and language acquisition. The ability to perceive and understand speech, especially in the presence of background noise, varies widely (Ching, et al., 2018). Speech production outcomes also vary widely, with children with hearing loss reported to have age-appropriate speech in some studies and no intelligible speech in other studies, and some studies showing faster acquisition of speech sounds of children with cochlear implants than their hearing peers (Iyer et al., 2017; Cupples et al., 2018; Crowe et al., 2012). Vocabulary acquisition can also pose difficulties for children with hearing loss, who have been reported in many studies to have smaller receptive and expressive vocabularies than their hearing peers (Nittrouer & Caldwell-Tarr, 2016). Similar findings have been reported in relation to children's syntax and global language skills (Nittrouer & Caldwell-Tarr, 2016; Cupples et al., 2018). On average, compared to their hearing peers, children with hearing loss also show poorer academic outcomes across a range of content areas and measures (Luft, 2017; Qi & Mitchell, 2012; Vosganoff et al., 2011; Traxler, 2000). Difficulties with language significantly impact on the child's ability to engage in social relationships, education, and later to participate in the workforce (Garramiola-Bilbao & Rodríguez-Álvarez, 2016; Luft, 2017; Marschark, 2018).

In summary, a congenital or early acquired hearing loss risks language delay and in turn a wide range of developmental difficulties and negative outcomes later in life. However, children with hearing loss is a heterogeneous group and several factors influence the associations between hearing loss, language delay, and life outcomes.

9.2.3 Pragmatic Skills of Children with Hearing Loss

Many children with hearing loss show difficulty with pragmatic skills. Development of pragmatic skills is highly influenced by communication experiences and language skills, both of which are likely to be different for children with hearing loss compared to their hearing peers. While phonology, semantics, morphology, and syntax are frequently examined for children with hearing loss, the pragmatic language skills of these children are less frequently investigated. Generally, findings indicate the pragmatic skills of children with hearing loss are delayed or disordered in comparison to their typically developing hearing peers, although this is not always the case (Paatsch et al., 2017; Toe et al., 2016).

9.2.3.1 Early Social Interactions

Early social interactions form the basis of later social communication and pragmatic development. Parent-child interaction helps to develop skills such as joint attention, reciprocity, and turn-taking that are fundamental to the development of later language-based pragmatic skills. The majority of children with hearing loss struggle to communicate with their hearing parents, which can result in child and parent frustration, and disruption to the type and amount of communicative interactions that occur between them (Barker et al., 2009). Differences in parent-child interaction style have been examined in some research. Compared to deaf parents of deaf children and hearing parents of hearing children, parents of children with hearing loss have been found to be more controlling in their communicative interactions, leading to different encounters with pragmatic language for these children (Barker et al., 2009; Vaccari & Marschark, 1997).

Rinaldi et al. (2013) investigated the early pragmatic skills of two-year-old children with cochlear implants. Only three of the 12 children had scores within the normal range on both subscales of the Social Conversational Skills Rating Scale (Bonifacio & Girolametto, 2007), with the other children scoring below the normal range. Smaller vocabulary size was also associated with poorer pragmatic skills. The authors of this study refer to the reciprocal relationship between language skills and engagement as an explanation. That is, children with better language skills are more able to engage with others in conversation and adults are more likely to engage with children with better language skills in conversation, giving these children more opportunities to practice and improve their pragmatic language skills. However, parents of children with hearing loss also use strategies different from those of parents of children with typical hearing which focus on making an interaction as successful as possible, such as gaining a child’s attention before communicating and use of touch (Lichtig et al., 2011).

Taking a different approach, Weisel et al. (2005) examined the interactions of four two-year-old children with hearing loss and their peers when they were in a regular preschool setting and a special education preschool setting. They found that the children with hearing loss used different strategies to interact with peers in the two settings. Strategies such as “moving closer, object-related social acts, and neutral touch” (p. 166) were used more often in the regular preschool with hearing peers, whereas “signing, direct entrance into play or interaction, and head turning in search of a partner” (p. 166) were used more often to interact with their peers with hearing loss in the special education preschool. Further, for three of the four children there was a greater number of attempts to initiate social interactions with hearing peers in the regular preschool, but initiation attempts were more successful in the special education preschool when used with their peers with hearing loss (Weisel et al., 2005). However, children with hearing loss may still have fewer opportunities to engage in social interactions than their hearing peers, even in integrated preschool settings (Duncan, 1999).

9.2.3.2 Social Communication and Pragmatic Language

The pragmatic skills of children with hearing loss have been investigated from several different perspectives, including use of pragmatic language, peer relationships and social skills, conversation skills, Theory of Mind (ToM), and high-level language skills. What is immediately evident from even a cursory look at the literature is that there are large differences in the skills of children in these areas both within and across studies, and that many different factors have been associated with differences in the pragmatic skills of children with hearing loss. These factors include degree of hearing loss (Shoeib et al., 2016; Lichtig et al., 2011), age (Bebko et al., 2003; Goberis et al., 2012; Nicholas, 2000), and language skills (Nicholas, 2000).

Several studies have taken a broad approach to pragmatic language, conducting investigations using tools that are designed to tap a wide variety of pragmatic skills and behaviours. There is a general trend that children with hearing loss have more difficulties in these areas than their hearing peers. Bebko et al. (2003) and Goberis et al. (2012) both reported, using different instruments, that children with hearing loss experienced delays in the mastery of pragmatic skills. Bebko et al. (2003) considered use of language across all communication modes using the Language Proficiency Profile-2 (Bebko & McKinnon, 1993) in 63 children with hearing loss aged 3–14 years. The study found that hearing children achieved ceiling scores at much younger ages than their peers with hearing loss, with children with hearing loss also showing greater variability than hearing peers across many age groups.

Goberis et al. (2012) used the Pragmatics Checklist (Goberis, 1999) to examine the skills of 126 children with hearing loss and 109 children with typical hearing aged between 2 and 7 years. They report that by 3 years of age children with typical hearing had reached the 75% criterion level for 44% of items and 100% of items by 6 years of age. For children with hearing loss, at 6 years of age 7% of items were at criterion level and only 69% of items were at criterion level with children who were 7 years of age, the oldest included in the study. The items that were not at criterion level for children with hearing loss at 7 years were "(1) provides information on request, (2) repairs incomplete sentences, (3) ends conversations, (4) interjects, (5) apologies, (6) request clarification, (7) makes promises, (8) asks questions to problem solve, (9) asks questions to make predictions, (10) retells a story, (11) tells four- to six-frame picture story in right order, (12) creates original story, (13) explains relationships between objects-action-situations, and (14) compares and contrasts" (p. 304).

Shoeib et al. (2016) investigated the pragmatic skills of 27 children with different degrees of hearing loss aged between 6 and 13 years with a mean age of identification of hearing loss of 2.6 years. Using the Arabic versions of the Test of Pragmatic Language (Alduais et al., 2012), and the Pragmatic Profile and Observational Rating Scale from the Clinical Evaluation of Language Fundamentals (Alduais et al., 2012), these investigators showed that children with hearing loss had lower pragmatic skills than their hearing peers. Children with moderate-severe or severe hearing losses also performed more poorly than those with mild or moderate losses.

Most et al. (2010) used the Pragmatic Protocol (Prutting & Kirchner, 1987) checklist to examine 29 different pragmatic behaviours, categorised as verbal, paralinguistic, and nonverbal communication. Their study involved analysis of videos of the spontaneous conversations of 24 children with hearing loss and 13 hearing peers aged between 5 and 9 years. Children with typical hearing were observed to use significantly more appropriate pragmatic behaviours. The majority of pragmatic behaviours were rated as inappropriate for at least one child with hearing loss, which occurred far less often for children without hearing loss. For 10 of the behaviours, over half of the children with hearing loss were rated as having inappropriate behaviour. These behaviours were: speech act pair analysis; turn-taking response; turn-taking interruption/overlap; turn-taking feedback to speakers; turn-taking adjacency; turn-taking contingency; turn-taking quantity/conciseness; lexical selection specificity/accuracy; lexical selection cohesion; and intelligibility. This occurred on only two behaviours for the hearing children: turn-taking interruption/overlap; turn-taking feedback to speakers. Thagard et al. (2011) investigated the pragmatic skills of a cohort of 81 children with hearing loss in grades K-8 in regular education settings in the United States. They found that regardless of the communication mode used, better pragmatic skills were associated with better academic outcomes. It should be noted that findings such as this could be confounded by many factors, including children's level of language skill.

Several studies have focused on the functions of language used in social interactions by children with hearing loss. Dammeyer (2012a) adopted a longitudinal perspective, examining the pragmatic development of three 5-year-old children with cochlear implants until they were 8 years old. In this study, the three children were observed playing together several times a year, and their interactions were quantitatively and qualitatively coded. Across the 4 years the children rarely engaged in communicative repair strategies. There was no change in the number of intelligible words produced in each sample over the 4 years of observation, although parent ratings of all children's speech intelligibility increased. Poor speech intelligibility was related to difficulties with maintaining verbal interactions and responding to others.

Nicholas (2000) investigated the development of communication acts in children aged 12–54 months of age, with a focus on informative/heuristic functions of language. Informative/heuristic functions of language represent a higher level of intentional communication, where the purpose of communication has developed from instrumental functions, such as giving directions, into informational or heuristic types, such as receiving and providing information. The use of intentional communicative acts significantly differed for children of different ages (older children used more) and for hearing status (hearing children used more). Regarding the communicative function of these intentional communicative acts, the communication of the children with hearing loss was more often categorised as non-informative (e.g. directive, repetition/imitation) from 12 months through to 54 months than their hearing peers. The children with hearing loss also used far fewer functions from the information-bearing categories (e.g. statement, response, question) than their hearing peers in the early stages of development, from 12 to 30 months. Overall, the proportion of intentional communication acts that performed informative/heuristic functions was significantly higher at all ages for hearing children and the variance across participants reduced more quickly for the hearing children. The percentage of intentional communication acts that performed informative/heuristic functions was also significantly correlated with children's skills in other areas of language for children with and without hearing loss.

Good pragmatic skills are necessary for being able to conduct a conversation smoothly and effectively. This aspect of pragmatics includes conversation skills, communication breakdown, and communication repair. This area of pragmatics has increasingly been a focus of research as the spoken language skills of many children with hearing loss have improved. In terms of the use of pragmatic skills to initiate, maintain, and repair conversations, research points to both areas of strengths and difficulties.

Lederberg and Everhart (2000) examined the conversational interactions of 20 dyads of children with hearing loss and their mothers. The dyads were observed longitudinally when the children were aged between 22 and 36 months. Compared to hearing peers, children with hearing loss were less skilled in maintaining the topic of a conversation, asked fewer questions, gave more instructions, and produced more utterances in which the pragmatic function of the communication was unclear. On the whole, the mothers of the children with hearing loss and typically hearing children behaved similarly, with the authors attributing differences to the delayed language skills of the children with hearing loss. Toe et al. (2007) reported some similar findings in their investigation of the conversation skills of 18 children aged 6–16 years with hearing loss while participating in a 10-minute conversation with a familiar teacher. It should be noted that all children used spoken language to communicate, but there was also an overall average language delay of two-and-a-half years within this group. In this study, like in that of Lederberg and Everhart (2000), the children asked few questions. The younger children, compared to the older children, also rarely initiated conversational turns.

The difficulty of children with hearing loss in identifying and repairing communicative breakdowns has often been reported. Jeanes et al. (2000) investigated the skills of 40 children aged 8–17 years with profound hearing loss who used spoken or signed communication (Signed Australian English). Children participated in a barrier task where they had to describe shapes and diagrams to a hearing peer. The focus of the investigation was the requests for clarification and responses to requests for clarification of the children with hearing loss. The group who used signed communication made significantly fewer specific requests than the children who were hearing or children with hearing loss who were spoken language users. Both groups with hearing loss made more non-specific requests for clarification than the hearing group. In terms of providing appropriate responses to requests for clarification, there was a significant difference in the number of appropriate responses provided by the signing and hearing groups, but no significant difference between the other groups. Also, while the hearing and spoken language groups utilized similar repair strategies, the signing group was much more likely to use less effective repair strategies, such as repeating the misunderstood utterance, which was a strategy rarely used by the other groups.

Tye-Murray (2003) investigated the skills of 181 children aged 4–5 years with cochlear implants in holding a spoken language conversation with a clinician. She reported that compared to their hearing peers, the children with hearing loss spent much more time in silence and experienced significantly more communication breakdowns. These experiences were more frequent for children with hearing loss who were not enrolled in oral-only education environments (i.e. in education environments were signed communication was utilised) and/or those who experienced poorer speech intelligibility and receptive language skills.

However, difficulties with communication breakdown and repair are not always the case for children with hearing loss. Conversational breakdowns were rarely observed by Toe et al. (2007), which the authors attributed to both the functional language skills of the group and the fact that the teacher and the child in each dyad were familiar with communicating with each other. Ibertsson et al. (2009) found in their study of teenagers with cochlear implants that these children produced significantly more requests for clarification than their hearing peers, although they confirmed previously presented information and requested elaboration less often.

Church et al. (2017), however, report that difficulties in conversation were only evident for the 10 children in their study who used cochlear implants, when analysis went beyond the use of broad measures such as initiation, turn, and clarification requests. Using conversation analysis techniques, Church et al. (2017) found that there were largely no differences in the self- and other-initiated repair strategies used in conversations between children with hearing loss and a hearing peer that they themselves selected as a conversation partner (i.e. a friend). However, Church et al. (2017) identified many instances in which the children with hearing loss engaged in “open class other-initiated repair” (p. 54), meaning that they “raised some problem with what the other child/speaker said (other-initiated repair), but did not specify what the problem was” (p. 54), such as by saying “huh?”. Such interactions were associated with both a failure to hear the previous utterance of the conversation partner and failure to understand the previous utterance of the conversation partner. They also identified instances of where communication breakdown was not addressed by either peer in the interaction, leading to poorer engagement and continuation of the topic of discussion. This situation was never observed where hearing peers were in conversation, as they always identified and addressed the source of the breakdown.

Using an experimental design imitating a game of Trivial Pursuit, Marschark et al. (2007) and Toe and Paatsch (2010) examined the communication breakdown and repair strategies of two groups of children with hearing loss. Marschark et al. (2007) examined college-aged students with hearing loss, comparing dyads of students who both used sign language, both used spoken language, or one used signed and one used spoken language. Toe and Paatsch (2010) examined dyads of 7- to 12-year-old children where each dyad contained a child with hearing loss who used spoken language and a hearing peer matched by gender and grade level. In both experiments each participant was required to read/sign a question aloud, and the partner was to repeat the question verbatim, ask for clarification if required, and answer the question. Marschark et al. (2007) reported that across all groups, requests for clarification (i.e. identifying and acting to repair a communication breakdown) were rare and that the sign language dyads understood more questions than the spoken language dyads. Overall, there was poor comprehension across groups and difficulties in comprehension were rarely addressed in an effective manner. Toe and Paatsch (2010) reported that the children with hearing loss required significantly more repetitions of the questions, but sought clarification more often than the children without hearing loss, and in fact answered more questions correctly than their peers without hearing loss.

9.2.3.3 Theory of Mind and Higher-Level Language

Theory of Mind (ToM) is a key aspect of social cognition. It describes an individual's ability to understand that others have mental states such as knowledge, desires, emotions, intentions, and beliefs that influence their behaviour and that may vary from one's own knowledge, desires, and beliefs. In relation to pragmatic skills, ToM skills play an important role, along with language, in being able to engage with others and understand their mental states through social communicative acts. While difficulties with ToM were traditionally considered a hallmark feature of autism spectrum disorder (ASD), many groups of children with different profiles in terms of social and linguistic skills have now been shown to experience differences, and often delays, in their ability to pass traditional measures of ToM development, such as perspective-taking and false-belief tasks.

Studies investigating ToM in children with hearing loss began in the 1980s, with the general findings of studies in the 1980s and 1990s being that children with hearing loss who had hearing parents demonstrated significant delays in ToM development (Morgan, 2015). One such study was that of Peterson and Siegal (1995) who examined the ToM skills of children with profound hearing loss aged 8–13 years who used sign language. These investigators used a false-belief story paradigm. They found that 65% of the children with hearing loss could not pass the task which was typically mastered by children without hearing loss at 4–5 years. Since this time there has been much interest in investigating ToM in children with hearing loss, particularly because ToM can be considered to exist at the junction of language and cognition, and children with hearing loss represent a population with a wide variety of language skills without the co-occurrence of cognitive impairment. The heterogeneity of children with hearing loss as a population has been reflected in the diverse findings of ToM experiments.

Language ability has often been found to be related to the ability to pass ToM tasks. However, it must be remembered that language ability does not stand alone as a single skill but can be a mediator for other factors that could impact on ToM development such as skills and experience in socialization, conversation, and executive function (Hutchins et al., 2017; Marschark et al., 2019). ToM can also be a consequence of factors related to language access, such as parental hearing status, age of access to language, and degree of hearing loss. Better skills in vocabulary (Hutchins et al., 2017; Fujino et al., 2017), syntax (Jones et al., 2015; Most et al., 2010), and pragmatic skills (Hutchins et al., 2017) have all been frequently associated with more advanced ToM skills. Having deaf parents, that is, parents who are fluent users of a signed language, has also been associated with an absence of delay on ToM tasks compared to hearing peers (e.g. Woolfe et al., 2002; Schick et al., 2007), supporting the argument that it is not deafness *per se* that influences the development of ToM. Again, possibly related to language skills, children who receive cochlear implants at younger ages have also been shown to have less delay in developing ToM skills (Sundqvist et al., 2014; Hutchins et al., 2017). Differences have also been attributed to the assessment measures used (Hutchins et al., 2017; Peterson et al., 2005), but this will not be further discussed here.

In summary, children with hearing loss are at risk of pragmatic language difficulties. Issues can be identified from early in the child's life and throughout childhood and in all aspects of pragmatic language. Social, cognitive, and structural aspects of language are affected. However, children with hearing loss can often be supported successfully in their pragmatic language development. For some, the best approach is visual communication starting early in life with visual strategies in the social interaction with caregiver and, at a later point, the learning of sign language. For others, it is about supporting spoken language development with the use of technology (hearing aids or cochlear implants) and intensive, early, and ongoing rehabilitation programs in using spoken language.

9.3 Children with Vision Loss

9.3.1 Introduction to Vision Loss

Vision loss is the limitation of one or more functions of the eye or the visual system. It includes impairments in the clarity of vision (acuity), the range of what is seen (visual field), and the perception of light and colour. The World Health Organization (2018b) estimates that globally there are 1.3 billion people living with some form of vision loss. Most of these people are over 50 years of age. Vision loss is extremely heterogeneous in terms of the part/s of the visual system that are impaired, the functional impact of the impairment, and the age at which the loss occurred. As with hearing loss, vision loss can be either congenital or acquired. Here, we will focus on congenital vision loss or vision loss that is acquired in early childhood.

There are many different schemas for classifying degree and type of vision loss, residual visual function, and impairment of peripheral or cerebral vision structures. Visual acuity is often indicated as a fraction, for example 6/18 (or 0.3 or 33%), which means what a person with no vision impairment can see from the distance of 18 meters, a person with vision impairment can see at 6 meters. 6/6 (or 1.0 or 100%) is no vision impairment, 6/18–6/60 is moderate impairment, 6/60–3/60 is severe impairment, and worse than 3/60 is categorized as blindness. Blindness is often further categorized into those with or without light perception and the ability to detect form (Greenaway & Dale, 2017). For visual field, degree is used. A visual field of a minimum of 60 degrees either side of the vertical meridian and 20 degrees above and below horizontal is, in some countries, the requirement for driving.

The aetiologies of childhood vision loss are numerous and diverse. Many aetiologies are associated with other impairments or disabilities. Globally, the most common aetiologies of significant vision loss in childhood are cataracts, cerebral dysfunction, corneal scarring, glaucoma, and retinal disorders, although the principal causes of vision loss vary greatly across regions (Solebo & Rahi, 2014). Vision loss can be linked to genetic syndromes, illnesses such as diabetes, or caused by infections, intoxication, or other damage to the visual system. Significant vision loss in childhood is a low incidence disability. Estimates from the UK report an annual cumulative incidence of 6 per 10,000 children (birth to 16 years), with the highest incidence occurring at birth (Solebo & Rahi, 2014). While vision loss is quite rare among the general population, it is quite common among children with disability, with around 75% of children with a diagnosed disability (e.g. neurological disorder or genetic syndrome) also having vision loss (Rahi & Cable, 2003).

9.3.2 Communication of Children with Vision Loss

While the impact of hearing loss on the development of communication is obvious for children with hearing loss, the impact is subtler for children with vision loss. The development of communication and language is based on children learning how to understand and interact with the physical world, and the cognitive skills that develop as part of this understanding and interaction (Tadić et al., 2010). The inability to access the visual world means that children with vision loss often have differences in their development of motor skills, mobility, exploration, play, and social interaction, all of which interact with communication and language development (Bathelt et al., 2019; Greenaway & Dale, 2017).

There are many early developmental and cognitive skills that sighted children acquire prior to, or simultaneously with, communication and language, such as joint attention, turn-taking, emotional responsiveness and recognition, and social interaction (Greenaway & Dale, 2017; Herrera, 2015; Tadić et al., 2010). For children with vision loss, the inability to access and interact with the physical world in the way that sighted children do means that development of these foundation skills can occur more slowly and/or differently, with children with vision loss showing a high likelihood of developmental differences, delays, difficulties, disorder, and deceleration (Dale et al., 2019). For example, turn-taking is more difficult to learn when the child's participation cannot be supported by facial expressions and body movements. Another example is joint attention. Without the ability to watch an adult looking at a toy car and pointing to a toy car, it is more difficult for the child with vision loss to link the word "car" to the object. More attention to use of tactile cues and extra time are often needed to successfully establish joint attention. Disrupted access to visual aspects of communication has greater implications for the development of pragmatic language than for the development of structural aspects of language, such as phonology, morphology, semantics, and syntax. Most non-verbal human communication is based on vision, such as facial expression indicating if a statement is sarcastic, and body movement showing a person's intention or attitude. It is, therefore, much more difficult for children with vision loss to learn how to read other people's intentions and "between the lines" messages.

Idiosyncrasies in the development of structural elements of language have been identified as common in children with vision loss (for a detailed review, see Sak-Wernicka, 2017). The types of language elements involved are most often related to complex language constructs that describe elements of the physical world which are inaccessible to children with vision loss. An example is deictic pronouns ('I' and 'you'), which children with vision loss have difficultly acquiring accurately as the meaning of the pronoun changes depending on the context. If the child says "I am eating an apple" and the father replies "Yes, you are eating an apple" the 'I' and 'you' refer to the same person. If a sibling then says "Why does she get an apple and I don't," the 'she' now refers to the person that 'I' and 'you' referred to previously and 'I' refers to a new person. Without access to the visual context which provides gaze, pointing, and clear information about who actually has the apple and who does not, it is much more difficult to grasp this type of language, making the pronouns involved in describing this confusing for a language learner. Therefore, many, but not all, children with vision loss show difficulties with pronoun use (Pérez-Pereira, 1999). Equally, children with vision loss have been reported to have difficulties with applying language to concepts such as colours, aesthetic terms (e.g. beautiful), spatial terms (e.g. behind), quantity (e.g. more), time (e.g. today), size (e.g. long), qualities (e.g. old), and other deictic terms (e.g. there) (Sak-Wernicka, 2017).

In terms of development of structural language skills, some children with vision loss (without any additional disabilities or developmental concerns) may develop strong language skills, in some cases superior to those of their sighted peers. Because these children have restricted access to non-verbal communication, they must rely on structural language use and, as a result, some might develop better skills in this area. It should be noted, however, that research describing the language outcomes of children with vision loss without additional disabilities and intelligence within the typical range is rare. Tadić et al. (2010) sought to examine the language skills of older children with congenital vision loss, who had verbal intelligence scores within the normal range. They examined the skills of 15 children with vision loss aged between 6 and 12 years using the Clinical Evaluation of Language Fundamentals-3 (Semel et al., 2000). Children with vision loss outscored their sighted peers on all four subtests (Word Classes; Listening to Paragraphs; Recalling Sentences; Word Associations) and performed significantly better on the Recalling Sentences subtest and Total Language Score.

Peltzer-Karpf (2012) described how the early language of children with vision loss seemed most vulnerable, with delays in phonological learning, lexical acquisition, and morphological and syntactic development. She reported that the developmental trend was similar for children with vision loss and typically sighted children, but that children with vision loss experienced delays initially. Funnell and Wilding (2011) described a longitudinal case study of a child with visual agnosia where difficulties in expressive vocabulary, but not receptive vocabulary, were identified in the preschool years and persisted through the school years. While this is a very positive outlook on the language and communication skills of children with vision loss, it must be remembered that many children with vision loss also have additional needs and that these additional needs often involve disruption to communication and the acquisition of language (Mosca et al., 2015).

9.3.3 Pragmatic Skills of Children with Vision Loss

There is great variability in the pragmatic language skills of children with vision loss, with most children showing marked difficulties in this area (Greenaway & Dale, 2017). Any discussion of the pragmatic skills of children with vision loss must start by considering the close links between vision loss, social communication and pragmatic language difficulties, and ASD. The incidence of ASD is greatly increased for children with a primary diagnosis of vision loss, with the incidence being estimated at 48%, 31 times higher than for children without vision loss (Jure et al., 2016). Part of the higher prevalence is linked to shared aetiologies causing vision impairment and ASD, but in other cases some of the autistic symptoms might be linked to social and communicative barriers related to vision loss itself.

Children with congenital vision loss are susceptible to social communication difficulties that are superficially extremely similar to those displayed by sighted children with ASD (Tadić et al., 2010). Hobson and Lee (2010) followed nine congenitally blind children who were diagnosed with ASD from childhood and into adolescence and showed how improvement in the children's social interaction skills significantly decreased their autistic symptoms. Therefore, the accurate diagnosis of ASD in children with vision loss is complex, a fact that is not helped by the general lack of appropriate assessment tools for diagnosing ASD in children with vision loss (Absoud et al., 2011; Greenaway & Dale, 2017; Parr et al., 2010). This presents some practical difficulties for describing the pragmatic skills of children with vision loss as to which differences in pragmatic skills relate purely to the experience of vision loss and which are associated with ASD. The reason for this overlap may lie in differences in early social interactions and to a lack of access to and/or engagement with the social world that belies the experiences of both children with vision loss and children with ASD. However, here we focus on research describing the pragmatic skills of children with vision loss and no diagnosed additional needs.

9.3.3.1 Early Social Interactions

The underpinnings of social responsivity, social interaction, and social communication lie in early infancy. The absence or impairment of vision in these foundational stages of development may be the source of the difference and difficulties that may follow for children with vision loss. The development of early social communication skills is particularly vulnerable for children with vision loss due to the importance of developing behaviours such as eye contact, facial expression, gaze, imitation, and gesture, all of which are heavily reliant on vision (Dale et al., 2014). Early parent-child interactions typically use vision to develop joint attention, which is another important pre-cursor to social communication (Greenaway & Dale, 2017). The absence of vision tends to make children less responsive and expressive, and less likely to initiate interactions and to share meanings during play. It also tends to make their communicative intent and focus of attention more difficult for parents to interpret (Greenaway & Dale, 2017).

Children's lack of visual access and changes to the way children engage with their parents means that parents need to adopt different strategies to engage with their children, to initiate and maintain interactions, and to share joint attention in ways that do not involve vision. Parents of children with vision loss tend to compensate for the lack of shared visual gaze with additional vocal and tactile behaviours and communication (Dale & Salt, 2008; Greenaway & Dale, 2017). Rattray and Zeedyk (2005) undertook intensive observation of mother-child dyads with children aged between 6 and 18 months and in which the children and mothers differed in their vision status. They identified occurrences of communication through touch, vocalizations, and facial orientation. In the interactions that they observed, infant and maternal touch and vocalisation most often occurred in the context of joint attention. The use of facial orientation in communication in periods of shared attention was, however, more variable and impacted on by the vision status of the child and the mother. In terms of verbal communication, Pérez-Pereira and Resches (2008) examined the early verbal interactions between mothers and children in dyads where the children were either sighted, partially sighted, or blind when the children were aged between 22 and 40 months of age. They reported that the mothers of the children who were blind used more directives and talked more, used descriptors more often to orient the child to a world that they could not see (e.g. characteristics of objects, describing events taking place, locations of objects), and used more strategies to include the child in the conversation.

Developmental setbacks are an additional risk to the early social communication development of young children with vision loss (Dale et al., 2019; Dale & Sonksen, 2002). Developmental setback is defined as the plateauing or regression of skills and has been observed in approximately one-third of children with profound vision loss (Dale et al., 2019). Risk factors for developmental setback include level of vision loss, age, and sex, with the risk highest for children with greater vision losses, children who were male, and children aged between 2–3 years. Such setbacks tend to impact on development globally, and particularly on communication and pragmatic skills (Dale et al., 2019; Dale & Sonksen, 2002).

9.3.3.2 Social Communication and Pragmatic Language

As could be expected, and already mentioned, children with vision loss display difficulties with non-verbal aspects of social communication, such as use of eye contact, gaze following, using and recognising facial expressions, and using gesture (Greenaway & Dale, 2017; Sak-Wernicka, 2017). Greenaway and Dale (2017) pointed out that conversation involves more than an exchange of words but also "tone of voice, facial expressions, eye contact, gesture and posture" (p. 460). Without access to much of this information during the conversation, a child with vision loss has less information about their conversation partner, such as their "emotion, motivation, level of interest and intended meaning" (p. 460). This can negatively impact on their ability to initiate and sustain conversations and minimise communication breakdowns. It has also been suggested that although children with vision loss can access features such as tone of voice, that this alone is not sufficient for making accurate judgements about emotional state (Dyck et al., 2004).

A number of studies have quantitatively compared the social communication and pragmatic language skills of children with vision loss to those of their sighted peers. James and Stojanovik (2007) used the Children's Communication Checklist-2 (Bishop, 2003) to examine the skills of eight children with vision loss aged 12–17 years. As a group, these children showed wide variance in their skills across all subscales (language structure, pragmatics, autistic type behaviours). Over 50% of children achieved scores below the tenth percentile for semantics, coherence, inappropriate initiation, use of context, non-verbal communication, social relations and interests. Tadić et al. (2010) described the structural language and social communication of 14 children with vision loss aged between 6 and 12 years. Children with vision loss scored significantly more poorly than their sighted peers on semantics, inappropriate initiation, stereotyped language, use of context, non-verbal communication, social relations, and interests. While these differences were statistically significant, it should be noted that the mean scores of the children with vision loss were mostly within the normal range, except for non-verbal communication and social relations.

Pijnacker et al. (2012) investigated 24 children aged between 6 and 13 years, also using the Children's Communicative Checklist-2 (Bishop, 2003). The mean scores for children with vision loss were all within the normal range for all subtests. Significant differences between the scores of children with vision loss and their sighted peers were observed only for the subtests non-verbal communication and inappropriate initiation. Together, these studies show a varied picture of the social communication and pragmatic language skills of school-aged children with vision loss without any additional diagnoses. Obviously, these studies all represent small cohorts of children and all showed great variability within their samples, demonstrating the need for further research in this area.

Pragmatic differences in the expressive language of children with vision loss can occur in the form of the use of echolalia and/or formulaic language (Greenaway & Dale, 2017). Echolalia is the non-meaningful repetition of language and has commonly been reported as a feature of the language of both children with vision loss and children with ASD (Greenaway & Dale, 2017). Echolalia has been described in ways that reflect vastly different viewpoints, from "a maladaptive, functionless behavior that should be extinguished or decreased" (Stiegler, 2015, p. 750) to "a functional adaptation that reflects a gestalt learning style and leads to more generative communication" (Stiegler, 2015, p. 750). The echolalic behaviours of children with vision loss have often been considered from the latter viewpoint. From this view the repetition of language in phrases and structures is an important step in language learning and communication. After the repetition has occurred, the child will analyse the word or phrase to determine its internal structures and to identify lexical and syntactic substructures (Pérez Pereira, 2014).

Echolalia and use of formulaic language have also been considered as a means by which children with vision loss can use language to initiate and maintain communicative interactions. Kitzinger (1984) identified a number of different pragmatic functions that repeated and echolalic utterances used in interactions between sighted adults and a 3-year-old child who was blind could have. These included: clarification; confirmation; indication that a response was required; dissatisfaction; agreement; and self-direction. It could be argued that for sighted children, non-verbal communication such as nodding or observing the reaction of the speaker/listener would be used in the context where this child used echolalic or repeated utterance.

9.3.3.3 Theory of Mind and Higher-Level Language

The relationship between ToM and pragmatic skills has been extensively researched for children with social communication difficulties, showing strongly that deficits in one are associated with deficits in the other. The link between ToM and pragmatic skills is thought to be that difficulties with ToM affect the ability to interpret intended meanings (Greenaway & Dale, 2017; Sak-Wernicka, 2016). Traditionally, children with vision loss have been found to have delays and deficits in ToM skills compared to their sighted peers up to 7 years (Brambring & Asbrock, 2010). Reasons considered for these delays include lack of visual co-orientation, difficulties with joint attention, and decreased ability to observe the emotional states of others (Brambring & Asbrock, 2010). It has been debated as to whether difficulties acquiring ToM is due to these children's vision loss or is part of the autistic-like behaviours seen in many children with vision loss. However, more recent thinking has moved away from the classification of this difficulty to suggest that traditional false-belief tasks, the ways in which they have been administered, and the cohorts of children they have been administered to may have overestimated the difficulties children with vision loss have with ToM (Greenaway & Dale, 2017; Brambring & Asbrock, 2010).

Brambring and Asbrock (2010) used false-belief tasks that were more accessible to the 45 children with vision loss in their study. They found that children with vision loss were able to solve first-order belief tasks at an age much younger than previously reported, showing a delay of only 2 years. Begeer et al. (2014) found that the type of vision loss children presented with explained variance in their ToM skills. In this study, the skills of children who had ocular blindness, which involves only the non-retinal parts of the eye, were compared to the skills of children with ocular-plus blindness, which involves the retina and/or parts of the optic tract and/or brain. The children with ocular blindness performed similarly to their sighted peers in this study, while those with ocular-plus blindness performed significantly worse than both comparison groups. The authors suggest that this finding shows it is not a lack of vision that is associated with slower ToM development, but neural factors. Pijnacker et al. (2012) considered second-order false belief tasks in their study of children with different degrees of vision loss, again failing to find differences in the performance of children with vision loss and their sighted peers.

Understanding of mental states is important to development of ToM. Children with vision loss are unable to access visual information about mental states, such as through facial expression, as well as their sighted peers. However, information about mental states may be supplemented in other ways. Tadić et al. (2013) observed the mothers of sighted children and children with vision loss reading a story to their child. They found that the mothers of children with vision loss engaged in more elaboration of the story and made significantly more references to the mental states of the characters in the story than the mothers of the sighted children.

In terms of non-literal language, children with vision loss have been found to have similar skills to those of their sighted peers (Edmonds & Pring, 2006; Pijnacker et al., 2012). Edmonds and Pring (2006) examined the ability of children with vision loss without additional needs to answer literal and inferential questions based on texts that were presented in print/braille or auditorily. The 17 children with vision loss (aged from 7 to 11 years) showed comparable performance to their sighted peers on answering inferential questions, and the children with vision loss outperformed their sighted peers when responding to literal questions from a text presented auditorily. Pijnacker et al. (2012) investigated the ability of 24 children with vision loss (aged between 6 and 13 years) to understand non-literal utterances in the context of a story. The children with vision loss performed similarly to their sighted peers on this task, with an effect of age observed for both groups, such that older children were more accurate in interpreting the meaning of non-literal language.

In summary, structural language learning is affected to a much lesser degree for children with vision loss as compared to children with hearing loss. For children with vision loss, linguistic challenges relate mainly to pragmatic language skills. This can be seen in the delay that many children with vision loss have in passing ToM tests compared to their sighted peers. However, additional disability and assessment artefacts for ToM tasks might explain a large amount of variation in skills, further illustrating that this association is not simple. The pragmatic language difficulties that children with vision loss experience might, for some, be due to additional intellectual disorders and for others due to different experiences in social interaction and language use. From reviewing the literature, it is clear that more research is needed to learn about pragmatic language development in the different subgroups of children with vision loss.

9.4 Children with Deafblindness

9.4.1 Introduction to Deafblindness

Deafblindness, or dual sensory loss, is the combination of vision and hearing loss. While different definitions are used, the degree of impairment of vision and hearing function in this population ranges from functional impairment to total loss of function. Children with deafblindness are commonly considered to form two different subgroups: congenitally deafblind (CDB) and acquired deafblind (ADB). Children classified as having CDB present with congenital impairment of both hearing and vision, while children with ADB were born with intact vision and/or hearing and lost function in these senses either suddenly or progressively, and simultaneously or sequentially.

The causes of deafblindness are extremely varied. Common aetiologies of CDB include infections (e.g. Rubella, Cytomegalovirus) and genetic disorders (e.g. CHARGE syndrome, Cornelia de Lange syndrome), with the most common cause of ADB being Usher syndrome (Dammeyer, 2010b). Deafblindness is a low-incidence disability, especially in children. Reported rates of incidence and prevalence in children vary greatly, mostly due to variation in definitions. Dammeyer (2010b) reported the prevalence of deafblindness in a national sample of children in Denmark. Prevalence was estimated at 1:15,000 when all children with deafblindness were considered, and 1:19,000 when only CDB children were considered. Wittich et al. (2012) conducted a chart review of three organisations providing services for people with dual sensory loss in Montreal, reporting that less than 7% of the cohort examined was aged less than 18 years.

9.4.2 Communication of Children with Deafblindness

Most children who are deafblind, especially those with CDB, will experience severe delays in communication and language development and many do not develop the ability to use symbolic language (Bruce, 2005). However accurate, this is a very broad statement that generalizes a population that is extremely heterogeneous along many dimensions (Ask Larsen & Damen, 2014). As a group, deafblind children vary greatly in terms of levels of impairment, functioning, and additional disability (Nelson & Bruce, 2016). Three especially crucial areas of individual difference that impact on differences in communication and language skills will be discussed here: sensory function; additional disabilities; and communication modality. Firstly, children present with different levels of functional hearing and vision which impacts on their ability to access communication and learn language through vision and/or hearing. As there may be deterioration of function in either or both senses, children's ability to access communication through vision and/or hearing may also decrease over time.

Secondly, deafblindness commonly occurs as part of a larger diagnosis that can involve complex medical conditions (e.g. CHARGE Syndrome), intellectual impairments (e.g. Down syndrome), movement disorders (e.g. cerebral palsy), and developmental disorders (e.g. ASD) (Dammeyer, 2011). The National Center on Deaf-Blindness (2018) reported that for their 2017 national survey in the United States approximately 87% of deafblind children had one or more additional disabilities, and over 40% of children were reported to have four or more additional disabilities. Thirdly, deafblind children use a range of communication modalities. Dammeyer and Ask Larsen (2016) described the communication modality used by a sample of 71 children aged 3–18 years with CDB. They reported that 39% of children communicated using a visual sign language, 32% used an oral language, 23% used a tactile sign language, 18% used idiosyncratic signs, 24% used an augmentative/alternative communication system, and 41% used pre-verbal communication. It should be noted that many children in this sample used more than one communication mode.

As for the level of communication and language proficiency developed by deafblind children, there is again, of course, great diversity. Few studies are available that characterize the communication competence of groups of deafblind children. Data reported by Dammeyer and Ask Larsen (2016) describing 71 children with CDB showed that communication skills fell into three groups. A small group of children did not show any language delay and the majority of these children communicated using tactile sign language, sign language, and/or oral language. However, 41% of children were at a pre-verbal level of communication and 42% had delayed communication. Overall, few children had a vocabulary of more than 60 words/signs. Wiley et al. (2013) reported on the language skills of 91 deafblind children who had received cochlear implants and were aged between 6 months and 8 years. A third (32%) of these children's receptive language skills were at the lowest reported level, which was sound detection, although 22% were at the highest reported level of being able to follow functional directions related to objects. In terms of expressive language skills, nearly half (49%) were at the lowest level (sound production) and only 12% were at the highest level (using complex sentences). Overall, the authors reported that language skills were related to the children's stage of development rather than the age at which they received their implant.

Considering the long-term communication and language development of these children, information is also available about people with CDB and ADB when they reach adulthood. Dammeyer (2010a) described the communication of 35 CDB adults with, and 82 CDB adults without Congenital Rubella Syndrome, reporting that both groups had significant difficulties with communication. Dalby et al. (2009) examined the communication of 88 adults with CDB, finding that only 16% of participants reported that they were understood by others and only 6% reported that they could understand others.

9.4.3 Pragmatic Skills of Children with Deafblindness

As CDB children's communication delays are severe, a lot of the existing research focus on how to support pre-verbal communication by use of tactile strategies. There is very little information available concerning the pragmatic language skills of deafblind children. However, some studies do describe the development of early skills that are built on in pragmatic language development, and these shall be the focus here.

9.4.3.1 Early Social Interactions

Development of the early building blocks for later pragmatic skills is particularly challenging for children who are deafblind, especially those who are CDB. These include the development of a range of communicative functions (e.g. labelling, requesting), turn-taking, and joint attention (O'Neill, 2007; Greenslade et al., 2019) in symbolic and/or non-symbolic communication. Difficulties in developing these skills relate to the restrictions in communicating by use of tactile and bodily languages and decrease or absence of information from the peripheral sensory environment. For example, establishment of joint attention when only the tactile modality is available can be challenging. As well as reducing children's ability to engage in social communicative acts with others, deafblindness also reduces children's access to social and contextual cues in their environment to interpret the meaning of these behaviours in their communicative partners (Bruce et al., 2004).

General comments on the communication of deafblind children that could relate to pragmatic skills are common, however research data specifically describing pragmatic skills is much rarer. Jaiswal et al. (2018) conducted a scoping review examining the participation experiences of deafblind adults. They commented that "adults with congenital deafblindness had limited engagement with others in society and experienced severe difficulties in social interactions" (p. 16). Regarding communicative functions, Rødbroe and Souriau (1999) commented that imperative communication (communication to obtain an object/action) is the function of communication most often used and received by deafblind people and that they often lack the opportunity to engage in communication for other purposes, such as declarative communication (exchanging thoughts/ideas). This lack of access and opportunity is compounded by children using non-symbolic and/or idiosyncratic behaviours and having other behaviours that make interpretation of communicative intent and meaning difficult for a communication partner (Cascella et al., 2015).

9.4.3.2 Social Communication and Pragmatic Language

Despite under-representation in the literature, there is some research concerning pragmatic skills in deafblind children. Here studies are described which examine three different aspects of pragmatics: engagement; communicative function; and communicative repair. Preisler (2005) used videos to examine the longitudinal communication development of six children with severe vision impairment and profound hearing loss, who were aged between 6 months and 3 years at the beginning of the study. Qualitative descriptions of child and parent interactions showed that deafblind children found ways to communicate and socially engage with their parents using facial expressions, movement of their arms, legs, and body, and vocalisations. Parents also communicated and socially engaged with their children, most often using speech, but also by means of vocalisations, signs, and body movements. Preisler (2005) interpreted these data to show that the deafblind children follow the same progression of communication skills as do typically developing children, some of which relate to early pragmatic development: social games and shared attention and actions. Social games were often based on turn-taking and body movement in which movement, touch, and sound were important elements. To different degrees, children were also able to show that they wanted an action performed or a game played. This shows the development of a new pragmatic function as they began to communicate their desires and their desire to interact with their parent.

Again using video recordings, Bruce et al. (2004) examined the range of communicative functions that were realised by three CDB children and their teachers. The children were aged 7, 8, and 12 years and all communicated intentionally at pre-symbolic or early symbolic levels (one- to two-word utterances). Across the 25 different communicative functions observed, there was an imbalance between those used by the teacher and those used by the student. Across all three cases, the functions asking questions, conveying affection, and giving directives were almost always the exclusive domain of teachers. By contrast, protesting, physical cooperation, and calling were almost always the exclusive domain of children. In addition, Bruce et al. (2004) noted that in many instances the teachers often used more than one communicative function within a single utterance, with the children not given the opportunity to process or respond to the different functions separately. The authors suggested that such teacher behaviour could be confusing for the child and may not provide a good role model for the child to model their communicative functions on.

Finally, Cascella et al. (2015) examined communication repair strategies of seven CDB children aged 4–8 years who used speech and sign language. They found that six of the children engaged in communicative repairs using strategies such as repetition, repetition and addition, and repetition and recast. The children who were able to walk independently were reported to also use more advanced repair skills. Repair strategy use was not associated with any of the other participant characteristics examined (e.g. age, joint attention, or communication skill level). The authors suggested that this association may be related to the fact that children who can move independently may have more opportunities to "experience typical discourse patterns" (p. 144).

9.4.3.3 Assessment and Intervention for Pragmatic Skills

Assessment and intervention of pragmatic skills in deafblind children is another area in which there is little research and few evidence-based resources. Assessment of the communication and language skills of deafblind children can be extremely difficult and subjective. Due to sensory impairment, co-occurring conditions, and significant communication impairments, standard measures of communication and language are generally not appropriate for use with deafblind children (Dammeyer, 2016). Further, many deafblind children who have developed intentional communication skills do so through modalities for which few formal assessments are available, such as visual sign language (Crowe, 2021) and tactile sign language (Dammeyer et al., 2015). One commonly used communication assessment for children who are deafblind is the Communication Matrix (Rowland, 2019) which examines communicative function as well as level. Observation of social interaction has also been recommended (Dammeyer, 2012b).

Regarding intervention, it has been clearly stated by many sources that without intensive intervention a child with CDB will not develop communication or language (e.g. Nelson & Bruce, 2016; Bruce, 2005; Dammeyer & Ask Larsen, 2016). Many have also stressed that due to the limited ability of deafblind children to observe and engage with their environment, intervention for children who are deafblind is reliant on direct and individualized instruction by professionals who are experienced in working with deafblind children and who are able to implement the best possible, research-based interventions to meet the needs of the child and their family (Bruce et al., 2016). Aligned with this, the use of visual and hearing assistive devices to increase access to the environment, communication, and language has yielded positive results for some children (e.g. Dammeyer, 2008; Wiley et al., 2013).

Narrowing the focus of intervention to pragmatic communication skills, Luckner et al. (2016) clearly stated that comprehensive intervention programs to address children's communication should include a focus on both the children's early social interaction and pragmatic skills and the skills of communication partners. This begins from the earliest stages of communication development, as early developing skills related to later pragmatic skills have been described as markers of the movement towards the development of symbolic linguistic representations, including joint attention to objects and to others, and imitation (Bruce, 2005; Rødbroe & Souriau, 1999). To this end, the work of Jan van Dijk and colleagues (e.g. Janssen et al., 2003, 2004) in the Netherlands has been instrumental in demonstrating that deafblind children, even those with severe multiple disabilities, can join others in engaging in favoured movements and activities, and that this can be the basis for establishing communication and early pragmatic skills (Nelson & Bruce, 2016). Others suggest deliberate strategies for providing opportunities to model different communicative functions and offer opportunities for deafblind children to communicate for a variety of purposes. For example, Bruce et al. (2004) suggests using sabotage-like techniques, such as forgetting to put juice in a child's cup, to give the child the opportunity to use communication to request or protest in a meaningful context that can be easily understood by all engaged in the situation.

Modifying the communication skills and styles of communication partners has also been used as a means of increasing the impact of intervention for deafblind children. Bruce et al. (2004) noted in their observations of the communicative functions used by teachers with their deafblind students that teachers often expressed multiple functions without pausing for the student to process or respond to each function. They noted that this could confuse children and that a student's opportunity for learning and communicative success would be maximised by expressing one function at a time and giving the child time to process each utterance and respond if they wanted to. Janssen et al. (2003) proposed a diagnostic intervention model which focused on educators' skills in recognising a child's communicative attempts/acts, adjusting their behaviours, and adapting the context of interactions to encourage positive communicative behaviours. In evaluating this approach, they found that the quality of educators' interactions with children greatly improved, and that the deafblind children they worked with also used more interactive behaviours. When attuning to children's behaviours, it has been noted that communication partners should look beyond how certain pragmatic functions look for typically developing children to how they could look for deafblind children. For example, while joint attention is often expressed through eye contact for typically developing children, deafblind children often express this through touch or actions, for example, moving towards a communication partner or placing a hand on the communication partner's body (Bruce et al., 2016).

In summary, functional hearing and vision impairment creates significant barriers for pragmatic language acquisition. Where the child with hearing loss can rely on vision for communication and the child with vision loss on hearing, the child with deafblindness has to communicate by use of the proximal senses such as touch. In general, most children with deafblindness, and especially those with congenital deafblindness, show severe pragmatic language delays. Supporting the pragmatic language skills of children with deafblindness can be challenging, but important, especially in the early and pre-verbal stages. Even pre-verbal social interaction and communication—such as joint attention—can be very difficult to establish for the child and the partner. Accordingly, research shows that few children develop symbolic language.

9.5 Summary

Pragmatic language difficulties are one of the characteristic difficulties of children with sensory loss. Sensory loss creates a situation in which a child is restricted in his/her interaction with the social context and, if not supported appropriately, this can lead to language delay and disorder, especially among those with congenital or early acquired sensory loss. However, it is important to note that pragmatic language difficulties are not an inevitable consequence of sensory loss. In other words, if children are well supported in their language and social development by use of hearing and vision aids, language support programs and/or by use of other communicative means (such as sign language), children with sensory loss will not necessarily experience pragmatic language difficulties. One example is cases of deaf children who have deaf parents who are sign language users. Studies have shown that such children do not show any pragmatic language difficulties compared to children without hearing loss (e.g. Schick et al., 2007).

In contrast, deaf children of hearing parents often do show pragmatic language delays (e.g. Meristo et al., 2007). Though this point has been debated (see Marschark et al., 2019), research findings generally point to children with sensory loss who have been properly supported having a much lower risk of pragmatic language delays and difficulties. However, the best way to support children with sensory loss can be difficult to identify and enact. Finally, it should also be acknowledged that children with sensory loss sometimes communicate differently, and that this does not indicate pragmatic difficulties *per se*, e.g. use of sign language or pragmatic conventions typical to Deaf culture. Research into the pragmatic language use and development of children with sensory loss is an important topic of research, not only to understand how best to support individual children, but also as a source to better understand human languages.

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Chapter 10
Selective Mutism

Gino Hipolito and Maggie Johnson

10.1 Introduction

This chapter will describe the clinical features of selective mutism (SM) along with common co-morbid conditions. It will review the epidemiology of SM and the reasons for the variation of prevalence in SM studies. It will explore the genetic, temperamental, neurodevelopmental and environmental risk factors that contribute to the aetiology of this heterogenous population. The conceptualisation of a disorder not only affects how the sufferers and those around them understand and consequently experience the condition (Johnson & Wintgens, 2015), it also influences assessment and drives the development and use of interventions. An incorrect conceptualisation can lead to inappropriate assessments and ineffective interventions which can waste clinical time and resources, exacerbate the SM, and increase the distress experienced by those living with SM and their families. Therefore, the chapter examines three conceptual views of SM that have emerged in the literature: SM is a variant of social anxiety disorder; SM is a phobia; or SM is a condition with underlying pragmatic language difficulties. The chapter argues that SM is a phobia of the expectation to speak to certain people. It will address the challenges of assessing a child or young person with SM particularly in the area of language and pragmatics. Lastly, the chapter will explore how to differentially diagnose and treat an additional pragmatic language disorder in the SM population.

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10.2 Clinical Features

Selective mutism is classified as an anxiety disorder in the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association, 2013) and the 11th revision of the International Classification of Diseases (ICD-11; World Health Organisation, 2018). According to these diagnostic and classification systems, individuals with SM display the following characteristics:

- A consistent failure to speak in specific social situations where talking is expected (e.g. to a teacher at school or peers in social situations) while the individual talks freely in other situations (e.g. to parents at home).
- This pattern has continued for over a month, not including the first month of a new environment such as school.
- The lack of speech impacts on the individual's education, work or social interactions.
- The failure to speak is *not due* to the limited knowledge of, nor discomfort with, the spoken language required in the specific social situation. Nor is it better accounted for by a communication disorder or condition resulting in transient mutism such as separation anxiety, schizophrenia or a psychotic episode.

In addition, it is recognised that individuals with SM may have a comorbid condition, such as another anxiety disorder or communication disorder, or be exposed to a second language. The co-existing issue will have different characteristics, onset and course to SM, with different implications for intervention, but should not affect the overall management of SM (Johnson & Wintgens, 2016). Steffenburg et al. (2018) argue that their findings point to the need to look out always for symptoms of autism spectrum disorder (ASD) in the SM population, as a dual diagnosis can be overlooked, particularly in girls (Posserud et al., 2018).

DSM-5 gives contradictory advice regarding a dual diagnosis of SM and ASD. Several pointers are provided regarding differential diagnosis. Children with SM generally have normal early development and language skills and, even in settings where the child is mute, social reciprocity is not impaired. It is emphasised that SM should be diagnosed only when a child has an established capacity to speak in *some* situations but not others. In contrast, early development is affected in ASD and the use of language for reciprocal social communication is impaired across *all* settings. Yet, despite the clear differences between SM and ASD, it is stated on page 195 of DSM-5 that SM does not occur exclusively during the course of ASD, suggesting that the ASD condition can account for the unique characteristics of SM.

ICD-11 is clear, however: SM and ASD are discrete conditions which can stand alone or co-exist. In ICD-11, ASD is listed as an 'exclusion' to SM, thereby acknowledging some similarity in symptoms (exclusions serve as a cross reference and help to delimit the boundaries of a category). Exclusions draw attention to the need for caution when making an accurate diagnosis, while still allowing for comorbidity (SM is similarly listed as an exclusion to developmental language disorders).

Kotrba (2015) concurs and argues that the distinguishing feature between SM and other disorders is that the child with SM can talk in some situations, but is prevented from using the same quality or quantity of speech in other situations due to anxiety. Thus, a child could have a dual diagnosis of ASD and SM if they speak in one situation (albeit inappropriately) but not in a more public setting.

The average age of onset for SM ranges between 2.7 to 4.1 years (Viana et al., 2009; Steffenburg et al., 2018). However, it is a concern that children are not referred for treatment until they are much older (Standart & Le Couteur, 2003). Kumpulainen et al. (1998) found that children were being referred between the ages of 6 to 11 years with the average age being 9 years. The researchers suggested that this lag between onset and receiving professional help may be due to the children being perceived as shy in the early years. Cohan et al. (2006) pointed out that the mutism becomes more apparent when children start attending school as they face greater pressure to speak to their teachers and peers.

10.3 Epidemiology

There is limited research on the prevalence of selective mutism and no known incidence of the disorder at present. Prevalence figures for SM range between 0.03% and 2% in the paediatric population (Bergman et al., 2002; Karakaya et al., 2008; Kopp & Gillberg, 1997; Kumpulainen et al., 1998; Sharkey & McNicholas, 2012). This variation is due to the type of study, the age of the children and the different criteria used to diagnose SM. Studies using clinical populations tend to yield lower figures than community populations. This may be due to children with SM not coming to clinical attention because they do not cause disturbance in the classroom and the disorder is not perceived as problematic in the home (Sharkey & McNicholas, 2008).

Generally, SM has a higher prevalence in young children and decreases in adolescence and adulthood. The prevalence is approximately 1 in 140 below 8 years (Bergman et al., 2002; Elizur & Perednik, 2003) and approximately 1 in 550 in population samples up to 15 years (Kopp & Gillberg, 1997; Sharkey & McNicholas, 2012). Sutton (2013) estimates an adult prevalence of 1 in 2400 (0.04%). Johnson and Wintgens (2016) advise that most studies are unlikely to include the ‘low profile’ presentation of SM, that is, children who, despite their anxiety about talking, manage to give minimal responses to people such as teachers. These low profile children meet the criteria for SM as they consistently fail to speak in other situations and show a marked lack of initiation.

Another issue impacting on the variation of prevalence is the different criteria for SM used in studies. Kumpulainen et al. (1998) used DSM III-R criteria (American Psychiatric Association, 1987) which may explain the higher prevalence of 2%. The authors acknowledged that the degree of impairment required for a diagnosis of SM in DSM III-R is vague and that this can affect the prevalence rate. Karakaya et al. (2008) loosely based their criteria on DSM IV-TR (American Psychiatric

Association, 2000) but with a narrower interpretation. They based the SM behaviour on environments (e.g. school) rather than specific social situations. Children with SM may speak to certain people in a specific situation in school (e.g. a peer, one-to-one) but not in another (e.g. small group or whole class). The criteria in the study also assumed that children with SM "cannot establish friendships" which is not a requirement of DSM IV-TR. Consequently, the prevalence was very low at 0.033%. Interestingly, the schools in the study identified 526 pupils as having symptoms of SM (0.82% of the sampled school population or 1 in 122). However, only 21 were diagnosed with SM by the researchers. No reason was given for this significant discrepancy.

Gender and bilingualism are also pertinent to prevalence studies in the SM population. While there are exceptions (Elizur & Perednik, 2003; Karakaya et al., 2008), in most studies SM is found to be more common in girls than in boys (Cline & Baldwin, 2004; Dummit et al., 1997; Kristensen, 2002; Kumpulainen et al., 1998; Steinhausen & Juzi, 1996). Sharkey and McNicholas' (2008) sample had an even higher ratio of girls to boys at 4:1. Elizur and Perednik (2003) found that children with bilingual immigrant backgrounds were four times more likely to have SM than those with monolingual native backgrounds.

10.4 Aetiology

Children with SM are considered to be a heterogenous group with no single identifiable cause. Scott and Beidel (2011) describe SM as having multiple aetiological factors which contribute uniquely as well as synergistically to the development of the condition. Several narrative reviews have summarised risk factors that can lead to SM such as genetic, temperamental, neurodevelopmental and environmental factors (Muris & Ollendick, 2015; Scott & Beidel, 2011; Sharkey & McNicholas, 2008; Steinhausen & Juzi, 1996; Viana et al., 2009). These categories of risk are common for anxiety disorders (Muris & Ollendick, 2015).

Studies have suggested that individuals with SM have a strong family history of SM, shyness and anxiety (Kristensen & Torgersen, 2001; Remschmidt et al., 2001). Stein et al. (2011) found that the common variation in a gene known as *CNTNAP2* has been associated with a risk of SM. This same gene has been correlated with the developmental language delayed component of autism and various forms of developmental language disorder. The study also found an association between the gene and social anxiety related traits (behavioural inhibition and social anxiety) in a separate sample of young adults.

Children with SM often display the features of 'behavioural inhibition' early on in their lives (Gensthaler et al., 2016; Muris & Ollendick, 2015). This temperament construct is defined by the habitual tendency to show persistent fearfulness and avoidance during confrontation with novel people, situations, and objects (Kagan, 1998). Children with behavioural inhibition in their early years show reticence in the presence of unfamiliar adults and lack of spontaneous speech with unknown people. These characteristics describe a child with selective mutism and occur during the typical age of onset of the condition (Muris & Ollendick, 2015).

Many children and young people with SM have comorbid neurodevelopmental conditions such as speech and language disorders, motor delays, sensory processing difficulties, intellectual disabilities and autism (Kristensen, 2000, 2002; Steffenburg et al., 2018). Notably, there is a high occurrence of communication disorders in children and young people with SM ranging between 10–50% in studies (Cleator, 2015). Children with SM also have comorbid ASD at a higher rate than the general population (Kristensen, 2000). In a retrospective study, Steffenburg et al. (2018) found that 63% of those diagnosed with SM in their clinic between 2003 and 2014 had comorbid ASD. It is important to note that this is a clinical sample and a retrospective study. Therefore, it is unlikely that this statistic can be generalised to a community SM population. The presence of neurodevelopmental delays/disorders that co-exist in the SM population may place extra demands on the child during everyday activities. These demands may increase the child's vulnerability to stress and anxiety particularly in a child predisposed with a behaviourally inhibited temperament (Muris & Ollendick, 2015). Moreover, SM may conceal developmental problems, resulting in adults not adjusting demands to the individual's level. This may consequently compound the stress and anxiety within the child.

Edison et al. (2011) observed that parents of children with SM tended to be more overprotective, controlling and granted less autonomy during their parental interactions than those with non-anxious children. This can reinforce the child's behavioural inhibition and limit the opportunities for the child to take risks, particularly communicative risks. This is a common dynamic found in the childhood anxiety literature (Muris & Ollendick, 2015). The transition from home to the preschool or school environment can be anxiety provoking for many reasons such as separation from parents, increased language demands, and increased interaction demands with adults and peers. It is during this period that many children are observed to have selective mutism. It is possible that either the selective mutism behaviour has become more apparent in this environment or this setting may have acted as a trigger for the behaviour.

Despite established links with comorbidity and environmental stressors in the literature, there is no suggestion that *all* children with SM present with any single factor. The factors can, therefore, be regarded as contributory rather than causal (Johnson & Wintgens, 2001, 2016).

10.5 Conceptualisation of Selective Mutism

Selective mutism is considered to be an anxiety disorder (American Psychiatric Association, 2013; World Health Organization, 2018). However, the nature of the link between anxiety and the failure to speak has not been agreed. One challenge is the lack of consistency in researchers' accounts of anxiety levels experienced by children who have SM (Black & Uhde, 1995; Bogels et al., 2010; Yeganeh et al., 2003, 2006; Young et al., 2012). The correct conceptualisation of a disorder influences how the sufferers and those around them understand the condition. It impacts on how sufferers are viewed and treated by others and dictates the types of interventions being developed and used. Incorrect conceptualisations may lead to inappropriate interventions which at best may be ineffective and at worst may maintain or exacerbate the mutism. Bearing these points in mind, we discuss three views of SM that have emerged in the last 25 years.

10.5.1 Selective Mutism as a Variant of Social Anxiety Disorder

Some researchers have suggested that SM is a variant of social anxiety disorder (SAD) rather than a stand-alone condition (Bergman et al., 2002; Black & Uhde, 1995; Bogels et al., 2010). This view has been largely driven by the apparent high comorbidity of SM and SAD (Bergman et al., 2013; Black & Uhde, 1995; Kristensen, 2000), with some studies identifying 100% of SM samples also fulfilling SAD criteria (Chavira et al., 2007; Dummit et al., 1997; Oerbeck et al., 2014; Vecchio & Kearney, 2005). The hypothesis has been rejected by DSM-5 and ICD-11. But it is worth revisiting the debate as it is a common misconception that SM is on a continuum with social anxiety or shyness.

In this view, not speaking is a natural form of social avoidance for younger children who have SAD. Bogels et al. (2010) suggest that SM is a functional behaviour learned as an avoidance strategy, similar to how feigning illness successfully allows children to avoid going to school. Scott and Beidel (2011) further suggest that SM may represent a behavioural variant of SAD where nonspeaking is an avoidance strategy secondary to the experience of extreme anxiety.

There are several flaws with this theory. Firstly, SAD cannot explain all cases of SM as there are children with SM who are not socially anxious. As stated in DSM-5, ‘children with this disorder may be willing or eager to perform or engage in social encounters when speech is not required.’ Omdal and Galloway (2008) interviewed six adults who had SM. They also observed five children with SM and interviewed their parents. Four of the six adults who had SM reported that they had not felt socially anxious from an early age, even though they did not speak. One adult shared, “I joined everything, but I was quiet and different because I didn’t speak... Still, I never felt on the outside of the group.” None of the five children with SM appeared to be socially anxious in the interactions in home, school or kindergarten. On the contrary, the children were highly independent and actively used body language, facial expressions and gestures. Two parents described their children with SM as highly social and eager to meet new people. One parent described their daughter with SM as “very positive, very giggly, usually in a good mood. She seldom gets excluded in the neighbourhood. She almost always participates.”

In Omdal's (2007) sample of adults interviewed, Linda, aged 48, reported that she did not characterise herself as being worried and socially anxious when she was young. Maria, aged 60, moved to another country at the age of 19 and started talking there and discovered that she was actually extroverted and not withdrawn. Roe (2011) asked 10 to 18-year olds with SM to report their perceptions of themselves. Characteristics such as being sociable, talkative and humorous were reported more often than being introverted or shy. In Albrigtsen et al.'s (2016) study, teenage twins described how they were eager to engage in football matches and other group activities where they could participate non-verbally. The self-reported descriptions of adults and young people and observations of children with SM in these studies do not fit the profile of SAD where the individual consistently avoids social situations or endures them with intense fear or anxiety (American Psychiatric Association, 2013; World Health Organization, 2018).

Secondly, the mean age of onset of SM does not coincide with the much older age of onset for children with SAD which is typically between the ages of 11 and 13 (Wong, 2010; American Psychiatric Association, 2013). Most children who develop SM early would not have the cognitive ideation that is core to SAD (i.e. the fear of being judged negatively by others leading to humiliation, embarrassment, rejection or offending others). Thirdly, it is understandable for SM and SAD to share similar symptoms since both anxiety disorders involve social situations. However, similarities between different conditions do not mean they are on the same continuum. Binge eating disorder and bulimia nervosa are both eating disorders involving binge eating, but the differences between the two conditions prevent one being considered a variant of the other. The challenge, therefore, is to establish a clear difference between SAD and SM. It should be led by the distinguishing features when considering a dual diagnosis, rather than focusing on the similarities.

When Walker and Tobbell (2015) interviewed four adults with SM, they acknowledged that there were similarities between SM and SAD but there were also significant differences. They suggest that SM and SAD comorbidity exists because of the low discriminatory power of the diagnostic criteria rather than extant psychological similarities. One adult, Ben, described how his difficulties resulted in avoidant behaviour, "At work I'd just hurry from place to place or try to look busy, so no one would talk to me, you know, small talk? That way I could avoid looking like an idiot, sometimes someone would catch me and I'd try to mutter something at them. I think they gave up after a while." Ben's fear may not be considered to be excessive or out of proportion as would be the criteria for SAD (American Psychiatric Association, 2013; World Health Organization, 2018). Rather, it may be considered to be a reasonable attempt to avoid the distress of undesired mutism.

In Johnson and Wintgens' (2015) paper, Meg blogs, "I have selective mutism but I don't agree that it's extreme anxiety causing it. If anything, it's the other way around, mutism causes the extreme anxiety. It's terrifying not being able to say what you need to say." Ben and Meg describe how their social anxiety was caused by their SM rather than the other way around. In common with SAD they avoided social situations but for different reasons; they feared the expectation to speak, rather than negative judgement. This compliments Omdal and Galloway's (2008) and Albrigtsen et al.'s (2016) findings that social situations did not provoke anxiety if non-verbal communication was acceptable.

Further examples of people's experience with SM link the expectation to speak to an involuntary frozen response. Rachel recalls: "It's like this absolutely horrible feeling when you'd almost rather die than to utter a word in front of certain people. When you're feeling so afraid of it that your whole body feels like it's physically frozen inside, you don't want someone pushing you, because there's nothing worse in the world than the thought of having to speak" (Johnson & Wintgens, 2015). Alison explains, "It's feeling as if your throat is physically locked when you're put in a position in which you are expected to speak and want to speak, but can't" (Sutton & Forrester, 2016). Sharry (2015) writes "When I stood in the classroom for the first time and that anxiety hit me, I had no say in the matter, it just struck. The same happened at every subsequent expectation to speak." None of these examples mention a fear of the social setting, the people they were trying to address or negative judgement, suggesting that their silence was not related to social anxiety. They had been conditioned to fear the act of speaking itself.

What emerges is a clear distinction between research papers that conceptualise SM based on descriptions or observations of children's *behaviour* and those that use self-report. The behaviour of individuals who have SM, SAD or extreme shyness looks very similar when the social situations expect or require verbal communication; reduced eye contact, lack of engagement or total avoidance of the situation (Johnson, in press). It is not until individuals are asked how they feel and what they are trying to avoid, that different drivers become apparent. Only the individuals who have SM explain their behaviour in physical rather than emotional or cognitive terms; they *could not speak*. Significantly, evidence in studies that reveal high rates of SM and SAD comorbidity have relied on the interpretation of parents, teachers or researchers and when children's views have been included they have been limited to completion of standardised questionnaires (Sharp et al., 2007). This creates a skewed and biased view. Understanding of the SM condition is a reflection of the research methods used to investigate it (Walker & Tobbell, 2015). Without the views of those who suffer SM, it is impossible to have a clear and balanced picture.

It is, therefore, suggested that the high rates for comorbid SM and SAD reported earlier in this chapter are regarded with caution, particularly in younger children. Nonetheless, there is a link between SM and SAD that cannot be ignored. As the individual with SM grows older, they are highly likely to develop SAD (Sutton, 2013). Speaking becomes more important in the education setting and peer relationships. Failure to speak can leave the child feeling powerless and susceptible to negative feedback (verbal and non-verbal) from others. Over time, mounting isolation and actual or perceived rejection affect their self-image and sense of being negatively evaluated by others (Johnson & Wintgens, 2015). Just as Meg described earlier, social anxiety is generally secondary to the SM rather than the other way round, and individuals with SM are at risk of developing SAD if their SM is not addressed at an early age (Omdal & Galloway, 2008).

10.5.2 Selective Mutism as a Phobia

An alternative way of viewing SM is as a specific phobia of the expectation to speak to certain people (Johnson & Wintgens, 2001, 2015, 2016). Omdal and Galloway (2008) describe SM as a specific phobia of expressive speech. They argue that SM fulfils the criteria for specific phobia in DSM IV (American Psychiatric Association, 1994). Johnson and Wintgens (2015) demonstrated how SM continued to meet the criteria in the recent DSM-5 (American Psychiatric Association, 2013). SM and specific phobias also share the same aetiological factors and respond in the same way to the same types of intervention.

According to DSM-5, in contrast with SAD where the individual is fearful or anxious about the possibility of being negatively judged, phobias are not associated with a specific cognitive ideation. Most sufferers of SM do not understand why they do not talk. This inability to explain a reaction over which the individual has no control is consistent with the lack of cognitive ideation that characterises phobias. In Roe's (2011) study, the most common response to telling others about having SM was, "I want to talk but can't and don't know why. It's not a conscious choice." Alison reflects, "It's not being able to explain why you don't talk… you don't fully understand it yourself" (Sutton & Forrester, 2016).

Experimental studies have demonstrated that individuals with phobias are hypervigilant in their perception of threat cues, a phenomenon known as attentional bias (Ollendick & Muris, 2015). Leanne blogged in 2016, "Every single person who has SM will have experienced hypervigilance to some degree. When given a psychological evaluation, I was found to have pretty severe levels of hypervigilance which was not surprising to me. For people who have SM, hypervigilance most often occurs when it comes to being extra alert about who is around. I will not be able to talk until I am completely and 100% sure that there is no one around that I am unable to talk to" (Leanne's Selective Mutism Awareness Month Blogs, 2016).

Ollendick et al. (2002) described phobia pathways as being multi-determined with genetic, temperamental and environmental factors (e.g. parental psychopathology, parenting practices and individual conditioning histories) influencing the development and maintenance of childhood phobias. This mirrors the developmental psychopathology framework for SM proposed by Viana et al. (2009) and Johnson and Wintgens's (2016) 'Factors contributing to the development of SM.' Johnson (in press) argues that only by viewing SM as a phobia itself can the development and course of SM be explained within such a heterogeneous population.

This is further borne out by the response of SM and specific phobias to intervention. Firstly, both conditions respond negatively to *pressure* and *avoidance* which Johnson and Wintgens (2015, 2016) describe as 'maintaining factors.' It is well established that a phobic individual must be allowed to face their fear *at their own pace.* Pressure to do so before the individual feels ready will trigger the fight, flight, freeze response. As their anxiety or sense of panic escalates, the individual becomes angry and oppositional, tries to escape or freezes on the spot. All these reactions are seen in SM when the individual is expected to speak, dependent on individual personalities and circumstances. Students may suddenly swear at their teacher in panic, refuse to go to school or stand immobile, unable to talk. The fear they experience strengthens their conviction that the phobic stimulus is a threat.

Unfortunately, avoidance provides great relief which has a similarly negative effect, making it harder to face their fear in future. In SM it is often, but not exclusively, parents who enable their children to avoid the expectation to talk by answering for them (Yeganeh et al., 2006; Manassis, 2009) or excusing them from social events. It is natural to want to reduce a child's distress, so the children's fearful reactions and care-givers' protective behaviours reinforce each other over time. Maintaining factors have more influence in SM than other phobias, because few people recognise SM as a phobia. Pressure is applied in subtle ways that most children thrive on, but as Cline and Baldwin (2004) note, the very efforts made to help children settle into new environments and socialise can actually reinforce and maintain SM.

Secondly, successful phobia interventions all utilise an exposure element. Stimulus fading is often the method of choice (Shabani & Fisher, 2006). This technique gradually exposes an individual to the fear-evoking stimulus. As the individual acclimatises and the fear response fades, the stimulus is increased until it no longer has the same effect. The behavioural strategies used most often in published research for SM intervention in Zakszeski and DuPaul's (2017) review were contingency management, shaping, hierarchical exposure and stimulus fading. Consistent with their view of SM as a phobia of expressive speech, Omdal and Galloway (2008) developed a successful programme of graded in vivo flooding, involving both teachers and parents in school and community settings working towards small goals agreed with the child to help them overcome their fear.

Johnson and Wintgens (2001, 2015, 2016) utilise general strategies to de-pressure communication at school and in the community, alongside a small steps approach involving desensitisation and graded exposure to help children face their fear of talking rather than avoid it. Although a variety of techniques is included in their resource manual, a stimulus fading approach is preferred if circumstances permit, as this usually provokes the least anxiety. The individual is encouraged to talk in a comfortable situation (e.g. alone with their parent) while another person gradually approaches and eventually joins in. The child or young person is first exposed to using their voice *in front of* the new person, then *with* the new person and finally, the parent or other conversational partner withdraws, leaving the child talking comfortably to the new person without support. As with all phobia management, Johnson and Wintgens argue that an essential part of the process is talking openly, calmly and positively to children about their fear, giving them a feeling of control over their anxiety.

Treating SM as a phobia yields positive results. But perhaps the most compelling argument for viewing SM as a phobia is the response from parents and individuals themselves at Johnson and Wintgens' training sessions. The most common feedback is that viewing SM as a phobia finally makes sense of SM behaviour and allows staff and parents to plan a way forward. D. J. Sharry was introduced to this explanation after visualising his SM of 30 years duration as a tiger lurking in the pit of his stomach, ready to pounce at any moment and hold him in the grip of fear. Not long afterwards he wrote to one of the authors, “There has been one phrase I think I’ve repeated to myself every day and that is, ‘It’s a phobia.’ That was a sort of revelation to me. Take today for instance. A work colleague passed on an unwanted box of chocolates. I started unwrapping them but then realised I’d have to ask everyone else in the office if they’d like to have one. I immediately had what I now know is a phobic reaction and my first impulse was to put the chocolates away. But then it dawned on me that I was just avoiding speaking so I said to myself, ‘This is a phobia.’ I spoke and offered the chocolates around.” Sarah-Jane, aged 50, writes in a similar vein: “Sometimes I still freeze. When this happens, I don’t panic – I stop and acknowledge that it is just a reminder of my SM, take a breath and start again.” (Johnson & Wintgens, 2016).

10.5.3 Selective Mutism as a Condition with Underlying Pragmatic Language Difficulties

Pragmatic language ability refers to the use and understanding of language in a social context and involves higher-level reasoning, narrative and conversational skills. The language skills of children who have SM have been investigated in the last 15 years, leading to the suggestion that pragmatic language difficulties may have a role in SM (Cummings, 2014). Cummings also notes that most interventions for SM aim to reduce children’s anxiety about speaking using social-pragmatic skills rather than structural language skills.

Klein et al. (2013) found that their cohort of children with SM had age-appropriate receptive and expressive vocabulary and understanding of narratives. However, 42% of the children with SM exhibited a significant expressive narrative language deficit, even when tested by their parents. Klein et al. (2017) found 68% of the children with SM in their treatment study exhibited a similar narrative language profile. Klein et al. (2013) suggest that children with SM may have an expressive language formulation problem. They hypothesize that these subtle difficulties may make the children anxious due to their perceived inadequacy in generating novel thoughts and converting them into decontextualised language such as narratives. However, they also recognise that SM may affect the development of pragmatic skills rather than the other way round. The more a child avoids speaking, the less likely they are to overcome anxiety in speaking situations, which may exacerbate the SM condition and prevent the practice needed to develop discourse and other higher-level language skills (Klein et al., 2013, 2017; McInnes et al., 2004).

As discussed in Sect. 10.4, children with SM may have comorbid disorders that involve pragmatic difficulties (e.g. language disorders, ASD). However, Klein et al.’s figures are higher than expected, given other findings. Several factors need to be taken into consideration. The high comorbidity rates are partly explained by the fact that Klein et al. used a narrative assessment. Earlier studies often focused on less challenging, lower-level language abilities. In their 2013 study, Klein et al. also recognised that the scores could have been negatively affected by the office setting in which the assessments took place. They suggested that some children with SM would be reluctant to participate in testing in an office setting, despite only their parents being present, and that some would not be as relaxed as they might have been in a more familiar setting. This is borne out by McInnes et al.'s findings (2004) that a group of children with SM scored better with their parents on a story-telling test at *home* than in unfamiliar surroundings. Consequently, Klein et al. could not be certain that they obtained every child's true language ability in their study.

Finally, parents are not necessarily the best people to obtain a true picture of their children's oral narrative skills. The current authors' clinical experience indicates that children who do *not* have SM usually take formal assessment tasks more seriously with a professional. Being assessed by parents is an unnatural situation that goes beyond the boundaries of most parent-child relationships, and children tend to display less perseverance than with their teachers and adults in more formal settings. McInnes et al.'s (2004) findings support this. While the children with SM did better on a narrative task with their parents at home, a group of children who did *not* have SM performed better in terms of narrative length and clause complexity in the more formal *clinic* setting, which presumably conveyed a greater sense of occasion and perhaps reminded the child of being at school. We, therefore, suggest that for children with SM, a parent assessment of narrative (pragmatic) ability is likely to yield better results than that of an unfamiliar professional, but may still provide an underestimate of the child's true ability.

Even with the higher comorbidity rates found in some studies, it is clear that not all children with SM presented with pragmatic difficulties. This rules out the suggestion that a pragmatic impairment underlies SM. However, all these children struggle to communicate in social contexts, so they may *appear* to have pragmatic difficulties. Perhaps what we are seeing in many cases is the effect of anxiety (Cummings, 2014; Manassis et al., 2007).

Klein et al. (2013) acknowledge that most children with SM exhibit a communication *performance* problem rather than deficits in their communication *competence*. Although children with SM presented with significant differences in some language tests as compared to controls, their average scores in the standardised language assessments were still within the normal range (Manassis et al., 2007; McInnes et al., 2004; Nowakowski et al., 2009). When the very nature of SM is a fear and anxiety around speaking, the performance deficit will be magnified the more the child is expected to speak. Tests that require a non-verbal response will provoke the least anxiety, followed by tests requiring a single word response, with tasks requiring a narrative response provoking the most anxiety. Comprehension tasks which require the child to point to the answer, rather than speak, are therefore likely to yield the most accurate results.

This was demonstrated by Klein et al. (2013) where the children performed best with an unfamiliar assessor on a picture vocabulary test which required a non-verbal (pointing) response. At the other end of the scale, assessment of expressive narrative is likely to yield the least reliable results. This appears to be borne out by the finding that children with SM perform better in a single word vocabulary test than in tests requiring longer responses (Klein et al., 2013, 2017). Furthermore, in both these studies, the children performed within the average range overall on a test of narrative comprehension which requires a single word or sentence response, but yielded a below average score overall when required to tell a story. The bias towards poor oral skills suggests a deficit in performance skills, for if they had a pragmatic disorder (impaired competence) we would expect to see a similar deficit in both oral and receptive narrative skills.

It is also of note that in Bergman et al.'s (2013) pre- and post-treatment randomised control trial, the narrative ability of children with SM improved with *behavioural* techniques rather than pragmatic therapy. This again suggests that before treatment, the children's restricted communication behaviour stemmed from their anxiety about talking, rather than a pragmatic disorder.

Johnson and Wintgens's (2016) model of confident talking reflects the effect of anxiety on children who have SM. The resulting performance deficit is captured by their stages of one-to-one interaction (Table 10.1) and the 'bystander' effect. As the child's anxiety increases, spoken language reduces from the free-flowing connected sentences required for pragmatic discourse and higher-level reasoning, to isolated sentences, phrases, single words and, ultimately, silence. Moreover, when talking to a parent, the child is wary of being overheard by others to the point of hypervigilance, as described by Leanne earlier in this chapter. The presence of others, even in an adjacent room, typically prevents children from relaxing and talking freely because, by revealing to a bystander that they can talk, there is more chance of being expected to speak to that person. The current authors argue that either or both of these factors reduce the child's speech output to measured, economical responses and can account for the apparently high level of pragmatic difficulties identified in the Klein studies. In other words, children and young people with SM present with similar social communication difficulties as those with pragmatic language disorder, but for different reasons.

Table 10.1 Stages of one-to-one interaction in the SM population (Johnson & Wintgens, 2016)

Stage	How child presents with a person outside their comfort zone
0	Absent
1	Frozen
2	Participates in shared activity without communicating
3	Communicates without talking (e.g. via gesture or writing)
4	Talks in earshot through parent or close friend
5	Uses voice (this includes reading aloud and rote speech which are not conversational in nature)
6	Responds with single words
7	Responds with sentences
8	Uses connected sentences
Generalisation	Talks freely in groups and public places, initiates and maintains conversation

10.6 Differential Diagnosis of Selective Mutism and Pragmatic Language Disorder

To the extent that impaired pragmatics can be an artefact of the way in which children with SM are assessed, it raises the important question of how clinicians can undertake a differential diagnosis of SM and pragmatic language disorder. This will require an understanding of both the similarities and distinguishing features of the two conditions, together with a range of assessment procedures which are not compromised by speech anxiety. Five areas of shared difficulty are discussed below, together with pointers for differential diagnosis. Suitable assessment procedures are discussed in the next section.

10.6.1 Deficits in or Complete Absence of Speech Acts

Pragmatic language disorder, also known as social communication disorder in DSM-5 (American Psychiatric Association, 2013), is a neurodevelopmental disorder affecting the child's ability to process, analyse and assimilate pragmatic information. Children with pragmatic disorders have impaired communication *competence* – difficulty making appropriate judgements about the use and interpretation of language in a given social context – while children with SM have a person-dependent fear of talking, leading to impaired *performance*. Greetings and initiated utterances are particularly difficult for them, because they invite further conversation. Their inability to override their fear of talking and initiate interaction is often to their detriment, e.g. a child might wet themselves rather than ask to use the bathroom or be reprimanded for something they did not do; a teenager might not apply for a place at college.

Distinguishing features to look for in assessment:

- A detailed case-history will reveal a different pattern of language development. Children with SM typically develop language appropriately within the home setting. Children with pragmatic disorder have never had effective social communication. They develop clear, fluent speech but are often unable to be specific about what they want, need and feel.
- Children with SM display an age-appropriate range of speech acts, but only use them in situations where they are able to speak freely without anxiety, i.e. when alone with people in their comfort zone. For example, they may wait for help at school, rather than ask for it, but at home they do not hesitate to request help when needed. Children with pragmatic disorder display a limited range of speech acts across *all* settings. It is the formulation of the speech act which causes difficulty, regardless of who is present.
- The difficulties experienced in the classroom by children with SM are practical rather than linguistic in nature and can be addressed relatively easily via practical

means. For example, they can ask for help via the friends they talk to in private. Children with pragmatic disorders require specific teaching around communicative intent and social interaction rather than a change of communicative partner.

- With pragmatic disorder, the child's verbal communication is usually inappropriate rather than absent. For example, they may go straight to stating what they require without a preliminary greeting or conversational opener to set the scene for the listener. Children with SM fail to talk when out of their comfort zone.
- Children with pragmatic disorder have no difficulty expressing themselves by taking action or vocalising. For example, they might help themselves to cake, push a child who is about to take their toy, laugh aloud at a TV programme or cry out in annoyance when they cannot have what they want. Children with SM hate to draw attention to themselves as this often leads to conversation or questioning. Consequently, they often remain immobile and passive, rather than spontaneously taking action or using non-verbal means to indicate their needs. They may be completely silent with certain people, unable to laugh or cry aloud, even when in great pain.
- When there is clearly no pressure to talk, children with SM can relax and communicate effectively through other means (e.g. gesture, writing, picture symbols). By using an alternative communication system, they are able to avoid talking and control their anxiety. Children with pragmatic disorders find it very difficult to communicate without speaking. Quite apart from their lack of pragmatic knowledge, they would need to actively suppress their natural urge to speak – an urge which children with SM do not have outside their comfort zone. Just as a person with a phobia of dogs needs no will power to stay away from dogs, children with SM need no will power to stay silent. On the contrary, they may be willing or eager to perform or engage in social encounters when speech is not required (e.g. nonverbal parts in school plays) (American Psychiatric Association, 2013).

10.6.2 Poor Conversational Skills

The expectation to talk to certain people triggers a panic reaction for children with SM so they do not attempt to initiate or maintain conversations. They are often too tense to smile, nod, point, write or vary intonation, and avoid making eye contact when they fear this will invite a question or conversation. Children with pragmatic disorders have not worked out the *rules* of effective communication, e.g. how to take turns in conversation, use intonation to convey sarcasm or use eye contact to ensure you have someone's attention before speaking to them.

Distinguishing features to look for in assessment:

- Children with pragmatic disorder display poor conversational skills across all settings. Children with SM display age-appropriate conversational skills, both verbal and non-verbal, with people in their comfort zone.

- When outside their comfort zone, children with SM cannot respond reciprocally with an even balance of comments, questions and responses, but may minimally respond with one or two words if the other speaker leads the conversation through questioning. They usually have more success reading aloud, as this does not involve a conversational partner; the good reader can focus purely on producing voice and getting to the end of the passage. However, body tension, flat intonation, reduced volume and strained voice quality (sometimes a whisper or nasal tone) will reveal their high level of anxiety about talking. Children with pragmatic disorder do not display the same body tension, whispering or altered voice quality. They may miss conversational cues and say nothing when a response is expected, or dominate conversations with their own choice of topic. Flat or inappropriate intonation and incongruous pause breaks reveal their lack of understanding when reading aloud.
- Children with SM converse freely with certain people but stop or lower their voices and become visibly tense when they register the presence of someone outside their comfort zone. This may also occur at home in the presence of close friends or second-degree relatives. Children with pragmatic disorder are not wary of being overheard when engaged in conversation and do not display sudden changes in body language, volume or verbal output when aware of bystanders.

10.6.3 Difficulty Answering Higher-Level Verbal Reasoning Questions

Children with pragmatic disorders have impaired verbal reasoning. Their use and understanding of both spoken and written language is affected by difficulty using contextual clues to follow implied meaning, draw conclusions and make inferences. Children with SM generally have adequate language skills but find higher-level verbal reasoning questions difficult because more speech is required to provide a satisfactory answer. Their anxiety increases when longer explanations are required and when additional reasons may be needed to support their answer, e.g. when discussing feelings, when ambiguity or alternatives exist with a risk of getting the answer wrong, and when the listener might hold a different opinion. They prefer simple factual questions that can be answered with a single word or short phrase, effectively closing the conversational exchange.

Distinguishing features to look for in assessment:

- Children with SM may not always answer higher-level verbal reasoning questions or non-factual questions which require guesswork, e.g. ‘What could he be thinking?’; ‘What might she say next?.’ However, most children with SM would be able to answer these questions in a comfortable situation, e.g. at home with parents. Children with pragmatic disorder do not present this contrast.
- Children with SM who have good literacy skills can perform better when they write their answers to questions requiring higher-level reasoning skills. Children

with pragmatic disorder have problems with reading comprehension and display verbal reasoning difficulties in both the written and spoken word.

- Children with SM may appear to have difficulty discussing abstract ideas such as feelings but can do so when the need to talk is removed. They are able to share such information using questionnaires such as Johnson and Wintgens (2016) ‘All about me,’ ‘Worrying thoughts’ and ‘Reactions of family/friends/staff,’ and can rank or rate statements to show the extent to which they agree with them.
- Children with pragmatic disorder struggle with certain verbally-biased activities, regardless of whom they are working with. In contrast, children with SM may be able to complete an activity if paired and sensitively positioned with a friend they feel comfortable with.
- Children with pragmatic disorder have a literal understanding of language. Their parents will be able to provide examples and are often aware that they have to provide very explicit instructions to avoid misunderstanding. Parents of children with SM usually report that there is nothing wrong with their child’s language or understanding. Their sole concern is that there are very few people that the child talks to.
- Both children with pragmatic disorder and those with SM may be able to read aloud to their teacher. However, they will typically fail to answer questions about the text, particularly questions that involve inference and reasoning (see stages 5–8 in Table 10.1). The child with pragmatic disorder will present the same difficulties when questioned by their parent and show a lack of narrative cohesion in their written work. In contrast, the child with SM will do well with their parents and display age-appropriate literacy skills in their written work.

10.6.4 Poor Narrative Skills

Children with SM need to be completely free of their fear of talking before they can speak with ease to a particular person and produce a cohesive detailed narrative. Talking to parents will be equally anxiety-provoking if they are concerned about being overheard or recorded. Children with pragmatic disorder lack the necessary verbal reasoning and listener awareness to produce cohesive narratives.

Distinguishing features to look for in assessment:

- Parents of children with SM will report no difficulty following their children’s narratives, e.g. accounts of an event at school or a film they have seen, whereas parents of children with pragmatic disorder will have to ask questions to establish the sequence of events.
- Pragmatic assessments will reveal significant qualitative differences. Children with SM will tend to omit details within an overall grasp of narrative structure, while children with pragmatic disorder will do one or more of the following: give too much detail, recall events in the wrong order or out of context, use

co-ordinating and subordinating conjunctions inappropriately, combine semantic errors with relatively sophisticated vocabulary.

10.6.5 Avoidance of Social Situations

The children's difficulties may lead to negative experiences of social interactions, which they consequently try to avoid. For example, children with pragmatic disorders lack the skills of negotiation and rapid two-way or group conversation that rule playground politics with their peers. They may, therefore, opt out, preferring to play with younger children, animals and adults who do not challenge their language skills in the same way. Children with SM fear more than the extremely unpleasant sensations of panic that are triggered by the expectation to talk. They experience an extra layer of social anxiety regarding the possible consequences of *not* talking – they might be told off, laughed at, questioned or suffer the humiliation of everyone staring at them. This anxiety can be eliminated through avoidance, and it is not uncommon for children with SM to act out of character in order to avoid the situations they dread. Research links their oppositional behaviour to communication anxiety rather than to an underlying oppositional disorder (Sharp et al., 2007).

Distinguishing features to look for in assessment:

- Children with pragmatic disorder will want to avoid linguistically challenging interactions and activities where they feel excluded. The pattern of avoidance relates to the type of activity and their understanding of how to participate. Children with SM display a pattern of avoidance that relates to the people present and whether or not they will be expected to talk.
- With pragmatic disorder the child may not enjoy certain situations but they do not display the signs of extreme anxiety displayed by children with SM, i.e. body tension and reduced motor movements, frozen facial expression, inability to speak.
- Provided the difficulty speaking has been openly discussed with the child in a supportive manner, children with SM can be very articulate and specific about why they do not want to attend certain events. For example, they may hate being expected to say 'Hello' and 'Thank you' to their grandparents. Children with pragmatic disorder struggle with reasoning in general and find it hard to give focused explanations, despite their apparently well-structured and clearly articulated expressive language.

10.7 Assessment

In view of the risk of comorbid developmental and anxiety disorders in children with SM, a comprehensive multi-modal assessment which includes a speech and language therapy assessment has been recommended (Dow et al., 1995; Manassis et al., 2007; Shriver et al., 2011; Wong, 2010). However, being assessed by strangers can be extremely anxiety provoking for children with SM, and it is important to consider that scores from formal assessments tend to underestimate their ability (Kotrba, 2015). Indeed, if not carefully and sensitively managed, there is a risk that attempts to secure a spoken response through direct assessment will further reduce children's willingness to engage with adults outside their comfort zone (Cleator & Hand, 2001; Cline & Baldwin, 2004).

Johnson and Wintgens (2001, 2016) argue that since children with SM have anxiety around speaking, it is best to avoid putting them through the stress of formal assessment unless it is deemed necessary. They suggest two levels of assessment: core and extended. The core assessment requires minimal interaction with the child and is usually sufficient to make a diagnosis of SM and to rule out the existence of comorbid conditions. If comorbid conditions, such as pragmatic language disorder, are suspected from the core assessment, an extended assessment is required, working directly with the child.

10.7.1 Core Assessment

The core assessment of SM for children and young people involves gathering information from significant adults and observation of the child interacting with others in order to establish the child's speaking habits and possible maintaining factors, gain an overall picture of the child's social and emotional profile and ascertain the general levels of the child's language, learning and development. Experienced observers will not be reliant on standardised assessments to confirm age-appropriate pragmatic skills. Supplementary information can be gained from older children and young people themselves, but this is generally done after a diagnosis of SM has been made using the core information.

10.7.1.1 Speaking Habits

The child or young person's speaking habits will confirm a diagnosis of SM, provide a baseline description of speech behaviour and inform targets and approach to intervention. They are established through the reports of parents, carers and school staff and observations of the individual in different situations, using what Shriver et al. (2011) coined the 4 Ws:

- **W**here, **W**hen and **W**ith whom does the child speak?
- **W**hat form of communication does the child use? For example, does the child use gestures, writing, sounds, whispering or short responses?

The child's speaking habits are recorded using a format such as the Summary Grid of a Child's Speaking Habits across Settings (Cline & Baldwin, 2004) or the Record of Speaking Habits (Johnson & Wintgens, 2016). The latter emphasises the situational nature of SM by including three different contexts: speaking one-to-one; speaking one-to-one in the presence of bystanders (public places); and speaking as part of a group. It is the marked contrast between what parents and people outside the child's comfort zone see that differentiates SM from other communication difficulties and disorders.

The impact of the child's restricted pattern of talking in different settings can be measured using the Selective Mutism Questionnaire (SMQ)© and School Speech Questionnaire (SSQ) with parents and staff, respectively (Bergman, 2013). These questionnaires provide a baseline measure of the extent to which the mutism interferes with the child's education and relationships and can be used to evaluate progress.

10.7.1.2 Maintaining Factors

Maintaining factors are behaviours and events that reinforce and maintain SM. Examples of maintaining behaviours include pressure to speak (through questioning, persuasion, bribery, praise, demands, or expressions of anxiety, disappointment or disapproval), over-correction of the child's speech, speaking for the child and comforting the child when they withdraw (as opposed to reassuring them and supporting them to participate).

It is important to identify and address the family and school's role in strengthening and prolonging the mutism (Bergman et al., 2013; Cline & Baldwin, 2004; Oerbeck et al., 2014) for if the maintaining factors are not eliminated or significantly reduced, SM will take much longer to resolve (Johnson & Wintgens, 2016). Johnson and Wintgens (2016) provide two questionnaires to assist in identifying maintaining factors: the 'Checklist of possible maintaining factors' for the child's teachers and care givers, and the 'Reactions of family/friends/staff' which is completed by older students.

10.7.1.3 Gathering Information from Parents and Carers

To develop a clear view of the child's presentation, contributing factors and possible ways forward, the clinician gathers the following information from parents and carers:

- The presenting problem
- The child's speaking habits
- The family context
- The child's strengths and interests
- Screening questions about the child's general development, speech and language development, social communication skills, motor development, sensory issues, learning, anxieties and behaviour. This will indicate whether further assessment or onward referral is required. For specific questions to screen for pragmatic difficulties, see Table 10.2.

Table 10.2 Indicators of appropriate pragmatic language development

Area	Examples of suitable screening questions and considerations
Communicative functions	When your child is with people they talk to easily, does he/she communicate for a variety of reasons, e.g. to greet them, get their attention, comment on interests, request or reject things and express emotion?
	For 5 years and older: Can your child explain how to play a new game? Can he/she tell you about something that just happened or a story of a book, film or TV programme?
	Does your child ask for help if he/she is having difficulties with an activity?
	For 7 years and older: Can your child tell and understand jokes with double meanings, e.g. “what do dogs do when watching a DVD?” “they press paws”?
	For 9 years and older: Can your child use and understand sarcasm, e.g. “I’m so happy my teacher gave me all this homework just before the holidays”?
Response to communication	When your child is with someone he/she feels comfortable with, does he/she respond appropriately to requests and comments?
	For 5 years and older: Does he/she respond appropriately to indirect requests, e.g. “I need help with tidying up”?
	For 7 years and older: Does he/she respond appropriately to abstract language, e.g. idioms such as “get your skates on” and “keep an eye on your sister”?
Conversation	When your child is with someone he/she talks to easily, does he/she initiate conversation or activities with them?
	When a conversation of interest starts, can he/she keep it going or does the conversational partner have to direct it?
	For 5 years and older: If your child is trying to tell you something and you don’t understand, does he/she use other ways to help you understand, e.g. gives another explanation, uses gesture/body language to demonstrate what he/she is saying?
	Does your child ask for clarification when he/she doesn’t understand something, e.g. “what does that mean?”?
General interaction	When your child is with someone he/she feels comfortable with, does he/she use appropriate eye contact?
	For 5 years and older: If your child speaks comfortably to a few adults and children, is there a difference between how he/she speaks to them?
Interaction with comfortable conversational partner(s)	Observation considerations: Does the child display a range of communicative functions and respond appropriately in the social context? Do they adapt their communication to the person they are talking to? Do they follow appropriate rules of conversation and storytelling? Can older children make inferences and understand nonliteral or ambiguous language?

Structured parent interviews such as the Diagnostic Interview (Kotrba, 2015) and the Parent Interview Form (Johnson & Wintgens, 2016) may be useful. Home video-recordings of the child talking in anxiety-free situations can also give valuable information regarding their speech, language and social communication skills (Bergman, 2013; Johnson & Wintgens, 2001, 2016; Kotrba, 2015).

10.7.1.4 Gathering Information from School

Most children with SM experience difficulties at school so it is important to ascertain how the child is functioning in that environment by exploring:

- Teachers' concerns
- The child's speech, language and communication
- The child's social interaction, temperament and behaviour
- The child's ability, attainment and interests
- Assessment results, current support the child is receiving and other professional involvement
- How the child's SM is managed at school
- The school's experience with SM

A structured format such as the Primary or Secondary School Report Form (Johnson & Wintgens, 2016) may be useful.

10.7.1.5 Observation of the Child or Young Person

Behavioural observation is the 'linchpin' of assessment and intervention of SM (Kearney & Vecchio, 2006) and must include observations made by parents. If children with SM are aware of an unfamiliar observer, their communication *difficulties* may be observed but not their strengths or true ability. This can lead to misdiagnosis if the observing professional is not equally conversant with the nature of SM, communication disorders and ASD (Wintgens, 2015). Therefore, observation of language deficits should always be accompanied by questions about how the child performs in a similar situation with people they talk to easily.

There are several ways to observe the child or young person in various settings:

- Video-recordings
- Observational logs completed by parents and staff
- Observations by a professional of the child managing their school day and interacting with peers, family members and the professional

Observation sessions are used to note:

- How family and staff handle the child's SM or other communication difficulties
- Communication *competence*: the child's communication and interaction skills with people they talk to easily

- Communication *performance*: variability in the child's spoken language output across different settings (speaking habits)
- Signs of anxiety within the child
- The child's level of communication comfort during interactions with different people in different settings, using scales such as
  - The stages of social communication comfort scale® (Shipon-Blum, 2012)
  - The stages of one-to-one interaction (see Table 10.1)
- The child's response to strategies designed to remove pressure and facilitate communication in children who have SM (see Sect. 10.7.2.1)

10.7.2 Extended Assessment

Information obtained from the core assessment may require further investigation by the appropriate professional or service in areas such as hearing, overall development, learning, fine or gross motor skills, sensory issues, speech, language, social communication and possibility of ASD.

10.7.2.1 Strategies When Meeting the Child

When meeting children with a view to conducting an assessment, it is important to build a positive rapport so that the child is comfortable in the examiner's presence, and to reduce their anxiety by openly addressing their speech anxiety (Johnson & Wintgens, 2001, 2016; Kotrba, 2015). Children must not feel under any pressure to speak. Rather, they need to know they can speak in their own time, as and when they feel ready. They might prefer just to talk to their parent during their visit. Depending on the age of the child, rapport can be built through play-based interaction or an explanation of what will happen during the session. Johnson and Wintgens recommend following the progression set out in Table 10.1. Throughout the session, the examiner gauges the child or young person's comfort level and nudges them towards the next level, creating opportunities for the child to speak rather than demanding that they do. For example, questions requiring a non-verbal response such as nodding or pointing (stage 3), are not asked until the child is sufficiently comfortable to physically engage in a non-verbal activity (stage 2). If the child talks to the parent in the presence of the examiner, it will be safe to ask questions through their parent (stage 4). If not, the examiner pulls away and can even leave the room, giving the child a chance to relax and talk without an audience, before returning to repeat the process.

Other things to consider during rapport-building are:

- Consciously relax and maintain a smiling countenance and friendly, open posture. Children with SM are extremely sensitive to other people's body language

and a blank or worried facial expression will convey disapproval, sadness or anxiety and raise the child or young person's anxiety level.
- Limit direct eye contact with the child with SM and focus more on the materials and games used in the session. Although this may seem counterintuitive to appropriate social interaction modelling, it makes the child feel more relaxed and more likely to speak during the interaction. Direct eye contact can make the child feel uncomfortable, especially after asking a question.
- Position yourself alongside the child rather than opposite. This is less threatening as it naturally reduces eye contact and the feeling of being scrutinised.
- Choose activities that the child enjoys or is good at. This information is gleaned from parents in earlier conversations or the child can be asked to bring an activity of their choosing to the session.
- Make comments and ask rhetorical rather than direct questions initially. Tag questions are useful to convey a sense of reciprocal chat prior to two-way interaction (e.g. "That's too big, isn't it?," "Wow, that was fast, wasn't it?"). Professionals can also think aloud and ask themselves questions (e.g. "I wonder what goes in here?").
- Leave pauses to give the child opportunities to respond during the activity or play.
- Self-rated questionnaires such as 'The Communication Rating Scale,' 'Talking to Strangers' and 'Reactions of Family/Friends/Staff' (Johnson & Wintgens, 2016) can be helpful to understand how the young person experiences SM while demonstrating that the professional values non-verbal communication.

10.7.2.2 Methods of Assessing the Child's Pragmatic Language Skills

There are several ways a professional can minimise anxiety as being a confounding factor in assessments. The same principles apply, regardless of the aspect of language, learning or development that is being explored. But for the purposes of this chapter, four options are considered in the context of pragmatic language assessment. The first three approaches rely on parental input while the last method involves direct assessment. In practice, a combination of approaches is used.

1. The parent observes the child's interactions in situations where they speak freely over a period of a week to record a range of different speech acts (see Sect. 10.6.1). Parents can use Johnson and Wintgens (2016) 'Record of independent social functioning and assertiveness' to help identify and write down examples of how the child uses language for different purposes, e.g. to request an item, direct an activity, ask for help or express disagreement. The record form is supplemented by video-recordings of the child engaged in relaxed purposeful spoken communication with friends or family members. Situations that might be suggested to video include the child teaching a communicative partner how to complete a task, problem-solving in a shared activity such as putting up a tent or completing a jigsaw or involved in an activity where they are likely to need adult help. The examiner is looking not only for a range of speech acts in the video

samples, but also the child's capacity to be sensitive to the communication needs of others, be a good listener and both respond and initiate in conversation (Fey, 1986).

2. This approach again uses video-recordings of the child interacting in different situations with a comfortable communication partner but a more formal element is introduced by conducting a conversational analysis of the video samples (e.g. The Conversation Analysis Profile, Fey, 1986) and/or completing a comprehensive pragmatic interview schedule such as the 'Pragmatics profile of everyday communication skills in children or school-aged children' (Dewart & Summers, 1995). This qualitative assessment allows the professional to investigate the child's interactions by drawing upon the observations of parents, teachers and other carers, and is divided into different aspects of pragmatics: communicative functions; response to communication; interaction and conversation; and contextual variation.
3. The third approach entails training the parent to administer formal pragmatics assessments as in the Klein et al. (2013) study. However, there are several considerations when using this method. Firstly, the parent needs to strictly follow the test administration guidelines in order to preserve the validity of the test scores (e.g. not giving extra cues or veering from the test script). Secondly, the administration of the assessment must be observed by the professional either live (in another room through a one-way mirror) or from a video or audio-recording. This ensures that the assessment is administered appropriately and allows the professional to correctly interpret and score the results. If the professional stays in the room while the parent administers the assessment, the child may still feel reluctant to speak freely impacting on the reliability of the results. Thirdly, assessment needs to take place in an anxiety-free environment in order to obtain a typical sample of the child's pragmatic skills. It has been suggested that the most favourable location is the child's home (Cleator & Hand, 2001; Klein et al., 2013; McInnes et al., 2004). If this is not practical, parental assessment in a school or clinic facility will yield more representative results if the child first spends time alone with their parent in that setting to establish comfortable conversational speech. At the time of assessment, they will need to be assured of no interruptions and located in a room that is well away from casual bystanders. It is not clear if these precautions were taken in the Klein et al. study (Klein et al., 2013).

   Fourthly, as discussed earlier in this chapter, having a parent administer a formal assessment does not always guarantee best results. The authors extensive clinical experience indicates that many children *without* SM perform better with the practitioner than for a parent. Parents are often aware of this dynamic and suggest that they leave the room to improve the child's performance. Therefore, parents should always be asked if they feel that the child or young person has fully applied themselves during a parental assessment, and whether in their opinion, the responses reflect the child's true ability. A less formal approach could yield more representative results. For example, parents could ask higher-level reasoning questions during play activities or in the guise of riddles or puzzles.

Suitable questions include Marion Blank's level 3 or 4 questions (Blank et al., 1978), and items from the Canterbury and Thanet Verbal Reasoning Assessment (Johnson, 2012).

4. The final approach involves the professional building rapport with the child or young person and gradually moving them towards comfortable verbal communication in order to conduct a formal face-to-face assessment. Once the child is communicating comfortably with the professional non-verbally (stage 3), assessments which require the child to point to their answer or provide a written response are possible. This can work well for simple vocabulary tests (Klein et al., 2013; McInnes et al., 2004) and it might be tempting to adapt receptive pragmatic tasks (e.g. narrative comprehension) in order to accept a written rather than spoken response from literate individuals. However, for the best results in assessments which require higher-level reasoning, inference and working memory, the professional should delay assessment until the child is less anxious and both thinking and speaking freely, i.e. at stage 8 (connected sentences) or even better, stage 8+ (spontaneous conversation). Until then, narratives and explanations are likely to be short and lacking in detail and, therefore, not representative of true ability.

   It may be quicker for the child or young person to build rapport at home (Cleator & Hand, 2001). However, as found in Cleator and Hand's and Klein et al.'s (2013) studies, this can take time. If the child has not spoken after several sessions, it may require the use of stimulus fading in order to facilitate speech, e.g. the sliding in technique (Johnson & Wintgens, 2001, 2016). The child talks to their parent alone initially and the professional gradually approaches until the child can accept their presence without anxiety. Verbal activities with increasing communication 'risk' are then introduced until the child can speak freely. The professional can now administer formal pragmatics assessments involving functional language use, narrative skills and higher-level verbal reasoning. For suggestions, see Table 2.1 in Chap. 2.

10.8 Treatment

SM intervention is categorised into four different approaches in the literature: systems, behavioural, psychopharmacological and psychodynamic approaches (Cohan et al., 2006; Zakszeski & DuPaul, 2017). Systems approaches focus on developing the understanding of SM in the significant people in the child's life such as family, school staff and peers. They target the behaviours of those around the child which perpetuate the SM and the avoidance behaviour of the child. Behavioural approaches use techniques to reduce the child's anxiety around talking and facilitate and reinforce successful speaking in different situations. These techniques include contingency management, shaping, hierarchical exposure, stimulus fading, cognitive restructuring, prompting, modelling and social skills (Zakszeski & DuPaul, 2017).

Psychopharmacological approaches use medication to reduce anxiety. Østergaard's (2018) systematic review compared cognitive behavioural therapy approaches, medication alone and combined approaches and found that the behavioural approaches were considered to be the first choice while medication and combination therapy were considered to be more appropriate for persistent cases of SM. Kumpulainen (2002) suggests that psychopharmacological therapy cannot be the first choice of intervention. However, if other methods are not effective then medication can be included as part of the treatment. Psychodynamic approaches try to understand the origins of SM in the child's unconscious. In the paediatric context, play and art therapies are often used.

Cohan et al. (2006) investigated the literature from 1990 to 2005 and found that behavioural and cognitive behavioural treatment modalities were the best supported. Pionek Stone et al. (2002) performed a meta-analysis of 114 treatment studies and found that behavioural interventions are more effective than no treatment at all for individuals with SM. Psychodynamic and systems treatment approaches did not have enough data to make the same claim. Zakszeski and DuPaul (2017) reviewed the literature from 2005 to 2015 and found that most of the papers used more than one approach. More than half the papers used both a systems and behavioural approach as utilised by Johnson and Wintgens (2016). Their intervention has been coined the '24/7' approach (Johnson, in press) as families and schools are empowered to help the child or young person face their fears through non-invasive strategies which are implemented in everyday situations as the opportunity arises.

10.8.1 The 24/7 Approach (Johnson & Wintgens, 2016)

Johnson and Wintgens' method incorporates a systems approach to precede and accompany direct behavioural work. The systems element involves educating the family and school about SM, planning and implementing changes in the home and school to eliminate the maintaining factors, openly acknowledging the child's speech anxiety and building the child's confidence and independence. By eliminating the maintaining factors, the aim is to establish a comfortable environment, increase participation rather than avoidance and create opportunities rather than demands to speak. No targets are set for the children; they are simply nudged towards talking by incrementally changing the adult's interaction style. Staff learn to speak to the child or young person without raising their anxiety (initially by avoiding direct questions, accepting other modes of communication and asking questions through parents and friends) and gradually introduce direct questions at the child's pace. Parents learn to support their child to answer other people rather than answering for them, and help their child tolerate being overheard by others through informal stimulus fading and reassurance. It is essential that speech is preserved as the child's default mode with their family, rather than relying on private whispering and gesture when others are present. For many children and young people, the systems element is enough to help them feel sufficiently relaxed to speak to key individuals. Pre-school children may even go on to speak freely in other situations.

When SM persists after systems changes, direct behavioural work is required to reduce the child or young person's anxiety sufficiently to facilitate progress. Here, through a process of target-setting and record-keeping, the child is fully aware of the small steps they are taking. Activities are carefully chosen, observing the level of communication risk in order to minimise the child's anxiety. Low-risk activities have a clear structure and involve rote-learnt or factual information at single-word level before short phrases and sentences. High-risk activities involving time pressure, ambiguity or uncertainty, longer explanations and conversation are left until last.

Direct behavioural work initially focuses on establishing speech with a key individual who has built non-verbal rapport with the child. This person is ideally available in situations where the child or young person is unable to speak, e.g. a teacher, teaching assistant or learning mentor. If a comfortable conversational partner such as a parent is available, then stimulus fading or 'sliding in' is usually recommended but if not, other stimulus fading techniques such as 'lone talking' or 'the telephone sliding in technique' can be utilised. Other behavioural techniques which do not require the presence of a conversational partner are based on the principle of shaping. This involves 'shaping' the individual's behaviour through a series of changes that gradually approximate to the desired behaviour. In the context of SM, a small-steps progression is used to shape the individual's communication from gestures to sounds, syllables, words, sentences and finally conversation or from a non-interactional verbal activity such as leaving voice messages or reading aloud to reciprocal conversation.

Once the child or young person is speaking to the new person alone, focus shifts to four areas of generalisation: increasing the range of conversational partners; developing interaction in groups; talking in public places; and independent social communication. The demands are carefully increased for each of the four areas in a co-ordinated manner, with parents supporting generalisation in the community.

10.8.2 Integrating Pragmatic and SM Interventions

If assessment confirms that the child or young person has a pragmatic language disorder or areas of pragmatic language difficulty, a speech and language therapist/pathologist would need to be involved if not already. Professionals will need to explicitly teach the areas of pragmatics outlined in Sect. 10.6, while considering the level of communication risk.

Giddan et al. (1997) suggested that remediation of speech and language problems can only be addressed once speech has been initiated in the therapy setting. The authors of this chapter further suggest that the individual needs to be at sentence level (stage 7, Table 10.1) to begin work on speech acts, verbal reasoning and non-literal language, and stage 8 when working on various discourse interventions (e.g. narrative, expository, conversational skills). Although it may seem intuitive to work on non-verbal social communication skills such as eye contact at stage 3, clinical experience indicates that individuals find these activities overwhelming until stages 7/8 when they are feeling more comfortable about talking in general. Kelly (2016) suggests that development of self-awareness and self-esteem precedes work on non-verbal and verbal social skills, and these foundational areas of social skills can be addressed if the child is not yet talking in sentences.

Many aspects of pragmatics are covered in the social communication area of generalisation for SM (Johnson & Wintgens, 2016). This involves gradually moving from 'safe' planned activities to high-risk spontaneous interaction in order to initiate requests and commands, share and seek information, correct misunderstandings and cope with a variety of topics (see Table 10.3).

Collaboration between speech and language therapists, parents and teachers is key to implementing the strategies for more effective communication at home and school (Adams & Lloyd, 2007). Parents have the in-depth knowledge about their child's social communication skills, which is necessary to plan detailed and personalised intervention (Baxendale et al., 2013). Teachers and teaching assistants have the valuable insight of the child's functioning in the classroom and school environment. Working together, pragmatic intervention can be tailored to the child's level, integrating explicitly taught skills into real-life situations (Baxendale et al., 2013). Most importantly, speech and language therapists can ensure that the deficits in pragmatics are treated within the framework of SM intervention. Otherwise, the pressure of targeting social communication skills could be a maintaining factor, prolonging the selective mutism and limiting the effectiveness of the pragmatic intervention.

10.9 Summary

Selective mutism is an anxiety disorder which impacts an individual's social interactions, education and work. The individual may display limited speech acts, lack of initiation, imbalanced turn taking, lack of eye contact or gestures to support interaction, avoidance of social interaction or difficulties answering higher-level verbal reasoning questions. In most children with SM, these difficulties arise from their anxiety to speak. However, due to the high comorbidity rate of communication disorders in this underserved population, there will be some individuals with both SM and a pragmatic language disorder. Therefore, it is important to screen for these difficulties during an assessment and, if indicated, use the methods discussed in this chapter to further explore the child or young person's pragmatic skills. If the child does have comorbid SM and pragmatic language disorder, then deficits in pragmatics need to be treated within the framework of SM intervention.

Table 10.3 Promoting effective social functioning

<table>
<tr><th colspan="6">Social functioning</th></tr>
<tr><td colspan="6">Within a small-steps programme, activities gradually move from:
LOW RISK → HIGH RISK</td></tr>
<tr><td colspan="3">Prompted →
Child responds to a question, cue or prompt</td><td colspan="3">Child-initiated
Child initiates the interaction by asking a question, giving an instruction or gaining someone's attention</td></tr>
<tr><td colspan="3">Factual content →
Child's contribution will not be challenged and is known to be correct. No other answers are possible</td><td colspan="3">Emotive content
Child's contribution may be challenged or require clarification. Content involves personal information or opinion, explanation or doubt (e.g. more than one answer)</td></tr>
<tr><td colspan="2">Structured →
Agreed turn-taking sequence; set language; clear rules for activity and task completion; no initiation required other than taking turn.</td><td colspan="2">Semi-structured →
Structured turn-taking, but some turns may take longer than others; language may vary; rules are in place but unpredictable duration; some initiation may be required, e.g. to cue next person, stake a claim (e.g. 'Uno!','Bingo!'), correct other people or give/seek clarification.</td><td colspan="2">Unstructured
Talking is not in fixed turn-taking sequence and may require 'calling out' (e.g. Snap!); language generated by participants; activity itself may be structured but interaction follows usual conversational and social rules and may involve negotiation or improvisation; initiation required for balanced participation.</td></tr>
<tr><td colspan="3">Planned →
Taking place within a designated programme session or rehearsed with known content and fixed end-point</td><td colspan="3">Spontaneous
Taking place as part of everyday routine, i.e. involving other people and/or time pressure, or introduced without prior warning</td></tr>
</table>

(Source: The Selective Mutism Resource Manual second edition, © Maggie Johnson & Wintgens, 2016. Reproduced with permission of Informa UK Limited through PLSclear)

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Chapter 11
Attention Deficit Hyperactivity Disorder

Reinie Cordier, Sarah Wilkes-Gillan, and Natalie Munro

11.1 Introduction

> Jeremy is in Year 1 and is delightful, but he can be tiresome. His teacher describes him as "busy," and wishes that he would just stop talking! Jeremy blurts out random answers even before thinking about them, and he finds it incredibly hard to listen to any story that his teacher reads to the class. The other children in the class tolerate Jeremy—they get frustrated when Jeremy doesn't listen to them. Jeremy says he has friends to play with, but he doesn't pick up on their suggestions of what to play or when it is time to change or adapt the play. He seems to flitter from one play friend to the next. Jeremy's parents worry about his social communication and whether he will develop and maintain friendships at school.
>
> In another Year 1 class, Rachel sits and constantly looks out the window. Her teacher describes her as "head-in-the-clouds" and wishes that she would "come back down to earth." Rachel rarely offers an answer to a question in class—her teacher taps Rachel's shoulder and says her name to get her attention. During story book time, Rachel looks like she is paying attention, but the teacher wonders if she is listening and understanding what is being read. Rachel used to have a friend at school, but she moved to another school far away. When asked to play, Rachel might join in for a little while, but then, to her friends, it looks like she loses interest in playing with them. Rachel's mother worries that Rachel is too shy and wants her to be more social.

You may have met children like Jeremy and Rachel. Even without a formal diagnosis of ADHD—excessive presentations of hyperactivity, impulsivity, inattention, or a combination of these behaviours (see the fifth edition of the *Diagnostic and Statistical Manual of Mental Disorders* (DSM-5; American Psychiatric Association,

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L. Cummings (ed.), *Handbook of Pragmatic Language Disorders*,
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2013) for specific criteria)—it is clear that these two children present with attention and social communication difficulties. A formal diagnosis in DSM-5 requires several behavioural indicators of ADHD to be evident for at least 6 months. Notably, these symptoms should negatively impact social, academic, or occupational activities, be present in more than one context (e.g. home or school) and be significantly more severe than expected for the child's developmental level (American Psychiatric Association, 2013). Interestingly, some ADHD symptoms such as "often talks excessively." "Often interrupts or intrudes on others" and "cannot wait for a turn in conversation" are also characteristic of social communication or pragmatic language difficulties. In this chapter, we aim to (a) describe the pragmatic language difficulties experienced by school-aged children with ADHD; (b) explain how pragmatic language can be assessed by means of carefully constructed, observational assessment; and (c) present intervention approaches and techniques that target pragmatic language abilities in children with ADHD. But first, we discuss what is known about the epidemiology and aetiology of ADHD, a complex neurodevelopmental disorder.

11.2 Background on ADHD

ADHD is a common childhood neurobehavioural disorder characterised by developmentally inappropriate levels of inattention, hyperactivity, and impulsivity. While we expect young children to express exuberance, rapid movement and inattention when presented with novel stimuli or while exploring a new environment, ADHD is marked by levels of inattention, hyperactivity and impulsivity that exceed the expected level of development (American Psychiatric Association, 2013).

11.2.1 The Core Symptoms and Subtypes of ADHD

The Diagnostic and Statistical Manual for Mental Disorders, fifth Edition (DSM-5) is a widely used diagnostic manual for ADHD (American Psychiatric Association, 2013). As stated in the DSM-5, the core symptoms of ADHD are inattention, hyperactivity and impulsivity. These symptoms vary in severity. Therefore, individuals with ADHD experience varied levels of impairment throughout their daily lives and across the lifespan (Caci et al., 2014). There are three subtypes of ADHD: (1) the predominantly hyperactive-impulsive subtype; (2) the predominately inattentive subtype; and (3) the combined subtype. In a meta-analysis of 86 studies across the world, Willcutt (2012) found that the predominantly inattentive subtype was the most common subtype of ADHD across all samples. The only exception was the predominately hyperactive-impulsive subtype, which was most common in preschool aged children (Willcutt, 2012).

11.2.2 *Prevalence*

In a comprehensive meta-analysis, the worldwide prevalence of ADHD in children and adolescents was estimated to be 5.3% (Polancyzk et al., 2007). This estimate varied widely among the 103 studies included in the review. The variance was due to a range of factors including different diagnostic criteria used to define ADHD, the method of assessing ADHD related symptoms, and the incorporation of multiple sources of information (Polancyzk et al., 2007).

In a more recent systematic review by Thomas et al. (2015) that included 175 studies and 179 ADHD prevalence estimates, the overall pooled estimate of the prevalence of ADHD was 7.2%. Prevalence estimates were found to be lower when using the third, compared with the fourth edition of the DSM and when studies were conducted in Europe compared to North America (Thomas et al., 2015). These prevalence rates were consistent with a meta-analytic review by Willcutt (2012) where the pooled prevalence rates of 86 studies were reported to be between 5.9% and 7.1% when including ADHD as defined by parent ratings, teacher ratings or a best estimate diagnostic procedure.

11.2.3 *Gender Ratio*

The prevalence of ADHD is well established to be higher in males compared to females (Nøvik et al., 2006; Willcutt, 2012). A meta-analysis by Willcutt (2012) found the ratio of ADHD to be consistently higher in males across a range of age groups spanning from 3 years to over the age of 19 years. Across the age groups the ratio of male: female was 1.8: 1 (3–5 years), 2.3: 1 (6–12 years), 2.4: 1 (13–18 years) and 1.6: 1 (19 years or older) (Willcutt, 2012). Results of the study also found that males were more likely to meet the criteria for all ADHD subtypes using the DSM-IV. Interestingly, a significantly higher proportion of females met the criteria for the inattentive subtype of ADHD, compared with the hyperactive or combined subtypes.

11.2.4 *Causes of ADHD*

There is no single risk factor identified in the research to be the cause of ADHD. Multiple causal factors are reflected in the heterogeneity of the disorder. This is indicated by a range of psychiatric comorbidities, varying clinical profiles, patterns of neurocognitive impairment, developmental trajectories, as well as a range of structural and functional differences in the brain (Luo et al., 2019).

There is research that acknowledges that ADHD may be associated with an underlying genetic component. In a study that pooled data from 20 twin studies, the mean heritability of ADHD was estimated to be 76%. The study compared data was from the US, Europe, Scandinavia and Australia (Faraone et al., 2005). These rates were similar to a study that used data from the Swedish Twin Registry, including 37,714 adult twins where the genetic contribution to ADHD was estimated to be 72% in adult twins (Larsson et al., 2014). There is also emerging research in the area of genetics that suggests there are potential biomarkers of ADHD, however, this research is not yet well established (Faraone et al., 2014).

11.2.5 Diagnosis

The National Institute for Health and Care Excellence (NICE) (2018) recommends that a diagnosis of ADHD should be made only by a qualified healthcare professional with training and expertise in the diagnosis of ADHD, such as specialist psychiatrists or paediatricians. The NICE guidelines further recommend that the diagnosis of ADHD be made based on the following considerations: (1) a full clinical and psychosocial assessment of the person; (2) a full developmental and psychiatric history; and (3) observer reports and assessment of the person's mental state. Further recommendations regarding diagnosis include that diagnosis should not be solely based on a rating scale or observational data, that ADHD symptoms should meet the diagnostic criteria listed in the DSM-5 or ICD-10 (for hyperkinetic disorder), and that part of the diagnostic process should include assessment of the person's needs, physical health, social and familial circumstances and coexisting conditions.

11.2.6 Co-morbid Conditions

In addition to the core characteristics of ADHD, children with ADHD can also experience a range of co-morbid conditions that further impacts a range of their day-to-day activities. In a recent study of 2447 children and adolescents where 650 (27%) were diagnosed with ADHD only, over 50% were reported to have learning disorders, over 20% had sleep disorders, almost 20% had oppositional defiant disorders, and over 10% had anxiety disorders. Other reported co-morbid conditions included intellectual disability, mood disorder, conduct disorder, tic disorders and autism spectrum disorders (Reale et al., 2017). ADHD is also associated with impairments in social functioning and communication skills that affect a child's performance in activities of daily living, including their ability to develop meaningful friendships and participate in school (Bagwell et al., 2001).

11.3 Pragmatic Language Defined

Just as ADHD is a multidimensional construct with historical debate regarding its definition, the same can be said about pragmatic language (Ariel, 2010). Pragmatic language—the social use of language—has traditionally been used to describe communication during interaction with another. The form of this communication could be verbal, non-verbal or paralinguistic (Prutting & Kirchner, 1987). Over time, scholars have included not only these communicative forms and the functions that were associated with them, but also behaviours that reflected social and emotional aspects of language (Adams et al., 2005). While pragmatic language definitions may vary, even within the chapters of this book, we adopt this multidimensional, conceptual definition of pragmatic language in recognition of the interconnectedness between pragmatic language, social cognition and emotional understanding (e.g. Fujiki et al., 2002; Matthews et al., 2018; St Clair et al., 2011).

It is important to study pragmatic language in ADHD because, although not causally related, pragmatic language difficulties are often co-morbid for children with ADHD (Camarata et al., 1999). In a systematic meta-analytic review, Korrel et al. (2017) found that children with ADHD had poorer pragmatic language skills compared with typically developing controls ($N = 315$; controls = 115, ADHD = 200; $p < 0.05$; r range: 0.27–0.71; weighted mean effect size = 0.98). This indicates that speech-language pathologists should be aware that children with ADHD are at high risk of experiencing pragmatic language problems and, as such, have a focus on the assessment and treatment of pragmatic language challenges in this population.

11.4 How Has Pragmatic Language Been Assessed in School-Aged Children with ADHD?

Various approaches have been used to assess pragmatic language abilities in children with ADHD. These include experimental studies, norm-referenced tests, checklists and observation methods. This section will examine the benefits and limitations of these approaches.

In the systematic meta-analytic review conducted by Korrel et al. (2017), four studies met the eligibility criteria of the review. Of these studies, two utilised parent and/or teacher (proxy-reported) pragmatic language checklists (Geurts et al., 2004b; Timler, 2014), while the remaining two studies utilised norm-referenced testing (Cadesky et al., 2000; Staikova et al., 2013). Standardised, norm-referenced testing of pragmatic language abilities has been criticised for its static and non-naturalistic approach (Gerber et al., 2012). There is also evidence that children with ADHD can demonstrate adequate knowledge (i.e. capacity) of pragmatic language on norm-referenced tests, but then display inappropriate use of those tested skills in unstructured contexts (Kim & Kaiser, 2000; Bignell & Cain, 2007), suggesting a deficit in performance typically seen in more naturalistic contexts.

Of the standardised tests that are available to assess pragmatic language, the majority focus on assessment at a capacity level, referring to the child having knowledge of pragmatic language (Westby & Washington, 2017). The International Classification of Functioning, Disability and Health (ICF) framework (World Health Organization, 2001) states that having this capacity is essential but is not always sufficient for participation or the performance of this knowledge. This distinction between capacity and performance is vital when considering the real-life implications of pragmatic language. A child may exhibit appropriate knowledge of pragmatic language in standardised tests, but be unable to perform this in conversations with peers or teachers (Westby & Washington, 2017).

The use of proxy-reported pragmatic language checklists, such as the Children's Communication Checklist (Bishop, 1998), may prove to be an efficient and valid method for the purposes of identifying pragmatic language difficulties (e.g. Geurts et al., 2004a). However, these types of instruments are not designed to provide the clinician with an in-depth understanding of the nature of the child's pragmatic language problems (Cordier et al., 2014). To some extent, the choice of assessment may reflect different purposes for that assessment. Denman et al. (2019) developed a taxonomy for a child language assessment that presents seven purposes of assessment. Pragmatic language assessments cover a very narrow range of those purposes and are mainly aimed at describing pragmatic language difficulties. Certainly within pragmatic language assessment, there has been a longstanding call for more valid measures of pragmatic language involving naturalistic observation (Adams, 2002; Russell & Grizzle, 2008).

In an integrated review which included studies involving typically developing controls or other clinical population comparisons, Green et al. (2014) found nine studies that directly measured pragmatic language abilities in children with ADHD. This included six studies that utilised observational ratings of structured, communication tasks and three using either a checklist or standardised, norm-referenced test. The six studies utilising observation focussed on referential communication tasks, storytelling, structured peer or adult conversation tasks, or structured tasks relating to a game such as 'Space Flight Game' with assigned peer roles of astronaut and mission control (King & Young, 1981; Whalen et al., 1979) or a 'TV Talk Show Host Game' with assigned roles of host and guest (Landau & Milich, 1988).

While these studies found pragmatic language difficulties in children with ADHD (described later), some of these tasks are also constrained because the child is provided with a hypothetical context, of which they may have little to no experience and yet they are being evaluated against it in that context (i.e. consider the role-play between the astronaut and mission control). In other tasks, such as referential listening, storytelling or adult-child conversation tasks, these activities are adult-directed. Adult-directed tasks offer structure, but again, it is unclear how the child performs beyond that carefully constructed task. This hampers our understanding of real communication and pragmatic language abilities. This also does not reflect who school-aged children predominantly interact with—their peers—not adult, research assistants! Jokes aside, the crucial sticking point here is the knowledge that children with ADHD have poor peer interaction and limited friendships (Normand et al., 2019; Marton et al., 2015).

This knowledge, therefore, predicates the need to better understand that context. In response to this need, our research team has developed a pragmatic language observation measure that is child-led and one that reflects naturalistic, unstructured, communication between peers during play. We will describe the development of this measure and what we have learnt about the pragmatic language difficulties of children with ADHD. As a starting point, we first summarise what is known about the nature of the pragmatic language difficulties of children with ADHD.

11.5 What Pragmatic Language Difficulties Have Been Observed in Children with ADHD?

Although varying with types of measurement, the difficulties children with ADHD may experience with pragmatic language have been widely reported. In a recent systematic review and meta-analysis of language difficulties in children with ADHD, Korrel et al. (2017) reported eight measures of pragmatic language, all of which found children with ADHD had significantly poorer performance than control groups. The studies also reported a strong relationship between ADHD and pragmatic language difficulties. An integrated review of 30 studies found that these difficulties often relate to excessive talking, difficulty adopting listener and speaker roles and inference comprehension (Green et al., 2014). These difficulties can be categorised by whether they relate to a deficit in either *capacity* or *performance* of pragmatic language.

11.5.1 Capacity

Findings relating to the capacity of pragmatic language refer to research that focused on measures that assess children's knowledge of pragmatic language, mostly using non-observational, standardised assessments. Staikova et al. (2013) found that, compared with typically developing peers, children with ADHD demonstrated difficulties with discourse management, narrative discourse and presupposition when shown pictures and read brief stories and, subsequently, asked to make inferences about the story and its characters.

Parents and teachers of children with ADHD commonly report capacity difficulties with inappropriate initiation, stereotyped conversation, use of conversation context, and conversational rapport when using both parent and teacher reports (Geurts et al., 2004a, 2004b; Helland et al., 2012; Väisänen et al., 2014). Using a parent-report questionnaire, Bruce et al. (2006) found that children with ADHD had difficulties carrying a conversation, interpreting what was said and being 'on track.' When measuring capacity, children with ADHD also demonstrate more, or different, difficulties with inappropriate initiation when compared with different clinical groups, such as autism spectrum disorder, specific learning disability and pervasive developmental disorder (Bishop & Baird, 2001), particularly when they have comorbid learning impairment or reading disability (Cohen et al., 2000). Surprisingly, when using similar methods, Kim and Kaiser (2000) found no differences between ADHD and typically developing peers in the capacity of pragmatic language, including receptive and expressive syntax, and semantics.

11.5.2 Performance

Measuring difficulties in performance of pragmatic language involves observing children with ADHD in a naturalistic setting. Across the research that has utilised observational assessment to investigate the pragmatic language performance of children with ADHD, one consistent finding is the presentation of poor verbal pragmatic language abilities. Firstly, conversation dyads (ADHD and typically-developing dyads compared to typically developing and typically-developing dyads) showed differences in the ADHD and typically-developing dyads' ability to sustain conversational reciprocity (Clark et al., 1988; Kim & Kaiser, 2000). Secondly, in game-type tasks requiring role assignment, children with ADHD were less able to adapt their verbal skills to meet the linguistic demands of the role they were playing (Landau & Milich, 1988; Whalen et al., 1979). Thirdly, children with ADHD displayed off-topic comments, frequent verbal interruptions and difficulties responding to verbal cues when taking on the listener role (Whalen et al., 1979).

When measuring the performance of pragmatic language, children with ADHD exhibit more difficulties overall when compared to typically-developing peers (Cordier et al., 2013, 2017; Wilkes-Gillan, Cantrill, et al., 2017a; Wilkes-Gillan et al., 2017b). Despite not finding any significant difference when measuring pragmatic language capacity, Kim and Kaiser (2000) did observe that children with ADHD experienced difficulties when adopting appropriate listener-speaker roles and giving feedback to speakers. This demonstrates the important distinction between capacity and performance; some children with ADHD may have adequate knowledge of pragmatic language but fail to perform it successfully when interacting with peers in naturalistic contexts. This difficulty with performance of pragmatic language can have social and emotional impacts for children with ADHD. Children with ADHD would often disagree and be less cooperative than their typically-developing peers (King & Young, 1981; Clark et al., 1988).

This section demonstrated the nature of the pragmatic language difficulties that children with ADHD experience. The reporting of these clinical differences may, in part, be explained by the nature of the measures used to evaluate their pragmatic language capacity and performance. The next section provides a summary of the types of measures used to evaluate the pragmatic language skills of children with ADHD.

11.6 Measurement of Pragmatic Difficulties

There is a wide variety of measures used to assess pragmatic language difficulties in children with ADHD. A common criticism of available measures is the heavy focus on measuring children's capacity for pragmatic language rather than performance in naturalistic settings. Moreover, the psychometrics of these measures are rarely evaluated or reported, casting doubt on the reliability and validity of assessing pragmatic language in children with ADHD. This section will detail common measures that have been used with children with ADHD and their psychometric properties, or lack thereof, which have been reported in research related to pragmatic language in children with ADHD (see Table 11.1). These measures are categorised into three groups: (1) standardised measures; (2) parent or teacher proxy-report measures; and (3) observational measures.

11.6.1 Standardised Measures

Standardised measures used to assess pragmatic language often include tasks that children are asked to perform, including explaining hypothetical situations in pictures, formulating sentences or drawing inferences about short stories. The Test of Pragmatic Language (TOPL-2; Phelps-Terasaki & Phelps-Gunn, 2007) asks children to make inferences from pictures and short stories. This test aims to measure the degree to which children can make meanings from context, make sense of facial expressions, and understand the emotional state of characters. However, when used in a sample of children with ADHD, no psychometric properties were reported (Kim & Kaiser, 2000; Staikova et al., 2013).

The Understanding Ambiguity Test (UAT; Rinaldi, 1996) aims to measure figurative or pragmatic interpretation of speech. The measure is administered through short-story dialogues that contain either an ambiguous phrase or a homonym. Each item can have a literal or figurative interpretation that is supported by context, and the child is asked to select from four pictures that represent correct and incorrect interpretations. This assessment has been used to measure understanding of figurative language in children with ADHD, however, no psychometric properties have been reported (Bignell & Cain, 2007).

The Test of Language Competence-Expanded Edition (TLC-E; Wiig & Secord, 1989) includes a sub-test that measures understanding of figurative language without supporting context. This is tested through several trials, each comprised of a spoken, non-predictive sentence context, for example, "Mum looks really low today," with four accompanying pictures. One picture represented the figurative interpretation, one the literal interpretation, and two represent foils. The child is asked to choose the picture that matches the sentence. Bignell and Cain (2007) also used this measure with children with ADHD, with no reported psychometrics.

11.6.2 Proxy-Report Measures

Proxy-report measures often involve parent and/or teacher ratings of children's capacity for pragmatic language. The most commonly used of such measures is the Children's Communication Checklist (CCC; Bishop, 1998) and the revised Children's Communication Checklist- 2 (CCC-2; Bishop, 2003). The CCC and CCC-2 involve both parent and teacher reports on domains such as speech, syntax,

Table 11.1 Reported psychometric properties of measures of pragmatic language used in studies of children with ADHD

Measure	Administration	Domains	Results
Pragmatics Observational Measure (Cordier et al., 2014)	Observational	Introduction and responsiveness Non-verbal communication Social-emotional attunement Higher-level thinking Negotiation	**Reliability** Cordier et al. (2014) Strong correlations between all items except 'discourse interruption' item Factor analysis revealed two factors After removing the item, unidimensional internal consistency: α = 0.98 Inter-rater reliability: Ranged from r = 0.887 and r = 0.999 Wilkes-Gillan, Cantrill, et al. (2017a) ICC: 0.77 Wilkes-Gillan, Munro, et al. (2017b) ICC: 0.83 Cordier et al. (2017) – ICC: 0.97 **Validity** Cordier et al. (2014) Construct: Positive associations between POM and PP items Criterion: Strong correlation between overall PP and POM item scores **Responsiveness** Cordier et al. (2014) Sensitivity: 79.7% Specificity: 89.6% Positive predictive value: 67.8% Negative predictive value: 94.1%

(continued)

Table 11.1 (continued)

Measure	Administration	Domains	Results
Pragmatics Observational Measure −2 (Cordier et al., 2019)	Observational	– Verbal aspects – Non-verbal aspects	**Item fit** Cordier et al. (2019) – Overall: 0.99 **Person fit** Cordier et al. (2019) – Overall: 0.97 **Dimensionality** Cordier et al. (2019) – Two distinct elements found (verbal and non-verbal) – Four items outside acceptable parameters and removed **Differential item functioning** Cordier et al. (2019) – Some items easier for typically-developing playmates and controls vs. ADHD – Some items easier for older children vs. younger – Some items easier for boys vs. girls – One item easier for girls vs. boys
Pragmatic Protocol (Prutting & Kirchner, 1987)	Observational	– Verbal aspects – Turn-taking – Non-verbal aspects	**Reliability** Prutting and Kirchner (1987) – Inter-rater: Pearson's $r = 0.94$ Cordier et al. (2013) – Inter-rater: 84% (overall); 83% (verbal aspects); 85% (turn-taking); 82% (non-verbal aspects) Kim and Kaiser (2000) – Appropriate behaviour: 92% to 100% (M = 97.9%) – Inappropriate behaviour: 80% to 100% (M = 93.6%)

(continued)

Table 11.1 (continued)

Measure	Administration	Domains	Results
S-MAPS (Wiig et al., 2004)	Observation or portfolio	– Adaptive social-emotional – Non-verbal communication – Language use – Social skills – Thinking style and creativity – Executive function	**Reliability** Cordier et al. (2013) – Inter-rater: 77% (overall) **Validity** NR **Responsiveness** NR
Children's Communication Checklist (Bishop, 1998)	Parent/teacher report	– Speech – Syntax – Inappropriate initiation – Coherence – Stereotyped – Use of context – Rapport – Social relationships – Interests – Pragmatic composite score	**Reliability** Bishop and Baird (2001) – Internal consistency: Ranged from α = 0.54 and α = 0.91 for parents; ranged from α = 0.65 and α = 0.92 for teachers – Inter-rater: Between parent and professional ratings ranged from $r = 0.30$ to $r = 0.64$ Bishop (1998) – Inter-rater: Pearson's $r = 0.80$ – Internal consistency: Ranged from α = 0.79 to α = 0.86 **Validity** Bishop (1998) – Correlated with test of language and IQ – Positive correlations between subscales **Responsiveness** NR
Children's Communication Checklist-2 (Bishop, 2003)	Parent/teacher report	– General communication composite – Social interaction deviance composite	**Reliability** Väisänen et al. (2014) – Internal consistency: Ranged from α = 0.56 to α = 0.90 **Validity** NR **Responsiveness** NR

(continued)

Table 11.1 (continued)

Measure	Administration	Domains	Results
Five to Fifteen (Kadesjö et al., 2004)	Parent/teacher report	– Motor skills – Executive functions – Perception – Memory – Language (incl. Pragmatics) – Learning – Social skills – Emotional and behavioural problems	**Reliability** Lambek and Trillingsgaard (2015) – Internal consistency: Ranged from α = 0.85 to α = 0.96 (parent report); α = 0.907 to α = 0.96 (teacher report) – Factor analysis revealed acceptable fit Kadesjö et al. (2004) – Internal consistency: Ranged from α = 0.86 to α = 0.96 – Inter-rater: Ranged from $r = 0.67$ to $r = 0.85$ – Test-rest: Ranged from $r = 0.74$ to $r = 0.91$ **Validity** Bruce et al. (2006) – Language domain correlated highly and significantly with executive function, memory, learning, perception, social skills and emotional/ Behavioural domains **Responsiveness** NR
Understanding Ambiguity Test (Rinaldi, 1996)	Short story dialogues with ambiguous phrase or homonym	– Understanding figurative interpretations of speech	**Reliability** NR **Validity** NR **Responsiveness** NR
Test of Pragmatic Language −2 (Phelps-Terasaki & Phelps-Gunn, 2007)	Asked to make inferences about pictures and stories	– Making meanings of context – Appreciation of facial expressions – Understanding emotional state of characters	**Reliability** NR **Validity** NR **Responsiveness** NR

Table 11.1 (continued)

Measure	Administration	Domains	Results
Test of Language Competence-Expanded Edition (Wiig & Secord, 1989)	Spoken, non-predictive sentence, with four accompanying pictures to choose	– Understanding of figurative language without supporting context	**Reliability** NR **Validity** NR **Responsiveness** NR

Key: α Cronbach's alpha, *r* correlation, *ICC* intraclass correlation coefficient, *M* mean, *NR* not reported

inappropriate initiation, coherence and use of context. At the time of development, Bishop (1998) reported strong inter-rater reliability between parents and teachers, and internal consistency ranging between 0.79 and 0.86. They found the test correlated positively with other tests of language and intelligence, as well as positive correlations between subscales. Using the CCC in a sample of children with ADHD, Bishop and Baird (2001) reported internal consistency ranging between 0.54 and 0.91 for parents and 0.65 and 0.92 for teachers. They found much weaker inter-rater reliability than previous reports when investigating correlations between parent and professional ratings of language (correlations ranged between 0.30 and 0.64). Using the revised CCC-2, Väisänen et al. (2014) report internal consistency between 0.56 and 0.90 in a sample of children with ADHD.

The five to fifteen (FTF; Kadesjö et al., 2004) is another parent- and teacher-report questionnaire. This measure includes many different domains, such as motor skills, perception and memory. Of interest, the language subscale includes some measurement of pragmatic language. Kadesjö et al. (2004) report internal consistency ranging between 0.86 and 0.96, as well as inter-rater reliability ranging between 0.67 and 0.85. When re-tested after 3 months, the FTF demonstrated acceptable test-retest reliability. Bruce et al. (2006) used the parent-report questionnaire with children with ADHD and reported the language domain correlated positively with domains assessing executive function, memory, learning, perception, social skills and emotional and behavioural domains. More recently, Lambek and Trillingsgaard (2015) reported strong internal consistency for both parent and teacher reports. They also conducted a factor analysis that demonstrated an acceptable model fit in a sample of children with ADHD.

11.6.3 Observational Measures

Although considered the best way to assess performance of pragmatic language in naturalistic settings, there are few observational measures of pragmatic language. The benefits to using such measures include the ability to use blinded or unbiased raters of a child's performance in the moment, rather than relying on recall from parents or teachers that may introduce bias due to their relationship with the child, as well as the retrospective nature of such questionnaires. Observational measures often use a trained expert to conduct the rating, which is advantageous over non-expert reviews conducted by parents or teachers. Despite these advantages, there is an identified lack of assessments using this type of measure, compounded further by the lack of reported psychometric testing of such measures.

The Pragmatic Protocol (PP; Prutting & Kirchner, 1987) is an observational measure, designed to assess verbal, non-verbal and paralinguistic aspects of pragmatic language. Reliability of this measure in a sample of children with ADHD has only been reported in terms of inter-rater reliability, which is consistently reported as between 80% to 100% agreement (Prutting & Kirchner, 1987; Cordier et al., 2013; Kim & Kaiser, 2000). The measure uses a rating scale of 'appropriate,' 'inappropriate' and 'not observed,' which has been criticised as creating a dichotomy that does not capture a range of performance (Cordier et al., 2014).

The Structured Multidimensional Assessment Profiles (S-MAPs; Wiig et al., 2004) is an assessment based on observation or portfolio. The S-MAPs was not specifically designed to measure pragmatic language but does include rubrics related to some aspects of pragmatic language. However, there is very little reported psychometric information on its use in children with ADHD, with only Cordier et al. (2013) reporting an inter-rater reliability of 77%. Recently, a new observational measure has been developed, known as the Pragmatics Observational Measure (POM; Cordier et al., 2014) and the revised version the POM- 2 (Cordier et al., 2019). Thus far, this measure has shown promise in reliably measuring performance of pragmatic language in children with ADHD.

11.7 The Development of the Pragmatics Observational Measure

As reported in Table 11.1, there is limited reporting of the reliability and validity of the few observational measures of pragmatic language. Until recently, the Pragmatic Protocol (PP; Prutting & Kirchner, 1987) was the only observational measure with reported psychometric properties, albeit limited. While the reliability of this measure is strong, the rating scale used creates an 'all-or-nothing' system that does not capture the wider range of performances that may be exhibited by children with ADHD in naturalistic settings. In addition, the PP did not include items that capture the social and emotional aspects of peer interaction.

If we were to adopt a more contemporary perspective of what pragmatic language is and what it involves, then we needed an observational lens to include both verbal and non-verbal aspects of pragmatic language, as well as socio-emotional aspects. When adopting this contemporary lens, the question is: Are other aspects of the pragmatic language abilities of children with ADHD affected? To answer this question, we needed to extend our view of what pragmatic language involves and, crucially, we also needed a measure that is psychometrically robust. In response to this, Cordier et al. (2014) developed the Pragmatics Observational Measure (POM).

The POM is an observational measure, at first designed to capture more nuanced performance of pragmatic language across five domains: (1) introducing communication and being responsive to social interactions with peers; (2) interpreting and using non-verbal communication; (3) understanding and using emotional reactions and intentions; (4) using higher-level thinking; and (5) appropriate negation techniques (Cordier et al., 2014). All 27 items are rated using a 4-point scale (1: rarely or never observed; 2: sometimes observed; 3: observed much of the time; 4: almost always observed). After revisions, the POM-2 now comprises of two domains: (1) verbal pragmatic language; and (2) non-verbal pragmatic language (Cordier et al., 2019).

In contrast to other observational measures, the psychometrics of the POM have been rigorously tested using both Classical Test Theory (CTT; Cordier et al., 2014) and Item Response Theory (IRT; Cordier et al., 2019) and the POM has demonstrated strong psychometric properties in support of being a reliable and valid measure of pragmatic language skills in children aged 5 to 11 years. Using CTT methods, the POM was found to have strong internal consistency ($\alpha = 0.98$). Factor analysis revealed one item, discourse interruption, which comprised a second factor. After removing this item, the measure was found to be unidimensional (Cordier et al., 2014). The measure also showed strong inter-rater reliability. In terms of validity, there were strong correlations between POM and PP items.

Using IRT, Rasch analysis revealed an appropriate and ordered rating scale, as well as excellent person (0.97) and item (0.99) reliability (Cordier et al., 2019). Dimensionality revealed two distinct elements, verbal and non-verbal pragmatic language. This precipitated the first revision of the POM to now encompass two dimensions, rather than the previous five domains.The POM was developed in a sample of children with ADHD, their typically-developing playmates and a control group, and has since been used multiple times to measure pragmatic language skills in children with ADHD (Cordier et al., 2013, 2017; Wilkes-Gillan, Cantrill, et al., 2017a; Wilkes-Gillan, Munro, et al., 2017b).

11.8 Pragmatic Language Interventions for Children with ADHD

Many psychosocial interventions have been developed with the aim of addressing the lingering social impairments of children with ADHD (Evans et al., 2008, 2014). These treatments vary by the mode of treatment delivery and treatment approach and include school-focused interventions, behavioural parent training, social skills training, summer treatment programs, and emerging approaches such as interventions involving play (Evans et al., 2014). While they are effective for improving some areas of performance, most of these psychosocial interventions have focused on social skill acquisition and have demonstrated limited effectiveness for improving social skills, of which communication skills are an essential component (Pelham & Fabiano, 2008; Young & Amarasinghe, 2010). Further, many interventions do not demonstrate maintenance of the treatment effect or there is limited follow-up to investigate maintenance of treatment effects (Evans et al., 2014).

The pragmatic language difficulties of children with ADHD are well documented as is the influence of pragmatic language on social competence. Pragmatic language has been found to be a strong mediator of the relationship between hyperactivity and problems with social skills (Leonard et al., 2011). Pragmatic language also mediates the relationship between inattention and poor social skills, albeit not as strong as the preceding factors (Leonard et al., 2011). Yet, despite strong evidence of the mediating effect of pragmatic language on children's social skills and the extent to which the pragmatic language of children with ADHD is affected (Cordier et al., 2017), there are surprisingly few interventions designed to address the pragmatic language of children with ADHD. Despite an extensive search, we could only locate four studies that reported on one intervention aimed at addressing the pragmatic language difficulties of children with ADHD. All four studies reported on varying pilot studies of a play-based intervention for children with ADHD. These studies were also included in a recent systematic review of play-based interventions for children with ADHD (Cornell et al., 2018). While the evidence from the four play-based intervention studies is preliminary in nature, there is emerging evidence that play-based intervention is showing promise in being effective in improving the pragmatic language skills of children with ADHD. See Table 11.2 for more detail on the four studies.

11.8.1 Participants

The studies included 23 children with ADHD, and the majority were boys. All participants were Australian school children aged 5–12 years.

11.8.2 Play-Based Approach

All the studies used a play-based intervention approach. Play is often used by health professionals in interventions to address a child's developmental needs within everyday contexts (Ginsburg, 2007; O'Neill et al., 2012). Play has been used as a therapeutic modality for children diagnosed with several developmental, emotional and behavioural disorders to enhance their social and communication skills (Cordier et al., 2009; Cordier & Bundy, 2009).

After comparing the peer-to-peer play interactions of 350 children, of which 112 had ADHD, Cordier et al. (2009) found that, overall, children with ADHD had significantly lower social play skills than the typically-developing children in the

Table 11.2 Summary of existing pragmatic language interventions for children with ADHD

Authors & study design	Participants	Approach	Ingredients of intervention	Domains of pragmatics	Main findings
Cordier et al. (2013) [a] *Pre-post-test design*	– 14 children with ADHD – 5–12 years (mean 7.6 years) – 10 males Australian	Play-based *(clinic based)* [b]	– Therapist modelling – Peer-mediation – Video-modelling – Parent involvement	– Pragmatic protocol (PP)—only three PP components rated: Verbal aspects-topic; turn-taking; and non-verbal aspects – Structured multidimensional assessment profiles (S-MAPs)—six dimensions: Adaptive social-emotional; non-verbal communication; language use; social skills; thinking style and creativity; and executive function	– The pragmatic language of children with ADHD improved significantly from pre- to post-intervention as measured by both the PP and S-MAPs – Both children with and without structural language difficulties improved significantly from pre- to post-intervention using S-MAPs – Only children with structural language difficulties improved significantly using PP
Wilkes-Gillan, Cantrill, et al. (2017a)[a] *Pre-post-test design*	– 5 children with ADHD – 5–12 years (mean 10 years) – 4 males – Australian	Play-based *(pilot parent delivered and 18-month follow-up)* [c]	– Therapist modelling – Peer-mediation Video-modelling – Parent involvement	– Introduction and responsiveness – Non-verbal communisation – Social-emotional attunement – Executive function – Negotiation *(measured by the POM)* – Children's communication checklist (CCC-2) subscales: Initiation; stereotyped language; use of context; non-verbal communication; social relations and interests	– Children maintained all skills gained 18-months following the intervention (POM) – Compared to a normative sample, children with ADHD remained below the average range on many aspects of communication skills 18-months after intervention (CCC-2)

Authors & study design	Participants	Approach	Ingredients of intervention	Domains of pragmatics	Main findings
Wilkes-Gillan, Munro, et al. (2017b) *Pre-post-test design* [a]	– 5 children with ADHD – Australian	Play-based *(comparison of therapist and pilot parent versions)* [b,c]	– Therapist modelling – Peer-mediation – Video-modelling – Parent involvement	– Introduction and responsiveness – Non-verbal communisation – Social-emotional attunement – Executive function – Negotiation *(measured by the POM)* – Children's communication checklist (CCC-2) subscales: Initiation; stereotyped language; use of context; non-verbal communication; social relations and interests	– Pragmatic language skills at the 18-month follow-up were not significantly different from those immediately after the therapist-delivered intervention, indicating skills had been maintained – 18 months after initial intervention, children still had lower pragmatic language skills than other children their age (CCC–2). Parent ratings indicated that children scored below average on four of seven CCC-2 subscales that reflect pragmatic language – The parent-delivered intervention had a large, significant effect on the pragmatic language skills of children from pre-intervention to 1 month post-intervention
Cordier et al. (2017) *Pre-post-test design*	– 9 children with ADHD – 5–12 years (mean 8.2 years) – 8 males – Australian	Play-based *(parent delivered)* [d]	– Therapist modelling – Peer-mediation – Video-modelling – Parent involvement	– Introduction and responsiveness – Non-verbal communisation – Social-emotional attunement – Executive function – Negotiation *(measured by the POM)*	– There was a significant improvement in the overall POM score for children following the intervention – Significant improvements with large effect sizes were found pre- to post-intervention and pre-intervention to follow-up

Key: [a]These studies report findings from the same participant sample; [b,c,d] These studies are from the same play-based intervention design: [b]Initial clinic-based design; [c]Pilot parent-delivered version; [d]Parent-delivered version control group. Cordier et al. (2009) then proposed a model for a play-based intervention aimed at enhancing the social play skills and empathy of children with ADHD. The conceptual model postulated how the characteristics of ADHD influence the play interactions of children with ADHD and is based on the premise that play is the natural context where children develop social and emotional skills. The model emphasised the need to: (1) capture children's intrinsic motivation through the natural context of play and interaction; (2) facilitate the development of interpersonal empathy; (3) include a regular, typically-developing playmate for friendship development; (4) involve parents to assist skill generalisation; and (5) include therapist-modelling to promote cooperative play between the dyad (children with ADHD and their playmate) and to support children with ADHD to use the target skills as natural peer interactions unfolded.

11.8.3 Active Ingredients of the Intervention

All the studies evaluating the play-based intervention contained four key active ingredients: therapist modelling; peer-mediation; video-modelling; and parent involvement. During 1-h clinic sessions, the therapist conducted a 20-min video-feedback session with the children. To promote consistency between the clinic and home, parents also joined the video-feedback session. Children were shown 3 min of edited video footage from the previous week of themselves playing. Green slides with a key message appeared before footage of desired social skills (e.g. Great sharing ideas) and red slides appeared when skills required improvement (e.g. We can listen to our friend). The therapist discussed the footage with the children, using key terminology to assist them in identifying positive "green" actions that would make their play more fun (e.g. You can help your friend). The therapist cued parents and playmates in the conversation (e.g. 'What do you think made that play so much fun?'). The therapist then supported the children to identify three key actions to remember before entering the playroom (video-feed-forward).

While engaging in mutually enjoyable, cooperative play for 25 min, the therapist modelled the desired pro-social skills: sharing; perspective-taking; problem-solving; negotiating; and responding to a playmate's verbal and non-verbal cues. Additional support was provided to help the children negotiate when disagreements occurred. Prompts and key terminology that linked back to the video-feedback session (e.g. 'Remember to talk to fix the problem') were used. The therapist also used gestures and key words to assist children in identifying the emotional states of their playmates (e.g. 'She's turning away—too rough!') and to highlight the consequences of their actions (e.g. 'If you play your friend's game, then they'll play your game'). Parents observed these sessions through a one-way mirror. The therapist then spent 15 min with the parent discussing how the skills and strategies in the session could be implemented at home.

The playmate involved in the intervention was a typically-developing playmate who regularly interacted with the child with ADHD (i.e. a friend from school or the neighbourhood or a sibling) and had well-developed social and communication skills. Peer-mediation techniques were used where the therapist provided the playmate with strategies for facilitating cooperative play and communicating clearly with the child with ADHD. The playmate also modelled desired social and communication skills to the child with ADHD through the naturally-occurring play interactions that took place in the intervention.

Parents received training on how to deliver home modules. Module allocation was based on children's baseline social play and communication skills and ongoing therapist observations. To deliver weekly home modules, the parent read a manual chapter and watched the corresponding video episode with their child. Parents engaged their children in a discussion about the fictional characters in the video who modelled pro-social behaviours in contexts familiar to children, such as taking turns on equipment at the park. Undesirable responses (e.g. yelling at a peer) were shown before the characters modelled how to repair the social interaction (e.g. problem-solving). During four of the weeks, parents were asked to facilitate a 40-min playdate at their home, inviting the playmate involved in the study. Parents used play cards and the terminology learnt during the intervention to give the children feedback before, during, and after the playdate. The cards were green (Great play! Keep going!), red (Let's stop and think), and purple (three things to remember).

11.8.4 Evidence from Main Findings of the Play-Based Intervention

For play-based intervention aiming to improve the pragmatic language skills of children with ADHD, the body of evidence provides some support for using a play-based approach to target pragmatic language outcomes. However, caution is required in drawing conclusions from the findings of these pilot studies. All the studies that contained a longitudinal component found pragmatic language skills were maintained when measured observationally. Further, all studies reported significant improvement in pragmatic language skills pre- to post-intervention when observational measures were used. However, no significant improvements were found when standardised language measures were used. Play-based approaches are particularly useful as they offer a naturalistic, age-appropriate, ecologically-valid context for promoting skill development as well as a context in which peers and parents can be incorporated into the intervention process (Bundy, 2012; O'Neill et al., 2012).

11.9 Directions for Future Research

Currently, a large body of research focuses on measuring the capacity for pragmatic language in children with ADHD. There is an identified need for large-scale, observational studies to identify all difficulties children with ADHD have with *performanc*e of pragmatic language. There is also a need for further refinement of measurements regarding the *capacity* for pragmatic language to include such assessments in naturalistic settings and to replace or augment parent and teacher proxy-reporting. Combining reliable and valid measurements of both capacity and performance of pragmatic language in naturalistic settings will help to develop profiles of pragmatic language difficulties for ADHD. Understanding the profiles of these difficulties will then, in turn, help to inform interventions. Currently, there is only a small body of preliminary evidence on one intervention that aimed to improve the pragmatic language of children with ADHD. Given the importance of pragmatic language on children's social interaction skills, there is need for further research into interventions aiming to improve the pragmatic language difficulties of children with ADHD. These interventions should aim to use more rigorous research designs (e.g. randomised controlled trials) with larger and more diverse samples of children and across a range of environments where children are required to use pragmatic language skills (e.g. home, school, community).

11.10 Summary

Children with ADHD experience significant pragmatic language difficulties, both in terms of the magnitude of difficulty they experience, as well as the quality of pragmatic language difficulties. They experience difficulties in pragmatic language in relation to capacity, that is, knowledge of pragmatic language as well as performance of pragmatic language, that is, using pragmatic language in naturalistic contexts.

Researchers have mainly used measures of pragmatic language capacity in observational studies to describe the pragmatic language difficulties of children with ADHD. The type of measures used were mainly parent and teacher proxy-report questionnaires and standardised, norm-referenced assessments of pragmatic language knowledge. Despite the advantages of using observational measures to assesses pragmatic language performance, there is a dearth of such assessments available. The POM-2 was developed and validated to address this need.

Very few studies have been conducted to date that evaluated the effectiveness of interventions aimed at addressing the pragmatic language of children with ADHD. This is particularly surprising against the backdrop of robust evidence of the mediating role of pragmatic language in the social skills difficulties of children with ADHD. We could only find four studies that evaluated a play-based intervention to address pragmatic language difficulties in children with ADHD. While the findings are promising, there is an urgent need for more studies using robust research designs.

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Part II
Complex Populations in Adulthood

Chapter 12
Right-Hemisphere Language Disorders

Emelia Lázaro García, Perrine Ferré, and Yves Joanette

12.1 Introduction

Individuals with acquired lesions of the right hemisphere can be compromised in their ability to understand and express pragmatic aspects of communication. This group of historically overlooked and underserved patients has presented clinicians and theoreticians with the possibility that an acquired brain lesion could interfere with pragmatic abilities. The goal of this chapter is to describe the pragmatic disorders that can arise following a lesion to the right hemisphere and that can be part of a more pervasive language impairment. After a short historical overview, we will describe the pragmatic deficits that may be present when the right hemisphere is damaged. Then, we will consider the latest evidence regarding incidence of pragmatic deficits following damage to the right hemisphere and the different clinical profiles that have been identified. The types of interventions currently available to minimize pragmatic deficits in individuals with right-hemisphere lesions will be discussed. Finally, we will discuss the importance of current knowledge to the modern concept of aphasia.

By the end of this chapter, we hope that readers will (1) understand the possible pragmatic impairments that can be present in adults suffering from right-hemisphere lesions related to stroke, head trauma, tumour, or any other aetiology; (2) grasp the incidence and the different profiles of such pragmatic deficits; (3) understand the newly developed intervention strategies that can be used to help individuals with pragmatic deficits communicate better; and (4) be aware of the ongoing discussions regarding the inclusion of such pragmatic deficits in the long-standing concept of aphasia.

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12.2 Historical Background

It has long been known that a brain lesion can impair the ability to speak. This observation was central in building interest in brain-behaviour relationships. According to Prins and Bastiaanse (2006), post brain-lesion-acquired 'speechlessness' episodes were reported as early as 1700 B.C. in an Egyptian papyrus. However, it was only at the end of the nineteenth century that pioneers such as Paul Broca (1863), Carl Wernicke (1874), and John Hughlings Jacksons (1878) started to truly understand the impact of acquired brain lesions on language abilities. It was then that the term 'aphasia' was introduced by Trousseau (1864). The term is still used today to refer to language disorders resulting from acquired brain lesions. The main discovery of the late nineteenth century—the one that would change forever the way we understand how our brain is organized for language and other cognitive functions—was the observation that aphasia essentially resulted from lesions to the left hemisphere in right-handers (Dax, 1865; Broca, 1865). In fact, the association between the left hemisphere and language constituted a major revolution in brain research. We know today that most cognitive abilities—including language abilities—are sustained by complex neurofunctional networks that are not equally distributed over the two hemispheres, resulting in what has been re-conceptualized as hemispheric cooperation (Sergent, 1994).

If the end of the nineteenth century is remembered as the time when the association between the left hemisphere and language was revealed, it is also the time when any role for the right hemisphere in language was ruled out. This discovery was reflected in nearly a century of belief that the right hemisphere played no role in language abilities. Thus, until the second half of the twentieth century, right-hemisphere lesions in right-handers were considered to have no impact on language abilities. However, converging lines of evidence from linguistics and clinical observations contributed to rehabilitating the role of the right hemisphere in some language abilities.

The first stream of evidence stems from the astute clinical observations of some clever clinicians (Eisenson, 1962; Weinstein, 1964; Critchley, 1962). Among others, these clinicians reported that, despite the absence of prototypical aphasic signs, individuals with a right-hemisphere lesion did not have full access to their communication abilities. Yet, this condition was difficult to characterize at that time. Theories of language were mostly limited to the components identified at the end of the nineteenth century: articulation, phonology, word finding and syntax. In the absence of a theoretical framework, Eisenson (1962) used expressions such as an impairment of the 'supra-ordinary' aspects of language.

Approximately a decade later, a second line of evidence provided the conceptual framework needed to refer appropriately to aspects of language that had hitherto been overlooked by traditional language theories. This framework captured for the first time the pragmatic dimensions of language. Initiated by theorists such as Grice (1975) and Searle (1979), the field of pragmatics started to emerge and to transform itself into a recognized component of linguistics (Cummings, 2009). In the same way that Lois Bloom and Margaret Lahey revolutionized the approach to language development and language disorders in children by focusing not only on the form of language but also on its content and use (Bloom & Lahey, 1978), the availability of theoretical frameworks of pragmatics transformed clinical impressions of language impairments in individuals with a right-hemisphere lesion. It is possible impairments of pragmatic components of language in adults with a right-hemisphere lesion that will be examined in the following sections.

12.3 Pragmatic Language Disorders in Adults with Right-Hemisphere Damage

A right-hemisphere lesion in a right-handed adult can be the cause of many impairments that can interfere with communicative abilities (Joanette et al., 1990). These include impairments of the processing—expression and/or comprehension—of the prosodic component of language and deficits affecting the semantic processing of words. However, in this chapter, the focus will be on the pragmatic deficits that can occur in individuals with such lesions. Thus, this chapter focuses on what can be thought of as one of the most characteristic language impairments that can occur following right-hemisphere damage (RHD). This is not to say that pragmatic deficits are pathognomonic of the language deficits in individuals with RHD. In fact, such deficits may also be present following traumatic brain injuries, in the early stages of dementia, in many psychiatric conditions, and in some individuals with aphasia. However, it is probably true that pragmatic deficits are best seen in isolation among RHD individuals. This is in sharp contrast to the impact of left-hemisphere lesions in individuals with aphasia, characterized by difficulties in one or more of the formal components of language (phonology, syntax, lexical semantics). On the contrary, RHD individuals do not have obvious problems with the form of language, but they do have difficulties with the context and use of language (Foldi et al., 1983). When present, pragmatic disorders in RHD individuals interfere with their ability to process the intention of communication, not its formal dimensions.

In this context, the current description of pragmatic language deficits in individuals with a right-hemisphere lesion will focus on discourse and, in particular, conversational abilities that call for the interpretation and encoding of the intention of communication.

12.3.1 Pragmatic and Discourse Abilities

Although pragmatics and discourse are frequently analysed separately in the literature, it is evident that, for discourse to take place, the role of pragmatics is fundamental. The ability to extract the gist of discourse, while reading a text or listening to a speech, and relate it to our own knowledge of the world is essential for appropriate behaviour and social interactions (Martin et al., 2018). The same happens with the planning and production of discourse. Discourse abilities allow the transmission of information from one speaker to another in the form of narrative, conversation or procedures (instructions). The exchange of information involves the expressive and receptive components of communication, as a message is transmitted or received. The most studied discourse forms are narrative and conversation, which can be particularly strongly affected by a lesion in the right hemisphere (Joanette et al., 1990; Wilson et al., 2018), notwithstanding the fact that they do not appear to be specific to this type of injury. In fact, some of these 'alterations' may also appear in 'normal' subjects or in other types of brain injuries.

Conversational discourse is almost invariably the first to develop in infants and the most used in everyday communication. This form of discourse is considered as a highly contextualized form of language use, and an intentional activity directed towards particular goals (Levelt, 1989). In this complex communicative situation, pragmatic abilities play a fundamental role and different deficits may arise in cases of RHD. In many such patients, the breach of the cooperative principle that regulates conversation is well known. Among other things, they show difficulties respecting speaking turns, adequately sustaining eye contact with their interlocutor, and controlling the progression and coherence of the topic during the exchange. Some also find it difficult to adapt their verbal production to the situational context and to the knowledge shared with each interlocutor (Chantraine et al., 1998). In this situation, patients produce inappropriate comments that may be redundant or, on the contrary, may disorient the interlocutor because they consider something to be common knowledge when it is not.

Alternatively, the narratives of patients with RHD are characterized as being frequently—on the expressive level—not very informative, although the number of statements produced may be similar to that of control subjects (Joanette et al., 1986). Characteristics such as a tendency to produce incoherent discourse, expressed through erroneous or absent anaphoric references, a tendency to digress or change topics tangentially, and the absence of thematic progression are part of the typical profile of RHD patients (Blake, 2006). At the receptive level, it is possible to observe difficulties integrating the set of elements in a story into a coherent whole in order to be able to make the necessary inferences to form an adequate understanding of the text. Patients also have difficulties processing some types of inferences. Comprehension problems are also manifested in difficulty rejecting incoherent interpretations of a text and in interpreting implicit information, for example, finding the moral of a fable. Failing to grasp the central idea of a speech, having problems suggesting a title for a story and even finding it hard to choose a phrase that summarizes the main theme are also common in patients with RHD (Joanette et al., 2008).

12.3.2 Pragmatic and Non-literal Language Abilities

Non-literal language abilities are important in social interactions because a large proportion of everyday communication is not transmitted directly, but rather makes use of pragmatic aspects of language such as indirect speech acts, irony, humour, sarcasm, and metaphor. Obviously these abilities are also fundamental in the context of discourse—mainly conversational—so it should be clarified that their presentation in a different section simply follows a 'didactic' decision. Non-literal language abilities, as their name implies, require the ability to process more than the literal meaning of an utterance in order to grasp the speaker's communicative intention (Sabbagh, 1999) in a given context, and to decide whether a sentence means what is said or more than what is said, as in the case of indirect request. We present these abilities in more detail below.

12.3.2.1 Interpretation of Indirect Speech Acts

The term 'speech act' describes an utterance in which there is an underlying intention to perform an act such as an order, a question, a promise, a wish or an assertion. Most of the time, speakers explicitly convey what they mean in an utterance, that is to say, the speaker's intention is explained by the linguistic form itself in a direct speech act. An example frequently used in the literature is when one person says to another 'Please close the window.' In this case, the message is issued as a direct speech act. Nevertheless, in daily communication it is not unusual for meaning to be conveyed indirectly, in which case the speaker's intention is not always explicit. For example, if a person enters a very cold room and says 'The window is open,' the speaker is not merely making an assertion, but is indirectly asking or suggesting to the listener to close the window because the room is cold. In this case, it is necessary for a hearer to be able to modify the literal interpretation in order to grasp the intention of the speaker in a given context. Different theories of speech acts (Searle et al., 1980; Gibbs, 1999, 2001; Clark, 1979) have been developed with the aim of reaching a better understanding of the psycholinguistic mechanism involved.

In sum, figuring out the speaker's intent requires the simultaneous integration of a lot of information, concerning the context, the speaker, previously acquired knowledge or knowledge we share with the speaker. Several studies have reported that RHD patients have difficulties in processing and understanding such indirect speech acts (Foldi, 1987; Stemmer et al., 1994; Vanhalle et al., 2000). Some authors have suggested that patients do not seem to have problems interpreting simple expressions, but they fail when a more elaborate interpretation is required, depending on the context or with non-conventional indirect speech acts (Stemmer et al., 1994). Other authors have suggested that deficits occur when the comprehension of the speech act demands a metacognitive analysis in an unfamiliar context (Vanhalle et al., 2000). The study of 'theory of mind' (ToM) in individuals with RHD has recently increased our knowledge of their difficulty processing communicative intention. We use this expression to denote the ability to form representations of other people's mental states and use these representations to understand, predict, and judge their statements and behaviours (Apperly, 2012). In this sense, the interpretation of indirect speech acts, which requires the ability to attribute mental states to others, is one example of ToM. According to this proposal, the problems patients with RHD have interpreting indirect speech acts could be conceived as a particular kind of disorder of ToM abilities (Champagne-Lavau & Joanette, 2009; Weed et al., 2010; Happé et al., 1999).

12.3.2.2 Processing of Metaphors

The use of metaphors is common in everyday language, and their processing, similarly to that of indirect speech acts, involves the recognition of an intention that is different from the literal meaning of the utterance (Joanette et al., 1990). As Bambini and Resta (2012: 41) point out, metaphor may be seen as a 'paradigmatically pragmatic phenomenon,' involving 'a gap between the conventional meaning of words and their occasion-specific use'—in other words, the distinction between pragmatics and semantics. According to Thibodeau and Boroditsky (2011), a metaphor can provide a succinct, vivid, and richly connotative description by drawing parallels between two distinct concepts.

The processing of metaphors is not limited to the lexical level, but can include sentences and even complete stories that ultimately express a message different from the literal meaning. Studies have focused on analysing the differences in the processing of conventional metaphors, also known as frozen, familiar or dead metaphors, versus new ones (Diaz & Eppes, 2018; Gagnon et al., 2003). The former may become lexicalized or embedded in semantics, similar to a definition of a single word because of their frequent use, so their processing may be quite similar to the comprehension of literal sentences. Alternatively, understanding novel instances of metaphoric language requires the dynamic integration of several distinct concepts. Factors that influence the comprehension of metaphors, such as novelty, comprehension difficulty, and the role of context have also been analysed in considerable detail (Diaz & Eppes, 2018; Bambini et al., 2014; Bambini & Resta, 2012).

Studies in patients with RHD have been particularly helpful in our current understanding of the impact of right hemisphere lesions on the processing of metaphors, despite the fact that there are still divergencies amongst studies. Among the earliest studies, Winner and Gardner (1977) observed that RHD patients choose images that represent the literal meaning of familiar metaphorical phrases more frequently compared to individuals with a left-hemisphere lesion as well as normal participants. However, the same RHD individuals were able to give a successful verbal explanation of these expressions, which lead Winner and Gardner to the conclusion that RHD individuals understand the metaphors, but they are not able to identify the appropriate situation in which a specific expression is relevant. Foldi et al. (1983) obtained similar results, emphasizing that there are no differences between RHD individuals and controls in terms of the verbal understanding of metaphors. Since individuals with right-hemisphere damage can also present visuo-spatial or visuo-perceptual problems, Joanette et al. (1990) suggest that this condition could interfere with such a verbal/picture matching task.

Other studies have focused on lexical analysis, considering the metaphorical meanings of polysemic words. After analyzing the performance of RHD patients and patients with left-hemisphere damage (LHD) in an auditory lexical decision task, Tompkins (1990) showed that in both groups of patients reaction time is longer for objectives preceded by a metaphorical word pair than a literal word pair, which suggests that different underlying processing is required for metaphorical meanings than for literal meanings. Brownell et al. (1984) studied the comprehension of the meaning of simple words in RHD patients and showed the tendency of these patients to privilege the literal meaning of metaphorical words. Another study focusing on the processing of the metaphorical meanings of words found a genuine difficulty in the ability of RHD patients to process such meanings (Gagnon et al., 2003). However, this study also reports difficulties in patients with LHD, thus minimizing the specificity of the result to the RHD patients, as this result could be due to other linguistic impairments in word processing. In summary, it is difficult to arrive at a clear picture of the impact of a right-hemisphere lesion on the ability to process metaphors or the metaphorical meaning of words, but there is general agreement that at least a sub-set of RHD patients can exhibit deficits in this area (see Sect. 12.4.2).

12.3.2.3 Appreciation of Irony, Jokes, Humour and Sarcasm

Humour, jokes, irony and sarcasm are also cases in which the speaker's intention cannot be extracted from the literal interpretation of the message. In these cases, the speaker's intention can only be understood if the particular context in which the communicative act occurs is taken into account. For example, when someone says 'How elegant you are!' to a person who is very poorly dressed, that particular communicative context determines the ironic meaning of the expression. Humour, jokes, irony and sarcasm can be considered as specific types of indirect speech acts. The fact that numerous studies have specifically examined these abilities constitutes a rationale for covering them in a separate section.

Clinical observations suggest that alterations in the comprehension of communicative intentions affect the understanding of humour and sarcasm in patients with RHD. For example, Kaplan et al. (1990) and Winner et al. (1998) have suggested that the problems these patients have distinguishing a joke or an ironic statement from a lie might be attributed to difficulties understanding the intentions of a story's protagonist. The ability to distinguish a joke from a lie, for example, requires an intact capacity to recognize that other people have beliefs that may differ from one's own. In the case of non-literal language, the listener must be able to distinguish what the speaker actually says from what he or she intends to convey. Therefore, a correct interpretation of an utterance's meaning relies on a correct comprehension of the speaker's intentions. To understand how a listener can interpret an ironic or false utterance, it is necessary to comprehend what the listener knows and what the speaker thinks the listener knows, thus requiring intact ToM ability. Such a complex processing of the speaker's intention can be referred to as requiring a meta-representational judgment (Gibbs, 1999).

Meta-representational judgment is also necessary for the interpretation of different types of figurative language such as sarcasm. Some studies have established a relationship between the alteration of executive functions and decreased ability to interpret sarcasm in patients with frontal lobe lesions (see, for example, McDonald & Pearce, 1996). In another study, Shammi and Stuss (1999) observed impaired abilities to process humour in two patients with right frontal damage, but also in a patient with left frontal lesions and three patients with bilateral frontal lesions. They concluded that the ability to process and understand humour is related to lesions of the anterior portion of the right frontal lobe, a conclusion that should be treated with caution, considering the limited number of subjects in the study and the variability in lesion sites. Nevertheless, it is noteworthy that the authors established a relationship between executive functions (working memory, mental flexibility, verbal abstraction) and appreciation of humour by all patients with damage to the frontal lobe.

The relationship between ToM abilities and executive functions in RHD patients with pragmatic impairments was examined by Champagne-Lavau and Joanette (2009). Their results suggested that the ability to understand pragmatic aspects of language is closely related to the ability to make inferences about other people's intentions (ToM). Even more interestingly, the authors reported that the association between ToM deficits and deficits in executive functions better correlated to the presence of pragmatic impairments than if there is only executive deficit.

In summary, patients with a right-hemisphere lesion can exhibit challenges in the pragmatic processing of non-literal discourse such as irony, humour, jokes and sarcasm. However, the underlying cognitive processes that account for such impairments are still to be clearly understood. Despite this lack of knowledge, it is important to be aware of the possibility of such impairments in this clinical population and to offer support to the patient and his or her family. The section concludes with brief discussion of two cases in which pragmatic impairments compromise everyday communication between patients and their families. In only one of these cases are the impairments correctly recognized and addressed in therapy.

Pragmatic Impairment: Clinical cases

When present following a right-hemisphere lesion, pragmatic impairments can interfere with everyday communication. It is only recently that clinical tools and proper training have allowed clinicians to recognize and support patients with pragmatic impairments and their families. Here are two examples of the impact on everyday life of pragmatic impairments in patients following a right-hemisphere stroke. The first case illustrates the presence of

(continued)

pragmatic impairments in a male patient. This patient's pragmatic impairments were not recognized and addressed. The second case describes pragmatic impairments in a female patient. The clinical management of this patient focused on her pragmatic impairments and the impact of these impairments on the patient and communication with her family.

CASE 1: Inappropriate clinical management of pragmatic deficits

In 1995, Joseph (not his real name), a 66-year-old man, suffered a right-hemisphere stroke. Although he could still talk, his wife and children found him "changed" and gradually lost pleasure and interest in communicating with him. They had noticed that since his stroke, he seemed to be not very attentive: he interrupted his interlocutors, went from one subject to another, and had difficulty following the topic of the conversation. In addition to problems complying with social rules in conversation, Joseph exhibited tangential discourse and comprehension difficulties, which made conversations disjointed. These pragmatic communication impairments gradually isolated Joseph from his relatives. Joseph consulted a speech-language pathologist at a rehabilitation center. The evaluation focused on the presence of an accompanying mild dysarthria, dysphagia, and facial palsy and concluded that Joseph had preserved abilities processing sounds, words and syntax. His discourse difficulties were not specifically noted and remained unaddressed.

CASE 2: Appropriate clinical management of pragmatic deficits

In 2007, Béatrice (not her real name), a 71 year-old woman, suffered a right stroke. Her husband reported she had "changed": she spoke a lot but her husband did not fully understand Béatrice's intentions, particularly the subtleties of her discourse. During her stay in a rehabilitation center, the speech-language pathologist evaluated Béatrice and then explained to the couple that these changes in communication behaviours were common after a right-hemisphere stroke. She proposed a more in-depth evaluation and a therapy program to reduce the impact of her pragmatic impairments on everyday communication, as well as counselling for her husband to use accurate strategies in conversation.

12.4 Clinical Perspectives on Communication Deficits in RHD

It is now relatively easy to have an idea of both the incidence and the characteristics of clinical profiles in aphasias linked to left-hemisphere damage. However, this is not true of pragmatic deficits. Factors such as the heterogeneity of clinical features, the limited availability of assessment tools that would allow one to describe the existence of pragmatic deficits, and the lack of consensus on the clinical labels to use to refer to patients with pragmatic disabilities make the epidemiological data unclear. There is still discussion of whether or not pragmatic disorders following an acquired lesion to the brain can be included in the clinical concept of aphasia. In our view, there is no doubt that pragmatic deficits are a language impairment and should be included in the clinical concept of aphasia (Joanette & Ansaldo, 2000; Joanette et al., 2018). For other readers, isolated acquired pragmatic disorders following RHD should be referred to as cognitive-linguistic deficits (Myers, 1999a). The existence of this debate over how isolated pragmatic deficits should be classified, and the absence of adequate clinical tools, have contributed to the fact that RHD patients with pragmatic deficits have been underserved for many years. This situation is even worse regarding the clinical interventions to be offered to such a clinical population. There are few intervention strategies for individuals with acquired pragmatic deficits and the field is still largely unexplored (see Ferré et al., 2018). Fortunately, a group of experts in the field was recently brought together by Margaret Blake with the aim of improving consistency in how language and communication problems in adults with RHD are diagnosed and treated (Blake & Johnson, 2019).

12.4.1 Incidence of Pragmatic Deficits in RHD

Currently, it is known that pragmatic deficits do not occur exclusively in cases of RHD (whether due to a vascular aetiology or a traumatic brain injury). Although less prototypical, some pragmatic difficulties can be observed in the presence of aphasia in cases of left-hemisphere stroke (Fridriksson et al., 2006; Glosser & Goodglass, 1990). In addition, some studies have shown that these difficulties are present not only in advanced dementias, for example, Alzheimer's disease, but also in early stages, such as mild cognitive impairment (Basaglia-Pappas et al., 2014).

Although the incidence of pragmatic disorders following RHD is difficult to estimate accurately, it is known that these disorders impair communication abilities and significantly limit an individual's interactions and social roles, regardless of aetiology. Moreover, these effects may persist for many years after the event that caused them (Ferré et al., 2012; Vallat-Azouvi & Chardin-Lafont, 2012). However, it is also recognized in clinical practice that not all patients with right-hemisphere lesions present such deficits. According to Joanette et al. (1991), approximately 50% of patients with RHD may be affected by one or more communication disorders. Particularly in the case of traumatic brain injury—a condition which usually involves both hemispheres—it is estimated that between 35% and 70% of adolescents and adults will have a cognitive impairment that impacts daily functioning after severe injury (Max et al., 1997).

Alternatively, it has been estimated that the proportion of communication deficits following a right-hemisphere stroke ranges from 50% in the general population (Benton & Bryan, 1996), to 78% in rehabilitation settings (Ferré et al., 2012). The manifestations of neurodegenerative diseases such as Alzheimer's disease must also be considered. In these cases, it is common for the early stages of the disease to affect the pragmatic components of communication. In fact, it is even possible that such disorders may be part of the initial or prodromal phase of neurodegenerative diseases affecting cognition (Basaglia-Pappas et al., 2014).

12.4.2 Profiles of Communication Disorders in RHD

A breakthrough in our understanding of right-hemisphere language disorder was achieved when it was proven that the left hemisphere did not have exclusive control over language abilities. The integrity of the right hemisphere is necessary for a number of language components, as shown by the descriptions of communication impairments in individuals with RHD (Joanette et al., 1990; Code, 1987). It is now widely accepted that a right-hemisphere injury can affect pragmatic, prosodic, lexico-semantic, and discourse aspects of communication (for a review see Myers, 1999a and Tompkins, 1995).

However, most studies have described deficits affecting these components of communication separately. Only very recently have attempts been made to explain their co-occurrence. To our knowledge, one of the first studies to consider, at least partially, the question of clinical profiles of communication impairments in adults with RHD was performed by Myers and her team (Blake et al., 2002; Myers, 2005). That study, which explored the relationship between perceptual integration deficits and verbal expression, reported the presence of some heterogeneity in the communication deficit profiles of the individuals evaluated, but was not able to clearly identify distinctive profiles. Another pioneering study on the subject was carried out by Joanette et al. (1991). After analysing the performance of participants with RHD, the authors found that not all such patients present with communication disorders. They also noted that, when communication impairments are present, the impaired abilities varied among participants, leading to heterogeneous profiles.

Once the possibility of exploring the existence of different clinical profiles was raised, different studies addressed the question in increasing depth (for a detailed review see Côté et al., 2007 and Ferré et al., 2009, 2012). In general, these studies have highlighted heterogeneity among individuals. Nevertheless, the existence of quite homogeneous subgroups of clinical deficits also emerges. Based on the results obtained, it has been possible to establish a potential taxonomy that considers two general aspects: (1) among RHD patients, there is always a subgroup of individuals who do not present with communication deficits; and (2) among those who are reported to have a communication disorder, distinct profiles are found. There is a group with massive, generalized difficulties, a group with mild deficits affecting mainly prosody and conversational skills, and a group with semantic and/or pragmatic deficits.

Although the site of the right-hemisphere lesion is not systematically correlated with the pattern of impaired communication abilities (subgroup) (Côté et al., 2007), the nature of the lesion (haemorrhagic vs. ischemic) still appears to impact conversational abilities differently (Lajoie et al., 2010). The time post onset has also been considered (Blake et al., 2002; Ferré et al., 2012). It does not appear to influence the distribution of profiles, suggesting that difficulties continue in the chronic stages after the RHD. However, studies by Ferré et al. (2009, 2012) were directed at possibly identifying communication profiles among RHD individuals in different groups of patients speaking Romance languages. Their results showed that the profiles observed in the different groups (coinciding with the previous studies) were influenced, at least partially, by demographic variables (age and education), but were only weakly linked to cultural origin. This suggests the existence of cross-cultural profiles.

Finally, the identification of clinical profiles has in turn raised the possibility that these profiles could be correlated with other cognitive disorders, including dysexecutive syndrome (mental flexibility, inhibition, shared attention mechanisms) (Myers, 2005) or ToM deficits (Champagne-Lavau & Joanette, 2009; McDonald, 2000). This possibility should be further explored in the near future.

12.4.3 Assessment and Rehabilitation Strategies for Communication Deficits in RHD

The communication disorders that can be experienced by RHD individuals have direct and obvious consequences for their day-to-day social activities. Interventions by speech and language pathologists (SLPs) are therefore crucial to adequately support RHD individuals and their families in dealing with their pragmatic communicative difficulties. Yet the level of evidence regarding RHD interventions has not achieved the high-quality standards required to formulate specific therapeutic recommendations. In the RHD population, evidence is still limited regarding the three major components of evidence-based practice: the scientific literature; clinical expertise; and client characteristics (Kennedy, 2013; Sackett et al., 1996). Still, clinical guidelines can be deduced from existing knowledge driven by each of these three components.

First, there is still a lack of scientific evidence regarding RHD and intervention strategies for pragmatic abilities. Sometimes, conclusions may be drawn without consideration of the aetiology of RHD, such as traumatic brain injury or stroke. Systematic reviews that address efficacy evidence for cognitive treatments in general (Cicerone et al., 2005; Finch et al., 2016) do not include data specific to the post-stroke RHD population, while traumatic brain injury has been more widely investigated. Although discourse characteristics are at least partially comparable between the two aetiologies (Coelho et al., 2003), their clinical manifestations are different (Penn, 1999). This can lead to confusion among clinicians, for example regarding frontal/executive disorders, which can appear concomitantly—although not consistently—following a right-hemisphere stroke (Monetta et al., 2006; Monetta & Pell, 2007; Ferré et al., 2013).

Regarding the pragmatic deficits that specifically characterize RHD individuals, the quality of available evidence regarding intervention is still maturing. An evidence-based systematic review of RHD-related communication treatments in 2013 identified only five studies that passed methodological quality criteria (Blake et al., 2013). This does not mean that no such interventions exist, but rather that most investigations in this field rarely manage to achieve controlled designs, such as a controlled trial or a controlled case study (e.g. ABA or multiple-baseline design). Nonetheless, single case reports can provide valuable information (Tate et al., 2013), especially considering that the constrained inclusion criteria that characterize controlled trials may limit generalization to the—generally complex—population encountered in clinical practice. Overall, the strength of the evidence is still low regarding the efficacy of pragmatic-conversational treatment following right-hemisphere stroke (Cappa et al., 2005). However, the preliminary evidence is encouraging and clinical guidelines can be deduced from the existing literature, as described below.

In addition, clinical expertise has only recently begun to emerge. Hitherto, communicative pragmatic disorders were mostly ignored in the health care continuum. Although this situation has now improved in many countries (Löfgren & Johansson, 2015), as has the availability of appropriate theoretical frameworks and assessment tools, inequities still exist (Côté et al., 2004; Hewetson et al., 2017). RHD individuals are often discharged without SLP-monitored therapeutic services and left to live with a communication disorder. For clinicians, the direct corollary is poor exposure to this clinical population. Consequently, clinicians may feel they lack expertise and confidence in dealing with this population. Clinical expertise is therefore scarce.

As for the third component of evidence-based practice, preferences of individuals with RHD can be hard to collect. Anosognosia or limited awareness and insight is one of the concomitant disorders frequently present after a right-hemisphere stroke. As a result, patients' motivation and adherence to the therapeutic project may be low, as may their ability to incorporate explicit strategies into everyday situations, thus compromising therapy success. In our experience, family members of RHD individuals can be solicited for this purpose, but they too tend to underestimate or exaggerate their relatives' pragmatic disorders, in comparison to conclusions drawn from an SLP's assessment.

Knowledge in this field is clearly still 'under construction' and SLP therapeutic practices designed for RHD pragmatic disorders will need time to achieve an evidence level that is comparable to that in the aphasic population. Nevertheless, even in the absence of strong evidence-based literature, some *clinical guidelines* can be derived from the existing knowledge.

First, it is crucial to remind readers that a *thorough, appropriate assessment* is the key to proper referral to SLP intervention services and to the establishment of therapeutic goals that matter to the patient and his or her family. Appropriate tools are tests and protocols that have been tailored to and validated on the population of interest. In this sense, assessment tools designed for general populations exhibiting pragmatic disorders are available today. They include, for example, the ASHA Functional Communications Measure (American Speech-Language-Hearing

Association, 1998) and the Functional Assessment of Communication in Adults (Frattali, 1998). However, pragmatic aspects of communication are not successfully assessed by traditional aphasia batteries. It is now accepted that highly complex or highly structured tasks will intensify the deficits and the heterogeneity observed in the performance of RHD adults with communication disorders (Marini et al., 2005).

It is worth noting that assessment tools for RHD are available in several languages. In English, three batteries have been published: the Burns Brief Inventory of Communication and Cognition (Burns, 1997); the Mini Inventory of Right Brain Injury—Revised (Pimental & Knight, 2000); and the Rehabilitation Institute of Chicago Evaluation of Communication Problems in Right Hemisphere Dysfunction—Revised (Halper et al., 1996). Recently, our group led the publication of the Montreal Protocol for the Evaluation of Communication (Joanette et al., 2017), the English version of a protocol first published in French and meant to evaluate possible communication deficits in RHD individuals (Joanette et al., 2004). This protocol is also available in several other languages, including Spanish (Ferreres et al., 2007), Portuguese (Fonseca et al., 2008; Kerr et al., 2015), German (Scherrer et al., 2017) and Italian (Tavano et al., 2012).

Second, interventions should be *centred on the person*, rather than on his or her impairment (Grant, 1990). This recommendation is obviously not exclusive to the RHD population, and will potentially enhance the therapeutic satisfaction of any group. Yet, the lack of awareness and knowledge of communication impairments that is so often encountered can limit therapeutic adhesion. A person-centred approach gives the individual and his or her family a chance to invest in the therapeutic process and maintain motivation. In this sense, therapeutic services should also extend to the person's direct environment (family, community) if that is his or her wish. A person-centred approach should guide the therapeutic process, from assessment to discharge.

Third, but not least important, therapeutic approaches should aim at a *generalization of treatment efficiency* to other items, activities or skills. This applies whether the therapy is task-specific or process-oriented (Myers, 1999b). The limitations of approaches that focus directly on the deficient process for the RHD population are that (1) the underlying processes impacting the ability need to be known and defined, which is often not the case with RHD individuals, as described above; and (2) their success relies in part on the hypotheses that the person is motivated to change his or her behaviours and is able to transfer the acquired knowledge to everyday activities.

Unfortunately, these hypotheses sometimes come up against the presence of anosognosia (Schmidt, 2014; Ylvisaker et al., 2005). SLPs can then favour approaches that *bypass or address deficit awareness issues*. Treatments that use implicit learning bypass awareness issues and have proven efficient in supporting comprehension of narratives and secondary lexical meanings, with some evidence of generalization (Blake et al., 2015; Tompkins et al., 2011; Tompkins & Scott, 2013). Alternatively, SLPs can use approaches that directly address the awareness issue. Metacognitive approaches are valuable tools to alleviate the limitations caused by reduced awareness levels in therapeutic efficiency (Kortte & Hillis, 2011; Schmidt, 2014; Ylvisaker et al., 2005; Cherney, 2006). These approaches incorporate both knowledge and regulation of cognition. Explicitly supporting self-criticism and self-regulation through strategy learning and coaching has been validated with traumatic-brain-injured individuals with pragmatic deficits (Dahlberg et al., 2007; Ylvisaker et al., 2005). Planning, self-judgement and problem-solving issues can, for example, be addressed through a structured, supervised process to identify and explore alternative actions to solve a problem (e.g. IDEAL, Bransford & Stein, 1993), as well as the use of feedback and modelling (Youse & Coelho, 2009). The use of visual feedback (pictograms illustrating expected or inappropriate pragmatic behaviours) or verbal feedback (positive or corrective reinforcement) is promoted (Fleming & Ownsworth, 2007).

However, knowing a strategy to regulate a behaviour does not mean it will be used in a given context. It is, therefore, essential to *present the therapeutic steps in a logical order* by having the patient experience a problematic situation, understand it, and train himself or herself to recruit alternative strategies. Gradually, more complex situations can be introduced. The items, the feedback, the context and the complexity can be varied: for example, varying the number of cues, the amount of shared knowledge, the type of inference (logical vs. social); practising in individual and group contexts; changing settings; and including significant others are all ways to increase the inherent complexity of a treatment (Ferré et al., 2011). A wide variety of training contexts will also favour generalization of the acquired strategies to new contexts. SLPs are encouraged to bring their therapy outside their offices and take advantage of pluri-disciplinary settings to train patients with different interlocutors and vary the practice environment. Ultimately and ideally, training of family members and community should be included in the discharge plan. Crucially, in some cases, problematic experiences only arise when the patient goes back to live in the community. Raising awareness and implementing strategies might, therefore, be much easier for the SLP and more natural for the person at this stage of the care continuum (Godfrey et al., 1993).

In a task-centred approach, the choice of activity to be trained for pragmatic disorders can be arduous. In the absence of client-suggested goals in a specific activity, *conversational discourse* can be a valuable choice to train discourse-pragmatic skills. Conversational discourse can be used simultaneously as a therapeutic goal (e.g. ‘manage an informative conversation with relatives on a familiar topic’) and a therapeutic tool (e.g. ‘choose a familiar topic of conversation and offer visual feedback to adjust the quantity and quality of the information online’) (Papathanasiou et al., 2011). During conversation, therapeutic metacognitive strategies include the use of audio or video recordings (Braden, 2014; Dahlberg et al., 2007; Youse & Coelho, 2009). This type of approach—metacognitive and focused on conversational skills during a conversational activity—has generated encouraging results with regard to the informativeness of the discourse (quantity and quality) and adaptation of non-verbal behaviours (e.g. eye contact, facial expressions) (Cannizzaro & Coelho, 2002; Cheippe & Magnard, 2016). Preliminary results from a pilot study also showed an impact on indirect speech act comprehension (Sigouin, 2007).

12.5 Right-Hemisphere Language Disorders, Pragmatics and Aphasia

The fact that a lesion in a right-handed individual's right hemisphere can result in disorders of the pragmatics component of language has now been ascertained. The purpose of this chapter was to describe those possible deficits, summarize the state of knowledge regarding their incidence and the clinical profiles of RHD individuals, and discuss some of the clinical options available to those individuals. There is no doubt that RHD individuals are now better recognized and integrated into the clinical practice of SLPs, to the benefit of those patients and their families. We still, however, need to reach a consensus on how to refer to RHD patients with pragmatic disorders. A simple and logical proposal would be to consider these deficits as new forms of aphasic semiology. Indeed, since the end of the nineteenth century, the definition of 'aphasia' has not changed. The term still refers to language deficits following an acquired brain lesion, irrespective of its aetiology or anatomical localization. However, since that time, the concept of language has evolved dramatically and now includes the pragmatic component (Stemmer, 1999). It would, therefore, serve the field and patients well if they were simply considered as having aphasia, given that aphasia in general is the expression of a deficit affecting language, which itself is the outcome of numerous cognitive mechanisms that may or may not be exclusive to linguistic abilities proper. In brief, the recognition that pragmatic disorders can impair the communication abilities of individuals with right-hemisphere lesions represents an important step forward in aphasiology, and certainly in the recognition of a population that has been underserved for a long time.

12.6 Summary

In this chapter, our goal was to give readers an overview of the pragmatic deficits most frequently observed following RHD. Based on the information and data presented, it is possible to see that the major advances in the theoretical frameworks of neuro- and psycholinguistics have been fundamental to the ability to identify and better understand these deficits and possible clinical profiles, which, as we have mentioned, seem to have transcultural scope. In the same way, although still at a preliminary stage, we have seen that a series of intervention strategies already exists to allow us to better support this long-neglected population. Finally, it must be stressed that there is still much to be done in this field. We consider it fundamental to address how we conceive of the relationship between right-hemisphere language disorders, pragmatics and aphasia, since this theoretical conception undoubtedly has an impact on practice, both in the clinic and in research on language/communication disorders. By extending and disseminating our knowledge, we can make sure that individuals with right-hemisphere lesions and possible language deficits will be better recognized, described and supported.

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Chapter 13
Psychiatric Disorders

Irene P. Walsh and Caroline Jagoe

13.1 Introduction

Psychiatry is a branch of medicine which focuses on the diagnosis, assessment, treatment and prevention of mental, emotional and behavioral disorders. Psychiatric disorders are those diagnostic categories that can be identified via a defined group of symptoms within clinical presentations and with reference to classification systems such as the Diagnostic and Statistical Manual of Mental Disorders-5 (DSM-5; American Psychiatric Association, 2013) and the International Classification of Diseases-11 (ICD-11; World Health Organization, 2018a). Psychiatric disorders include clearly defined conditions or 'syndromes' such as schizophrenia, mood disorders (e.g. bipolar disorders, depression), and often less clearly defined diagnoses such as anxiety.

What is common to many psychiatric disorders are disturbances of thinking, emotion and consequently sometimes behavior, which often impact a person's ability to socially and occupationally engage with their communities in optimal ways. Language and communication can also be affected in many different ways in people who experience psychiatric disorders, with the prevalence of such difficulties reported in the literature (Emerson & Enderby, 1996; Walsh et al., 2007). In Walsh et al.'s (2007) study the prevalence of discourse and communication difficulties among a cohort of people with a variety of psychiatric presentations was over 60%, indicating the pervasiveness of pragmatic language difficulty across this population. Pragmatic language difficulties have been described in specific psychiatric populations, for example, in schizophrenia (e.g. Langdon et al., 2002; Jagoe, 2015; Bambini et al., 2016) and in bipolar disorder (e.g. McClure et al., 2005). Pragmatic language difficulties associated with anxiety in autism spectrum disorder have also been described (Walsh et al., 2018).

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Our work in this area to date has also clearly identified that the person behind the diagnostic label is all-important. The psychiatrist and the diagnostic team must see beyond textbook accounts, exercising their ‘capacity for intuitive understanding of each patient as an individual’ (Cowen et al., 2012, p. 1). Respecting each patient as an individual is crucial to any understanding of communication skills or needs and, in turn, to a consideration of pragmatic ‘breakdown’ as a product of the interaction, as opposed to assuming breakdown as residing in one or other of the interlocutors.

Pragmatic language ‘breakdown’ is often assumed to be an inevitable part of the psychiatric diagnosis, and pragmatic language interventions have typically focused on the remediation of the individual’s communication difficulties and addressed the ‘impairment’. We will suggest that there is an under-recognised facet of complexity with regards to pragmatic language disorders in people with mental health difficulties. The complexity lies not only in the documented intrapersonal elements of cognition and pragmatic abilities but critically in the *interpersonal nature of pragmatic language* in the broader sense. Moving beyond the focus of the literature on the individual impairment, this chapter will attempt to situate the complexity as a dyadic one that resides in the interaction itself.

The chapter begins by examining some of the most commonly occurring conditions that are labelled as ‘psychiatric disorders’, including their prevalence, onset and symptoms. It then addresses pragmatic language disturbances within the context of people with a psychiatric disorder (or ‘mental health disorder’, a term used interchangeably here). Keeping the person or patient to the fore, we will then consider the ‘insider’s perspective’ and draw on first- or other-person accounts of pragmatic language communication struggles and challenges in everyday talk interactions. Only by hearing the ‘voice’ of the person in this regard can we truly appreciate the effects of pragmatic language breakdown and take steps to design and implement appropriate interventions for people with communication disorders (Kovarsky & Curran, 2007). We then discuss how the communication dynamic affected can be best understood by appreciating the dyadic complexity and sociability of the ‘interactional space’. We adopt two evaluative lenses through which to explore the nature of pragmatic language difficulties within the context of communication with people with psychiatric disorders. The first encompasses *cognitively* orientated theories of pragmatics and the second includes *sociolinguistic* theories. The latter proposes the idea of a process of ‘conversational sociability’ (Walsh-Brennan, 2001) as a means to reconstrue the interactional-talk space in this context. The chapter concludes with suggestions for how the interactional-talk space can be adapted and changed by the interlocutors, to render more successful pragmatic language interactions within the context of this complex and underserved population.

13.2 Psychiatric Disorders: Some Background

The following sections give a general overview of the most commonly occurring psychiatric disorders as experienced in adulthood: schizophrenia, mood disorders and anxiety.

13.2.1 People with Schizophrenia

The World Health Organization states that schizophrenia is a psychiatric disorder affecting 21 million people worldwide (World Health Organization, 2018b). Although the prevalence of schizophrenia in men and women is considered equal, the onset of the disorder occurs earlier in males, often in early adulthood (Sadock et al., 2015). Recent reports suggest that people with schizophrenia can account for a fifth of all admissions to psychiatric units and is second only to mood disorders such as depression (Daly & Craig, 2018).

Patel et al. (2014, p. 638) define schizophrenia as "a complex, chronic mental health disorder characterized by an array of symptoms, including delusions, hallucinations, disorganized speech or behaviour, and impaired cognitive ability". The illness can vary widely in its severity and clinical presentation across those affected. Arguments remain as to whether it can be called a 'clinical syndrome' to reflect the idea of comprising 'a number of disease entities', or if it is best considered a 'single disease entity' with variable clinical presentations (Carpenter, 2007, p. 199). Regardless of how it is defined in textbook accounts, this major psychiatric illness can have a significant impact on the wellbeing and daily functioning of the individual and that of their families (Birchwood & Jackson, 2001).

Schizophrenia can present in acute and chronic forms. The illness can have a trajectory of an acute phase leading to the more chronic stage of the illness, when acute symptoms abate, and chronic symptoms persist. The nature and timeline of this trajectory are highly variable. A person who may be deemed chronically ill can still experience episodes of acute symptomology (Frith, 1992), oftentimes necessitating hospital (re-)admission and reviews of medication. Across these acute and chronic phases of the illness, *positive and negative symptoms* (defined below) may be evident. As both categories of symptoms can co-occur in one individual at any one time, or throughout the course of the illness, descriptions of 'predominantly positive symptom' and 'predominantly negative symptom' presentations in schizophrenia are terms which have been found to be most useful in practice.

A person in the acute stage of schizophrenia can present with predominantly positive symptoms, including, for example, delusions, hallucinations and disorders of thinking (Cowen et al., 2012). Such symptoms have been described by Frith (1992, p. 10) as being "abnormal by their presence", reflecting "an exaggeration or distortion of normal brain function" (Moller, 2013, p. 347). *Delusions* are defined as disturbances in thought content described as "firmly held false beliefs that reasoning cannot correct and for which there is no support in reality" (Tasman & Mohr, 2011, p. 258). Delusions can vary in nature and form and include those that can be described as *persecutory* (e.g. a person believing that someone is persecuting or tormenting them), *grandiose* (e.g. a person believing that they are very wealthy, aristocratic or highly talented) or *religious* (e.g. a person believing they are God or have special religious powers). Delusions can significantly interfere with communication as content may be deemed inappropriate or irrelevant to the ongoing communication (Walsh, 2008a). Also characteristic of positive symptoms, *hallucinations* are defined by Tasman and Mohr (2011, p. 258) as "sensory perceptions with a compelling sense of reality" which can take a number of forms, including auditory (e.g. hearing voices) and visual (e.g. seeing things that are not there). Auditory hallucinations are most common and can interfere with communication as the person is distracted from a conversational interaction by having to attend or respond to his/her 'voices'.

Disorganised conversation is another characteristic positive symptom in which a person is overly talkative and persists on certain topics or themes while not taking account of a listener's confusion or disinterest. According to Hinzen and Rossello (2015, p. 1), the main features of positive symptoms can:

> …fall into place as failures in language-mediated forms of meaning, manifest either as a disorder of speech perception (Auditory Verbal Hallucinations), abnormal speech production running without feedback control (Formal Thought Disorder), or production of abnormal linguistic content (Delusions).

A person in the chronic stage of the illness is more likely to present with predominantly negative symptoms (Kirkpatrick et al., 2001). Frith (1992, p. 10) describes such symptoms as "abnormal by their absence", being "a diminution or loss of normal brain function" (Moller, 2013, p. 347). Negative symptoms are characterised by social withdrawal, lack of drive and emotional apathy (Cowen et al., 2012). Once again, communication is affected by these symptoms, not least by the effects of social withdrawal where *poverty of speech* (lack of spontaneous speech) and/or *poverty of content of speech* ('empty' speech, lacking in content) have long been considered characteristic (Andreasen & Grove, 1986). An "increased latency to respond" (Hogg & Hall, 1992, p. 176), where responses are markedly delayed, can also disrupt the flow of conversations, frequently leading to conversation breakdown. Therefore, a person with predominantly negative symptom schizophrenia can be very quiet (even persistently silent) and hard to engage in communication. Once engaged in conversation, a person with predominantly negative symptom schizophrenia may only give responses that are minimal and unelaborated, with the listener having to work hard to keep the conversation going. Hence, the social communication behaviour of a person with chronic schizophrenia may impress a hearer as "odd and stilted", with speech being "slow and its content ... vague and incoherent" (Gelder et al., 1996, p. 249).

In sum, Frith (1997, p. 13) describes communication in schizophrenia succinctly as follows:

> the abnormalities of schizophrenic language appear not to lie at the level of language competence but of language use. The problems arise when the patient has to use language to communicate with others.

13.2.2 People with Mood Disorders

13.2.2.1 People with Depression

Estimates from the World Health Organization (2018c) suggest that, globally, there are more than 300 million people of all ages living with depression. However, such a figure can only be considered a rough estimate given that (i) people with depression may go undiagnosed, and (ii) depression occurring co-morbidly with other conditions is often missed (Fenton et al., 2016). Symptoms such as depressed mood, loss of interest and pleasure, feelings of worthlessness, and diminished ability to think or concentrate (which demonstrate a change from typical functioning over a specific period of time), are included among the diagnostic criteria for major depressive disorder in DSM-5 (American Psychiatric Association, 2013). Impairment in social and occupational functioning may also be a feature. A less chronic or enduring form of depression—often termed, reactive depression—is that which may be "triggered by a recent event that, because of its acute nature, has a good prognosis" (Kellerman, 2009, p. 53).

France (2001a) explains that the language expression of a person with depression can be limited, conveying minimal information. Sadock et al. (2015) state that people with this diagnosis may have poverty of content of speech (not unlike that found in the person with negative symptom schizophrenia). Additionally, Sadock et al. (2015, p. 366) explain that a person who is clinically depressed may "respond to questions with single words and exhibit delayed responses to questions. The examiner may literally have to wait 2 or 3 min for a response to a question". Such delayed responses may have implications for conversational interactions where the 'to and fro' nature of turn-taking in conversation is key to successful pragmatic language interactions. Furthermore, a person with a clinical diagnosis of depression may experience withdrawal and/or a tendency to avoid social contact (e.g. Schelde, 1998), in conjunction with changes in social skills (Joiner & Timmons, 2008; Sergin, 2001), all impacting pragmatic language interactions to varying extents.

13.2.2.2 People with Bipolar and Related Disorders

People with bipolar and related disorders experience mood disorders which can include irritability, deep depression and mania (intense euphoria). This group of disorders constitutes one of the leading causes of disability worldwide (Krahn, 2011). Findings from the Global Burden of Disease Study 2013 (Ferrari et al., 2016, p. 440) found that there has been a steady increase in prevalence "from 32.7 million

Table 13.1 Features of the bipolar spectrum

Subtype	Description
Bipolar I disorder	Characterised by one or more manic episodes, alternating with major depressive episodes; person may experience feelings of an inflated self-esteem, reduced need for sleep, and may be over talkative and highly distractible; symptoms may be severe enough to impact work or social life, and symptoms may last for a few days to a few months; person may require hospitalisation
Bipolar II disorder	Characterised by a major depressive episode (current or past) and at least one hypomanic episode (i.e. a milder occurrence of mania) over a period of at least 4 days; symptoms may not be severe enough to cause social or occupational dysfunction or to require the person to be hospitalised
Cyclothymic disorder	Characterised by less severe symptoms than the bipolar disorders where the person experiences repeated episodes of hypomania and non-psychotic depression for a period of at least 2 years; the 'opposing manifestations of depression and hypomania' (p. 118) are seen in the following ranges within symptoms such as uninhibited social interaction or social withdrawal, sleeping too little or too much, or increased or diminished productivity in occupational activities

Note: Adapted from Tasman and Mohr (2011, p. 118)

cases of bipolar disorder globally in 1990 to 48.8 million in 2013; equivalent to a 49.1% increase in prevalent cases, all accounted for by population increase and ageing".

As for many other psychiatric diagnoses, there is a wide range of clinical presentations among those affected by bipolar disorder. Tasman and Mohr (2011, p. 117) describe a 'bipolar spectrum' which underpins the idea of such a range and severity. DSM-5 includes criteria for the following: Bipolar I disorder (formerly known as, and synonymous with, the term 'Bipolar Disorder'); Bipolar II disorder; and Cyclothymic disorder (see Table 13.1).

Both mania and hypomania have been further described by Sadock et al. (2015, p. 348) as "associated with inflated self-esteem, a decreased need for sleep, distractibility, great physical and mental activity, and overinvolvement in pleasurable behaviour". A manic phase may also be characterised by feelings of great creativity and grandiosity, which may include "ideas of self-importance" (Fenton et al., 2016, p. 116). Robillard and Hickie (2015) describe cyclothymia as characterised by episodes of hypomania which do not reach the threshold for diagnosis of bipolar disorder, along with periods of mild to moderate depression.

Regardless of the specific manifestation of the mood disorder within the bipolar spectrum, there can be varying impacts on pragmatic language and social communication. Abnormalities of speech and thought such as a fast speech rate, flight of ideas, distractibility and delusional beliefs may all be apparent in manic episodes. Speech patterns, in terms of changes in modulation such as pitch and volume, have been used to help identify and clinically assess mood states within the spectrum (Goodwin & Redfield Jamison, 2007; Sadock et al., 2009; Karam et al., 2014).

France (2001a, p. 69) comments that while experiencing mania, a person's speech may be:

> too rapid, too much (pressure of speech), unnecessarily loud and not taking into account the present social setting; it is, in addition, difficult to interpret and full of jokes, puns, plays on words and amusing irrelevancies. It may become theatrical and full of dramatic mannerisms and singing. Sounds rather than meaningful conceptual relationships may govern word choice.

It is not difficult to see, therefore, how an affected person's speech and language may contribute to a communication breakdown or miscommunication between speaker and hearer.

13.2.3 People with Anxiety Disorders

Kellerman (2009, p. 15) defines anxiety disorders in general terms as:

> any disorder in which a person cannot control or resist the experience of anxiety. Examples include obsessions and compulsions, intrusive thoughts, hysterical reactions, all sorts of phobias, posttraumatic stress, and panic reactions.

Anxiety is a normal human experience. However, when anxiety "begins to interfere in the person's life in a significant way, it is considered a disorder" (Barlow & Ellard, 2018, p. 179). Anxiety disorders are common in the general population, with one third of the population affected by an anxiety disorder during their lifetime (Bandelow & Michaelis, 2015). The prevalence of anxiety disorders is greater in women than in men (Sadock et al., 2015). Anxiety disorders may co-occur with other mental illnesses (Fenton et al., 2016), and indeed with communication disorders. Anxiety disorders have in common excessive fear and anxiety which result in behavioural disturbances. DSM-5 recognises several major anxiety disorders, with the main ones being Generalised Anxiety Disorder, Panic Disorder, Specific Phobia, and Social Anxiety Disorder (American Psychiatric Association, 2013). Previous versions of the DSM included Post-Traumatic Stress Disorder (PTSD) and Obsessive-Compulsive Disorder as anxiety disorders. However, DSM-5 has reclassified these in the more specific categories of trauma and stress-related disorders and obsessive-compulsive and related disorders, respectively. Table 13.2 outlines some of the subtypes of anxiety disorders, specifically those which may impact on communication.

The impact of anxiety disorders on communication has had little focused attention. The relationship between anxiety and communication is complex, as France (2001b p. 37) explains:

> There are those speech disorders which accompany anxiety disorders and those which result from them, and there are communication problems that result from social inadequacy and perhaps exacerbate an anxiety disorder. The dividing line between these three categories is difficult to define.

Some specific findings have been reported, for example, lower anxiety levels in people with social phobia are accompanied by decreased proportions of silent

Table 13.2 Major anxiety disorders

Anxiety disorder (DSM-5 criteria)	Description
Generalised anxiety disorder	Lifetime prevalence of 3.7% (Ruscio et al., 2017), with higher prevalence and impact in high-income countries; diagnosed when there is at least 6 months of excessive and ongoing worry and anxiety alongside other symptoms such as muscle tension, fatigue, agitation or restlessness, irritability, difficulties with concentration, and sleep disturbances
Panic disorder	Lifetime prevalence of 1–4%; characterised by recurrent panic attacks, that is, episodes of intense and sudden fear or discomfort involving somatic symptoms (such as palpitations, trembling) and fear (e.g. fear of dying) (de Jonge et al., 2018)
Specific phobia	Very common anxiety disorder with a lifetime prevalence of up to 12.5% (Kessler et al., 2005); characterised by an excessive fear towards a particular object or event; DSM-5 (American Psychiatric Association, 2013) identifies five types (animal type; environmental type; blood-injection-injury; situational; other)
Social anxiety disorder	Lifetime prevalence of 4%, with highest rates in high-income countries, the Americas and Western Pacific Regions (Stein et al., 2017); characterised by excessive fear of social or performance situations, fearing scrutiny or embarrassment

pauses and a decrease in the high-frequency elements in the speech signal (Laukka et al., 2008). These acoustic parameters are interpreted by the listener as signs of nervousness (*ibid*), a factor which would be likely to impact on the interaction. Communication and social difficulties may constitute a significant risk factor for developing social anxiety (Brownlie et al., 2016; Pickard et al., 2017) and some people with aphasia may experience what has been described as 'linguistic anxiety' (Cahana-Amitay, 2011). Perhaps the area which has been most thoroughly addressed is that of anxiety in adults with chronic stuttering. A systematic review of this literature concluded that adults with chronic stuttering display higher levels of both trait and social anxiety (Craig & Tran, 2014). Pragmatic disturbances may, therefore, result from primary anxiety disorders or disorders of communication may increase the risk of anxiety disorders (e.g. see Walsh et al., 2018).

13.3 Pragmatic Language Disturbance in People with a Psychiatric Diagnosis

The study of language and communication in adult psychiatric disorders has primarily had an 'impairment focus', with the location of the disorder or breakdown considered to reside in the individual with the diagnosis. Impairments across 'levels' of linguistic ability have been documented. These include changes in speech rate and prosodic features in people with depression, for example (e.g. Alpert et al., 2001; Cannizzaro et al., 2004; Garcia-Toro et al., 2000; Moore et al., 2004; Uekermann et al., 2008); semantic, pragmatic and discourse abilities in people with schizophrenia (e.g. Docherty et al., 2003; Jagoe, 2013; Marvel et al., 2004; Woods et al., 2007; Meilijson et al., 2004; Walsh-Brennan, 2001); and the processing of emotional prosody in people with bipolar mood disorder (e.g. Hoertnagl et al., 2015; Paris et al., 2018). Despite the broad spectrum of linguistic impact, there is general agreement that communication difficulties in this heterogenous population manifest most prominently in the domain of pragmatics.

Pragmatic disturbances in adults with psychiatric disorders have been documented across people with diagnoses of schizophrenia, including difficulties with pronoun use, cohesion and coherence (e.g. Elvevåg et al., 2007; Allé et al., 2015); managing irony and deceit (e.g. Parola et al., 2018); and comprehending non-literal forms of language (e.g. Kiang et al., 2007; Schettino et al., 2010; Mossaheb et al., 2014). While most work in the field has focused on schizophrenia, some evidence exists for pragmatic language difficulties in people with bipolar mood disorder (e.g. McClure et al., 2005) and pronoun use in depression (e.g. Rude et al., 2004; Pennebaker & Chung, 2013). Aside from a few key studies, the incidence and prevalence of pragmatic language disorders in these populations has had comparatively less focus when compared to other clinical presentations (Bryan & Roach, 2001; Emerson & Enderby, 1996; Walsh et al., 2007).

Pragmatic language disorders in people with a psychiatric diagnosis (and particularly schizophrenia) have largely been explained as manifestations of impairments in theory of mind (ToM) and/or executive function. There is a complex interplay described in the literature between ToM and pragmatics. Poor performance on tasks involving the interpretation of non-literal language or Gricean maxims have been documented (e.g. Tényi et al., 2002; Haas et al., 2015) and associated with impairments in ToM (e.g. Tényi et al., 2002) or inhibitory control (Deamer et al., 2019). In contrast to performance on assessment ‘tasks’, analysis of natural conversation has revealed evidence of intact pragmatics skills (e.g. Walsh-Brennan, 2001), as well as indirect evidence of ToM (e.g. McCabe et al., 2004; Jagoe, 2013, 2015). Impairment in executive function, with the ‘component’ skills of inhibition, sustained and selective attention, initiation and working memory, has also been provided as an explanation for pragmatic language disturbances. Executive function deficits may co-occur with pragmatic disruption. However, most research suggests that ToM is better correlated with pragmatic tasks (e.g. Langdon et al., 2002; Brüne & Bodenstein, 2005; Champagne-Lavau & Stip, 2010). Champagne-Lavau and Stip (2010) conclude that ‘pragmatic deficits cannot be completely explained by executive dysfunction’ (p. 293).

Regardless of whether the underlying cognitive substrate(s) of pragmatic disruption relates to executive function deficits, ToM impairments, or both, it is likely that difficulties in communication will impact on social relationships. Indeed, there is evidence that these pragmatic difficulties have an impact on social relationships (e.g. Deamer et al., 2019) and quality of life (Bambini et al., 2016). The insider perspective is critical in order to understand the experience of pragmatic disruption.

13.4 A Take on Pragmatics: The Insider's Perspective

Increasingly, in published literature in adult psychiatry (and related areas), we see space given to 'first-person accounts' of experiences of mental health disorders. For example, journals such as *Schizophrenia Bulletin* regularly publish first-person accounts (see, for example, Wisdom et al., 2008). Researchers have used such accounts to map changes to the 'consumer' experience of living with schizophrenia, for example, and service development or reform over time (Gumber & Stein, 2013). Other sources are (edited) books devoted to the topic of first-person experiences of mental illness (e.g. LeCroy & Holschuh, 2012; Cohen, 2008), along with biographies (e.g. McCloskey, 2011; Nasar, 2002) and autobiographies (e.g. Saks, 2007). These accounts often recount both the challenges of, and the steps to, recovery the individual has taken. The term 'recovery' has a very particular and powerful meaning within the context of mental health. Anthony (1993, p. 527) defines it as a means to develop "new meaning and purpose in one's life as one grows beyond the catastrophic effects of [psychiatric] illness".

More effective pragmatic language communication is often part of the 'recovery' equation and can be referenced in experiential accounts of psychiatric disturbance. The accounts often include some description of the struggle and frustration with communication, especially the challenge of using language in social interactions. In other, more formal textbook accounts, pragmatic 'errors' are often described without paying due regard to the communication context that is at the heart of pragmatic language usage. These first-person accounts speak of challenges in the pragmatic skills of 'real-time' conversation. The complex and dynamic nature of conversational interaction is often described within the challenges experienced in, for example, topic selection, appropriacy, turn-taking, and meaning-making. The ability to engage in 'small talk' is relevant here as an example which requires some of these 'real-time' pragmatic skills and has been identified as a difficult area for some people with psychiatric disorders (Gibbons & Butler, 1987; Barham & Hayward, 1995; Walsh et al., 2018).

The ability to engage in 'small talk' (i.e. 'phatic communion', as first described almost one hundred years ago by Malinowski in Malinowski, 1923), is an essential part of everyday pragmatic language functioning. The term 'phatic communion' describes participation in a type of talk in which the primary aim is to convey and share sociability; where interlocutors create "ties of union ... by a mere exchange of words" (Malinowski, 1923, p. 478). In phatic communion, utterances have the "direct aim of binding hearer to speaker by a tie of some social sentiment or other" (Malinowski, 1923). Some early accounts emphasise the so-called aimless quality of small talk by describing it as "empty" (Turner, 1973), whereas other, later accounts focus on its social value in establishing interpersonal relationships (Cheepen, 1988; Coupland et al., 1992; Coupland et al., 1994). More recently, linguists such as Deborah Tannen (Tannen, 1986) Deborah Cameron (Cameron, 2000) and Justine Coupland (Coupland, 2000) have written on the topic as an area for serious linguistic research. The social value of such talk is acknowledged by people with psychiatric disorders, as some of the first-person accounts below illustrate.

For some people with schizophrenia, the social isolation resulting from a lack of ability to engage in small talk is particularly striking. The following comments were made by people with schizophrenia who had moved to a community-based hostel from hospital. Although their opportunities for social interaction had increased significantly in their new community, they often reported feeling lonely, despite having the desire to engage in conversation (from Gibbons & Butler, 1987, p.351):

> I feel isolated, I've no idea why. It's not because people don't want to be friendly.
> Lonely, yes, but there's nothing to say to anyone.
> I should pay someone to talk to me.

Having 'nothing to say to anyone' and, moreover, the suggestion 'to pay someone' to engage with another in talk, underline the willingness but inability to engage in such pragmatic language interactions. For others with a diagnosis of schizophrenia, topic selection within small talk is of great import. The account below describes the frustration this person with schizophrenia experiences as the conversation tends to focus on medication and symptoms of the illness (Barham & Hayward, 1995, p. 57) and not the more social or 'safe topics' usually referenced in small talk interactions:

> It depresses you at times like that, you just don't want to talk about things like that [medication, symptoms] you want to talk about normal things everybody else talks about sex, drugs and rock and roll or something, or horse racing ... You want to break out of that mould of being part of a schizophrenic fellowship or whatever. It does get you down at times.

A person with anxiety disorder, as associated with autism spectrum disorder, explains that it is her ability to engage in turn-taking within small talk interactions that is the challenge: "*My (conversational) 'to and fro' doesn't swing like a pendulum'* (Walsh et al., 2018, p. 117). Moreover, this person gets at the nub of pragmatic language usage when she explains how the intent of an interlocutor's contribution to the conversation is often difficult to discern (Walsh et al., 2018, p. 117):

> Why wouldn't I be confused with language, people have their own meanings; People don't say what they mean and often they don't want to say what they mean.

A recurrent theme in many first-person accounts, then, is the desire to engage in social talk with others, an experience that often proves elusive to those with enduring psychiatric illness. What is also apparent from accounts such as these, is the fact that people with psychiatric presentations (e.g. schizophrenia) are often aware and have some insight into their own conversational difficulties (Walsh, 1997), an awareness that has implications for working with communication skills. Yet, as emphasised in the opening paragraphs of this chapter, pragmatic language breakdown (regardless of what clinical population is being studied) does not lie with one or other interlocutor in an interaction, but rather is a result of the dynamic of that interaction. Whether we 'hush' or 'hear' adults with psychiatric disturbances as they talk of their communication struggles is pertinent here, where the actions of the interlocutor can promote or inhibit successful conversations in clinical and non-clinical interactions and in troubles-telling contexts (Walsh, 2008a). This point is not lost on individuals with psychiatric disorders as they often identify difficulties as a product of the interaction itself, as the following extracts demonstrate:

> My own experience of paranoia can be succinctly described using language games. […] What I found was that my perception of how people conversed became reduced to the simple rules of a language game, and I was an unwilling participant in this game. (Anonymous, 2011)

> It is your responsibility as medical professionals to communicate well with us. We have schizophrenia. We are mentally ill and we can't always manage our interactions with other people. You must teach us how to communicate well with you. (Schneider et al., 2004, p. 574)

The accounts of experiences of people with psychiatric diagnoses, therefore, provide a useful lens through which to explore the intrinsically dyadic nature of the pragmatic language disturbances they experience.

13.5 Dyadic Complexity and Sociability: Where Pragmatics Meets Psychiatry

Complexity lies in the interactional space between speaker and hearer regardless of who has the diagnosis. Ferguson (1996, p.56) echoes this point when she talks of the 'interactive relationship' as messages are negotiated:

> It is possible to extend our notion of communicative competence beyond consideration of how competence is vested in the individual, by recognizing competence as arising from the interactive relationship of communication partners as they negotiate messages.

Furthermore, Thomas (1995, p. 22) discusses the notion of meaning-making as a dynamic process, one that lies at the core of pragmatic language communication:

> Pragmatics is meaning in interaction. This reflects that meaning is not something which is inherent in the words alone, nor is it produced by the speaker alone, nor by the hearer alone. Making meaning is a dynamic process involving the negotiation of meaning between speaker and hearer, the context of the utterance (physical, social and linguistic) and the meaning potential of an utterance.

Theories of pragmatics and social interaction have enabled discourse to be analysed through different evaluative lenses. Such theories have helped researchers to at least attempt to begin to unravel the complexities inherent in pragmatic language usage in clinical populations, with particular reference to the dynamics of the interaction. Two broad stances characterise the theoretical approaches taken with regards to dyadic interaction involving people with psychiatric diagnoses. The first encompasses cognitively orientated theories of pragmatics and the second includes sociolinguistic theories.

13.5.1 Cognitively Orientated Theories of Pragmatics

Perhaps the most extensively applied pragmatic theory is that of Bara's Cognitive Pragmatics Theory (see Bara (2010) for a full articulation of the theory). Cognitive Pragmatics Theory takes a neo-Gricean approach, based on the notion that communication is a cooperative activity (Bara, 2010). The idea of communication as a 'behaviour game' emphasises the cooperative, intentional and dyadic nature of human communication. Both linguistic and extralinguistic features are explicitly considered within Cognitive Pragmatics Theory. Cognitive Pragmatics Theory has a growing body of applied clinical literature that is based on the theory (e.g. Gabbatore et al., 2015; Bosco et al., 2018). A specific assessment tool, The Assessment Battery for Communication (ABaCO; Sacco et al., 2008), has been developed on the principles of Cognitive Pragmatics Theory. A clinical protocol, Cognitive Pragmatic Treatment (CPT), has also been developed (Sacco et al., 2016, p. 2) and applied in relation to people with schizophrenia. Cognitive Pragmatic Theory, therefore, offers clear clinical tools, with a theoretical foundation and emerging evidence base for application.

A second cognitively orientated theory which has been applied to pragmatic analysis within psychiatry is that of Relevance Theory. Relevance Theory (Sperber & Wilson, 1986/1995) is also a neo-Gricean theory, which considers communication to be reliant on a single overarching principle, that of relevance. In this approach, communication exploits the human tendency to attend to and process stimuli which are most relevant to them. This cognitive bias guides the hearer towards the intended interpretation of an utterance by expectations of relevance alone. Relevance Theory has had more limited clinical application. In terms of mental health disorders, it has been applied to communication interactions with people with schizophrenia (Jagoe, 2013, 2015), including considerations for assessment and intervention (Jagoe, 2020).

13.5.2 Sociolinguistic Theories of Pragmatics

A sociolinguistic evaluative lens, which allows a multi-theoretic approach to analysis of talk in interaction, has helped to uncover a process of 'conversational sociability' (Walsh-Brennan, 2001; Walsh, 2002/2003). Invoking the principles and analytic methods of Communication Accommodation Theory (Coupland et al., 1988, 1991), Frame Theory (Goffman, 1974; Tannen, 1993) and Politeness Theory (Brown & Levinson, 1987), it has been shown that rather than being pragmatically *disabled* in talk, people with psychiatric disturbance (i.e. in this instance, people with chronic schizophrenia) can be *enabled* to engage in the dynamic of successful conversational interactions. Walsh-Brennan (2001) argues that given the right conversational-linguistic 'environment', including an open and receptive interlocutor, people with communication disorders associated with psychiatric disorders can be supported to engage in successful pragmatic language interactions. Akin to the notion of small talk, then, is a process termed *conversational sociability*. This can be defined as the type of talk illustrated by:

> An integration of features of form and function, evident within and emergent from the discourse, where participants display a readiness or willingness to chat, converse, engage in social talk, marking friendliness and affability not a stiffness or formality in conversational interactions (Walsh-Brennan, 2001, p. 293)

The word 'process' is used here intentionally as opposed to 'genre', as the latter is perhaps too suggestive of a discrete entity which can be invoked at different stages of a conversation. A process suggests a potentially diffuse functional operation within discourse (see also Coupland et al., 1992; Coupland, 2000). The above definition places conversational sociability conceptually between Schneider's (1988) notion of small talk, as a form of socio-pragmatic competence, and Coupland's (2000, p. 5) interpretation of small talk being concerned with "the explanation of social functions which [it] may achieve for interlocutors". Although an approach to professional interaction based on conversational sociability may appear deceptively simple, adapting to this way of behaving in talk requires a multi-level revision of usual conversational practice.

13.6 Changing Conversational Practice to Address Pragmatic Complexity

People with psychiatric diagnoses represent a complex population in terms of pragmatic disruption. While the individual may display a complex presentation of pragmatic abilities and disabilities, complexity also lies within the interactional encounter. The focus of research, assessment and intervention has typically been on direct language therapy or 'training' for the individual with the diagnosis. A recent systematic review concluded "that pragmatics and discourse skills are skills that can be trained in patients with schizophrenia and that this training can be retained over time" (Joyal et al., 2016, p. 92). In an intervention study with people with schizophrenia, Bosco et al. (2016) used communicative pragmatic treatment, based on Cognitive Pragmatics Theory, and reported positive outcomes and retention of 'skills' 3 months post intervention. Despite these reports of success, the population has remained relatively underserved. The type of complexity that has been less often explored is pragmatic complexity, and specifically how the dyadic nature of pragmatics can be exploited to create change or influence interactions. In this approach, the interlocutor and communication environment become paramount. How might this complexity be addressed, and the population better served? The concepts arising from the theories addressed in Sect. 13.5 suggest possible features for clinical application.

Taking a cognitive pragmatic stance to the challenge of addressing pragmatic disruption might be informed by existing cognitively focused interventions. One such protocol is that of Metacognitive Training (MCT; Moritz & Woodward, 2007) which is specifically designed for people experiencing (or at risk of) psychosis. However, the structure and design would appear to lend itself to application for pragmatics intervention. MCT uses a combination of discussion, reflection and exercises aimed at targeting 'cognitive biases' or the systematic ways in which errors of judgement may occur:

> Exercises targeting each bias individually demonstrate the fallibility of human cognition in general […]. Personal examples of these biases expressed by MCT participants, and discussion of ways to counter them, serve to provide corrective experiences in a fun and supportive atmosphere, yielding obvious advantages over mere lecturing (Moritz & Woodward, 2007, p. 621).

Suggestions for adapting this protocol (Jagoe, 2020) involve group sessions broadly structured on the framework used in MCT: (1) Metapragmatic awareness tasks and normalising aspects of communication challenges; (2) Discussion of extremes of these difficulties that may be associated with psychiatric diagnoses; (3) Responses to challenges and implementation of new or alternative strategies; and (4) Implementation exercises within the session itself and as home practice.

Both Cognitive Pragmatics Theory and Relevance Theory emphasise the collaborative nature of communication. Assessment and intervention within Cognitive Pragmatics Theory have focused explicitly on the individual with the disorder. Although the application of Relevance Theory has largely focused on the individual with the diagnosis, some work has been done on considering the role of the communication partner within the interaction. This more dyadic focus has been undertaken, both in relation to schizophrenia and also within a non-psychiatric population, exploring interaction involving aided communication (Jagoe, 2015; Jagoe & Smith, 2017; Neuvonen et al. in submission). Despite their cognitive nature (and often assumed intra-personal focus), both Cognitive Pragmatics Theory and Relevance Theory explicitly consider engagement between speaker and hearer. The role of interlocutors (or communication partners) cannot be ignored and is an area of clear overlap with the approaches to conversational sociability described above.

Research on communication in psychiatric service provision, using conversation analysis, has demonstrated that it is possible to intervene to optimise patient-clinician communication, thereby improving outcomes (McCabe et al., 2016; Priebe & McCabe, 2008; Priebe et al., 2007). Addressing pragmatic abilities, then, should be considered from both sides of the dyad—the skills of the individual with the diagnosis (perhaps through an adapted MCT approach) *and* the skills of the communication partner.

While conversation partners may helpfully adjust their communication strategies to accommodate an individual assumed to be less communicatively 'competent' (Garcia et al., 2001), such an assumption also risks eroding successful engagement. Particularly in a disorder in which stigma is rife, an assumption of mental illness may lead to a subsequent assumption of 'incompetence' and an avoidance of engagement (Walsh, 2008b). Careful attention then needs to be given to how communication partners are supported to interact with a person with pragmatic disturbances due to a psychiatric diagnosis. Considering the truly dyadic nature of conversational sociability in interactions implies that professionals and carers are encouraged to:

1. be aware that conversational ability in people with psychiatric disorders may be masked or suppressed by the illness process, by their perception of the illness or the person, or by their inhibitive discourse style;
2. re-evaluate the power of sociable talk and to realise the important socio-relational (and hence transactional) function it serves;
3. be guided in ways to maximise effective conversational interactions; professionals particularly must be encouraged to exploit opportunities for sociable talk in the pursuance of therapeutic goals;
4. adopt this ‘way of behaving’ in talk in order to maximise the therapeutic gains which can be accrued.

Furthermore, the apparent meta-conversational (or meta-pragmatic) awareness exemplified by the first-person accounts of communication difficulties as part of a psychiatric diagnosis, can be a useful starting point for the exploration of reported conversational difficulties. ‘Talk about talk’ can be fruitfully incorporated into intervention programmes, building on this insight and enhancing awareness of the value of sociable talk and the relationship-building which can be achieved through its use. Listening to and not just hearing the voice of a service user is key, if optimal support is to be provided (Kovarsky & Curran, 2007). Those who work directly on communication skills can be encouraged to address conversational issues in meta-conversational or meta-pragmatic terms with their clients, while at the same time modelling and acknowledging conversational sociability in an essentially interaction-focused way.

13.7 Providing Opportunities for Talk, Sharing Information and Educating Others

Though seemingly obvious, providing opportunities for talk within mental health contexts is of significance. People with chronic mental illness may traditionally have experienced impoverished social environments (e.g. while hospitalised), where administration of medication and other healthcare activities may be main foci of interaction (France, 2001c). Moreover, in other institutional-like settings (e.g. high-support community hostels), the predictability of the daily routine can often inadvertently prevent meaningful, spontaneous conversational interactions from taking place. Despite the best efforts and intentions of busy healthcare staff, opportunities for talk may be severely limited without support. However, there have been improvements in these contexts of care as greater importance is given to the need for positive social interactions in the promotion of well-being and recovery, and the mental healthcare professional’s role in this regard (Slade, 2010).

Providing opportunities for talk is only one element of the wider remit of a therapeutic approach in working with people with psychiatric disorders. Professionals, such as speech and language therapists, have a responsibility to their colleagues to share their expertise in the area of communication and pragmatic language usage. Given that 'talk' is the medium of psychiatry, educating mental healthcare professionals about the value of engaging in conversational sociability, for example, might affirm, in some cases, what some professionals already do. But for those whose conversational style inhibits (or hinders) rather than facilitates (or helps) conversation to emerge, for those whose style of interaction enhances the sense of disability rather than exploiting repressed 'ability', significant gains can be had from carefully considering what conversation has to offer clinical interactions.

Families, too, are key stakeholders in conversational interactions within the context of communicating with people with psychiatric diagnoses. Families may have experiences of communication struggles as a psychiatric disorder gets in the way. They may also benefit from support to reconsider and exploit the value of sociable talk in the (re)building of relationships.

Information sharing, however, is not enough if professionals and carers are to adopt a different conversational 'way of behaving' with those they care for. Workshops and small group work, even direct work with conversational dyads, could be undertaken to highlight what is involved in a conversational sociability approach to interactions. As for direct work with clients, video demonstration and evaluation, along with role plays and in vivo interactions between clients and professionals can all be used to promote appropriate and effective interactions.

Hence, the very particular pragmatic language needs of people with psychiatric disorders can be addressed and interpreted by speech and language therapists. Speech and language therapists must share information with others about the complexity of pragmatic language interactions with people with psychiatric illness and the importance of adapting conversational styles. Interaction must be the focus of intervention. Clients, carers and professionals can all benefit from interventions that highlight the positive influence of attending to communication as a truly dyadic and dynamic process, which is not the responsibility of either the speaker or the hearer, but of the speaker and hearer in interaction.

13.8 Summary

This chapter examined the complexity of pragmatic ability and disability in people with psychiatric diagnoses. The literature predominantly points at a range of interrelated difficulties which are situated within the individual. We have suggested that an equally complex but under-recognised feature of pragmatic ability resides in the interactional space of talk. Pragmatic theories alongside socio-linguistic concepts help to understand the roles of both parties in the conversation. We suggest frameworks that inform ways of working alongside those with psychiatric diagnoses and the people within their communication networks.

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Chapter 14
Dementia of the Alzheimer Type

Angela Roberts, Marie Savundranayagam, and J. B. Orange

14.1 Introduction

The aim of this chapter is to provide an overview of the literature on pragmatic language abilities in individuals with dementia of the Alzheimer type. Dementia of the Alzheimer type is a common neurodegenerative disorder that compromises language and cognitive function, as well as other abilities. The chapter examines the prevalence, genetic and other risk factors, and the neuropathological features of dementia of Alzheimer type. Pragmatics is integral to the communication difficulties that are experienced by people with this form of dementia. The implications of cognitive, linguistic, and pragmatic impairments on conversation difficulties among individuals with dementia of the Alzheimer type and their families will also be addressed. The chapter concludes with a brief discussion of the impact of pragmatic language impairments on family caregivers and considerations for interventions in this area.

14.2 Dementia of Alzheimer Type: Some Background

According to the World Health Organization, the most common cause of dementia worldwide is Alzheimer disease, accounting for 60–70% of dementia cases in 2018 (World Health Organization, 2018). It is estimated that 44 million people have dementia of the Alzheimer type or a related dementia (Alzheimer's Association,

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2019). Dementia of the Alzheimer type is typically associated with older age. Less than 10% of all cases are classified as early onset (i.e. onset under 65 years of age) (National Institute on Aging, 2019).

Dementia of the Alzheimer type refers to the clinical manifestation (e.g. cognition, language, visual spatial skills, and behaviour symptoms) of an insidious form of dementia that results from the cardinal neuropathology of Alzheimer disease (i.e. neurofibrillary tangles, senile plaques, neuronal loss in hippocampal and cortical regions) (Rasmussen et al., 2018). The presence of the *APOE e4* allele is a significant genetic risk factor for Alzheimer disease (Jalbert et al., 2008; Rasmussen et al., 2018). Modifiable risk factors for Alzheimer disease and related dementias account for 35% of all causes. These modifiable risk factors include less formal schooling, hearing loss, midlife hypertension, midlife obesity, smoking, later-life depression, physical inactivity, low levels of social contact, and diabetes (Livingston et al., 2017).

Although the personal consequences of this form of dementia are serious and wide-ranging, the disease impact is not limited to the individual living with dementia of the Alzheimer type. As of 2019, more than 16 million people living in the United States were providing unpaid care for a family member with dementia of the Alzheimer type (Alzheimer's Association, 2019). Astonishingly, the estimated worldwide economic burden of dementia is estimated at more than one trillion US dollars, representing approximately 1% of global gross domestic product (Alzheimer Disease International, 2018). Growth in dementia cases and financial burdens are greatest in countries with the lowest gross domestic product per capita—further increasing the burden of living with Alzheimer disease (Alzheimer's Disease International, 2018). The global impact of dementia of the Alzheimer type, both present and projected, highlights the importance of (1) better understanding the nature of pragmatic language impairments and their role in the early detection of dementia of the Alzheimer type, and (2) increasing research that addresses the development of interventions that target pragmatic language impairments and their devastating impact on persons living with dementia of the Alzheimer type and their families.

14.3 Clinical and Neural Characteristics of Dementia of Alzheimer Type

Episodic memory changes early in the disease represent the prototypical clinical symptom profile in dementia of the Alzheimer type. Research suggests, however, the presence of affective, psychosis, and neuropsychiatric symptom clusters that represent potentially distinct syndromes (Cook et al., 2003; Jalbert et al., 2008). Within the neuropsychiatric symptom cluster, there is also evidence of an Alzheimer disease language-symptom subtype (Rogalski et al., 2016). Dementia of the Alzheimer type symptom profiles, severity, and disease trajectory are associated with a number of factors, including age of onset, sex, and the presence of one or more *APOE* e*4* alleles (Rasmussen et al., 2018).

Mean survival is estimated at 11.8 years from symptom onset (Jalbert et al., 2008). But even before the onset of manifest symptoms, dementia of the Alzheimer type can have a long pre-clinical stage. Studies suggest the possibility of subtle pragmatic language changes even in the pre-clinical stage of dementia of the Alzheimer type (e.g. Rapp & Wild, 2011; Szatloczki et al., 2015). While robust predictors of cognitive and language symptom progression in dementia of the Alzheimer type remain under investigation, research shows that declines in composite language scores occur at a rate of 0.71 standard units per year (Hebert et al., 2000). The observed heterogeneity of clinical profiles, and the possibility of a symptomatic pre-clinical stage, suggest that pragmatic language impairments are an important consideration at all clinical stages of dementia of the Alzheimer type.

Studies show that Alzheimer disease-related neuropathological changes to the bilateral entorhinal cortex, bilateral hippocampus head, and left medial perirhinal cortex underlie episodic and semantic memory deficits in persons with dementia of the Alzheimer type (Hirni et al., 2016). In addition to these salient neural changes, recent neuroimaging studies suggest that the superior longitudinal fasciculus may also be affected in those with mild cognitive impairment and early dementia of the Alzheimer type (Liu et al., 2011). These findings suggest the potential for disrupted connectivity among language critical structures in both left and right cerebral hemispheres (Bernal & Altman, 2010; Axer et al., 2013), substantiating clinical observations of language impairments in pre-clinical and early-stage disease. Recent systematic reviews by Szatloczki et al. (2015) and a special issue in *Aphasiology* (Rochon et al., 2018) offer current overviews of the speech and language production characteristics (i.e. form and content) among persons with dementia of the Alzheimer type that contribute to, and are complicated by, pragmatic language impairments.

14.4 Social Cognition Impairments in Dementia of the Alzheimer Type

Social cognition refers to cognitive processes that enable people to interpret the emotional and cognitive states of others and to use that information to interact with them (Elamin et al., 2012). Theory of mind (ToM) is conceptualized as an essential ability underlying social cognition. ToM has two levels of complexity: first-order and second-order. First-order ToM refers to the ability to infer what another person feels or thinks about an event in the world. Second-order ToM is the ability to infer a person's feelings or thoughts about another person's mental states. Social cognition and ToM involve individuals' abilities to use and to understand the multidimensionality of language situated in social contexts and, as such, are key foundational concepts of pragmatics.

ToM also includes affective and cognitive components (Brothers & Ring, 1992). Affective ToM refers to the ability to infer or to interpret the *emotional states* of others. It is often measured by Reading the Mind in the Eyes Test, where a participant must match emotions and mental states with photographs displaying the eye regions of faces (Baron-Cohen et al., 2001). On the whole, researchers have demonstrated no impairment of affective ToM in early-stage dementia of the Alzheimer type (Freedman et al., 2013; Kéri, 2014), but have demonstrated impairment in middle-stage dementia of the Alzheimer type (Castelli et al., 2011; Laisney et al., 2013).

One concern raised in the literature is that using measures such as the Reading the Mind in the Eyes Test to evaluate affective ToM does not consider all of the contextual information that is used in real-world language contexts to infer the emotional states of other people. To address this limitation, Sava et al. (2019) investigated the influence of dementia of the Alzheimer type on first-order affective ToM using contextual information that does not include visual facial cues. Moreover, they also examined the performance of participants with dementia of the Alzheimer type on inferring a range of positive and negative emotions, such as joy and disgust. Contrary to previous findings, Sava et al. (2019) found that participants with mild dementia of the Alzheimer type show worse performance on affective ToM than healthy younger and older adults. While their performance did not differ for anger, fear, and joy, they exhibited poorer performance for disgust, sadness, and surprise. Collectively, the literature suggests that context and available information may affect ToM processes in early dementia of the Alzheimer type. Additionally, it suggests that there may be specific emotional states that are more difficult for persons with early dementia of the Alzheimer type to infer when facial cues are not provided.

Cognitive ToM is the ability to infer the *thoughts* of others. It is typically assessed using first- and second-order false belief tasks (for a review of assessment methods, please see Sandoz et al., 2014). Among participants with early dementia of the Alzheimer type, investigators demonstrated that first-order cognitive ToM is preserved but that there are deficits in second-order level reasoning (Castelli et al., 2011; Cuerva et al., 2001). By contrast, researchers found deficits in first-order cognitive ToM among participants with middle-stage dementia of the Alzheimer type, suggesting that first-order reasoning may decline with increased dementia severity (Laisney et al., 2013). However, it has been suggested that second-order cognitive ToM tasks are more cognitively complex than are first-order tasks, which may explain why deficits in second-order level reasoning are more commonly reported than first-order ToM impairments in early dementia of the Alzheimer type. Consequently, many researchers have investigated whether ToM deficits are independent of, or linked with, other cognitive processes, such as executive function and episodic memory (Laisney et al., 2013; Moreau et al., 2013, 2016). Laisney et al. (2013) found that while language comprehension abilities did not influence performance, there were significant relationships among second-order false belief deficits and performance on tests of inhibition, verbal fluency, and working memory (backward digit span). These findings highlight the complex relationship between ToM and executive function in dementia of the Alzheimer type.

The findings from Laisney et al. (2013) are consistent with the domain-general or the integrated perspective, which contends that ToM abilities are supported by other cognitive functions beyond social cognition including executive function, memory, and language abilities (Bora et al., 2015; Ramanan et al., 2017). Because a number of tasks used to assess ToM rely heavily on verbal attention and short-term working memory abilities, parsing unique social cognition and pragmatic impairments from more global executive function and language difficulties in dementia of the Alzheimer type can be challenging (Castelli et al., 2011). For example, impairments on measures of executive function (e.g. verbal fluency, digit span) significantly predict ToM performance in participants with dementia of the Alzheimer type (Ramanan et al., 2017), providing support for the domain-general perspective. Support for the domain-general perspective also comes from studies showing that participants with dementia of the Alzheimer type perform relatively well on first-order false belief tasks (Freedman et al., 2013; Zaitchik et al., 2004, 2006) that typically have lower cognitive demands compared to their performance on more challenging second-order false belief tasks (Cuerva et al., 2001). Thus, it remains unclear whether ToM deficits are a core characteristic of dementia of the Alzheimer type or whether performance on ToM tasks reflect underlying executive function changes now thought to emerge early in the disease alongside episodic memory impairments (Baudic et al., 2006).

To date, investigators of two major studies examined the relationship between ToM and pragmatic communication among participants with dementia of the Alzheimer type. Cuerva et al. (2001) used a second-order false belief task, where participants with mild dementia of the Alzheimer type were asked to infer a person's belief about another person's belief. In the same cohort, they also assessed pragmatic abilities in the form of indirect requests and understanding conversational implications. The authors found a significant relationship between second-order false belief task performance and pragmatic impairments in conversation. Moreau et al. (2016) also assessed ToM in interactions using a referential communication task, in which participants with dementia of the Alzheimer type described five tangrams with the goal of directing the experimenter to arrange them in a particular order. Dementia of the Alzheimer type participants used very few definite references (a marker of mutual understanding with the interlocuter). Instead, individuals with dementia of the Alzheimer type used more indefinite references, a discourse behaviour associated with introducing new information and used typically in cases where the listener is assumed to have a low degree of shared familiarity with the speaker's frame of reference (Fodor & Sag, 1982). The prominent use of indefinite references in the Moreau et al. study suggests that ToM deficits in speakers with dementia of the Alzheimer type create ambiguity in conversations by impairing the speaker's ability to attribute knowledge to the listener.

Findings from studies assessing pragmatic abilities and spontaneous conversation highlight the significance of ToM deficits on relationships between participants with dementia of the Alzheimer type and their communication partners. ToM abilities are essential for maintaining conversations and relationships especially when topics include sarcasm, figurative language, and humour based on word-play and inference, among other socially contextualized forms of language. Moreover, ToM deficits are linked with pragmatic impairments, especially those that require persons with dementia of the Alzheimer type to derive meaning from context, recognize the relevance of what is communicated by another person, and interpret emotional states (Cuerva et al., 2001; Shany-Ur et al., 2012). Further, difficulties with interpreting shared knowledge or beliefs can lead to social withdrawal or apathy, modifiable risk factors for dementia (Livingston et al., 2017). Failing to understanding another person's feelings and/or cognitive perspective can result in perceived hurtful communication or behavioural difficulties, which cause distress in caregiving relationships (Savundranayagam et al., 2005; Savundranayagam & Orange, 2011). Because of their potential importance for conversation and social relationships for persons with dementia of the Alzheimer type, more work is needed to develop ideal assessment tools and paradigms that will allow clinicians and researchers to disentangle the complex interactions among executive function, episodic memory, and ToM impairments in dementia of the Alzheimer type and their cascading effects on the quality of and effectiveness of everyday pragmatic language interactions.

14.5 Figurative and Non-literal Language Impairments in Dementia of the Alzheimer Type

Despite the fact that metaphors and other forms of figurative language commonly appear in clinical neuropsychology and diagnostic batteries used in dementia of the Alzheimer type (Rapp & Wild, 2011), the robust examination of figurative language processing has received surprisingly little attention, compared to other areas of cognition and pragmatic language research in this population. Whilst early studies that reported the presence of figurative language impairments in dementia of the Alzheimer type (Chapman et al., 1997; Kempler et al., 1988) led to the belief that metaphor and idiom comprehension impairments are hallmarks of this form of dementia, a number of more recent studies have called into question the ubiquity of this finding (Amanzio et al., 2008; Papagno, 2001). Recent studies in dementia of the Alzheimer type have assessed figurative language abilities with increased granularity, including concrete and novel metaphors, as well as idioms with different gradients of transparency, decomposability, and plausibility. Under the premise that different types of figurative expressions reflect different cognitive processes (Gibbs & Richard, 1989; Gibbs, 2002), and thus varying degrees of right versus left hemispheric activation (e.g. Gradient Salient Hypothesis; Giora, 2003), a number of researchers have explored figurative expressions as a means of parsing semantic from executive and attentional control impairments in mild cognitive impairment and dementia of the Alzheimer type.

Amanzio et al. (2008) compared figurative language comprehension in early- to middle-stage individuals with dementia of the Alzheimer type to healthy adults by asking participants to generate verbal explanations for opaque idioms, and for conventional and novel metaphors. Individuals with dementia of the Alzheimer type were significantly more accurate when processing conventional metaphors than novel metaphors. Interestingly, individuals with dementia of the Alzheimer type differed from healthy adults only on the novel metaphor task. These findings are consistent with Papagno (2001) and suggest that in early-stage dementia of the Alzheimer type individuals are able to access lexicalized meanings of figurative expressions but are impaired in constructing meaning. Counter to Amanzio et al.'s findings, other recent studies examining opaque idioms showed that individuals with dementia of the Alzheimer type are more impaired than healthy controls, even in the early clinical stage (Papagno et al., 2003; Rassiga et al., 2009).

Of late, researchers expanded figurative language research in dementia of the Alzheimer type by asking whether these tasks can detect the disease in very early clinical states, and also whether severity of figurative language impairments are associated with disease progression. To this end, Maki et al. (2013) examined metaphor comprehension (conventional and novel) and also sarcasm comprehension in early-stage dementia of the Alzheimer type and amnestic mild cognitive impairment using a multiple-choice response format test (the Metaphoric and Sarcastic Scenario Test). Both dementia of the Alzheimer type and amnestic mild cognitive impairment groups showed greater comprehension problems for metaphor and sarcasm compared to the healthy adult group. Additionally, disease stage appeared to play a role in performance with the dementia of the Alzheimer type group being more impaired than the amnestic mild cognitive impairment group on both metaphor and sarcasm items. Post-hoc error analyses also showed qualitative differences between the dementia of the Alzheimer type and amnestic mild cognitive impairment groups, revealing that the dementia of the Alzheimer type group was more likely to select the literal interpretation response choice.

Other researchers have reported selective declines with disease progression. Using a within-subjects, longitudinal design in early-stage dementia of the Alzheimer type, Papagno (2001) found that only 10% of their cohort exhibited impairments in figurative language (opaque idioms, common/nominal metaphors). However, in a subset of participants re-assessed 6–8 months later there were significant declines in metaphors but not idioms. Consistent with Maki et al. (2013), qualitative analyses revealed that comprehension errors on idiomatic expressions were more likely to be literal in nature. Interestingly, metaphor errors tended to reflect insufficient information/confusions, suggesting that different forms of figurative language may be sensitive to impairments in different cognitive domains.

Collectively, these studies suggest that figurative language impairments may emerge over time as dementia of the Alzheimer type progresses but may not always be present in early clinical states. The literal interpretation bias for figurative expressions has also been reported by a number of researchers (Kempler et al., 1988; Maki et al., 2013; Papagno et al., 2003; Rassiga et al., 2009). Several authors interpreted this finding as being consistent with an executive control hypothesis and concluded that the tendency to select the literal response choice reflects difficulties inhibiting the literal interpretation that is concurrently activated with the non-literal interpretation (Maki et al., 2013; Papagno, 2001; Rassiga et al., 2009). This hypothesis is further reinforced by studies showing positive and significant correlations between measures of executive function and performances on figurative language tasks and assessments (Amanzio et al., 2008; Papagno et al., 2003; Rassiga et al., 2009). An alternative hypothesis for figurative language impairments in dementia of the Alzheimer type is that constructing meaning requires enactment of events through generating mental images, which may be affected by episodic and semantic memory impairments in early clinical-stage dementia of the Alzheimer type (Amanzio et al., 2008; Mashal et al., 2005). Still others favor the explanation that processing figurative expressions taxes both semantic and executive function resources, depending on the task context (Rassiga et al., 2009).

On the whole, the literature suggests that the diagnostic sensitivity of figurative language impairments in dementia of the Alzheimer type may be affected by the form of figurative language (i.e. novelty, transparency, decomposability), the format for evaluating figurative language comprehension (i.e. verbal explanation, multiple choice, sentence-picture matching), the severity of disease, and the task context (i.e. whether information regarding a competing literal interpretation is available). There are several challenges in considering this literature. One challenge is the number of different evaluation formats reported in the literature, ranging from multiple choice response questions (Chapman et al., 1997; Maki et al., 2013), verbal explanation paradigms (Cardoso et al., 2014; Chapman et al., 1997; Papagno et al., 2003), priming paradigms (Rassiga et al., 2009), paragraph comprehension (Cardoso et al., 2014), and sentence to picture or written proverb matching (Cardoso et al., 2014; Papagno et al., 2003; Rassiga et al., 2009). For example, in a within-subjects comparison Papagno et al. (2003) found that individuals with dementia of the Alzheimer type were significantly more accurate when providing verbal explanations for idioms than in a sentence-to-picture task. Additionally, using standardized measures typical of those reported in neuropsychology practice, Cardoso et al. (2014) found that individuals with amnestic mild cognitive impairment were more impaired than healthy controls when proverb comprehension was assessed through a written version of a proverb-matching task and when idioms were assessed through a verbal explanation task, but the groups did not differ when non-literal language (form not specified) was evaluated using a text passage comprehension task.

A second concern is the high variability observed on figurative language tasks for both healthy, older adults and individuals with dementia of the Alzheimer type, making it difficult to generalize and to reproduce results from many studies that have small sample sizes (Rassiga et al., 2009). Moreover, to date the majority of research in this area has depended on the clinical diagnosis of dementia of the Alzheimer type in isolation without ruling out other sources of cognitive impairment (e.g. vascular infarcts or white matter disease) or confirming the likelihood of Alzheimer disease pathology through blood-based biomarkers. Given the elevated potential for clinical diagnostic ambiguity in dementia of the Alzheimer type (Beach et al., 2012), increased diligence to confirming the likelihood of underlying Alzheimer disease pathology will be essential for drawing more direct relationships between Alzheimer disease pathology and figurative language impairments.

14.6 Conversation Impairments in Dementia of the Alzheimer Type

A normal everyday activity (Liddicoat, 2007), conversation is defined and studied in a myriad of ways by eminent social, linguistic, psychological and ethnomethodological researchers (e.g. Goffman, Goodwin, McGregor, Myllyniemi, Schegloff, ten Have, among others). It is beyond the scope of this chapter to summarize the multiple definitions or to compare and contrast analyses of conversation with conversation analyses. Other authors provide comprehensive, detailed descriptions and explanations of these constructs (e.g. see Goodwin & Heritage, 1990; Sacks et al., 1978; Sidnell & Stivers, 2013). For the purposes of this chapter, conversation refers to:

> A back-and-forth series of verbal and nonverbal exchanges between two or more participants who observe certain rules and also violate them in an irregular flow of speaker's and listener's turns, acceptable and unacceptable simultaneous activities, acoustic and visual pauses, and a number of other positive and negative behaviors within each turn, differentially oriented between speakers and listeners or among listeners, and conditioned by personality, situational context, and cultural background (Poyotos, 1982, p. 156).

In the earliest writings of Alois Alzheimer in 1906, communication and conversation difficulties were identified as a key and problematic feature of the syndrome that carries his name (Alzheimer, 1906). Alzheimer noted the following symptoms in his 51-year-old female patient, Auguste D., "… that although she spoke clearly and articulated well (i.e. speech production), she often stopped midsentence as if she were at a loss or indecisive as to whether she was saying the right thing." (Dahm, 2006, p. 906). Research into pragmatic language skills in conversation contexts has continued in earnest since those earliest observations. Examples of more recent and notable volumes and reviews of the wide-ranging pragmatic and conversational skills of persons with dementia of the Alzheimer type include Asp and de Villiers (2010), Guendouzi and Müller (2014), and Kindell et al. (2017).

Problematic talk in the form of conversation breakdowns (i.e. episodes where the sender and receiver unsuccessfully exchange a message/idea) and unsuccessful conversation repairs (i.e. failed actions taken to resolve a conversation breakdown) are common in dementia of the Alzheimer type (e.g. Guendouzi & Müller, 2002; Orange et al., 1996, 1998; Watson et al., 1999; Young et al., 2016). Language and cognitive impairments in dementia of the Alzheimer type lead to difficulties with turn management, stating messages clearly, understanding others, and marking referents, among other challenges (e.g. Orange & Purves, 1996). The source of problematic talk is not isolated to changes in the affected person's language, cognition, and pragmatic abilities. Problematic talk also results from family conversation partners' maladaptive responses to conversation difficulties (e.g. Guendouzi & Müller, 2002; Orange et al., 1996; Small & Perry, 2005; Savundranayagam & Orange, 2011; Young et al., 2016). These maladaptive responses have important implications relative to communication-related caregiver burden, the expression of responsive behaviours by persons with dementia of the Alzheimer type, and strategies for caregiver communication enhancement education and training programs, the latter of which are addressed later in this chapter.

Topic management difficulties among persons with dementia of the Alzheimer type include limited topic selection, reduced topic initiation, difficulty maintaining topics, ambiguous change/shift and reintegration of topics. These problems reflect impairments in the simultaneous integration of multiple cognitive, language, cultural, sensory and social systems and processes (Dijkstra et al., 2004). Early descriptions of topic management behaviours in persons with dementia of the Alzheimer type showed that they use topic maintenance utterances infrequently and make few topic changes, but that when they do shift topics it is done incoherently (Garcia & Joanette, 1994, 1997; Mentis et al., 1995). Mentis et al. (1995) described the passive nature of the topic management style of persons with dementia of the Alzheimer type wherein their conversation partners played prominent roles introducing and maintaining topics while simultaneously creating communication breakdowns when shifting topics. The influential nature of poor topic management by persons with dementia of the Alzheimer type impacts their conversational partners who relinquish topic development during overlapping turns when talking (Young et al., 2016) and during non-coherent topic shifts (Garcia et al., 2001). Interestingly, persons with dementia of the Alzheimer type (particularly in early and middle stages) pretend frequently to know the topic(s) or correct answers to questions during interactions, a discourse behaviour referred to as a “saving appearances response,” which can lead partners to underestimate the severity of the dementia (Matsushita et al., 2018).

Wray (1999, 2014) observed the increase in the occurrence of formulaic, overlearned forms of language in the context of rising processing demands. Formulaic language sequences are defined as:

> …a sequence, continuous or discontinuous, of words or other meaning elements, which is or appears to be, prefabricated: that is, stored and retrieved whole from memory at the time of use, rather than being subject to generation of analysis by the language grammar (Wray, 1999, p. 214).

Presumably, formulaic responses are used during topic management problems or during instances of conversational breakdown to optimize continuity within and to prolong the duration of social interactions. More recently, Hall et al. (2018) found that the familiar conversation partners of three persons with dementia facilitated topic extensions specifically when participants with dementia experienced topic management problems related to their declarative memory problems, and not during topic shifting. This suggests that conversation partners are sensitive to different types of conversation behaviours in dementia of the Alzheimer type and differentially apply facilitation ‘strategies’ in varying conversation contexts.

Williams et al. (2010) examined the co-occurrence of topic maintenance and conversational breakdown using multiblock discriminant correspondence analysis in the conversations of normal, non-cognitively impaired older adults and of persons with early and middle clinical-stage dementia of the Alzheimer type and family caregivers. They found that control dyads exhibited fewer significant associations between topic boundaries (i.e. change and shifts) and conversation breakdowns whereas early and middle clinical-stage dementia of the Alzheimer type dyads showed stronger (i.e. increasing) associations across the trajectory of the clinical stages which distinguished the three diagnostic groups. These findings align with the observations of showing partners' contributions that link topic shift and conversational problems in dementia of the Alzheimer type dyads. Additionally, family caregivers of persons with early clinical-stage dementia of the Alzheimer type and persons with middle clinical-stage dementia of the Alzheimer type contribute disproportionately more utterances that create miscommunication (Orange et al., 1996). These miscommunication-causing utterances have issues related primarily to lexical marking of referents, declarative memory problems and unsuccessful repair strategies containing elaborations, including lexical markers of tense and semantic content (Orange et al., 1996).

Overall, the literature on conversational breakdown and repair shows that persons with dementia of the Alzheimer type, regardless of clinical stage, signal misunderstandings (i.e. repair initiators) in a myriad of ways, attempt to repair their own errors and those made by their partner which results in both expedient resolution and unresolved conversation outcomes. The active contributions of persons with dementia of the Alzheimer type to the creation and to the resolution of misunderstandings reveal an inherent conversational competence that can be capitalized on in caregiver communication enhancement education and training programs. The heretofore identified conversational competencies of persons with dementia of the Alzheimer type can be used to dispel the myths and stereotypes that persons with dementia of the Alzheimer type are not competent conversationalists, reduce their communication predicaments, optimize their social engagement and participation which, in turn, will reduce their isolation, loneliness and responsive behaviours.

14.7 Impact of Pragmatic Impairments on Families

14.7.1 Impact of Pragmatic Impairments on Relationships

Pragmatic communication abilities are essential in the maintenance of one's identity in relation to another person. The most central relationship identities in caregiving relationships are spousal and filial identities given that most family caregivers are spouses/partners and adult-children. Pragmatic communication impairments associated with dementia of the Alzheimer type make it challenging for family members to maintain their initial relationship identities as spouse or adult-child (Dahm, 2006). According to Caregiver Identity Theory, negative and persistent changes in the quality of the relationship can be distressing for caregivers when they highlight a discrepancy between a caregiver's appraisal of themselves and their norms or standards for their relationship with the person with dementia (Montgomery & Kosloski, 2013). For example, a wife who experiences scolding by her husband with dementia may experience an internal conflict between her self-appraisal of what it means to be a good wife and/or a good caregiver in the context of the social feedback provided by her husband that is skewed because of his pragmatic communication impairments (Chan et al., 2010).

In dementia of the Alzheimer type, the relationship identity, which includes the initial familial role, often expands to include the caregiver role (Donnellan et al., 2015). Some caregivers find the changes to the initial familial relationship to be challenging to accept. This is true for both spousal and adult-child caregivers. For example, spousal caregivers experienced less mutuality and reciprocity in their relationships because their partners with dementia were less able to understand or to contribute to a conversation, and were too reliant on their caregivers for thinking and social interaction (Boylstein & Hayes, 2012; Quinn et al., 2008). The lack of meaningful social communication was the most frequently reported source of marital disruption by caregivers in a study on caring for spouses with dementia (Boylstein & Hayes, 2012). Spousal caregivers reported missing the rich conversations they had prior to the onset of dementia of the Alzheimer type. They missed talking about politics and their daily experiences, especially in cases where the family caregiver still worked. They also reported having to take the lead in initiating and maintaining conversations as a result of pragmatic communication impairments. Adult-children reported losing a part of themselves, when they can no longer seek advice and reassurance from their parent with dementia (Caron & Bowers, 2003). These studies show how pragmatic impairments highlight a discrepancy between and the current experience for family caregivers and the norms of their spousal (i.e. mutuality, reciprocity) or parent-child relationships (i.e. receiving advice and guidance).

14.7.2 Impact of Pragmatic Impairments on Behaviour

What disrupts relationships further are responsive behaviours. Responsive behaviours, which have been called behavioural problems or problematic/disruptive behaviours, are behavioural responses to unmet needs (Whall & Kolanowski, 2004). Responsive behaviours are the most common and consistent predictors of caregiver burden in dementia of the Alzheimer type (Chan et al., 2010; Savundranayagam et al., 2005, 2011; Savundranayagam & Orange, 2011; Savundranayagam & Montgomery, 2010). These behaviours can result from pragmatic communication difficulties (Savundranayagam et al., 2005; Savundranayagam & Orange, 2011). They include behaviours such as anger, agitation, or inappropriate sexual behaviour.

Pragmatically appropriate conversation feedback is an important aspect of relationship maintenance. Receiving negative feedback, in the form of responsive behaviours, can be frustrating for caregivers and diminish their level of closeness in conversations with their relative with dementia. For example, one caregiver reported being “often scolded…fiercely with foul language” by her husband with dementia (Chan et al., 2010). Moreover, pragmatically inappropriate responsive behaviours are often embarrassing and make it difficult for the caregiving dyad to engage in social interactions in public. Consequently, they can lead to greater social isolation for the dyad. For example, caregivers reported that their spouses with dementia would become aggressive when having difficulty engaging in conversation with other people (Tatangelo et al., 2018). As pragmatic communication abilities decline, persons with dementia of the Alzheimer type may respond to communication breakdowns or misunderstandings with responsive behaviours.

Repetitive questions or statements are responsive behaviours that violate expectations and rules around the social use of language. Repetitive questions have been reported as frustrating for family caregivers for the last two decades (Bourgeois et al., 1997; Boylstein & Hayes, 2012; Donnellan et al., 2015; Kuhn, 1998). Caregivers report being embarrassed publicly by their relatives' repetitive questions (Boylstein & Hayes, 2012). Repetitive questions can also lead to other forms of responsive behaviours (e.g. escalating anger, withdrawal) that may emerge in the context of a family member's frustrations with pragmatic language difficulties in dementia of the Alzheimer type (Savundranayagam & Orange, 2011). Therefore, it is important for caregivers to consider that the source of responsive behaviours in dementia of the Alzheimer type may be related to pragmatic communication difficulties. Interventions to support family caregivers in the use of effective communication strategies (discussed briefly later in the chapter) can help them to prevent responsive behaviours by improving social closeness experienced in the context of conversations.

14.8 Pragmatic Language Interventions in Dementia of the Alzheimer Type

Over the past three decades, there have been tremendous advances in the development of theories and frameworks to enhance the communication of older people with and without dementia. This includes the Communication Enhancement Model (Ryan et al., 1995) for use with older adults, the Communication Empowerment Framework (Morris et al., 2018) for adults with dementia, and person-centered communication (Love & Pinkowitz, 2013). Several key reviews have been published of communication interventions for persons with dementia and their caregivers (Alsawy et al., 2017; Egan et al., 2010; Hopper et al., 2013; Swan et al., 2018; Vasse et al., 2010). Communication enhancement education and training programs for family and health care professionals, some empirically based, have also been published (e.g. Critten & Kucirkova, 2019; Krause et al., 2009; Liddle et al., 2012; Roberts, 2018; Williams et al., 2018) as well as reviews of these programs (Eggenberger et al., 2013; Machiels et al., 2017; Morris et al., 2018; Nguyen et al., 2019).

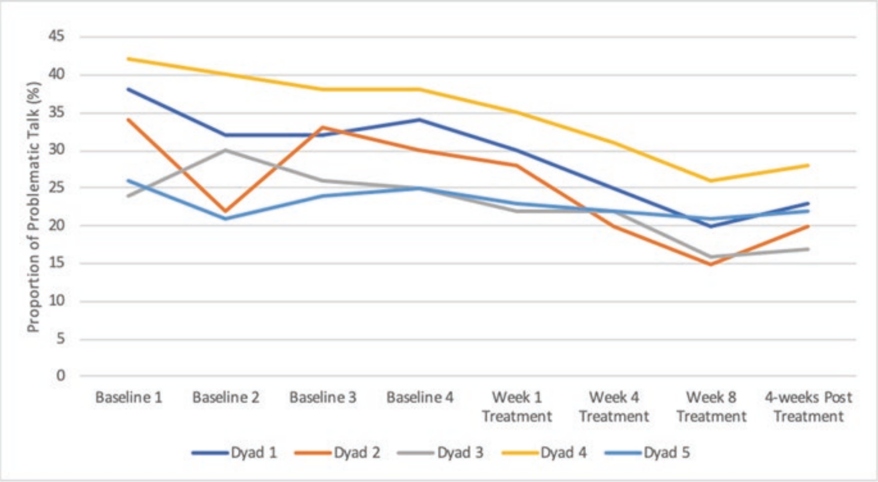
The overall thrust of the reviews is that direct interventions on communication enhancement for persons with dementia are premised on the fact they can learn, especially when facilitated by social interaction (Duff et al., 2013) or spaced retrieval principles (Hopper et al., 2005). Caregiver communication enhancement education and training programs typically adopt both didactic and experiential learning, with more recent interventions including the use of trained, simulated clients to demonstrate the skills of persons with dementia of the Alzheimer type and to offer hands-on experiences practicing and implementing evidence-informed strategies. To this end, novel approaches are emerging in this area that include on-line, asynchronous modules and video-based vignettes summarizing effective language and pragmatic communication approaches to persons with dementia. Others integrate new technology of tablet-based conversation memory notebooks and life storybooks (Dynes et al., 2018; Subramaniam & Woods, 2016) built off previously well-documented hardcopy materials to enhance novel conversation utterances and topics (Bourgeois, 1992).

To date, the majority of programs direct conversation strategy education and training efforts toward the healthy communication partner. A limited number of conversation strategy education and training interventions include the person with dementia as an active learner (Roberts, 2018; Small & Perry, 2012/2013). With few exceptions, family communication dynamic studies have examined communication partners' acquisition of strategy knowledge but have not examined treatment effectiveness specifically on pragmatic communication behavior changes between the person with dementia of the Alzheimer type and their family interlocutors.

Roberts' (2018) pilot study suggests that a dyadic conversation strategy education and training program that actively engages both the person with dementia of the Alzheimer type and their family interlocutor is feasible and results in real-world pragmatic communication improvements measured using conversation analysis approaches. Restructuring Dementia Conversations (RDC) is an 8-week education and training program. It integrates active learning techniques including conversation practice activities with on-line clinician feedback and synchronous, off-line reflection using video-review of conversations between the person with dementia and their family interlocutor. Baseline conversations are analyzed to identify dyad-individualized conversation strategies based on existing positive strategies/pragmatic behaviors that can be enhanced by training (e.g. collaborative conversation repair) and ineffective strategies/pragmatic behaviors that are reduced/extinguished through training (e.g. overlapping talk). Dyad-centered strategies taught in the program focus on message clarity, language structure, detecting conversation signals of success and breakdown, mutuality/reciprocity, among others. Treatment outcomes include both objective (proportion of problematic talk) and subjective (conversation rating scales) pragmatic communication measures. Proportion of problematic talk at the dyad level is calculated as the number of words involved in conversation breakdowns and unsuccessful conversation repairs divided by the number of total words.

Pilot data from a group of mild and moderate clinical-stage individuals with dementia of the Alzheimer type showed an average reduction in overlapping talk of 30%, a perceived reduction in conversation difficulties of 30%, an average 22% increase in the number of conversation turns initiated by the person with dementia, and an average 24% reduction in conversation breakdowns (Roberts, 2018). Below are RDC pilot data for the outcome measure proportion of problematic talk, collected during in-home conversation recordings from five dyads (Fig. 14.1). In general, participants show a reduction in the proportion of problematic talk that is maintained 4-weeks post treatment.

Despite advances, collectively, the extant literature leaves remaining questions regarding the efficacy of conversation training approaches on real-world conversations, pragmatic communication behaviours, and quality of life outcomes in persons with dementia of the Alzheimer type and their informal conversation partners. It is also unclear whether, and at what stages, the efficacy of conversation enhancement programs may benefit from including the person with dementia as an active participant in the intervention versus the recipient of strategies learned by their communication partners. Lastly, although interventions target pragmatic communication, currently few interventions measure outcomes using variables that reflect directly changes in pragmatic communication behaviors between persons with dementia and their interlocutors. As such, much more research is needed into interventions that are grounded more robustly within a pragmatic language framework.



Dyad 1: Male middle clinical-stage DAT age 77 + female spouse age 68
Dyad 2: Female early clinical-stage DAT age 72 + male spouse age 74
Dyad 3: Female early clinical-stage DAT age 89 + son age 63
Dyad 4: Female middle clinical-stage DAT age 52 + male spouse age 56
Dyad 5: Male early clinical-stage DAT age 68 + male spouse age 67

DAT = Dementia of the Alzheimer type
Proportion of problematic talk = the number of words involved in conversation breakdowns and unsuccessful conversation repairs divided by the number of total words

Fig. 14.1 Proportion of problematic talk multiple baseline single case study trial of restructuring dementia conversations

14.9 Summary

Research in the area of pragmatics, communication and dementia of the Alzheimer type over the past several decades has yielded valuable new insights into the complex relationships among linguistic, cognitive, social and behavioural systems and processes. Research findings to date show that persons with dementia of the Alzheimer type retain selected foundational elements of pragmatics that impact their everyday conversations. These include, but are not limited to, several levels of theory of mind, understanding some conventional metaphors and idioms, participating actively in conversational topic management and conversational breakdown and repair strategies, and displaying linguistic and behavioural features that show personhood and 'self' are present, albeit in diminished form. Nonetheless, the expanding foundation of research in pragmatics and persons with dementia of the Alzheimer type provides empirically and clinically important insights that can help family and formal caregivers understand the disease process and communicate effectively with persons living with this condition.

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Chapter 15
Parkinson's Disease

Marc D. Pell, Laura Monetta, Jonathan A. Caballero, and Valérie Coulombe

15.1 Introduction

Parkinson's disease (PD) is a neurodegenerative illness associated with dopaminergic cell loss in the brain which affects middle-aged and older adults (typically people over the age of 50 years). It is commonly known that people with PD (PwPD) experience difficulties with movement and a progressive decline in motor functions. The 'hallmark,' diagnostic signs of the disease are slowness of movement (bradykinesia), resting tremor, muscle rigidity, and changes in posture and gait. Most PwPD receive dopamine replacement therapy (levodopa or "L-Dopa") to alleviate their motor symptoms by restoring the abnormal function of dopamine-transmitting neurons in the brain.

In addition to motor disability, PD neuropathology progressively involves each of the principal domains of cognitive function: attention; memory; executive function; perceptual-motor function; language; and social cognition (American Psychiatric Association, 2013). The prevalence and evolution of cognitive deficits in PwPD is quite heterogeneous, although non-motor symptoms inevitably become more prominent with disease progression (Roheger et al., 2018). It is estimated that 30–40% of PwPD present with mild cognitive impairment at time of diagnosis (Yarnall et al., 2014) and the effect of pharmaceutical interventions that control motor-related symptoms, such as L-Dopa, may at times exacerbate cognitive symptoms in the disease. Ultimately, the majority of PwPD develop broader cognitive deficits, leading to dementia over the 15 to 20-year course of the disease (Aarsland et al., 2003; Buter et al., 2008).

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Research highlighting the psychosocial impacts of living with PD within the clinical profile of the disease is much more recent. As summarized more than a decade ago by Pell and Monetta (2008), the complex manner in which both motor and cognitive functions are altered by the disease plays a key role in understanding how PD affects interpersonal communication and social-pragmatic skills. Much has been learned in recent years. Many research teams are now actively studying how PD affects language use, identifying potential factors that contribute to 'pragmatic' communication abilities in PwPD. This chapter provides an update and analysis of this literature, while considering avenues for remediating pragmatic deficits.

15.2 Neuropathology and Clinical Features of PD

Parkinsonism refers to a spectrum of degenerative neurological conditions associated with the unilateral onset of extrapyramidal motor signs and symptoms, accompanied by variable changes in perceptual and (social) cognitive functions (Titova et al., 2017). Heterogeneity in the pathology and clinical presentation of PD, and how the disease specifically affects communication skills, reflects the diverse ways that major dopamine-producing sites in the midbrain degenerate, altering brain function through their connectivity with subcortical and cortical brain areas. In idiopathic PD, loss of dopamine-producing neurons in the substantia nigra pars compacta leads to dopamine insufficiency in the striatum via the nigrostriatal pathway. These changes produce extrapyramidal motor symptoms once dopamine loss attains critical levels (Haber, 2003).

Over time, cognitive abilities are affected as neural projections to the striatum, limbic structures, and prefrontal areas of the neocortex (among others) are functionally compromised. Denervation in the basal ganglia gradually impacts on a set of parallel, functionally separable basal ganglia-thalamocortical circuits (Alexander et al., 1986). Several of these circuits play a critical role in motivated behavior and a range of high-level cerebral processes that control cognition, decision-making, and adaptive social behaviour (Jiang, 2018; McNamara & Durso, 2018). Disruption of the limbic and prefrontal circuits, which project to areas of the dorsolateral or orbitofrontal prefrontal cortex of the brain, variably produce different 'frontal lobe signs' (Levy & Dubois, 2006). These deficits include difficulties in the organization of information to facilitate behaviour (executive functions, dorsolateral prefrontal circuit) and/or difficulties using and integrating emotional/limbic information from the environment (orbitofrontal circuit) (Alexander et al., 1986; Bonelli & Cummings, 2007). As PD neuropathology always begins on one side of the brain (left- versus right-sided onset) and the basal ganglia project to the opposite cerebral hemisphere, the way that cortical (especially frontal) brain regions are compromised in PD is further dictated by how neurological functions are functionally lateralized at the level of the neocortex. The side of disease onset can, therefore, add to the complexity of clinical symptoms experienced by many PwPD, especially processes involving language and social cognition.

Research demonstrates that both motor and cognitive deficits can be a source of problems that affect language use in PwPD (Pell & Monetta, 2008). Loss of motor flexibility and control produces other ubiquitous signs of PD that involve communication, such as facial masking (hypomimia) and slurring of speech (hypokinetic dysarthria). These conditions, which can play a critical role in interpersonal behaviour, are directly linked to reduced physiological support of the underlying musculature, restricting the typical range of facial and vocal-articulatory movements (Darley et al., 1969; Jankovic, 2008). Sensory-motor deficits are not the only source of communication difficulties in PwPD. Progressive changes in visuo-perceptual functions (Clark et al., 2010), attention and processing speed (Grossman et al., 2002; Jokinen et al., 2013) and memory (e.g. Kosutzka et al., 2019; Lewis et al., 2003; Monetta, Grindrod, & Pell, 2008b) can impede language processing and effective communication in significant ways, because the ability to appropriately use language in its social context rests on these basic mental functions. There is especially strong evidence that impairments in executive resource control, due to disruption of the dorsolateral prefrontal circuit, negatively impact on pragmatic language skills. Goal-directed behaviour is supported by executive functions, such as working memory (Gabrieli et al., 1996), attentional set-shifting (Monchi et al., 2004; Owen et al., 1993), and mental processes for flexibly planning and inhibiting both internal and external information (Brown & Marsden, 1991). Since many pragmatic abilities rest upon intact executive processing skills, the progressive decline in these functions can produce secondary 'cognitive-linguistic' impairments as the disease advances.

According to McNamara and Durso (2018), another reason that pragmatic deficits are prevalent in PD is because the disease affects key structures of what has been called the "social brain" (e.g. the ventral striatum of the basal ganglia, amygdala, insula, and ventromedial and dorsomedial regions of the prefrontal cortex). These brain regions are critically involved in a number of social functions, such as the perception of socioemotional cues and the ability to recognize intentions and mental states, allowing individuals to interpret the contextual significance of language. Given that the neuropathological course of PD affects brain systems devoted to language use and social cognition, it is not surprising that many PwPD encounter difficulties when interpreting the socially intended, 'pragmatically-appropriate' meanings of language (Monetta et al., 2009; Murray, 2008; Pell & Monetta, 2008; Vachon-Joanette et al., 2013). Moreover, the fact that PD is primarily associated with subcortical pathology with secondary effects on cortical operations, it is not surprising that many cortically-based language functions (e.g. phonological and morphosyntactic knowledge) are relatively spared in PD when compared to conditions defined by degeneration of the neocortex, such as Alzheimer's disease (see Auclair-Ouellet et al. (2017) for a review, and Chap. 14 in this volume).

Although research on the nature of pragmatic language disorders in PD is just beginning to mature, there can be no doubt that PwPD report serious concerns and frustration about changes in their ability to communicate effectively (Miller et al., 2008) and that these changes negatively affect their quality of life (Miller, 2017; Schwartz & Pell, 2017). Both PwPD and their communication partners identify changes in communication as a major barrier which limits control over their lives, their confidence, and independence (Miller et al., 2008). To better understand these issues, we summarize evidence of how PD affects pragmatic language abilities in two inter-related neurocognitive domains: verbal communication and social cognition (including conversational interactions). The next section considers how PD impacts on linguistic operations and the transmission of meaning through language (especially non-literal meanings). In Sect. 15.4, we then look at how broader changes in social cognition, such as emotion processing and perspective-taking (‘theory of mind’), contribute to pragmatic deficits in PD, and how PwPD engage in conversational interactions. We conclude the chapter by describing current therapeutic approaches used in PD, mostly to alleviate motor-related symptoms of the disease, while pointing to promising areas for targeted intervention of difficulties in social communication.

15.3 Effects of PD on Verbal Communication

When the effects of PD on verbal communication are broadly evaluated, research shows that PwPD have pronounced difficulties understanding the indirect, non-literal meanings expressed through language in many social contexts. The ability to process non-literal language was described by Austin and Grice, who pointed out that interpersonal communication requires sensitivity to linguistic and social cues which convey “speaker’s meaning” (Grice, 1975). Listeners must be able to distinguish what is said from what is intended in a particular linguistic or physical context. When non-literal meanings are communicated, listeners must go beyond the literal meaning by presumably inhibiting the literal one (“indirect access” theory; Grice, 1975). Other researchers argue that access to non-literal meanings is more direct in many situations and depends on the relevance and expectations created by an utterance (Gibbs, 1999; Giora, 1999; Sperber & Wilson, 2006). Either way, it can be said that the ability to infer the non-literal meaning of language, or intended speaker meaning, is not always straightforward. This ability typically relies on other aspects of cognitive performance, such as attention (e.g. to the relevance of different social cues), executive functions (e.g. to inhibit alternate meanings), and working memory (e.g. to hold multiple sources of information in memory during utterance interpretation).

There is evidence that PD affects the ability to interpret non-literal language in several ways. Many PwPD display abnormalities in the comprehension of emotional connotations of verbal cues and metaphorical expressions used in everyday conversations (e.g. “this pie is heaven”). They may also find it difficult to generate different types of inferences from oral or written discourse (i.e. to understand what is not said based on what is actually said), to detect verbal irony (e.g. sarcasm), and to recognize the pragmatic implications of indirect speech acts (e.g. interpreting a phrase such as “it’s really hot in here” as an indirect request to open the window).

In what follows, we examine each of these topics in more detail to arrive at a broader picture of how PD affects verbal communication and receptive language.

15.3.1 Emotional Connotations of Language

Unlike neurodegenerative conditions such as Alzheimer's disease, the semantic knowledge underlying concepts/word forms used in verbal communication remains largely preserved over the course of PD (readers are referred to Salmazo-Silva et al. (2017) for related research and a more detailed analysis). Nonetheless, many PwPD appear to experience difficulties when linguistic concepts refer to emotion, affect or inner motivational states.

The emotional associations encoded by language may refer to 'basic' emotion states (e.g. joy, anger; Ekman & Cordaro, 2011) or to affective properties of environmental stimuli, such as their valence (inherent pleasantness) or arousal (level of activation associated with stimulus appraisal; Palazova, 2014). For example, some words are strongly associated with discrete emotion categories, such as fear for "danger" or disgust for "mucus." Most words also have affective associations which are evaluated and understood along a continuum. The word "angel" typically has a positive valence, whereas "evil" is negative; "peace" is associated with low arousal, whereas "rollercoaster" has a much higher level of arousal. The processing of emotional words, including their pragmatic uses, emotional connotations, and associated motor and physiological responses, is governed by features of the emotional conceptual-semantic system (Abbassi et al., 2015; Macoir et al., 2019).

Research shows that PwPD are less accurate than age-matched healthy participants when asked to infer the discrete emotional meaning of written words (Wagenbreth et al., 2016) or written sentences, such as "I didn't make the team" (Paulmann & Pell, 2010). Also, their ability to characterize the affective properties of words often differs from healthy adults. For instance, one study reported that PwPD gave more neutral ('blunted') ratings to written words associated with both negative and positive concepts and displayed differential sensitivity from healthy adults in their intensity ratings (Hillier et al., 2007). However, PwPD appear to display a sensitivity to the affective relevance of words when evaluated using more implicit measures, such as when making lexical decisions, i.e. judging whether a word is real or not, or when neurophysiological responses to affective words are recorded (Borg et al., 2012; Castner et al., 2007; Dissanayaka et al., 2017; Wagenbreth et al., 2016).

Overall, these studies suggest that automatic semantic processing of emotional (and non-emotional) words is generally spared in PwPD, although these processes tend to be slower or delayed in many patients who have other cognitive processing impairments (Angwin et al., 2017; Dissanayaka et al., 2017; Iyer et al., 2019). Performance seems to deteriorate especially when PwPD are required to explicitly identify and retrieve information about emotional attributes of language (Coundouris et al., 2019), suggesting that cognitive task demands are an important factor in how PwPD use emotional information from language.

Another way that PD impacts on semantic processing in PwPD is in their ability to activate and appreciate the sensory-motor properties of (emotional) words. According to Embodied (Simulation) Cognition Theory (e.g. Shapiro, 2010), mental constructs are built from motor, perceptual, and bodily experiences that form a core part of semantic knowledge, details which may be critical for recognizing emotional language. Interestingly, studies show that relative to healthy controls, semantic processing in PwPD is slower and less accurate when processing action verbs versus objects (Cotelli et al., 2007; Fernandino et al., 2013; Salmazo-Silva et al., 2017). These effects may highlight the fact that words representing actions draw more heavily on 'embodied' motor-related semantic content and underlying brain systems, such as the motor basal ganglia-frontal loop, which are impaired in PD (see Smith & Caplan, 2018). Reduced access to 'embodied' semantic features that represent words of an emotional nature may also be predicted. Thus, there is preliminary evidence that (emotional) semantic processing in PD can be negatively affected by both cognitive and motor-related deficits in the disease.

15.3.2 Metaphorical Language

The use of metaphorical language is not just a poetic way to express oneself but an important mode of communication that people use every day (Cardillo et al., 2012; Kövecses, 1988; Lakoff & Johnson, 1980; Smith et al., 1981). In a metaphorical phrase, the intended meaning goes beyond the literal meaning of the words used. A person, object or other entity is described in terms of something that is conceptually distinct. For example, if someone says, "that baby is a doll," the speaker intends the hearer to infer that the baby shares certain properties with dolls, such as their cuteness. Thus, metaphor comprehension requires listeners to activate corresponding literal and/or figurative concepts in semantic memory and to switch between different attributes of the concept (mental flexibility) to select a contextually appropriate meaning (Bowdle & Gentner, 2005; see Holyoak and Stamenković (2018) for a critical overview of metaphor comprehension).

The ability of PwPD to process metaphorical versus literal meanings of language has been investigated in a series of studies (Berg et al., 2003; Monetta & Pell, 2007; Tremblay et al., 2014). For example, Monetta and Pell (2007) used a priming task to examine how people with and without PD activate the literal vs. metaphorical meaning of written utterances, such as "That tiny mosquito was a vampire." Results of these studies show that PwPD make more comprehension errors and respond more slowly to utterances containing metaphorical meanings than healthy controls. This is not the case for literal meanings (Berg et al., 2003; Monetta & Pell, 2007; Tremblay et al., 2014). In one study, the ability to activate metaphorical meanings was selectively impaired in PwPD who had executive impairments, such as reduced working memory, implicating the dorsolateral frontostriatal circuitry in successful metaphor comprehension (Monetta & Pell, 2007).

Other evidence suggests that metaphor comprehension in PwPD is linked to a decline in frontal lobe executive control that affects high-order language processing (Lewis et al., 1998; McKinlay et al., 2009; Monetta & Pell, 2007; Grossman et al., 2003; Miller, 2017). This hypothesis was explicitly tested by Tremblay et al. (2014). However, in contrast to most studies in the literature, they found no evidence that metaphor comprehension deficits were predicted by difficulties in executive control in PwPD. Moreover, when the authors looked at the contribution of potential linguistic variables that could influence comprehension (e.g. metaphor polysemy, or the extent to which metaphors had multiple meanings), there was again no relationship between the degree of polysemy and performance on the metaphor comprehension task (Tremblay et al., 2014). The authors concluded that PwPD may not adequately activate the multiple features underlying metaphorical concepts, leading to a misunderstanding of metaphorical relationships. Monetta and Pell (2007) speculated that in PD patients with working memory problems, processes underlying metaphor interpretation may simply be delayed. These data suggest that, among other potential factors, the role of executive functions in the comprehension of metaphorical meanings by PwPD should continue to be studied.

15.3.3 Generating Inferences from Discourse

An inference is any conclusion that someone draws about what is "not said" based on what is actually "said." While an inferential process is often necessary to understand the contextual relevance of speech acts and ironic comments, people draw various types of inferences about the meanings conveyed by discourse as they process conversations or narratives. For example, when the events of a story are not explicitly related to each other, readers generate a bridging inference to conceptually link the elements together to form a coherent internal representation of the story. Typically, this internal representation combines explicit linguistic information from the narrative with general world knowledge already held by the individual (McKoon & Ratcliff, 1992; Potts et al., 1988). Thus, successful generation of inferences will result in a story representation that involves both the explicit propositions contained in the text and implicit propositions inferred by the individual by bridging or elaborating information in the text.

Berg et al. (2003) presented a battery of tests evaluating pragmatic language abilities to adults with and without PD, including a test in which participants had to make inferences from narratives or dialogues of varying complexity. They found that difficulties generating inferences from discourse were more pronounced in PwPD when compared to a range of other "high-level" language tasks (comprehension of ambiguous sentences, metaphors, word definitions, etc.). This study suggests that the ability to make appropriate inferences from discourse is a particularly sensitive test of high-level language abilities in individuals affected by PD (Berg et al., 2003). Work by Monetta, Grindrod, and Pell (2008b) usefully elaborates on this topic. Using a standardized test, they evaluated how PwPD draw inferences from short stories based on explicit or implied information that touches on main ideas or details presented in the discourse. Results showed that only a subgroup of PD patients, those with working memory deficits, were significantly impaired in the ability to make correct inferences when compared to control participants. PwPD who had working memory problems found it especially difficult to make predictive inferences about implied details in the stories. These data suggest that PD is associated with problems generating inferences but in a selective manner, depending on the status of cognitive skills that support the inferential process.

The ability to generate alternative ideas about what could have happened based on a description of past events also relies on the ability to make inferences and to engage in causal reasoning. McNamara et al. (2003) looked at how well PwPD and healthy adults make ‘counterfactual inferences’ based on a brief context, while completing a battery of tests that included measures of general cognitive ability, social functioning, and specific measures of prefrontal functions (inhibition, interference, and planning). Results confirmed that PwPD are less able to generate counterfactual thoughts and performed at chance level on the test of counterfactual inferencing, an impairment that was related to frontal dysfunction in their participants rather than generalized cognitive impairment. These data underscore the relationship between prefrontal cognitive impairments and difficulties using verbal information to generate inferences and to reason, although much more work is needed in this area.

15.3.4 Understanding Verbal Irony

Another situation in which PwPD have difficulties understanding non-literal meanings of language is when processing ironic messages. When people communicate irony, they strategically highlight contextual, visual and/or vocal cues that accompany their utterance to convey a meaning that differs from what is said literally (Gibbs, 2000; Mauchand et al., 2020). Understanding irony involves an inferential process that considers multiple sources of information about intended speaker meanings (e.g. lexical, contextual, emotional and prosodic sources). People must detect the relevance of cues that could point them to the intended non-literal (ironic) meaning of an utterance while considering shared knowledge held by the speaker and listener for the ironic meaning to be detected (see Gaudreau et al., 2013). Neuropsychological studies of various brain-damaged populations show that the ability to understand verbal irony is highly correlated with both the ability to make inferences about another person’s thoughts (theory of mind) and an individual’s executive resource capacity (Champagne-Lavau & Joanette, 2009; Martin & McDonald, 2005; Mo et al., 2008; Monetta et al., 2009). Thus, intact executive functions and social cognitive skills, such as perspective-taking, may be prerequisites for understanding verbal irony.

The ability of PwPD to understand verbal irony was investigated by Monetta et al. (2009). PwPD simultaneously heard and viewed stories which ended in a lie or an ironic statement, which could only be differentiated by correctly inferring what the protagonist of the story knew in relation to another story character. Compared to healthy adults, PwPD were less able to judge whether a final statement of the story should be interpreted as a joke (irony) or a lie. In addition, PwPD were less accurate than controls in answering true/false questions about the second-order beliefs held by story characters (i.e. to decide what one character thinks about what another character is thinking). In a follow-up study, Pell et al. (2014) presented videotaped vignettes of people in everyday social interactions to a new group of adults with and without PD. Participants had to infer whether the final statement of an interaction was sincere, a lie, or a sarcastic comment (i.e. verbal irony). The authors also presented tasks measuring theory of mind, emotion processing, and cognitive performance. Results showed that PwPD were impaired in the ability to infer "enriched" social intentions, such as irony, sarcasm or lies, from non-literal remarks made in the videotaped dialogues. In contrast, they showed similar capacity to healthy participants to recognize emotions and social intentions meant to be literal. Difficulties drawing complex social inferences were significantly correlated with limitations in working memory and executive functioning in PwPD (Pell et al., 2014).

When put together, the limited studies of verbal irony processing in PwPD suggest a failure in non-literal interpretation abilities and a specific decline in the ability to make second-order mental state attributions while generating inferences. As suggested by the literature on metaphor comprehension and the ability to generate broader inferences during verbal communication, the extent to which frontal lobe signs are present in PwPD seems to act as a strong predictor of difficulties understanding ironic language and making mental state attributions that allow PwPD to detect non-literal meanings and intentions.

15.3.5 Understanding Indirect Speech Acts

Another feature of non-literal language that can be problematic in PwPD is the ability to understand indirect speech acts—an utterance in which one speech act is performed indirectly by performing another (Searle, 1975). When someone says, "Can you give me my coat?," typically the speaker is not questioning the listener's physical capacity to give the coat; rather, they are requesting that the listener perform a specific action (i.e. to pass the coat). In everyday life, indirect speech acts are commonly used to reject proposals and to make requests and are frequently tied to politeness considerations in discourse. Understanding indirect speech acts requires an individual to build a conceptual model of the situation and to be aware of the speaker's intentions in order to make a proper inference (Ackeren et al., 2012). Interpreting indirect speech acts relies on an inferential process, rather than a learned association between form and meaning (Holtgraves, 1994).

McNamara et al. (2010) examined the processing of indirect speech in PwPD in two different experiments: one using an "on-line" sentence verification task, in which participants read scenarios and then responded as quickly as possible to utterances which sometimes represented an indirect interpretation of the previous utterance; and a second "off-line" experiment, in which PwPD read scenarios and then had to write down their interpretation of indirect utterances. Results showed that PwPD were significantly slower than healthy controls to activate the indirect meanings of speech acts (Experiment 1), but that they were not less accurate in interpreting the intended meaning of indirect utterances in the off-line task (Experiment 2). These findings imply that PwPD were largely capable of inferring the social relevance of indirect speech acts, but that the speed of activating indirect meanings is slower due to the disease (McNamara et al., 2010). Interestingly, the authors reported that PwPD were overly confident in their interpretations and were often unaware of errors in interpretation. This observation fits with early reports that PwPD sometimes over-estimate their social-pragmatic abilities (McNamara & Durso, 2003; Berg et al., 2003).

To corroborate and extend these results, Tremblay et al. (2012) evaluated whether PwPD with and without depressive symptoms could accurately identify the meaning of indirect speech acts in French, using an indirect speech acts subtest from a normed battery (Protocole Montréal d'Évaluation; Joanette et al., 2004). Results were consistent with the off-line results reported by McNamara et al. (2010), indicating no significant impairment in indirect speech act comprehension in PwPD relative to healthy controls, and no evidence that depression influenced performance (Tremblay et al., 2012). Collectively, these studies suggest that the comprehension of indirect speech acts—which are often used as a 'face-saving device' in conversations—is relatively spared in PwPD when compared to other forms of non-literal language use. Still, it seems likely that reductions in the speed of activating indirect meanings can emerge in certain processing environments and cause difficulties for some PwPD, affecting how indirect speech acts and other forms of non-literal language (e.g. metaphors) are processed during social interactions (McNamara et al., 2010; Monetta & Pell, 2007).

15.3.6 Summary of PD and Verbal Communication

In summary, there are clear indications that PD impacts negatively on the comprehension and use of non-literal meanings encoded by language in many verbal contexts studied to date, in the absence of a breakdown in linguistic abilities per se. Processing metaphorical expressions, drawing inferences from discourse, and understanding ironic, contrafactual intentions of a speaker are frequently problematic in PwPD, although these deficits are heterogeneous among those affected. PwPD who have significant deficits in executive resource capacity, such as working memory, seem most at risk for experiencing difficulties in non-literal language use. These effects are due, at least in part, to a progressive decline in the normal functioning of the basal ganglia-prefrontal circuits. General reductions in cognitive processing speed, which delay the time course for activating non-literal meanings of language and using them during the comprehension process (e.g. for metaphors or indirect speech acts), are also likely to play a role in how PD affects verbal communication and receptive language in real-life settings. In addition, PwPD may have selective difficulties retrieving the experiential ('embodied') features associated with concepts, including affective words, due to a reduced neurophysiological activation for those semantic properties, caused by sensory-motor and cognitive deficits in the disease.

It can be said that the main way that PD affects the contextually appropriate use of language, at least as it involves non-literal communication, is by reducing the cognitive resources and control needed to engage in high-level language processing. Failure to understand non-literal language may have important consequences for well-being and social outcomes as the use of indirect forms of speech in everyday life plays an important social regulatory role. Indirect forms of communication allow strengthening social bonds and permit a sophisticated management of relationships and communication, often minimizing or avoiding potential conflicts and risky situations (Pinker et al., 2008; Rothermich & Pell, 2015). Future studies should look even more closely at the relationship between non-literal language abilities in PwPD and cognitive changes brought on by the disease.

Our review also underscores that pragmatically appropriate communication rests on specific aspects of social cognition, such as the ability to adopt perspectives and to process nonverbal cues that interact with language to guide meaning (particularly when social intentions are non-literal). To fully illuminate how PD affects pragmatic skills, it is thus important to analyze how PD affects social cognition, as developed in the next section.

15.4 Effects of PD on Social Cognition

Social cognition includes the ability to detect, interpret, and appropriately respond to socioemotional cues (social perception) and to attribute affective and mental states to other people (theory of mind). The ability to decipher nonverbal cues in social situations is critical for interpersonal communication to be successful. Nonverbal displays convey different types of emotive information which can guide a listener's interpretation of what the speaker actually means by their utterance (e.g. to be ironic). Understanding how PD affects nonverbal communication and the perception of socioemotional cues is, therefore, vital to describing the functional competence of PwPD in daily life and to understanding how social cognition contributes to high-order language processing.

15.4.1 Social Perception

Social perception is linked to emotive communication, the various ways that people exchange information related to their emotions, affective disposition and mood, attitudes and interpersonal stance, and the effects of these cues on others (Caffi & Janney, 1994). While emotive information can be transmitted verbally (e.g. “I’m very angry!”), more typically humans communicate this information through their facial expression, voice (e.g. shouting), and/or body (e.g. clenching fists), emphasizing the importance of having effective social perception skills. Nonverbal signals can accompany or take the place of speech. Either way, accurate perception of socioemotional cues is crucial for rewarding social interactions (Elfenbein & Ambady, 2002). Deficits in social perception have been linked to reduced quality of life and increased social isolation in several different clinical populations, including autism, traumatic brain injury, as well as Parkinson’s disease (McDonald & Flanagan, 2004; Miller, 2017; Schultz, 2005).

Social perceptual processing relies on cortical and subcortical regions of the ‘social brain’ (McNamara & Durso, 2018) that are directly affected by PD neuropathology. Degeneration of the nigrostriatal and mesocorticolimbic dopamine systems impacts negatively on the insula, frontal lobes, and other key structures of the limbic system (e.g. amygdala) that are critically involved in emotion processing (Péron et al., 2012; see Coundouris et al. (2019) for a recent meta-analysis). Evidence that PD is associated with difficulties in the evaluation and recognition of socioemotional cues has accumulated steadily over the past 25 years (Borg et al., 2012; Gray & Tickle-Degnen, 2010; Pell & Monetta, 2008). Below, we discuss how deficits in emotion processing manifest in PwPD as a function of the communication channel being used (facial vs. vocal displays) and their social ramifications.

15.4.1.1 Facial Expressions

Faces convey information about a person’s identity (e.g. age, race), their emotions, and are used to make a variety of social inferences about another person (e.g. whether they are trustworthy or kind). Recent meta-analyses conclude that the social perception of faces is impaired in PwPD (Gray & Tickle-Degnen, 2010; Coundouris et al., 2019), although the nature and source of these difficulties is not always agreed upon.

One of the many non-motor signs of PD is a decline in visuo-perceptual functions (Clark et al., 2010), operations which contribute to the ability to process faces and other visual social cues. Several studies have concluded that PD is associated with basic difficulties in the visual scanning and structural encoding of faces (Clark et al., 2010; Dewick et al., 1991; Garrido-Vásquez et al., 2016; Marneweck et al., 2014). These deficits may be more pronounced when visual tasks involve ‘configural’ (i.e. holistic) as opposed to componential processing of information from faces (Cousins et al., 2000; Narme et al., 2011). According to this research, specific difficulties in the structural analysis of faces could lead to impairments in a broad array of face processing conditions, including the ability to recognize emotions and other social traits from faces.

Other studies imply that face processing skills break down in more selective ways and are not always accompanied by basic visual-perceptual difficulties. In studies of emotional face processing, there is evidence that PwPD can accurately encode faces to discriminate their identity but fail to correctly recognize the emotion conveyed by facial expressions (Kan et al., 2002; Sprengelmeyer et al., 1995; Gray & Tickle-Degnen, 2010). Others report difficulties in the recognition of specific emotions, such as disgust, fear and anger (Gray & Tickle-Degnen, 2010; Pell & Leonard, 2005; Sprengelmeyer et al., 2003), although deficits are not always restricted to negative emotions (Argaud et al., 2018; Sedda et al., 2017). Further evidence suggests that PwPD can implicitly process emotional information from faces in a typical manner, as measured by priming effects, but are impaired in explicitly naming the emotional expression (Schwartz & Pell, 2017; Wagenbreth et al., 2016). In a recent study that looked beyond emotions, the ability to infer social traits from faces, such as dominance or competence, was also impaired in PwPD (Hirai et al., 2019).

Still other studies have shown that many PwPD, even those who demonstrate other types of emotional processing impairments, display intact face processing skills on a wide range of tasks (Adolphs et al., 1998; Borod et al., 1990; Pell & Leonard, 2005). Different suggestions have been made to explain these discrepancies across studies (Argaud et al., 2018; Gray & Tickle-Degnen, 2010; Pell & Monetta, 2008). For example, it has been proposed that face processing deficits are heterogeneous but more pervasive in advanced stages of PD as visual-perceptual deficits become more prevalent, affecting how socioemotional attributes of faces are understood (Dewick et al., 1991; Hipp et al., 2014). The presence of executive dysfunction in PwPD (notably, attention abilities, verbal and visuospatial working memory and verbal fluency) seems to influence the magnitude of emotional face processing impairments, although deficits have also been observed in cognitively intact patients (Pietschnig et al., 2016; Argaud et al., 2018; Alonso-Recio et al., 2014; Assogna et al., 2010). Hypomimia (facial masking) has also been put forward as a factor to explain impairments in face recognition when perceptual abilities are relatively spared (Marneweck et al., 2014). According to the Embodied Simulation Theory, hypomimia could disturb how emotions are recognized by hampering the ability to covertly mimic (i.e. simulate) the expressions of interaction partners (Lotze et al., 2008; Gray & Tickle-Degnen, 2010). This explanation bears similarities to why many PwPD are impaired in the processing of action and emotion words that draw upon embodied semantic information (Dreyer et al., 2015).

In face processing tasks, the nature of stimuli presented (e.g. intensity of expression, dynamic vs. static expressions, unimodal vs. multimodal expressions) also seems to have an impact on social perception skills in PwPD. To understand how the intensity of facial expressions influence emotion recognition, one study manipulated photographs to produce increasingly more subtle expressions of anger, disgust, fear, happiness, sadness and surprise (Buxton et al., 2013). It was found that

PwPD could accurately identify prototypical emotional displays but were impaired at recognizing more subtle social expressions. Other work suggests that PwPD demonstrate greater accuracy on facial emotion recognition tasks when presented dynamic vs. static expressions, especially when emotional cues are encountered in ecologically-enriched contexts such as natural social scenes (Pell et al., 2014; Schwartz & Pell, 2017). Thus, while PwPD still seem to experience difficulties processing information from dynamic facial stimuli (Garrido-Vásquez et al., 2016), it is possible that social perception in PwPD is facilitated when redundant emotional cues are available in more than one communication channel (e.g. facial cues combined with vocal and linguistic cues; Paulmann & Pell, 2010). Additional research that considers how PwPD use socioemotional information from faces in dynamic, socially-enriched processing environments that resemble daily living is needed to elaborate on these ideas.

15.4.1.2 Vocal Expressions

Like faces, information conveyed by tone of voice (speech prosody) reveals a speaker's identity, their emotions, and a host of other interpersonal features. However, unlike faces, meanings in the voice cannot be captured in a static image. They are specified over time by modulating the pitch, loudness, vocal quality, and rhythmic properties of an utterance (Pell & Kotz, 2011).

Reduced physiological support for respiration, phonation, and articulation produce a characteristic dysarthria in PwPD, restricting the ability to produce articulatory movements underlying speech segments in linguistic communication. Beyond individual speech sounds, when PwPD talk they display reduced loudness of speech and a monotonous voice that lacks typical pitch variation ('monopitch'; Goberman & Coelho, 2002). PwPD are less effective in communicating suprasegmental contrasts in speech that are expressed at the syllable or utterance level, obscuring pragmatically rich meanings such as emphatic stress and vocal emotion contrasts (Cheang & Pell, 2007). Research shows that PwPD have difficulties expressing emotional prosody spontaneously or when imitating a speaker model (Schröder et al., 2010) and their attempts to communicate emotional contrasts and emphatic stress in speech are often misinterpreted by listeners (Pell et al., 2006). Difficulties in vocal expression in PwPD can be largely attributed to motor symptoms associated with akinesia and rigidity in phonation, articulation and respiration (Péron et al., 2012). In addition to motor disturbance, PwPD may have diminished access to limbic information for the modulation of speech, interfering with their ability to successfully convey emotions in the vocal channel (Arnold et al., 2014; Möbes et al., 2008).

The social perception of voice information is also disturbed in many individuals with PD (Coundouris et al., 2019). Evaluation of socioemotional features of the voice involves procedures for acoustically structuring the auditory input, detecting the socioemotional significance of the cues, and like other nonverbal cues, analyzing this information in relation to what a person is simultaneously saying. Research shows that PwPD fail to identify the meaning of utterances spoken with different emotional inflections (Pell, 1996; Pell & Leonard, 2003; Péron et al., 2012; Schröder et al., 2010) and to use vocal cues to infer a speaker's mental state, such as how confident they are in a statement (Monetta, Cheang, & Pell, 2008a). In these tasks, PwPD often appear to have greater difficulties recognizing negative emotions, particularly disgust and fear (Dara et al., 2008; Paulmann et al., 2008). Many studies have linked difficulties in the processing of vocal emotion expressions to concurrent cognitive dysfunctions (Breitenstein et al., 2001; Gray & Tickle-Degnen, 2010; Rektorova et al., 2016) and to left-sided dominant motor symptoms in PwPD, i.e. right hemispheric brain dysfunction (Garrido-Vásquez et al., 2013; Stirnimann et al., 2018). In addition, the presence of emotion-specific impairments in PwPD may be dictated by brain dopamine levels, depending on the stage of the disease and a patient's medication status (Buxton et al., 2013).

As was true for facial expressions, the precise way that voice perception breaks down in PwPD is still not well understood. Some authors argue that discrimination of acoustic features in PwPD is spared and that deficits arise due to impaired mapping of acoustic features onto emotional or attitudinal representations during social perception (Paulmann et al., 2011; Pell, 1996; Scott et al., 1984). Other researchers suggest that auditory perceptual abilities, such as the ability to detect changes in vocal parameters such as speech rate, are disturbed by the disease (Breitenstein et al., 2001; Troche et al., 2012). Pell and Leonard (2003) hypothesized that basal ganglia dysfunction in PD disturbs basic timing operations that underlie vocal expression processing, hampering the ability to derive meaning from sequential properties of sensory events and to respond to meaningful changes in these stimuli. Along these lines, Jaywant et al. (2016) proposed that disconnections between the basal ganglia (caudate) and the superior temporal cortex progressively reduce the sensitivity of PwPD to socially-relevant meanings conveyed by biological motion. According to this idea, difficulty extracting (spatio)temporal properties of human motion, including dynamic voices and visual stimuli, is a major determinant of social perception deficits in PwPD. Such deficits could be exacerbated in individuals with reductions in cognitive resource capacity, such as low working memory, due to further limitations in the ability to hold sequential cues in memory (Pell & Leonard, 2003).

15.4.2 Perspective-Taking/Theory of Mind

Adopting a theory of mind is often essential to correctly interpreting socioemotional cues and to decoding pragmatically complex messages. Theory of mind (ToM), mentalizing, and perspective-taking are all terms that refer to how people attribute mental states—intentions, beliefs, affect or knowledge—to oneself and others (Frith & Frith, 2003; Premack & Woodruff, 1978). A distinction can be made between affective ToM, which is a more automatic process for representing another person's emotions and feelings, and cognitive ToM, which is a more controlled process for representing another person's beliefs and intentions (Poletti et al., 2012). While not all agree on the psychological mechanisms that allow people to form representations of another person's mental and affective states (Apperly, 2008; Bodden et al., 2010; Freedman & Stuss, 2011), it is broadly agreed that ToM is a critical skill in human social cognition (Brüne & Brüne-Cohrs, 2006; Carlson et al., 2013; Hughes & Leekam, 2004) and contributes in a substantive manner to social-pragmatic competence (Schnell et al., 2016; Wang et al., 2006).

Neuroimaging studies reveal that many of the brain regions activated when people make mental state attributions (e.g. medial prefrontal cortex, anterior cingulate, temporal parietal junction; Mahy et al., 2014; Luyten & Fonagy, 2015) are susceptible to disruption in PD. While both affective and cognitive ToM rely on prefrontal projections to the basal ganglia, affective ToM predominantly engages the orbital frontal-striatal (ventral) system which prepares limbic and emotional information for behavioral responses. In contrast, cognitive ToM involves the dorsolateral fronto-striatal circuits which help to sequentially organize sensory and motivational information (Bonelli & Cummings, 2007). In terms of the relationship between ToM and social perception, implicit ToM tasks, such as when people make attributions about another person based on their external features (face or vocal expression), are more strongly associated with affective ToM. Additional brain regions are likely involved in more controlled (cognitive) ToM tasks, in which mentalizing draws upon external cues as well as internal representations of the self and others (Luyten & Fonagy, 2015). This emphasizes that different tasks used to evaluate ToM in PwPD rely on other cognitive processes to varying degrees and need to be taken into account (see Poletti et al., 2011). Nonetheless, given the established role of the basal ganglia/frontostriatal circuitry in mentalizing, social perception, and executive functions, and the co-dependence of these functions during real-life communication, it is not surprising that evidence for ToM deficits in PwPD is rapidly accumulating (Bodden et al., 2010; Costa et al., 2013; Freedman & Stuss, 2011; Monetta et al., 2009; Poletti et al., 2012; Saltzman et al., 2000; Yu & Wu, 2013; Bora et al., 2015).

Research shows that ToM difficulties tend to worsen at later disease stages of PD (Bora et al., 2015; Yu & Wu, 2013) and often co-occur with executive processing deficits. For example, Fabbri et al. (2018) looked at how PwPD used ToM to perform individual actions and to coordinate behaviour with others to achieve a common goal. They found that only patients with more marked cognitive ToM deficits showed impairments in a joint action task, whereas the influence of affective ToM deficits on performance was less evident. In another study, Costa et al. (2013) investigated the effects of executive dysfunction on ToM using a custom task requiring PwPD to evaluate socially inappropriate responses in short conversational vignettes (similar to the faux-pas test). They found impairments in ToM only in PwPD who presented executive dysfunctions in relation to a healthy control group, pointing to the likelihood that perspective-taking abilities in PwPD are predicted, at least in part, by their executive resource capacity (Bora et al., 2015; Monetta et al., 2009; Pell et al., 2014). Along these lines, the current literature suggests that PwPD experience more pronounced difficulties for cognitive versus affective ToM (Bora et al., 2015; Poletti et al., 2011; Schwartz & Pell, 2017), and tend to show impairments for cognitive ToM at earlier stages than affective ToM (Poletti et al., 2011; Yu & Wu, 2013). Further research on this topic is clearly needed. It should increasingly use tasks that capture pragmatically-challenging situations encountered in real life, using naturalistic scenarios in which a failure to correctly adopt the speaker's perspective could lead to adverse social consequences (Bora et al., 2015; Rothermich & Pell, 2015).

15.4.3 Conversational Interactions

The ability to engage in discourse, while seemingly routine, involves multiple, interacting processes whose use is governed by pragmatic knowledge (Johansson et al., 2019; Roberts & Post, 2018). Conversation is the most typical discourse context in daily life. It involves discourse planning, topic initiation, and the selection, sequencing and tailoring of relevant information concepts that correspond to the message or other properties of the interactional/discourse frame. Nonverbal cues play an important role in how people process the linguistic message, attribute mental and affective states to their conversational partner, and understand their social intentions.

It seems likely that changes in non-verbal expressivity—motor-related reductions in facial movement (masking), blinking, smiling, and vocal expression (Pitcairn et al., 1990)—contribute in a major way to impressions of reduced informativeness and (impaired) pragmatic competence in PwPD during spontaneous language production. PwPD have reduced emotional behavioral outputs (e.g. less expressive facial expressions and reduced abilities to express emotional prosody) and these changes tend to have negative or unintended consequences which affect their participation in social settings (Jaywant & Pell, 2010). It has been noted that PwPD participate less in conversation overall (Johansson et al., 2019), show reduced conversational initiation and turn-taking (Hall et al., 2011), and tend to produce fewer, shorter utterances (Murray, 2000). Their spontaneous speech may contain fewer correct information units than healthy adults of similar age and education (Bayram et al., 2019; Roberts & Post, 2018; Murray, 2000; see also Cummings et al., 1988). Moreover, PwPD often demonstrate an increased duration of pauses in their speech, particularly as the motor severity of PD increases, disrupting the overall flow of speech (Alvar et al., 2019; Ash et al., 2012; Illes, 1989; Smith & Caplan, 2018).

The (often mistaken) impression that PwPD are less engaged and interested in the social context, the lack of typical interactive behaviors (e.g. facial mimicry; Kang et al., 2019), and a reduction in filled pauses (Alvar et al., 2019) are all factors that reduce the overall naturalness of speech. These alterations simultaneously promote a variety of negative social impressions of PwPD and reduce the desire of others to interact with them as competent social partners (Jaywant & Pell, 2010; Hemmesch et al., 2009; Hemmesch, 2014; Schwartz & Pell, 2017; Tickle-Degnen et al., 2011). According to some research, negative impressions could have an even greater impact on women living with PD due to sex-related stereotypes of how women and men typically express themselves socially (Tickle-Degnen et al., 2011; Wang & Tickle-Degnen, 2018). This literature underscores a number of variables that should be carefully considered when assessing the social-pragmatic capacity of PwPD based on their spontaneous language production.

An intriguing but relatively unexplored feature of conversational behaviour that may be associated with PD is a reduction in the use of formulaic language. Expressions such as idioms ("I went out on a limb for you"), swear words ("dammit") and a broad range of conversational speech formulas ("Really!," "No way!," "We'll see!") are considered "fixed" expressions in a language and are omnipresent in conversational interactions. Interestingly, these formulaic expressions, which consist of specific words produced in a certain order and with a stereotyped intonation, are primarily used to convey non-literal and often affective meanings (Van Lancker Sidtis & Sidtis, 2018a, 2018b). Current neurolinguistic models propose that speech formulas are represented as holistic expressions and rely on a basal ganglia-right hemisphere network during speech production (in contrast to cortically-based, linguistic functions underlying generative language). Subcortical brain areas are involved in formulaic language because associated motor and verbal gestures are highly stereotyped, overlearned, and affectively-laden (Van Lancker Sidtis & Sidtis, 2018a, 2018b).

Research by Van Lancker Sidtis and colleagues reveals that conventionalized phrases and routines, which can serve as the 'glue' for spoken interactions, are produced less frequently by PwPD. Compared to healthy individuals, PwPD display a lower frequency of formulaic expressions in their spontaneous speech, while retaining the ability to understand formulaic impressions (Van Lancker Sidtis et al., 2015). This contrasts with the pattern of difficulties observed in other neurodegenerative populations such as Alzheimer's disease, where impaired cortical but intact basal ganglia functioning yield an increased production of formulaic expressions with decreased comprehension performance (Van Lancker Sidtis et al., 2015). Further research which looks more closely at the notion of formulaic or 'automatic' speech and how it is affected by PD represents a promising direction for advancing knowledge of pragmatic communication and conversational abilities in PwPD.

15.4.4 Summary of PD and Social Cognition

Intact social cognition is an essential element of social functioning and quality of life (Hasson-Ohayon et al., 2017). Research on PD clearly demonstrates that the ability to detect and assign meaning to nonverbal cues that have interpersonal significance during communication (social perception) is impaired, as is the ability to attribute mental states to others. These impairments can be partly explained by a reduction in executive control functions, which tend to covary with decreased performance in social cognition tasks. However, as difficulties in emotion perception and ToM are also observed in the absence of cognitive dysfunction (Coundouris et al., 2019; Fine et al., 2001), PD can have a direct impact on social cognitive functions. New studies suggest that difficulties in social perception can be partially predicted by the lateralization of PD motor symptom onset, with greater deficits in individuals with predominantly left-sided symptoms (i.e. right-hemisphere dysfunction; Coundouris et al., 2019). Denervation of circuits projecting from the basal ganglia to the right cerebral hemisphere may lead to more pronounced social perception deficits due to the privileged involvement of right hemisphere brain regions in processes underlying social cognition, such as emotion processing (Pell, 2006). At present, it may be concluded that social perception deficits manifest to a relatively similar extent when meaning is conveyed by facial or vocal expressions (Coundouris et al., 2019), although the cause of difficulties in each channel may be different.

A better understanding of how PD affects processes underlying social cognition should help to pinpoint the source of negative social outcomes that are frequently described by individuals living with the disease. For example, it appears that differences in how PwPD express themselves (i.e. alterations in vocal and facial expressions) negatively affect their social opportunities and outcomes by promoting false impressions of what they mean and how they feel (McAuliffe et al., 2017; Miller et al., 2008; Tickle-Degnen et al., 2010; Jaywant & Pell, 2010; Schwartz & Pell, 2017). When conversational partners misattribute the cause of expressive abnormalities in PwPD to negative mental states or attitudes (e.g. that they are bored, angry, disinterested in the conversation), they are likely to behave differently towards PwPD and/or to avoid further social contact, fundamentally altering how the social context evolves. Similarly, if PwPD fail to appropriately evaluate socio-emotional cues and to take the perspectives of their conversational partner into account, this is likely to promote negative inferences and influence the types of social decisions that are made by those participating in the conversation (Lee & Harris, 2013). As research continues, raising awareness of how social cognition is altered in PD, and how these changes can lead to various misunderstandings, could be a starting point to facilitate healthier, more supportive interactions with PwPD.

15.5 Remediation of Pragmatic Communication Deficits in Parkinson's Disease

In light of evidence demonstrating the impact of PD on verbal communication and social cognition, what types of interventions are available to remediate pragmatic deficits in PwPD? Clinical and experimental treatments for PD with potential effects on social-pragmatic skills include dopamine pharmacological treatments, subthalamic nucleus deep brain stimulation (STN-DBS), non-invasive brain stimulation, as well as behavioral remediation approaches (cognitive remediation/speech-language pathology).

To date, the goal of most pharmacological/surgical treatments has been to improve major motor symptoms brought on by PD (e.g. tremor, rigidity), which have the potential to improve areas of pragmatic performance that rest on intact motor functioning (e.g. mitigating negative social impressions linked to reduced non-verbal expressivity). Unfortunately, while these treatments are often successful in alleviating cardinal signs of motor disability in PwPD, they do not always improve motor-related difficulties in speech and voice (Brabenec et al., 2017) and can have negative repercussions on (social) cognition (Auclair-Ouellet et al., 2011; Combs et al., 2015; Cools et al., 2001, 2006). For example, due to the so-called "overdose effect," pharmacological approaches with dopaminergic therapy in the early stages of PD may be toxic for socioemotional functions subserved by the mesocorticolimbic pathways which are still relatively spared (Argaud et al., 2018; Péron et al., 2014). These issues are discussed in more detail below, and new directions for behavioral interventions are considered.

15.5.1 Dopaminergic Therapy

Dopaminergic antiparkinsonian medications (levodopa or L-Dopa), which redress the dopamine imbalance within the basal ganglia, have long been administered to ameliorate PD motor symptoms. These medications seem to have positive effects on certain aspects of language performance. Of note, there is an improvement in semantic processing when motor imagery is involved (e.g. processing of action words), which correlates with motor improvement in the on-medication condition (Péran et al., 2013; Roberts et al., 2017). These improvements can be attributed to increased motor functions impacting on systems for perception, action and introspection during semantic processing (Dreyer et al., 2015; Fernandino et al., 2013).

Dopaminergic agents to control motor symptoms inevitably affect patients' cognitive functioning. A systematic review of the effects of dopaminergic therapy on cognition in PD revealed beneficial chronic effects of levodopa on memory encoding and retrieval, mental flexibility, planning and verbal fluency, but deleterious effects on inhibitory control (Roy et al., 2018). According to the L-Dopa overdose hypothesis (Gotham et al., 1986), restoring dopamine within the basal ganglia can have a positive influence on certain cognitive functions when dysfunctional projections to the cortex are re-established (e.g. within the dorsolateral prefrontal cortex), while overdosing intact circuits, such as the orbitofrontal loop, and altering their functions (Gotham et al., 1988; Roy et al., 2018). This idea fits with current evidence that optimally medicated PwPD exhibit moderate deficits in social perception of facial and vocal cues, whereas patients in a hypodopaminergic state (i.e. off or withdrawn from medication) are relatively unimpaired (Coundouris et al., 2019). The extent to which PwPD display an emotional bias in processing negative facial and vocal stimuli has also been linked to dopaminergic therapy and stage of the disease (Lundqvist et al., 2017; Pell & Monetta, 2008; Péron et al., 2014). Similarly, difficulties in the processing of temporal information during communication seems to vary according to the level of dopaminergic medication (Jones et al., 2008).

There are still many unanswered questions about how dopaminergic therapy influences pragmatic language abilities in PD. Medication-related changes in pragmatic performance may be secondary to cognitive changes induced by dopaminergic therapy, may be brought on by the "overdose" of dopamine in certain brain circuits that are otherwise functionally intact (e.g. those involved in social perception), or may be absent. Researchers point out that there is significant variability in the progression of dopamine loss in striatal, limbic, and cortical regions among PwPD (Titova et al., 2017) and, therefore, significant variability in the effects of levodopa on brain structures that support (primary or secondary) functions involved in social communication and contextualized language processing among individuals with PD. Further investigations into this topic are clearly warranted.

15.5.2 Deep Brain Stimulation

Deep Brain Stimulation (DBS) is a surgical procedure in which an electrode is implanted in the brain to stimulate regions involved in movement, such as the subthalamic nucleus (STN). It is an increasingly utilized therapeutic option for PwPD. The beneficial effects of STN-DBS on motor symptoms are well established, but its effects on cognition, high-level language abilities and emotion processing are still controversial. In one study, Tremblay, Macoir, et al. (2015a) compared metaphor comprehension 'on' and 'off' STN-DBS. Results indicated that electrical stimulation did not have any effect on metaphor comprehension or any other language functions evaluated in this study (i.e. verbal fluency, lexical functions and semantic processing). Other studies have failed to detect changes in facial or vocal emotion recognition after STN-DBS in PwPD (Aiello et al., 2014; Albuquerque et al., 2014; Berney et al., 2007; McIntosh et al., 2015). By contrast, other studies have reported either reduced executive abilities (e.g. verbal fluency and inhibition) or an improvement in executive functions (e.g. working memory and psychomotor speed) under stimulation in PwPD (see Halpern et al. (2009) or Martínez-Martínez et al. (2017) for a review on the subject). Given the relationship between executive brain functions and communication skills, the effect of possible changes in cognition on pragmatic functions after stimulation need to be carefully considered (Brück et al., 2011).

Some research suggests that emotional processing deficits and cognitive dysfunctions fluctuate across time after the DBS implant. The surgical procedure was found to influence patients' cognitive functions and emotional processing in the first months after the surgery, followed by a return of the impairment profile within the pre-DBS range one year later (Aiello et al., 2014; Auclair-Ouellet et al., 2011; Zangaglia et al., 2009). In addition to transient effects of DBS surgery, studies comparing the 'on' and 'off' stimulation states indicate that there was no reduction of executive or pragmatic language abilities, leading to the conclusion that deficits may be secondary to other causes than electrical stimulation, such as the microlesion induced by the surgical procedure (Aiello et al., 2014; Halpern et al., 2009; Tremblay, Macoir, et al., 2015a). It is worth noting that some studies have reported detrimental effects of DBS on language, including impairment in grammatical processing (Phillips et al., 2012). Other studies have reported a specific reduction in facial recognition of fear (Le Jeune et al., 2008) and a cognitive bias towards negative vocal emotions (Péron et al., 2010), which might alter similar mechanisms that are affected by dopaminergic medication. Further studies examining the effects of DBS surgery on communication skills in PwPD will be needed to fully understand its combined effects on cognitive, linguistic, and affective functions.

15.5.3 Non-invasive brain Stimulation

There is a growing literature on non-invasive brain stimulation, such as direct, alternating, or intermittent transcranial magnetic stimulation, as a new treatment option for PwPD (see Chen and Chen (2019) for a recent review). Non-invasive brain stimulation activates cortical neurons by inducing electrical current generated through a rapidly changing magnetic field to facilitate or inhibit the synaptic connections, leading to a change in cortical excitability. For the treatment of PD non-motor symptoms, most studies using non-invasive brain stimulation have focused on the treatment of PD-related depression or cognitive dysfunction (Chen & Chen, 2019). Much less attention has been devoted to the treatment of pragmatic deficits. In a notable exception, Tremblay, Monetta, et al. (2015b) ran a double-blind sham-controlled case study of a PwPD. For the first time, it was shown that intermittent transcranial brain stimulation of the right dorsolateral prefrontal cortex improved metaphor comprehension in association with an increase of the right hemisphere motor excitability (Tremblay, Monetta, et al., 2015b). While these findings remain tentative until larger participant samples are studied, this work motivates new research that attempts to stimulate brain regions that contribute to brain networks underlying social cognition and pragmatic language functions.

15.5.4 Behavioural Remediation of Pragmatic Deficits

The behavioral remediation of pragmatic deficits in PwPD is still in its infancy. One behavioural approach which could have beneficial impacts on pragmatic communication in PwPD is for the treatment of hypophonia (weak voice). For example, the Lee Silverman Voice Treatment (Ramig et al., 2001) has proven its efficacy as an intensive treatment approach which targets speech functions by increasing vocal loudness and the amplitude of motor output, while focusing on self-monitoring of vocal functions. This treatment could be used to improve specific pragmatic abilities by enhancing expressiveness in behavioural output, such as increasing facial expressivity (Spielman et al., 2003) and reducing monotonicity (Ramig et al., 1995). Improving the expressivity and naturalness of their speech in social interactions could help to eliminate some of the negative impressions made about PwPD and lead to more successful and rewarding social outcomes for those living with the disease.

Given that many pragmatic difficulties experienced by PwPD are due to underlying changes in cognition and/or executive functioning, another logical approach to remediation is to focus on the cognitive origins of pragmatic symptomatology, by improving executive functions that support high-level language usage. In recent years, researchers have shown that cognitive rehabilitation—defined as non-invasive treatment for cognitive impairment based on theoretical models of restoration, compensation, and optimization of the cognitive functions that target particular cognitive skills—helps to mitigate cognitive deficits in PwPD and benefits patients in their daily lives (see Biundo et al., 2017; Díez-Cirarda et al., 2018). To date, cognitive rehabilitation programs have focused on attention (Cerasa et al., 2014), inhibition (Nombela et al., 2011), and in training multiple executive functions simultaneously (Foster & Hershey, 2011; Díez-Cirarda et al., 2018). Research has also shown that during the remediation/compensation of cognitive deficits, the caregiver could play an important role in applying strategies which ensure a successful behavioural intervention (Habermann & Davis, 2005).

Despite evidence that therapies targeting executive functions are often effective, their impact on pragmatic communication in PwPD has not yet been addressed. This constitutes an obvious area for future work, as the overlapping relationship between executive functions, communication, and social cognition and their shared neurocircuitry would predict beneficial effects in all these areas. Another issue to be studied is the optimal intensity of treatment. In a randomized control study, Tickle-Degnen et al. (2010) demonstrated that increasing the number of hours of self-management rehabilitation enhances quality of life (including social communication) in PwPD. In that study, PwPD took part in group discussions and other daily life activities to communicate and exchange ideas on where they could apply the learned strategies. Results demonstrated that participants who had been trained more hours focusing on self-management and social communication showed greater benefits in pragmatic ability. Further studies are, therefore, warranted to determine the optimal approach and optimal times of exposure to treatment during cognitive rehabilitation to observe positive effects on pragmatic functioning in people living with PD.

15.6 Summary

This chapter highlights ways that idiopathic Parkinson's disease affects verbal communication and aspects of social cognition that support language usage and effective interpersonal behaviour. Our analysis underscores that PD is defined not only by a decline in motor and cognitive functions but important difficulties that affect social cognition and communication. Communication difficulties in PwPD are often heterogeneous and can arise due to motor problems (e.g. slurring of speech) or as a secondary consequence of cognitive alterations (e.g. impaired executive functions). In addition, social-pragmatic deficits can emerge as the direct result of dopamine-related changes that impact negatively on regions of the 'social brain,' particularly regions in the prefrontal cortex, which allow people to successfully inter-relate and communicate in everyday life. An important area of difficulty for people with PD is to understand different types of indirect meanings and to generate social inferences based on verbal, nonverbal, and contextual cues. Adults with PD also have marked deficits in the perception of socioemotional cues from facial and vocal expressions and are less able to use these and other cues to infer another person's mental state. Although still in their infancy, our discussion touches upon therapeutic approaches which could be used to mitigate the effects of pragmatic impairments and allow individuals with PD to participate more fully in the social environment, promoting greater independence and well-being.

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Chapter 16
Multiple Sclerosis

Antonio Carotenuto, Rosa Iodice, and Giorgio Arcara

16.1 Introduction

Multiple sclerosis (MS) is a chronic inflammatory disease of the central nervous system (CNS). It is characterized by the accumulation of demyelinating plaques throughout the brain and spinal cord and axonal loss, resulting in progressive disability in most affected individuals. Specifically, MS patients will progressively develop both physical and cognitive disability, which might include deficits in language and communication. This chapter will summarize complex clinical aspects of MS, including epidemiology, diagnosis, disease course, and prognosis, and will focus on recent evidence and advances in our understanding clinical symptoms underlying the overall disability caused by the disease. Among MS-related symptoms, cognitive impairment is becoming a fundamental element. It reflects pathological changes throughout the CNS and allows us to forecast disease evolution and prognosis. Pragmatic impairment can complicate the clinical picture and negatively impacts activities of daily life but is still little investigated. In this chapter, we focus our attention and research on pragmatic abilities in MS.

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16.2 Epidemiology, Aetiology and Pathophysiology

The incidence and prevalence of MS are increasing over time in both developed and developing countries (Browne et al., 2014). Unfortunately, the underlying cause is still to be determined. Based on the most recent data, the prevalence of the disorder is 33 per 100,000 people, with a high level of variability from country to country. The highest prevalence is in North America and Europe (140 and 108 per 100,000 people, respectively), whereas the lowest prevalence belongs to Asia and sub-Saharan Africa countries (2.2 and 2.1 per 100,000 people, respectively) (Belbasis et al., 2015). Moreover, within continents, there is significant regional variation. In Hong Kong, the prevalence is 0.77 per 100,000, whereas in Iran the prevalence is 85.80 per 100,000 (Eskandarieh et al., 2016).

Prevalence data fueled the hypothesis that some environmental factors might play a key role in the pathogenesis of MS. For example, it is widely accepted that prevalence is directly associated with latitude. MS prevalence increases with latitude and areas with a temperate climate far from the equator have a higher prevalence compared to countries closer to equator (Ascherio & Munger, 2007a). Moreover, the incidence of MS decreases with migration from high to low latitude areas, and vice versa, when occurring during childhood and adolescence (Beretich & Beretich, 2009). Probably, people living closer to the equator are more exposed to sunlight that promotes the production of vitamin D, levels of which are closely linked to disease severity (Gasperoni et al., 2019).

Some other modifiable and non-modifiable demographic features are associated with the pathogenesis of MS over time. MS is more common in females than in males, with a female-to-male sex ratio of 3:1 in most developed countries (Orton et al., 2006). In addition, considering that dizygotic twins have a higher risk of MS compared to siblings, intrauterine environment might impact on the future likelihood of developing MS. Nevertheless, it is unclear whether this is due to common environmental exposures or epigenetic mechanisms or if these factors are playing a joint role (Ramagopalan et al., 2010).

Environmental, genetic, and epigenetic factors have a causal role in MS and potentially interact with modifiable risk factors (Olsson et al., 2017). Diet, obesity in early life, and cigarette smoking are associated with MS and worsen the disease course over time (Marrie, 2004). In addition, exposure to infectious agents during the life course also affects the risk of developing conditions involving the immune system such as MS. For example, Epstein-Barr virus exposure acts as a potential causative trigger for MS (Ascherio & Munger, 2007b). Despite encouraging epidemiological studies and plausible hypotheses, a direct pathological role for Epstein-Barr virus and other infectious diseases in MS is yet to be confirmed.

Genetic factors also have a putative role in the development of MS. For monozygotic twins the concordance rate of MS is approximately 18% compared to 3% in siblings (Willer et al., 2003). While highly appealing, a single causative gene for MS has never been reported and MS might be more likely to have a polygenic predisposition. In line with this model, the major histocompatibility complex region confers approximately 20% of the genetic susceptibility to MS, with the HLA-DRB1*15:01 allele accounting for the greatest risk (International Multiple Sclerosis Genetics Consortium et al., 2011; Patsopoulos et al., 2013). On the other side, HLA-A*02:01 appears to be protective.

Moving further, genome-wide association studies have identified more than 100 single nucleotide polymorphisms associated with an increased risk of MS. Most of these single nucleotide polymorphisms are somehow related with immune functions (International Multiple Sclerosis Genetics Consortium et al., 2013). Specifically, genetic studies underline a dysfunction of the adaptive immune system, which consists of T and B lymphocytic cells, leading to the development of MS (Gay et al., 1997). In MS the auto-immune process targets specific CNS antigens (the so-called 'autoantigens'), recruiting T and B cells, which migrate from the blood to the CNS. Having penetrated the CNS through the blood-brain barrier, activated T cells undergo clonal expansion, secrete pro-inflammatory cytokines, stimulate microglia and astrocytes, and propagate a cascade of inflammatory events, ultimately resulting in the destruction of myelin (demyelination), oligodendrocyte loss and axonal damage (Patani et al., 2007; Patrikios et al., 2006).

Demyelination is the pathological hallmark of MS. It is associated with axonal loss, gliosis and, sometimes, remyelination. Pathological studies reported that demyelinating lesions display considerable heterogeneity depending on the age and activity of the plaque. Acute lesions are characterized by a high number of inflammatory cells infiltrating the lesions, an indistinct margin edema, demyelination and, to a lesser extent, axonal damage. Chronic, inactive plaques show more pronounced axonal loss with a lack of inflammation and, sometimes, an ongoing remyelinative process (Patrikios et al., 2006). Revealing the mechanisms leading to axonal loss is extremely intriguing since it might lead to novel therapies aimed at halting disease progression. Amid different putative biological processes, neuronal energy deficit linked to mitochondrial dysfunction, the loss of myelin trophic support, and cytoskeletal disorganization of chronically demyelinated axons are correlated with permanent axonal loss (Ciccarelli et al., 2014). Axonal or neuronal loss (referred to as neurodegeneration) is particularly relevant because it is the main underlying mechanism of permanent physical and cognitive disability in MS.

16.3 Clinical Features and Course of MS

Early stages of MS are usually characterized by acute episodes of neurological deficits, known as relapses. Common deficits include visual disturbance, motor weakness, sensory changes, incoordination, gait disturbance and bladder dysfunction. Disabling fatigue is common and can be difficult to manage. Physical disability in MS is primarily measured by the Expanded Disability Status Scale (EDSS) (Kurtzke, 1983). The EDSS allows clinicians to assess neurological disability in each of eight functional systems (pyramidal, sensory, cerebellar, visual, bladder/bowel, brainstem, cerebral/mental and 'other'). EDSS also provides information about a patient's ambulation ability. An EDSS score of 6.0 or higher indicates that the patient requires at least unilateral support for mobility.

Cognition is also impaired in about 50–70% of MS patients, with direct negative consequences on everyday life (Chiaravalloti & DeLuca, 2008). Processing speed, attention, and working and episodic memory are the major cognitive domains affected by MS (Chiaravalloti & DeLuca, 2008). Diffusion MRI studies in MS patients show reduced fiber tract fractional anisotropy in white matter networks connecting several cortical areas (Dineen et al., 2009). Importantly, these abnormalities were also found in normal appearing white matter of MS patients, underlining the contribution of pathophysiological mechanisms beyond clinically manifest demyelination. Therefore, the disconnection between different cortical regions throughout the brain caused by demyelinating lesions over the fiber bundles contributes to cognitive impairment.

Apart from this subcortical disconnection syndrome, gray matter abnormalities are increasingly detected in imaging of MS patients and are likely to contribute to cognitive dysfunctions (Rocca et al., 2015). Old age and being male are two factors that appear to be related to greater cognitive impairment (Chiaravalloti & DeLuca, 2008). Cognitive deficits are also found nearly twice as often in patients with progressive forms of MS (PPMS and SPMS) than in RRMS (Ruano et al., 2017). A meta-analysis indicated that MS patients not only experience quantitative but also qualitative differences in cognitive profile compared with healthy controls, with a higher proportion and magnitude of executive deficits in progressive MS (Zakzanis, 2000).

The clinical course in MS is extremely variable. Most patients diagnosed with MS undergo a relapsing-remitting (RR) course in which patients experience attacks of neurological dysfunction with partial or complete recovery. Usually, within 20 years from diagnosis, the majority of patients diagnosed with RRMS will develop a secondary progressive disease course (SPMS) (Scalfari et al., 2010). This phenotype is characterized by an insidious accrual of physical disability, independent of the relapses. About 15% of patients develop a progressive course without experiencing relapses from the onset of the disorder. This is primary progressive MS (PPMS) (Scalfari et al., 2010).

The diagnosis of MS is based on the integration of clinical, imaging, and laboratory findings. To fulfill diagnostic criteria, clinical expertise is necessary to demonstrate objective evidence of dissemination in time and space of lesions typical of MS (Thompson et al., 2018) and, importantly, to exclude other neurological conditions, such as vascular pathology, infectious disorders of the CNS, and other autoimmune conditions, particularly acute diffuse encephalomyelitis (Brownlee et al., 2015).

Over the last decade, there has been a significant increase in the development and approval of novel disease-modifying treatments for RRMS patients. In general, treatments target neuroinflammation and could have an indirect effect on neurodegeneration. However, their efficacy for slowing down disability accrual and reducing the development of brain atrophy in clinical trials has been moderate at best and, up to now, only one disease-modifying treatment (ocrelizumab) has been shown to be effective in patients with PPMS (Montalban et al., 2017). Although disease-modifying treatments prevent the occurrence of relapses, they do not treat symptoms associated with MS, such as spasticity, bowel-bladder dysfunction, cognitive impairment, and vision deficits. The large range of treatments available, while welcome, also makes determining treatment plans more complex. To guide the pharmacological treatment of MS, guidelines have been developed by the European Committee for Treatment and Research in Multiple Sclerosis and the European Academy of Neurology (Montalban et al., 2018).

16.4 Pragmatic Abilities in Multiple Sclerosis

MS symptoms also sometimes include language impairment (Renauld et al., 2016). Most studies of language abilities in MS have assessed language functions indirectly, through confrontation naming and verbal fluency tests embedded in neuropsychological batteries (Murdoch et al., 2003), or through tests that evaluate basic aspects of language such as the Token Test (Luzzatti et al., 1991; Friend et al., 1999). However, MS patients rarely exhibit impairments in confrontation naming and verbal fluency and full-blown aphasia is documented only in sporadic cases (Achiron et al., 1992). Also, naming and fluency involve cognitive processes such as executive functioning (Kennedy & Murdoch, 1989). Some studies have tested specific linguistic abilities such as auditory comprehension, reading and comprehension of phrases (Friend et al., 1999). They have revealed a wide range of communicative impairment associated with MS. All these language tasks (naming, fluency, auditory comprehension, reading and comprehension of phrases) rely primarily on lexical knowledge, semantics, and the syntax of sentences.

There are other aspects of language that go beyond core linguistic functions like naming. Context-dependent aspects of language include the ability to produce discourse, engage in conversation, and understand the non-literal meaning of language (Bambini, 2010; Bambini & Bara, 2012). These high-level language abilities require cognitive and pragmatic skills. In MS, comprehension of ambiguous sentences and metaphors, and narrative speech may be impaired (Arrondo et al., 2009; Lethlean & Murdoch, 1997). However, these deficits are only seldom detected and a clear theoretical background underlying the impairment is still lacking. High-level language impairment may be a consequence of impairment in executive functions (Renauld et al., 2016; Arrondo et al., 2009). The understanding and use of metaphor, narrative discourse and other aspects of pragmatics should be examined as part of a comprehensive language evaluation. Pragmatic language skills are still largely unexplored in MS.

To date, only one study has extensively assessed pragmatic abilities in MS using a standardized test, the Assessment of Pragmatic Abilities and Cognitive Substrates (APACS; Arcara & Bambini, 2016; Bambini, Arcara, Bechi, et al., 2016a). It shed light on the relationship between pragmatics and cognitive/psychosocial features. Carotenuto and collaborators reported that MS patients displayed normal performance on the Token Test, indicating that basic language skills were intact. However, pragmatic impairment was evident in 55% of MS patients (Carotenuto, Cocozza, et al., 2018b). Pragmatic impairment was independent of disease duration and disease severity, suggesting that the quantity of pathological damage, calculated as lesion load throughout the brain and the extent of brain atrophy, only minimally influenced pragmatic abilities. Conversely, the type of pathological damage, especially in terms of lesion mapping throughout the brain and demyelinating lesions in specific fibre bundles, was crucial in determining impaired pragmatic abilities.

Other findings from the same study were that the ability to communicate the main aspects of everyday situations, as assessed in the Description task in APACS, were preserved in MS as opposed to the ability assessed in the Interview task, where MS patients failed to provide the appropriate amount of information and produced less essential information than control subjects and were under-informative in consequence. This finding confirms the results of an earlier study that explored quantity, lexical and syntactic complexity, and global thematic flexibility in spontaneous language production in MS (Arrondo et al., 2009). In addition to production impairment, MS patients also performed poorly in the Narratives section of APACS, which consists of a task assessing the comprehension of short stories. Moreover, MS patients displayed reduced performance in tasks that specifically explore the comprehension of non-literal language, such as Figurative Language 1, Figurative Language 2, and Humor. This revealed a significant impairment in the comprehension of non-literal meanings in these patients.

As with other neurological populations, it appears likely that pragmatic impairment in MS is related to executive dysfunction. Executive functions are involved in cognitive control processes including self-regulation, planning, and task management. Impairment of executive functions can lead to specific pragmatic deficits. For example, impairment in planning and monitoring discourse produces aberrant discourse structure, while disinhibition may impair the ability to use indirect and subtle language devices such as hints (Martin & McDonald, 2003). In MS, global cognitive impairment, assessed through the Brief Repeatable Battery (BRB)—a standardized neuropsychological battery widely used to assess cognition in MS patients—was not associated with pragmatic impairment (Amato et al., 2006). However, when exploring the association between each individual test of the BRB and APACS, pragmatic abilities were linearly associated with semantic fluency as assessed through the Word List Generation (WLG) (Carotenuto, Cocozza, et al., 2018b). As verbal fluency is usually taken to be a measure of executive functions (Henry & Crawford, 2004), this association clearly confirms the crucial role of executive deficit in pragmatic abilities. This finding is in line with previously reported associations between executive functions and pragmatics in other clinical population (e.g. patients with traumatic brain injury) (Martin & McDonald, 2003; McDonald & Pearce, 1995; McDonald et al., 2006).

Even if verbal fluency is often considered to be a test of executive functions, other language and cognitive abilities also influence fluency performance (Shao et al., 2014). For example, verbal fluency also involves vocabulary knowledge, and skills of lexical search and retrieval (Morere et al., 2012). Therefore, the relationship between WLG and pragmatics might reflect, on the one hand, the association between executive function and pragmatic abilities and, on the other hand, the impact of expressive language abilities on communicative competence. Unexpectedly, the other tests of the BRB that target verbal memory, visual memory, information processing speed, attention, and working memory seem not to be associated with pragmatics. One could speculate that pragmatics is not influenced by basic cognitive functions like memory and attention. However, since the data are limited, caution should be taken in drawing such a conclusion and further studies are needed to confirm these results. To summarize, pragmatic deficits might be present in MS patients even in the absence of general cognitive impairment and may be overlooked by clinicians who test their patients for global cognitive impairment.

Effective communication also relies on intact social cognition abilities. Social cognition refers to a wide array of mental operations that underlie social interactions. It is a multi-domain ability that includes empathy (Vollm et al., 2006), social perception of emotions from prosody, facial expressions, and body gestures (Calder & Young, 2005), and theory of mind (ToM) (Abdel-Hamid et al., 2009). The latter component refers to the ability to attribute emotion and intention to other people, known in the literature as affective and cognitive ToM, respectively (Frith & Frith, 2006). Carotenuto and colleagues demonstrated that pragmatic abilities are directly associated with ToM (Carotenuto, Cocozza, et al., 2018b). A large body of literature has shown that the ability to interpret social information plays an important role in shaping communicative behavior in patients (Martin & McDonald, 2003; Champagne-Lavau & Joanette, 2009). Moreover, a recent meta-analysis showed that MS patients have significantly lower performance in social cognition and, specifically, in ToM compared to healthy controls (Cotter et al., 2016; Pottgen et al., 2013; Banati et al., 2010).

The association between pragmatics and ToM is further confirmed by the fact that theoretical models of pragmatics highlight the important role of ToM in supporting the recognition of communicative intentions, especially when interpretation is required to move beyond the literal meaning and to infer the speaker's communicative intentions based on context (Sperber & Wilson, 2002). Carotenuto and colleagues showed that ToM was specifically linked to the comprehensive domain of pragmatics but not to pragmatic production (Carotenuto, Cocozza, et al., 2018b). Therefore, a social cognition deficit cannot fully account for the pragmatic impairment in MS, especially when considering discourse production and conversation. This suggests that both pragmatics and social cognition should be assessed in MS to reveal the contributions of underlying cognitive, linguistic and socio-cognitive factors to these communicative domains.

Pragmatic impairment is also associated with well-known psychosocial symptoms in MS such as depression and fatigue. Fatigue and depression are selectively related with discourse production skills, possibly reflecting the difficulty of engaging in conversation and social interactions when a person is experiencing these symptoms (Krupp et al., 1989). These findings have important clinical implications. To the extent that social cognition and pragmatics are essential for appropriate social interactions (Montel & Bungener, 2007), deficits in these cognitive domains might have a significant adverse impact on quality of life and interpersonal communication. Previous research on neurological patients has indicated that the competence in conversational discourse correlates with social integration and quality of life (Galski et al., 1998). Similarly, data on psychiatric patients has shown a relation between pragmatic abilities and quality of life (Bambini, Arcara, Martinelli, et al., 2016b). Phillips and colleagues only focused on emotion perception when they confirmed an association between ToM and quality of life in MS (Phillips et al., 2011). This leaves open the question of whether pragmatic deficits have an impact on quality of life. This point remains unexplored and deserves further investigation.

16.5 Neuropathology of Pragmatic Impairment in Multiple Sclerosis

To understand the neuropathology of pragmatic disorders in MS, it is useful first to emphasize that specific brain areas and networks are associated with pragmatic abilities. Initially, clinical evidence (mostly from neuropsychological studies conducted between the 1970s and 1990s) suggested that impairment in communicative behavior and in pragmatics (the latter understood as prosody) was mostly associated with damage to the right cerebral hemisphere, whereas communicative disorders such as aphasia were associated with damage to the left hemisphere. Thus, it seemed that a clear dichotomy could be drawn: left hemisphere lesions cause aphasia while right hemisphere lesions cause pragmatic impairment. This conclusion, however, was not completely supported by subsequent studies that relied on neuroimaging techniques such as functional MRI (fMRI) (Eviatar & Just, 2006; Lee & Dapretto, 2006; Bambini et al., 2011). These studies demonstrated bilateral activation throughout the brain during pragmatic tasks such as metaphor, idiom, and irony comprehension (for a meta-analaysis, see Bohrn et al., 2012).

Importantly, the activation of brain areas has been shown to depend on the nature of the pragmatic task and on specific characteristics of the experimental stimuli used in a study. For example, Mashal and Kasirer (2012) reported activation of the right homologue of Wernicke's area only for novel and salient metaphors. Despite heterogeneity of results, there is considerable agreement that pragmatic abilities involve several areas of the brain, not all of which occur in the right cerebral hemisphere. This finding has been confirmed by several sources of evidence and, today, pragmatic impairment is described not only after right hemisphere damage but in a large array of different pathologies (Cummings, 2017).

One recent proposal for the neuroanatomical basis of pragmatic abilities is the one hypothesized by Catani and Bambini (2014), who sketched a neuroanatomical model for Social Communication And Language Evolution and Development (SCALED). The SCALED model stresses the importance of white matter tracts connecting different areas throughout the brain, and the role of the networks created by these tracts. The authors identify five different networks associated with language and communication. Among these networks, one has a pivotal role for pragmatic abilities: a bilateral temporo-parietal network connecting Wernicke's area in the temporal lobe and Geschwind's area in the parietal lobe (Catani & Bambini, 2014). However, it is important to stress that besides a temporo-parietal network, successful pragmatic competence also requires the integrity of other connections, particularly those between parietal and frontal lobes. The involvement of fronto-parietal connections is fundamental both for correct linguistic analysis of the input (e.g. relying on Broca's area) and for higher-order functions such as executive functions (in areas of the dorsolateral or medial frontal cortex).

The central role of networks is of crucial importance for patients with MS, in whom brain lesions might interrupt white matter fibers. Alterations in white matter integrity (that is, structural connectivity) in MS have already been associated with alteration of functional connectivity, which can be measured in vivo with advanced techniques such as the functional MRI or magnetoencephalography (MEG). Studies with these techniques converge in indicating that decrease of white matter integrity (as compared to controls) might be associated with either an increase or a decrease in functional connectivity in resting-state networks (Lowe et al., 2008; Hawellek et al., 2011). These apparently paradoxical results were explained in two ways: a compensatory mechanism that might not completely succeed in compensating for neuroanatomical damage leading to pragmatic impairment, or a maladaptive role with a further negative impact on pathology, reflecting the loss of flexibility in functional interactions across distant areas of the brain.

Importantly, higher functional connectivity is not always associated with more advanced disease stage or anatomical damage. It is important to stress that results also depend on the methodology used to analyze the data. For example, Tewarie et al. (2014) used MEG and applied methods from Graph Theory to study change in the topology of brain networks in patients with MS. They found that patients with MS experienced loss of hierarchical structure, that is, a less complex structure in connectivity, as compared to healthy controls (Tewarie et al., 2014). In another study, investigators found that MS was associated with both an increase and decrease in MEG connectivity as compared to controls, and the direction of the effect depended on the frequency band (e.g. delta, theta, or alpha) of electromagnetic signal taken into account (Schoonheim et al., 2013).

Considering the crucial role of the temporo-parietal network in pragmatics, Carotenuto, Arcara, et al. (2018a) conducted a resting-state fMRI study on MS patients to assess whether functional connectivity of the temporo-parietal junction (TPJ) towards the rest of the brain was associated with pragmatic performance measured by a comprehensive pragmatics battery, the APACS (Arcara & Bambini, 2016). This study aimed to explore the neuropathological changes associated with pragmatic impairment in MS. The analyses showed that higher connectivity between the right TPJ and paracingulate cortex was associated with better pragmatic performance. A similar trend (not as strong as for the right TPJ) was found also for the left TPJ. The role of TPJ connectivity also highlights the important association between pragmatics and ToM. Several studies suggest that TPJ is a crucial hub where different sources of information converge, such as past experiences, an agent's description of intention, and perception of the stimulus.

The TPJ is also involved in several cognitive functions such as attention, memory, and low- and high-level language abilities (Carter & Huettel, 2013). Both ToM and pragmatics rely on many of these cognitive aspects and so involvement of the TPJ was somehow expected (Bambini et al., 2011; Schuwerk et al., 2017). As for the paracingulate cortex, this area was already associated with executive functions, supporting the close relationship of these cognitive skills in supporting pragmatic abilities. In addition, both the TPJ and the paracingulate cortex are parts of the so-called default-mode network. As the default-mode network seems to be involved in complex social problem solving (Mars et al., 2012), pragmatic abilities might be associated with default-mode network connectivity. Finally, pragmatic abilities are associated with functional connectivity between the TPJ and the paracingulate cortex over both the left and the right cerebral hemispheres, partly confirming the importance of a bilateral network for pragmatics.

16.6 Therapeutic Options for Pragmatic Impairment in Multiple Sclerosis

Since the characterization of pragmatic deficits in MS is still at an early stage, it is difficult to suggest potential treatments for MS patients suffering from pragmatic impairment. An important consideration concerns the initial assessment of the deficit. Given that the prevalence of pragmatic deficits in MS is quite high—55% according to Carotenuto, Cocozza, et al. (2018b)—a specific rehabilitation treatment might be extremely useful.

One of the techniques that has gained ground in the last decade as a treatment for neurological symptoms with well-defined neuroanatomical substrates is Transcranial Direct Current Stimulation (tDCS). In tDCS a small current is applied to the scalp via (typically, a pair of) electrodes. This current can lead to changes in neuron excitability that can promote neural plasticity. Several studies have directly tested the efficacy of tDCS on specific symptoms, such as cognitive or physical deficits (e.g. Ayache et al., 2016; Mattioli et al., 2016). Results have been inconsistent. For example, they showed either an ameliorative effect for tactile deficits (Mori et al., 2013) or no effect for spasticity (Iodice et al., 2015). Whether this technique may be successfully employed, either alone or in combination with other treatments, for pragmatic impairment in MS is still uncertain. However, the TPJ and other relevant areas found in the study by Carotenuto, Arcara, et al. (2018a) could be candidate targets for stimulation with non-invasive brain stimulation techniques like tDCS. Before viewing tDCS as a potential treatment for MS, several aspects should be better defined with dedicated studies, such as the stimulation set up and parameters, because these details can significantly influence the efficacy of the treatment.

Caution should be exercised in the utilization of tDCS as a treatment for pragmatic impairment in MS as several studies have shown that applying tDCS can lead to an increase in widespread connectivity (Polanía et al., 2011; Vecchio et al., 2016; Pellegrino et al., 2018). This is not necessarily a positive effect. Indeed, MS patients might experience a generalized increase in connectivity associated with lower cognitive functioning and disease severity, resulting from maladaptive mechanisms in response to demyelinating lesions. Thus, an increase in connectivity may even be detrimental to patients with MS.

An alternative to neuromodulation treatments is represented by behavioral treatments that focus on rehabilitation such as treatments used for other pathologies (e.g. see Gabbatore et al. (2015) for patients with traumatic brain injury). Behavioral treatment for pragmatics seems to be promising and has been found to improve communicative abilities in several pathological populations such as TBI (Sacco et al., 2016) and schizophrenia (Bosco et al., 2016).

Several studies offer neural and behavioral evidence of generalized positive effects from video-game training on cognitive control abilities of older adults. These studies show performance improvement that is comparable to the one observed in younger adults who are habitual action video-game players (Anguera et al., 2013). Although reports of beneficial effects in cognition following video-game training in the MS population are relatively rare (Prosperini et al., 2015), games could provide a potential rehabilitation tool for improving cognitive and pragmatic functions based on the hypothesis that playing action video games can modulate the activity of brain regions serving both motor and cognitive domains.

It should be emphasized that pragmatic treatments face practical problems such as the time that is required for treatment. Currently, pragmatic treatments typically last about 3 months and require frequent sessions that occur at least twice a week (Gabbatore et al., 2015; Bosco et al., 2016). Given the amount of time required, it is important to assess whether a behavioral treatment for patients with MS should be focused on pragmatics or should be focused on other symptoms with higher impact for patients' quality of life such as depression or fatigue (Benedict et al., 2005). However, since there are no studies directly addressing this issue, there is a need for investigations that explore the impact of pragmatic impairment on the quality of life of MS patients, and whether an improvement in pragmatic abilities in these patients may lead to an improvement in their mood and quality of life.

16.7 Summary

Although pragmatic deficits in patients with MS are still relatively unexplored, the few available studies that have investigated these deficits have served to highlight their frequency and extent. However, the impact of these communicative deficits on the everyday lives of patients with MS, and how physicians might modify their interactions with patients who have these deficits remains to be evaluated. Patients may be unable to interpret aspects of language such as metaphor and irony. This can compromise their understanding of a wide range of messages communicated by physicians, from evaluation and diagnosis to treatment and support. It would also be beneficial to explore further the neuropathological basis of the pragmatic impairment in MS with a view to developing therapeutic opportunities including behavioral therapy and electrical brain stimulation. This would result in an improvement in patients' social interactions and serve to reduce the economic burden of MS on society.

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Chapter 17
Amyotrophic Lateral Sclerosis

Valentina Bambini and Mauro Ceroni

17.1 Introduction

Amyotrophic lateral sclerosis (ALS) owes its name to the major clinical and neuropathological findings indicating neurodegeneration of, respectively, the peripheral and the central motor neurons. The disease shows a spectrum of clinical patterns: variable duration (from 1 to 15 years), variable onset (spinal or bulbar), extremely variable involvement of the peripheral and the central motor neurons, from Primary Lateral Sclerosis to Progressive Muscular Atrophy, making almost each case unique. For many years, neurologists believed that people with ALS would be completely lucid even as their body experienced progressive, and eventually total paralysis. It is now well recognized, however, that a number of cognitive, behavioral, social, and pragmatic deficits may be observed in 50% of ALS patients, and 15% of cases reach criteria for a diagnosis of fronto-temporal dementia (FTD) (Crockford et al., 2018; Goldstein & Abrahams, 2013; De Marchi et al. 2021).

This chapter reviews findings that have contributed to our understanding of pragmatic impairment in ALS. But first, it examines in Sect. 17.2 the epidemiology and aetiology of ALS as well as criteria that are used to diagnose cognitive and linguistic impairment in ALS. Section 17.3 reviews structural aspects of language in ALS,

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including speech, grammar and the lexicon. This section provides an overview of the general linguistic profile of patients with ALS, highlighting the linguistic domains that are most impaired, and considering also the specific domain of action/verb semantics. Section 17.4 focuses on pragmatics. Although the literature is modest, it can be grouped into two domains: one dealing with difficulties in producing pragmatically appropriate discourse, and the other dealing with difficulties in comprehending implicit meanings. Studies in these two domains are reviewed in Sects. 17.4.1 and 17.4.2, respectively, with emphasis placed on tasks and assessment tools and examples of patients' communicative behavior. Section 17.5 deals with the broader issue of communication and conversation in everyday life. A summary of the chapter's contents is offered in Sect. 17.6, together with some recommendations for future studies.

17.2 ALS: Some Background

The epidemiology of ALS has been the focus of several studies. An epidemiological meta-analysis of studies published between 1995 and 2011 reported that in Europe the median prevalence and incidence of ALS were 5.40 and 2.08 per 100,000 person-years, respectively (Chiò et al., 2013). Although studies from outside of Europe are scant, it seems that prevalence and incidence figures are lower in populations of mixed ancestral origin compared to European populations, with further differences in age of onset (Zaldivar et al., 2009; van Es et al., 2017). Granted these differences, studies reported a worldwide median prevalence of 4.48 and a standardized incidence rate of 1.68 per 100,000 person-years (GBD 2016 Motor Neuron Disease Collaborators, 2018; Marin et al., 2017). Median age of onset is 65 years in populations of European ancestry, whereas onset is approximately 10 years earlier in genetically heterogeneous populations (van Es et al., 2017). The male to female ratio is estimated to be around 1.5:1 (Manjaly et al., 2010), while the lifetime risk of ALS is 1 in 472 in women and 1 in 350 in men (Alonso et al., 2009). According to some studies, however, sex ratios might change with age and across countries, as well as over time (Logroscino et al., 2010; Longinetti & Fang, 2019; Manjaly et al., 2010).

The genetic basis of ALS and its relationship to FTD is the focus of an increasing number of studies. Investigators have discovered more than 30 different genes linked to familial ALS (Renton et al., 2014), which indicate that ALS is a clinically and genetically heterogeneous multidomain neurodegenerative disease. Moreover, investigators have discovered gene mutations that give rise to ALS as well as to FTD and to combinations of both, namely TARDP, C9orf, FUS and other rarer genes (Saberi et al., 2015). This underlines the relationship between ALS and FTD and confirms the idea that these conditions are the extremes on the phenotypic spectrum of a single disease (Burrell et al., 2016; Ferrari et al., 2011). The spectrum includes individuals with ALS with cognitive impairment who nevertheless do not meet the criteria for dementia (ALS-ci), individuals with behavioral impairment who nevertheless do not meet the criteria for dementia (ALS-bi), individuals with both cognitive and behavioral impairment without dementia (ALS-cbi), and individuals with features of both ALS and FTD (ALS-FTD) (Woolley & Rush, 2017).

The complexity and heterogeneity of ALS profiles makes neuropsychological assessment in this population especially important (De Marchi et al. 2021), as reflected in the most recent consensus criteria for the diagnosis of fronto-temporal dysfunction in ALS (Strong et al., 2017). This new version of the consensus criteria expands on a previous version (Strong et al., 2009), by incorporating more fine-grained distinctions of cognitive domains and the idea of a fronto-temporal spectrum disorder of ALS. While the old version focused especially on executive dysfunction, the new version acknowledges the importance of also considering impairments in social cognition, language and pragmatics. Based on the 2017 consensus criteria, a diagnosis of ALS-ci is to be made when there is evidence of either executive impairment (which could include social cognition) or language impairment (which could include pragmatics) or a combination of the two. More specifically, citing from Strong et al. (2017):

> *A diagnosis of ALS-ci depends on evidence of either executive dysfunction (including social cognition) or language dysfunction or a combination of the two.*
>
> *Executive impairment is defined as:*
>
> *(1) Impaired verbal fluency (letter). Verbal fluency deficits must control for motor and/or speech impairments to be valid.*
>
> *OR*
>
> *(2) Impairment on two other non-overlapping measures of executive functions (which may include social cognition).*
>
> *Language impairment is defined as:*
>
> *(1) Impairment on two non-overlapping tests (which could include pragmatic function).*

What is relevant for this chapter is the definition of language impairment as impairment in two non-overlapping tests, *which could include pragmatic function.* This is important for two reasons: (1) it shows that the presence of pragmatic impairment in ALS is now fully acknowledged; and (2) it classifies pragmatic impairment as part of language impairment, which in turn is classified as part cognitive impairment. This does not imply that pragmatic impairment depends on impairment in other cognitive domains, but rather that pragmatic impairment is one of the forms in which cognitive impairment can appear in the highly heterogenous ALS population. Indeed, the relationship between language, pragmatics, and executive problems has been one of the most investigated topics in the literature, along with the relationship with motor symptoms.

17.3 Speech, Grammar, and Lexical Semantics in ALS

The most prominent language-related problem in ALS is speech production (Orange & Hillis, 2012). Although speech is considered a 'peripheral' component of the faculty of language, as it lies at the interface with motor and sensory domains

(Hauser et al., 2002), speech problems impact other language aspects, such as discourse productivity and conversation management, and in turn affect communication effectiveness and quality of life (Felgoise et al., 2016). Any study of language in ALS should thus start by considering speech, and should control for speech production impairments and motor deficits in assessing other linguistic aspects (Strong et al., 2017). Concerning 'core' language aspects, the systematic review of the literature conducted by Pinto and colleagues (Pinto-Grau et al., 2018) identified 57 studies on ALS and language conducted between January 1975 and August 2017. Seven domains of linguistic abilities were considered in the review: (a) verbal expressive language (i.e. discourse productivity); (b) verbal fluency (i.e. word generation); (c) confrontation naming; (d) semantic processing; (e) auditory comprehension; (f) action verb processing vs. object noun processing; and (g) reading and writing. Overall, the review pointed to expressive problems at the level of both grammar and semantics and suggested that there might be linguistic difficulties in ALS that are independent of speech impairment and impairment in executive functions.

17.3.1 Speech and Dysarthria

Dysarthria indicates the condition where, due to damage to the central and/or peripheral nervous system, speech movements are impaired in speed, force, range, direction and timing (Orange, 2009). It is estimated that 80–95% of patients with ALS exhibit severe dysarthria during disease progression (Linse et al., 2018; Tomik & Guiloff, 2010). Studies have also documented the co-occurrence of dysarthria and apraxia of speech, a condition where there are impairments in the ability to plan or program commands that direct speech movements (Duffy et al., 2007).

Dysarthria in ALS usually results from the involvement of both upper and lower motor neurons and is described as mixed spastic-flaccid dysarthria. Typical features of speech in ALS are imprecise consonants (i.e. slurred speech sounds), hypernasality, harsh voice quality, slow speaking rate, monopitch and short phrases (Orange & Hillis, 2012). Assessment of speech includes sound error profiles (in tasks such as single and multiple sound production and repetition, single and multiple syllable word repetition, etc.), rate of speech, and intelligibility scores.

Many patients exhibit loss of adequate speech even at the onset of the illness (Linse et al., 2018). A longitudinal study showed that speech remained adequate for 18 months from the first bulbar symptoms, but speech quality rapidly declines during the course of the illness (Makkonen et al., 2018). Speech deteriorates more in the bulbar group compared with the spinal group and the difference remains significant at follow-up. In some cases, dysarthria might lead to complete speechlessness, i.e. anarthria (Orange & Hillis, 2012). Because of these problems, 60% of patients need augmentative and alternative communication methods (Makkonen et al., 2018), such as eye-tracking-based computer devices and brain-computer interfaces. These technologies offer the possibility of maintaining independent communication even in advanced stages of ALS (Linse et al., 2018).

17.3.2 Grammar and the Lexicon

To sketch a global profile of the linguistic domains of grammar and the lexicon in ALS, it is useful to start from the study of Taylor and colleagues, who ran an extensive language assessment in 51 patients with ALS and matched controls (Taylor et al., 2013). They created a composite measure of language skills, based on a series of tests assessing fluency, grammar, and lexical/semantic knowledge, among which were the Cookie Theft Picture (from the Boston Diagnostic Aphasia Examination; Goodglass & Kaplan, 1972), the Test of Reception of Grammar 2 (Bishop, 2003), the British Picture Vocabulary Scale II (Dunn et al., 1997), and the Pyramid and Palm Trees Test (Howard & Patterson, 1992). Using the 5% cut-off obtained from the control sample, the authors showed that 43% of the patients were classified as impaired in the composite language measure. Among the tests where patients mostly failed were the Test of Reception of Grammar (35%) and the British Picture Vocabulary Scale (49%). In parallel with the language assessment, the authors also ran an extensive assessment of executive functions, reporting that 31% of patients were impaired. Interestingly, although there was a significant correlation between language impairment and executive impairment, and executive scores accounted for 44% of variance in language scores, the two domains were to some degree distinct. The authors concluded that language and executive deficits do not always co-occur in ALS (see also Goldstein & Abrahams, 2013).

Another study performing a broad language assessment is the one by Tsermenteli and colleagues, where a sample of 26 patients with ALS and controls was evaluated using syntactic and semantic tests as well as an examination of connected speech (Tsermentseli et al., 2016). Results indicated that patients' performance in some semantic tests was spared, such as single-word naming, but there were major deficits in other semantic tests (such as action/verb semantics) and in syntactic measures. They also found that syntactic measures discriminated between patients and controls and argued that syntactic difficulties are the predominant feature of the linguistic profile of ALS.

While some studies argue that syntactic impairment is predominant, the literature offers robust evidence of impairment in the semantic domain too. Studies focusing on lexical/semantic aspects showed that problems can affect naming, but also extend to semantic knowledge. For instance, Leslie and colleagues evaluated lexical/semantic skills through a variety of tests (Leslie et al., 2014), and showed that approximately one third of a sample of 17 patients with ALS were impaired in the naming scores. The same frequency of impairment was also reported in semantic knowledge, which was calculated using non-verbal test scores (i.e. scores that required pointing responses) such as word comprehension and semantic association, thus minimizing the impact of dysarthria. These data indicate that semantic deficits are also common in ALS, and they are relatively independent of the severity of motor impairment. Interestingly, the study of Leslie et al. also included patients with ALS-FTD and with semantic dementia (SD), which allowed them to identify the following gradation of impairment: controls > ALS > ALS-FTD > SD. In all clinical groups, semantic deficits were associated with atrophy of the anterior temporal lobe, which is a region known for its pivotal role in semantic processes (Lambon Ralph et al., 2016).

17.3.3 Action Language

Because of the motor nature of the dysfunction in ALS, the literature has paid considerable attention to the domain of action language. The aim of studies in this area is to investigate whether patients with ALS are impaired in processing language that expresses actions, and whether this impairment is related to alterations of the motor cortex (Bak & Chandran, 2012; Grossman et al., 2008). A series of studies of Bak and colleagues demonstrated that patients with ALS have more pronounced difficulties when dealing with action verbs than when dealing with objects nouns, as revealed through different tasks such as word-picture matching, naming from drawings, and picture-picture matching (Bak & Hodges, 1997, 2004; Bak et al., 2001). Grossman et al. also offered neuroanatomical data, showing that the impairment in action words correlated with atrophy in the motor cortex (Grossman et al., 2008). Difficulties with verbs were reported also in production. For instance, Cousins et al. (2018) tested 28 patients with ALS in the Cookie Theft Picture description task and showed that, compared with matched controls, individuals with ALS produced less agent body verbs, i.e. verbs where the body is the agent of the action (e.g. “the boy is *stealing* the cookies”), but an equal amount of theme verbs, i.e. verbs where the body is the recipient of the action (e.g. “the boy is *falling*”). Moreover, this dissociation correlated with grey matter atrophy of the premotor cortex.

Papeo and colleagues, however, placed less significance on the claim of a specific problem in representing actions (Papeo et al., 2015). Indeed, Papeo et al. remarked that studies on nouns vs. verbs have often confused grammatical and semantic aspects: by comparing nouns referring to objects and verbs referring to actions, what the literature showed might simply be a higher difficulty of verbs as a grammatical class, rather than of actions as a semantic domain. They argued that, if there is a specific difficulty in action representation in ALS, then the problem should arise not only with verbs but also with manipulable object nouns (e.g. *pen*). They tested 21 patients with ALS in a series of tasks involving action verbs and manipulable-object nouns and found that patients performed better with nouns than verbs. This speaks against a problem in action representation *per se*. Moreover, the noun-verb asymmetry was in the range observed in the normal population. Based on additional tests, the authors argue that the higher difficulty with verbs might be explained in terms of difficulty with the representation of the logical/temporal sequence of motor events, which belongs to the domain of executive functions. The debate on these aspects is of course still open, as it relates to the broader issue of grounded cognition, and the link between action and language.

17.4 Pragmatics in Amyotrophic Lateral Sclerosis

Pragmatics in ALS has received very little attention from researchers to date. A search on Pubmed conducted in March 2019 (when the first version of this chapter was written) using the keywords “amyotrophic lateral sclerosis” and “pragmatics” returned only two results. One is a paper by Bambini and colleagues (Bambini, Arcara, Martinelli, et al., 2016b). In this study, the authors performed a comprehensive assessment of pragmatic skills in ALS, using the Assessment of Pragmatic Abilities and Cognitive Substrates (APACS) test, a standardized tool for the evaluation of expressive and receptive pragmatics in Italian (Arcara & Bambini, 2016). The other is a paper by Fisher and colleagues (Fisher et al., 2017), which deals with pragmatics in everyday communication and evaluated perceived communicative skills as assessed through the La Trobe Communication Questionnaire (see Sect. 17.5). The same search performed in May 2021 (at the time of proof-correction) returned just one more result, namely Bambini et al. (2020b) on humor comprehension. Searching for “amyotrophic lateral sclerosis” and a more specific pragmatic aspect, i.e. “figurative language”, returned again just one paper, the article by Bambini, Arcara, Martinelli, et al. (2016b). When we used “amyotrophic lateral sclerosis” and “discourse” as keywords, the search returned two other papers: Ash et al. (2014) and Roberts-South et al. (2012). Although these studies were not explicitly framed in a pragmatic perspective, they focused on expressive abilities in producing discourse, taking into account aspects that are often included in the pragmatic domain, such as measures of discourse coherence and informativity. The same searches with “motor neuron disease” did not return any other relevant paper. A search through other sources highlighted two other papers dealing with pragmatics: a paper by Strong et al. (1999) was retrieved in review articles on language in ALS (Orange & Hillis, 2012; Pinto-Grau et al., 2018) and includes the evaluation of discourse production; a paper by Staios et al. (2013) was found while examining the literature on social cognition in ALS (e.g. Strong et al., 2017) and is a study of sarcasm comprehension.

As is evident from the outcome of the search described above, i.e. *six* articles in total, the literature on pragmatics in ALS is still in its infancy and is more concerned with difficulties in production than in comprehension. Studies on production mostly applied discourse analysis to structured production tasks, often employing the Cookie Theft Picture description task (on the role of this task in identifying pragmatic markers of neurodegenerative diseases, see Cummings, 2019), whereas comprehension was assessed through different types of comprehension questions. Moreover, the assessment of pragmatics was often accompanied by the evaluation of other linguistic and cognitive aspects. Albeit limited in number, these few studies consistently indicate that ALS is associated with impaired narrative expression and comprehension of non-literal meanings, possibly with an overall frequency of impairment—36% based on Bambini, Arcara, Martinelli, et al. (2016b)—that is comparable to the one observed for language problems. Also, the available evidence suggests that pragmatic problems are dissociable from impairment in cognitive or socio-cognitive skills, and that they are to some extent distinct from impairment in structural language aspects (vocabulary and grammar).

17.4.1 The Pragmatics of Narrative and Other Discourse

In the study by Strong et al. (1999)—probably the first to consider pragmatics in ALS—a sample of 13 patients was evaluated at two different time points in order to characterize the neuropsychological profile of ALS longitudinally. Among the tests, patients were administered an interview task and the Cookie Theft Picture description task. The outcome of these tasks was evaluated in terms of discourse units, fluency, repetitions, self-corrections, and other conversational aspects. The study showed that, at the second time point, individuals with ALS produced significantly fewer self-corrective utterances compared with controls. Although not fully discussed by the authors and not considered in terms of pragmatics, this suggests altered discourse communication over time. Indeed, reduced self-correction and thus an increased presence of uncorrected errors has also been reported in other clinical conditions such as Alzheimer's disease and Parkinson's disease (McNamara et al., 1992).

More recently, a study by Roberts-South et al. (2012) assessed the description of the Cookie Theft Picture over a period of 24 months in 16 non-demented patients with ALS and a group of matched controls. Several discourse measures were used, including discourse productivity measures (such as number of words and utterances), discourse efficiency measures (such as the number of correct information units, i.e. words that were intelligible in context, accurate in terms of picture/topic content, and relevant to picture/topic content), and content units (different novel concepts used to describe the picture). Results showed that discourse efficiency was more vulnerable than discourse productivity. Although there were no differences between patients and controls in total words or total utterances, there were subtle differences in discourse content, with patients being less effective than controls in conveying content. The authors argued that a possible reason might be found in the high presence of revisions and reformulations (e.g. [He is] [she was climbing] she was reaching for a cookie), which was also compromised in the study by Strong et al. (1999), or the presence of "empty words" (e.g. The [thing] it is somewhat a [you know thing]), which did not contribute to content. Reduced discourse efficiency was observed at all time points, while there was no impairment in a range of standardized language measures (like vocabulary and grammar).

Difficulties in the pragmatics of narrative discourse also emerged in the Frog Story task, where participants are asked to tell the story in the children's picture book "Frog, where are you?" (Mayer, 1969; see also Ash et al., 2006). Ash and colleagues showed the Frog Story to 26 patients with ALS and a sample of controls (Ash et al., 2014). The narratives were scored for local connectedness (paying attention to adverbials, referential expressions such as pronouns and determiners, and statements of cause and effect), global connectedness (whether the speaker acknowledges the point of the story, namely, that the frog found at the end is the frog that was present in the boy's room at the beginning), and maintenance of the story theme (i.e. searching for the frog). Differences between patients and controls were observed for local connectedness and for the maintenance of the story theme, but not for global connectedness. Although there was an association between narrative measures and executive scores, these differences were also observed in a subset of patients who did not have executive deficits. Moreover, the impairment was also observed in the subset of patients with no dysarthria. Thus, as shown in other studies, difficulties in discourse cannot be fully accounted for in terms of executive measures or motor impairments. The authors also investigated the neuroanatomical basis of discourse disruption. Local connectedness was associated with gray matter atrophy in right dorsolateral prefrontal and bilateral inferior frontal regions and with reduced fractional anisotropy in white matter tracts, specifically tracts mediating projections between prefrontal regions. These data suggest that difficulties in narrative abilities might be related to a disruption of the networks in the frontal lobes that are likely to support discourse-specific processes (Zacks & Ferstl, 2016).

A later study by Ash et al. (2015) of the same sample of participants and with the same task (the Frog Story) focused on fluency and grammar rather than discourse. It showed that patients were less fluent (in terms of quantity of speech, speech rate, and speech articulation errors) and committed more grammatical errors compared with controls. Speech rate and articulation errors were related to motor impairment, while grammatical errors were independent of motor difficulty. The investigation of neuroanatomical aspects revealed that grammatical problems were related to gray matter atrophy in left inferior frontal and anterior temporal regions and to reduced fractional anisotropy in white matter tracts (superior longitudinal and inferior frontal-occipital fasciculi). These networks do not fully overlap with those reported in the Ash et al. (2014) study, thus pointing to some degree of segregation between the brain structures supporting structural language and those supporting pragmatics.

The data from the study by Bambini, Arcara, Martinelli, et al. (2016b) confirmed and extended findings of problems in the pragmatics of discourse in ALS, by offering evidence from other discourse tasks, namely the autobiographical interview and the description of everyday scenes. More specifically, Bambini and colleagues used the APACS test, which consists of one section devoted to the assessment of the production of pragmatics and one to the assessment of the comprehension of pragmatics (Arcara & Bambini, 2016). Here, we will consider the outcome of the production section (see Sect. 17.4.2 for the outcome of the comprehension part of the APACS).

The APACS production section includes an interview task and a description task. In the interview task, patients are asked to talk about autobiographical topics (family, house, job, daily routine) for a duration of 5–10 min, and then the outcome is annotated for the frequency of occurrence of several communicative difficulties. These difficulties mainly affect the pragmatic level of language, specifically informativity and information flow. As measures of informativity, the APACS considers instances of over- and under-informativity, i.e. inability to offer the appropriate amount of information and to adhere to the Gricean Maxim of Quantity. Measures of information flow include topic shifts, i.e. abrupt change of topic indicative of difficulties with the Gricean Maxim of Relevance and altered order of elements in the discourse. Measures of discourse cohesion (such as lack of discourse referents) are also included. The frequency of communicative difficulties is then converted into a score. While the interview task aims at assessing the pragmatics of discourse, the description task aims at assessing communicative effectiveness more broadly, by measuring the ability to convey salient information. In the description task, patients are shown pictures of everyday situations and are asked to name the main elements, i.e. the agents, the actions, and the location, with the aim of evaluating the ability to identify and communicate salient information. For instance, one of the description items depicts a situation at the playground with children playing, and participants' descriptions are considered appropriate when the location (the playground), the main characters (the children), and the main action (playing) are spontaneously communicated. Scores focus on communicative effectiveness rather than on formal aspects (i.e. paraphasias are not counted as errors). Finally, a Pragmatic Production composite score is derived from the two production tasks (interview and description).

Bambini and colleagues reported data from 30 patients with ALS (after excluding three patients with dysarthria from the complete sample). They found that 12 patients (40%) were impaired in the Pragmatic Production score compared with the control sample: specifically, 14 patients (47%) were impaired in the interview task and 10 patients (33%) were impaired in the description task. Among all APACS tasks, the interview task was the one associated with the highest effect size in the group difference between patients and controls. Patients' speech was poorer than controls' speech in terms of informativity and information flow: patients were often over-informative, thus reducing thematic cohesion, and their narrative often lacked cohesion, with referents frequently missing. Table 17.1 shows an extract from an interview between an examiner (E) and A.S., a 61-year-old male participant (P) with ALS (A.S. is case 21 in Table 1 in Bambini, Arcara, Martinelli, et al., 2016b). A.S. has 11 years of education. He has limb onset, is 13 months post onset, and is rated 40/48 on the ALS Functional Rating Scale-revised form (Cedarbaum et al., 1999). Over-informativity and tangentiality are evident in turns 4, 10, and 12. Examples of missing referents are displayed in turn 10, with repetitive use of null-subject pronouns, not preceded by prominent discourse referents.

As in previous studies, Bambini and colleagues also explored the relationship between impairment in the pragmatics of discourse and executive functions. The latter were assessed with a battery of tasks including Phonemic Fluency (Carlesimo et al., 1995), Semantic Fluency (Novelli et al., 1986), the Wisconsin Card Sorting Test (Laiacona et al., 2000) and the Frontal Assessment Battery (Appollonio et al., 2005). Moreover, the authors also took into account the role of social cognition. This was evaluated with the Social Intelligence Battery (Prior et al., 2003), including measures of Theory of Mind, Normative Situations, Violations of Norms, and Appropriateness. A regression analysis showed that 27% of variance in the Pragmatic Production composite score was explained by executive scores, whereas in another regression social cognition scores accounted for 20% of variance in the Pragmatic Production score. A combined analysis including both executive functions and

Table 17.1 Extract from the APACS interview task showing the discourse production of a patient with ALS

1	E: Com'è fatta la sua casa?	E: How is your house?
2	P: Ma io c'ho una casetta piccola eh. Due locali…	P: Well I have a small house eh. Two rooms…
3	E: Un appartamento?	E: An apartment?
4	P: Si, due locali. Infatti nel bagno ho messo la doccia, che è più comoda eh. E poi l'ho personalizzata. Ah non c'ho… [cerca il telefono nelle tasche, per mostrare una fotografia] se no le facevo vedere.	P: Yes, two rooms. And in the bathroom I put the shower, which is more comfortable eh. Then I personalized it. Ah I don't have… [*A.S. looks for the cell phone in the pocket, to show a picture*] otherwise I would show.
5	E: Non importa. Quante stanze ha?	E: It doesn't matter. How many rooms does it have?
6	P: Ma allora c'è la sala e la cucina. Sai che ho fatto il muretto per dividere. Poi c'è la camera da letto e il bagno. Saranno to'… sui cinquanta metri quadri, più o meno.	P: Well, there is the living room and the kitchen. You know I built a little wall to separate. Then there is the bedroom and the bathroom. It's probably… about fifty square meters, more or less.
7	E: Vive in un condominio?	E: Do you live in a condo?
8	P: Sì, è un condominio, brava, che non è grande eh, siamo noi e quelli…, si sta bene.	P: Yes, it is a condo, correct, which is not big eh, it's us and those… it's good.
9	E: A che piano è?	E: What floor is it?
10	P: Io? Piano terra, anzi menomale piano terra lì hanno sbagliato, mi han fregato eh. Han detto che mi facevano un pezzo di giardino. Sì me l'han fatto ma piccolo. Sarà… [si alza e mima lo spazio del giardino] Qui c'è la casa. Il giardino è così. Invece sulla mappa sai cos'era? Era due volte di più. E poi lì hanno detto che hanno sbagliato e ho dovuto accettar lo stesso… perché avevo firmato... va be'. Poi io…	P: Me? Ground floor, indeed thank goodness, ground floor, there they* made a mistake, they* fooled me eh. They* said that they* would do a piece of garden for me. Yes they* did it but small. It must be… [*A.S. stands up and mimics the extension of the garden*] Here's the house. The garden is like this. But do you know how it was on the map? Twice as big. And then they* said that they* made a mistake and I had to accept anyway because I had signed… whatever. Then I… *Indicates the use of null-subject pronouns in original Italian*
11	E: Eh… dica?	E: Yes… what?
12	P: Poi io essendo grafico così, io in un centimetro ci faccio stare un casino di sopra, perché io sfrutto veramente… Essendo grafico faccio le cose giuste. Se guardi quelli di sopra che c'hanno la casa come la mia…	P: Then since I am a graphic designer so, in a centimeter I fit lots of things because I exploit it really… Being a graphic designer I do the right things. If you look at the people upstairs who have the apartment like mine…
13	E: Non c'è paragone?	E: There is no comparison?
14	P: Si vede proprio la differenza	P: Yes, you really see the difference.

social cognition showed that both predicted variance in Pragmatic Production (49% of variance in total), with a slightly higher effect size for executive functions than for social cognition. However, when looking at co-occurrence of deficits in different domains, no systematic association was found between pragmatic deficits and deficits in social cognition or executive functions. Specifically, only some of the patients with a deficit in Pragmatic Production were classified as impaired in executive functions or in social cognition. These findings suggest that executive function and social cognition skills are indeed linked to the ability to provide appropriate discourse in conversation. Executive functions possibly support aspects such as topic management (including avoiding repetitions and signaling topic shifts) and turn taking, whereas social cognition skills might help in understanding the interlocutor's perspective and informational needs. Yet, at least in ALS, pragmatic difficulties cannot be equated with dysexecutive behavior or social cognition impairment.

17.4.2 Inferring Non-literal Meanings and from Texts

Information on receptive pragmatic abilities comes mainly from the study of Bambini, Arcara, Martinelli, et al. (2016b), and is based on the outcome of the comprehension section of the APACS test administered to 33 non-demented patients with ALS. This section of APACS includes four tasks: Narratives, which evaluates the ability to understand explicit and implicit aspects of narrative discourse; Figurative Language 1, which evaluates the ability to understand figurative expressions (idioms, metaphors, and proverbs) presented in a minimal context through multiple-choice questions; Humor, which also uses multiple-choice questions to assess the ability to understand humorous utterances; and Figurative Language 2, which differs from Figurative Language 1 in that the figurative expressions are presented out of context and the task is based on an open response format, asking participants to explain the meaning of the expressions. Finally, a composite score is derived from the four comprehension tasks, which is named Pragmatic Comprehension.

The study showed that patients with ALS performed worse than controls in all comprehension tasks. Focusing on figurative language understanding, patients were impaired both when assessed with the multiple-choice response format (Figurative Language 1 task) and when assessed with the verbal explanation response format (Figurative Language 2 task). The Figurative Language 2 task was more difficult than the Figurative Language 1 task, as reflected in the higher effect size and higher frequency of the deficit: 9/22 (41%) patients impaired in Figurative Language 2 vs. 9/33 (27%) patients impaired in Figurative Language 1, based on the fifth percentile cut-off derived from the control sample (note that in Figurative Language 2 the $N = 33$ sample was reduced to $N = 22$ due to the exclusion of some patients for dysarthria or fatigue). The greater difficulty of the open response format has been observed in other clinical populations too (Arcara et al., 2019). It is probably due to the costs of planning and producing the response to the interlocutor, which increases the difficulty of inferring figurative meanings (Kalandadze et al., 2019).

The inspection of the responses given in Figurative Language 2 clearly shows the struggle of patients with ALS in organizing their responses and expressing abstract aspects of figurative meanings (see Table 17.2). Patients often repeated the target

Table 17.2 Examples of responses of patients with ALS in the APACS Figurative Language 2 task

Example 1, patient P.S.	E: “Certi ricordi sono spine”.	E: “Some memories are thorns”.
	P: Certi ricordi sono spine…	P: Some memories are thorns…
	E: Mhmh	E: Mhmh
	P: Perché… (tossisce)… la pace…	P: Because… [coughing]… the peace…
	E: Mi può ripetere, scusi?	E: Can you repeat please?
	P: Certi ricordi sono spine.	P: Some memories are thorns.
	E: Cosa vuol dire?	E: What does it mean?
	P: Sono falsi.	P: They are false.
	E: Sono falsi?	E: They are false?
	P: Mh sono spine.	P: Mh they are thorns.
Example 2, patient P.S.	E: “Certe voci sono trombe”.	E: “Some voices are trumpets”.
	P: Certe volte?	P: Some times? *[P.S. probably mistook the word ‘voci’ (‘voices’ in English) for ‘volte’ (‘times’ in English), as they both start with vo-]*
	E: “Certe voci sono trombe”, cosa vuol dire?	E: “Some voices are trumpets”, what does it mean?
	P: Eeh	P: Eeh
	E: Quando si dice quello è un trombone…che cosa vuol dire?	E: When one says “that guy is a big trumpet”… what does it mean?
	P: Vuol dire che… ha un problema.	P: It means that… he* has a problem.
	E: Ok.	E: Ok.
Example 3, patient Z.V.	E: Allora, “L’abito non fa il monaco”, cosa vuol dire?	E: So, “The vest does not make the monk”, what does it mean?
	P: Per me sì.	P: For me yes.
	E: Che cosa vuol dire?	E: What does it mean?
	P: Eh il vestito fa il monaco… per esempio… le persone vestite bene.	P: Eh the vest makes the monk… for instance… well-dressed people.
	E: Cosa vuol dire in generale “L’abito non fa il monaco”?	E: What does it mean, in general, “The vest does not make the monk”?
	P: Se una persona è… giusta… messa bene, sta anche bene	P: If a person is… right… well dressed, also looks good.
	E: Ok.	E: Ok.
	P: Per me!	P: For me!

expressions and offered wrong (see example 1) or vague responses (see example 2) or used concrete examples to explain the figurative meaning of a proverb (example 3). The examples in Table 17.2 are taken from the responses of two participants with ALS. P.S. is a 70-year-old male participant with ALS (P.S. is case 26 in Table 1 in Bambini, Arcara, Martinelli, et al., 2016b). P.S. has 5 years of schooling. He has bulbar onset, 15 months of disease, and is rated 37/48 on the ALS Functional Rating

Scale. Z.V. is a 72-year-old female participant with ALS (Z.V. is case 24 in Table 1 in Bambini, Arcara, Martinelli, et al., 2016b). Z.V. has 5 years of schooling. She has bulbar onset, 6 months of disease and is rated 43/48 on the ALS Functional Rating Scale.

Other preliminary data by the same group (Bambini et al., 2018) showed that patients have more difficulties in understanding and explaining metaphors expressing mental characteristics (e.g. "Grandparents are pillars") than physical or behavioral characteristics (e.g. "Lifeguards are lizards"), which suggests that greater difficulties are experienced when dealing with more abstract contents. It is interesting to note that difficulties in ALS have also been reported in the domain of logical metonymy or aspectual coercion (Baggio et al., 2016), i.e. when linguistic elements trigger a shift in meaning, as in the case of temporal modifiers that change the meaning of a verb (for instance, the modifier "for two hours" in a sentence like "She repaired the TV for two hours" indicates an event which continued for 2 h without conclusion). In a task where participants were asked to accept or reject sentences that involved aspectual coercion, patients with ALS exhibited an abnormally high acceptance rate compared with controls, i.e. patients accepted more instances of coercion than controls, likely due to a problem in representing the semantic and contextual constraints that govern the application of coercion. Although aspectual coercion does not strictly classify as pragmatic, these findings are indicative of difficulties in the broader domain of meaning shift and contextual adjustment that are not limited to figurative language.

Returning to the study by Bambini, Arcara, Martinelli, et al. (2016b), patients also failed in the Narratives task included in APACS. In this task, participants are orally presented with short stories that are similar to everyday news (e.g. about a robbery of a jewelry store or about commuters' protest at delayed train service). They are then asked questions about main or more detailed aspects of the texts, either explicit or implicit. The frequency of impairment was 45% (15/33), indicating a widespread problem in text-based inferences. These difficulties in comprehending narrative discourse seem to correspond to difficulties in dealing with the production of conversationally appropriate discourse. This suggests that impairment of discourse is not linked to a specific modality but is 'overarching' (Grodzinsky et al., 1983), i.e. involving both expressive and receptive language.

Finally, patients exhibited difficulties in understanding humor, as assessed through the Humor task in APACS. This task includes seven items, each presenting a brief story that requires respondents to select the funny ending among three options: one humorous; one straightforward; and one a non-sequitur. For instance:

A man goes into an umbrella shop and sees that the price of an umbrella is 10 pounds. So he asks the shopkeeper: "What could I get for less?" And the shopkeeper replies:

Humorous ending: *"Wet."*
Straightforward ending: *"We don't have any umbrellas for less than that."*
Non-sequitur ending: *"It's nearly lunch time."*

In this task, A.S. (whose interview is reported in Table 17.1) selected the non-sequitur ending. He did the same for two of the other seven items. At group level, patients performed significantly worse than controls, with 7/33 (21%) showing impairment based on the fifth percentile cut-off derived from the control sample. For further evidence on humor impairment in ALS, see the more recent Bambini et al. (2020b).

Sarcasm, which is considered a more aggressive form of ironic communication, is also a challenge for patients with ALS. In a study by Staios et al. (2013), participants were presented with the Social Inference-Minimal subtest of The Awareness of Social Inference Test (TASIT; McDonald et al., 2007). This consisted of video vignettes of actors producing sincere, sarcastic, or paradoxically sarcastic statements, followed by a series of yes/no questions about what the actor was doing, saying, thinking, and feeling. For instance, in one video the two actors have the following conversational exchange:

Michael: "Well, congratulate me!"
Gary: "What for?"
Michael: "I've got a date with Anne."
Gary: "Anne!"
Michael: "Come on, don't be jealous."
Gary: "Sure, I'm jealous"

And then the questions were presented, for instance, asking whether Gary thinks that dating Anne is a good thing. Patients with ALS did not differ from controls in answering questions about sincere statements, but they were impaired in answering questions about sarcastic and paradoxically sarcastic statements, with no differences between patients with bulbar and limb onset. The difference remained significant after controlling for executive skills. Interestingly, this study is framed in the context of social cognition, a domain which has been described as largely impaired in ALS (Bora, 2017; Cerami et al., 2014). Indeed, the ability to understand humor, irony, and sarcasm is considered by some in pragmatic terms and by others in social cognition terms (Bischetti et al., 2019; Martin & McDonald, 2003), which leads us to the next point, i.e. the relationship between pragmatics and other cognitive domains.

The study by Bambini, Arcara, Martinelli, et al. (2016b) also addressed the relationship between impairment in the domain of pragmatic comprehension and impairment in other cognitive domains. As observed for the pragmatics of discourse production (see Sect. 17.4.1), results confirmed that several cognitive dimensions are related to impaired pragmatic comprehension but that these dimensions do not fully explain pragmatic comprehension difficulties. Regressions showed that 40% of variance in the Pragmatic Comprehension composite score was explained by executive scores, whereas social cognition (together with age) explained 63% of variance in pragmatic scores. A combined analysis including both executive functions and social cognition showed that only social cognition was a significant predictor of Pragmatic Comprehension. When examining co-occurrence of deficits in the different domains, no systematic association was found between deficits in Pragmatic Comprehension and deficits in social cognition or executive functions.

These data are indicative of a strict relationship between pragmatic inferential skills and social cognition skills, although the two domains cannot be viewed as overlapping (see also Bosco et al., 2018), at least when considering a comprehensive pragmatic comprehension measure such as in APACS. Conversely, the role of executive skills in comprehending pragmatic aspects of language seems limited. Executive functions are less central for pragmatic comprehension compared to the role that they play in the domain of pragmatic production.

17.5 From Pragmatics to Use: Social Communication and Conversational Patterns

The current section adopts a more ‘applied’ perspective, moving from the pragmatics of language—intended as the ability to produce adequate discourse and to infer implicit meanings—to the use of language in social communication by people living with ALS. While studies on the pragmatics of discourse described in earlier sections focused on linguistic characteristics such as informativity and coherence of speech, another relevant issue is how people with ALS use their pragmatic skills in everyday contexts and whether difficulties in pragmatics compromise daily communication. One tool that captures pragmatic skills as used in daily life and social communication is the La Trobe Communication Questionnaire (Douglas et al., 2000), a 30-item questionnaire of perceived verbal communicative abilities via self- and informant (e.g. a spouse) report. The La Trobe Communication Questionnaire assesses communicative behaviours and aspects related to the Gricean maxims, such as initiation/conversational flow (speak too slowly), disinhibition/impulsivity (speak too quickly), conversational effectiveness (give completely accurate information, in accordance with the Gricean Maxim of Quality), and partner sensitivity (hesitate, pause, or repeat yourself, thus contravening the Maxim of Manner).

Using this tool, Fisher and colleagues evaluated communicative skills in a sample of 32 patients with ALS without significant cognitive impairment and 24 healthy controls (Fisher et al., 2017). In addition, each participant nominated a close other (generally spouse, child or sibling) as informant. Self-reports did not reveal any difference between patients with ALS and controls. Within patients, results were similar for bulbar and limb onset subgroups and there was no effect of clinical variables. However, informants rated patients with ALS significantly lower than controls in communicative abilities, specifically in initiation, effectiveness and partner sensitivity (but not disinhibition). This indicates that patients with ALS do have difficulties in social communication related to pragmatic skills, and that they are mostly unaware of such difficulties.

Other studies have adopted a broader approach to communication, considering how people with ALS participate in a conversation and acknowledging aspects related to dysarthria and augmentative and alternative communication (AAC) strategies and technologies. As pointed out in Bloch and Tuomainen (2017), we cannot assume that unintelligibility automatically results in limited interaction: despite poor speech sound intelligibility, some people with ALS remain interactive and participative in conversation thanks to AAC. Fruitful insights come from the use of conversation analysis, a qualitative approach that focuses on features of interaction (e.g. turn taking) and problems in talk (Bloch & Wilkinson, 2009). For instance, by using conversation analysis to study the video-recorded natural conversation of two dyads with ALS over a period of 18 months, Bloch and Wilkinson (2009) were able to describe the main problems that recipients of dysarthric turns experience in everyday conversation with people with ALS. In some cases, recipients of dysarthric speech managed to identify the problem in a specific word in a prior turn and to fix it through repair in subsequent turns. In other cases, the problem was more complex, and the recipient could not identify the specific aspect in the dysarthric speaker's turn that was the source of the problem, which resulted in a longer repair sequence. Overall, these studies indicate that conversation in dysarthria is not simply a matter of intelligibility of single words, but rather it involves a fragile understandability, which must be closely monitored and negotiated between the conversational partners. Indeed, evidence also indicates that speech intelligibility does not predict the level of impairment in the interaction in a systematic way, suggesting that conversation contains elements that are not directly related to speech intelligibility but rather concern the patterns of the interaction (Bloch & Tuomainen, 2017).

After seeing a range of pragmatic impairments and how these impairments affect everyday language and conversation, one should ask if all this in turn impacts patients' quality of life. There is indeed evidence, coming from a range of different clinical populations, that pragmatic language disorder and functional communication difficulties affect quality of life (Cummings, 2014). For instance, the APACS total score is one of the predictors of quality of life scores in schizophrenia (Bambini, Arcara, Bechi, et al., 2016a). Studies on more specific aspects of pragmatic competence also highlight a link between quality of life and discourse productivity (Bowie & Harvey, 2008) and between quality of life and metaphor comprehension (Adamczyk et al., 2016; Bambini et al., 2020a). Unfortunately, there are currently no studies of how pragmatics impacts quality of life in ALS. The only evidence of a relationship between language and quality of life concerns speech (Felgoise et al., 2016). By using the speech items of the ALS Functional Rating Scale, a popular measure of ALS severity that includes, among several other domains, the evaluation of speech in a range from 4 (normal speech) to 0 (loss of useful speech), the authors showed that there is a relationship between functional abilities for speech and quality of life (assessed with the ALS-Specific Quality of Life Questionnaire; Simmons et al., 2006). Notably, not only individuals with complete loss of speech but also individuals with early symptoms of speech impairment rated their quality of life as significantly lower than individuals with no speech impairment, indicating that the impact of communicative difficulties on quality of life occurs early in the disease course.

17.6 Future Research Priorities

Several priorities for future research are suggested by this discussion. The first priority is there is a pressing need to undertake more research on pragmatics in ALS and to conduct studies of larger samples of participants. This research should attempt to define more clearly the pragmatic profile of people with ALS, the incidence of pragmatic language disorder in ALS, the progression of pragmatic disorder and the presence of co-morbid symptoms. Further studies are also needed to define the role of pragmatic impairment in the ALS-FTD spectrum. Most studies considered in this review included individuals with ALS who did not meet criteria for dementia. However, there is also evidence of pragmatic difficulties in FTD (especially the behavioral variant), including the domains of discourse (Ash et al., 2006; Rousseaux et al., 2010), non-literal language (Orange & Hillis, 2012), pragmatic inference from scalar terms (Spotorno et al., 2015), humor (Clark et al., 2015) and sarcasm (Rankin et al., 2009). Research is needed to compare the profile of pragmatic language disorder in ALS (distinguishing also its sub-phenotypes) to that of ALS-FTD and pure FTD. This is consistent with the view of Roberts et al. (2017) who argue that “there is a growing imperative to develop cohesive and comprehensive profiles of pragmatic performances in non-AD dementia” which has for a long time been “plagued by disease heterogeneity, complexity of symptoms, as well as a myriad of diagnostic assessment protocols” (p. 348).

To better clarify patterns of pragmatic decline in ALS and in the ALS-FTD spectrum, the use of standardized tools for pragmatic evaluation is desirable. The diagnostic criteria of Strong et al. (2017) recommend a series of tests including the Pragmatic Protocol (Prutting & Kirchner, 1987), the Lille Communication Test (Rousseaux et al., 2001), various picture description tasks such as the Picnic Scene from the Western Aphasia Battery (Kertesz, 1982), as well as the APACS test. The APACS test is reccomended also in De Marchi et al. (2021). We support the recommendation to adopt standardized tests and we also highlight that the use of APACS has several advantages. First, it allows for comparisons with other clinical populations, since the test has been used to describe pragmatic language disorder across a wide range of conditions (Arcara et al., 2019; Bambini, Arcara, Bechi, et al., 2016a; Carotenuto, Arcara, et al., 2018a). Second, the choice of APACS allows for cross-linguistic and cross-national studies and comparisons—a highly relevant issue in the field of aphasia (Beveridge & Bak, 2011; Menn et al., 1996) and pragmatic disorder (Ferr et al., 2012). Indeed, the APACS test, originally developed in Italian, is currently being adapted and validated in a number of other languages (for preliminary data on the Flemish version, see Bambini et al., 2021).

The second priority for future research is that studies of pragmatics in ALS should also assess the impact that pragmatic impairment might have on quality of life. This would make it possible to devise strategies to help patients cope with those pragmatic difficulties that have the most adverse impact on daily functioning and social life. Communicative difficulties in ALS are not just speech difficulties (Bloch & Tuomainen, 2017) and, although we know very little about them, pragmatic difficulties are likely to play a prominent role in the quality of communication and social relationships of people with ALS. Conversation analysis, which provides a naturalistic evaluation of conversation, could nicely complement other assessment tools and help provide a more accurate description of pragmatic symptoms in daily life in order to direct intervention (Bloch & Wilkinson, 2009). We know that the relationship between communication difficulties and quality of life is not linear in ALS and that the early phases of disease have the greatest impact on well-being (Felgoise et al., 2016). It is thus key that communication intervention is provided as early as possible in the course of disease (Woolley & Rush, 2017), that it anticipates, prepares for, and adapts to communicative changes (Judge et al., 2018), and that it includes family members and care providers. Technological aids such as augmentative and alternative communication should be considered too, as part of a complex support process (Judge et al., 2018) that extends beyond unintelligible speech to pragmatic difficulties.

A third priority for future research is that pragmatics in ALS should also be linked with theory. Research in this field not only allows for better clinical assessment and improved treatment for patients and their caregivers, but also sheds light on theoretical issues such as the connections between motor systems (especially pyramidal) and cognitive and language skills that were once considered to be unrelated to motor function. Studies on communication in ALS can thus generate new perspectives in research into human brain evolution and the relationship between motor function and social life in human beings.

17.7 Summary

Today, it is well established that there are language difficulties in ALS, affecting especially grammar and lexical/semantic knowledge. Additionally, there is initial evidence that language difficulties in ALS might also affect pragmatics. Pragmatic language disorder in ALS includes deficits in the content and organization of narrative and other discourse, and impaired comprehension of non-literal meanings, with these difficulties extending to social communication and daily conversation. While data for ALS are limited, it seems that pragmatic language disorder cannot be fully accounted for in terms of executive function and social cognition impairments, although these cognitive domains do show some associations with pragmatics. As pointed out in the consensus criteria (Strong et al., 2017), it is possible that language impairment, including pragmatic impairment, 'may both contribute to the profile of ALS and also occur as part of a mixed cognitive profile that includes executive function impairments or social cognition impairments' (p. 7). As for neural substrates, the available evidence indicates that the disruption of networks in the frontal lobes, specifically the dorsolateral prefrontal cortex and inferior frontal cortex bilaterally, might underlie difficulties in discourse production in ALS. Concerning the receptive aspects, there are no studies investigating the neural underpinnings of pragmatic comprehension difficulties in ALS. Based on what has been reported for other neurodegenerative diseases (Carotenuto, Cocozza, et al., 2018b), it is likely that global pragmatic impairment in ALS is associated with damage to large-scale connections involving both hemispheres and extending beyond frontal to temporo-parietal regions.

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Chapter 18
Huntington's Disease

Charlotta Saldert, Ulrika Ferm, and Lena Hartelius

18.1 Introduction

> */…/ His little fedora isn't on straight.*
> *He jumps and jerks in his chair, as if possessed.*
> *Eyebrows dance beneath a wrinkling forehead.*
> *He smiles and frowns, smiles and frowns, smiles and frowns.*
> *His worried wife now ponders the skeletal frame*
> *of the once plump salesman whose handshake*
> *sold a thousand cars /…/.*
> Thomas D. Bird, MD©, Seattle, Washington
> (Bird, 1994, p. 179)

Huntington's disease (or chorea) is a hereditary, neurodegenerative disorder that results in progressive motor, cognitive, and neuropsychiatric impairments. The disease compromises language abilities and there is evidence that pragmatic aspects of communication are impaired in early stages of the disease. This chapter will first provide a brief description of the etiology and progression of the disease in terms of general motor and cognitive impairments. This is followed by an overview of research findings on speech and language difficulties, as these abilities are considered as basic prerequisites for pragmatic ability. In Sect. 18.4, pragmatic aspects of communication in Huntington's disease are described and illustrated with extracts of interviews and everyday conversational interaction. The chapter adopts an emergentist perspective on pragmatics, as presented by the linguist Michael Perkins. As such, different cognitive abilities and emotion processing involved in pragmatic ability are discussed in Sect. 18.5. The chapter ends with an overview of methods

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used for assessment of communicative ability in Huntington's disease along with a description of current research on intervention.

18.2 Epidemiology and Clinical Features

18.2.1 Etiology and Prevalence of Huntington's Disease

Huntington's disease is caused by an expansion of a cytosine-adenine-guanine (CAG) sequence within the huntingtin (HTT) gene in chromosome 4 (Kay et al., 2017). The mutation results in production of an abnormal protein that causes dysfunction and neuronal cell death in primarily the basal ganglia but also the thalamus and cerebral cortex. The condition has an autosomal-dominant pattern of inheritance which means that there is a 50% risk of transmission of the disease to the next generation. As such, there is a high frequency of affected relatives within a family. In affects both men and women. The disease can become clinically manifest at any age, from early childhood to old age. It tends to develop in middle age, with a mean onset age of 45 years (Pagan et al., 2017). The mean age of onset of so-called juvenile Huntington's disease is younger than 20 years. This form of the disease accounts for up to 10% of affected individuals.

Until the discovery of the mutation causing the condition in 1993, the diagnosis of Huntington's disease was based on motor symptoms and family history (Kay et al., 2017). Nowadays, a gene test can confirm the diagnosis. Structural imaging measures of striatal atrophy (e.g. functional MRI and PET imaging) may also track disease progression even in a premanifest stage of the disease (Ross et al., 2014).

Huntington's disease occurs in all populations worldwide but is more common in those of European ancestry. However, genetic differences and changing demographics result in geographic and ethnic variations in reported prevalence between populations and over time (Kay et al., 2017). Currently available prevalence estimates in Europe vary from 3.95 per 100,000 in Greece to 13.5 per 100,000 in the United Kingdom (see Kay et al. (2017) for a review). It is expected that increasing lifespans, less stigma of diagnosis, and better diagnostic methods will result in a higher prevalence of Huntington's disease worldwide in the future.

18.2.2 Motor Symptoms and Impaired Perception

The movement disorder in Huntington's disease can be divided into two main components (Ross et al., 2014). The first consists of involuntary movements in the form of chorea. Chorea is a type of dyskinesia characterized by involuntary, spasmodic movements especially of the limbs and facial muscles. It begins early and is characteristic of Huntington's disease. The second component involves impairment of voluntary movements, including incoordination, bradykinesia, and rigidity. This tends to dominate in cases of earlier onset of the disease, including juvenile Huntington's disease, and can become more prominent in the late stages of adult-onset Huntington's disease (Pagan et al., 2017; Ross et al., 2014).

As the disease progresses, the movement disorder results in a gait disturbance and imbalance. This leads to falls which contribute to increased morbidity in affected individuals. A motor speech disorder is also common (see Sect. 18.3.1). A swallowing disorder develops progressively and choking becomes more and more common. The primary cause of death in Huntington's disease is aspiration pneumonia, which has been estimated to occur in over 80% of individuals with advanced Huntington's disease (Ross et al., 2014). Eye movement abnormalities have been described and are believed to be related to the development of brainstem dysfunction (Pagan et al., 2017).

There is evidence of impaired perception. Visual cognition and perception have been found to be impaired in early manifest Huntington's disease. Coppens et al. (2019) reported that participants with Huntington's disease performed significantly worse than controls on visual perception tasks, including impaired recognition of animals and objects from sketched silhouettes. Alterations in central auditory processing have also been described. Profant et al. (2017) reported deficits in speech processing in cognitively demanding conditions such as speech in noise. Impaired sound source localization was also found.

18.2.3 Cognitive Decline and Emotional Effects

Although cognitive decline always occurs in Huntington's disease, there is variation in how the cognitive disorder manifests (Ross et al., 2014). Cognitive deficits in Huntington's disease include reduced cognitive processing speed, decreased attention, and impaired mental flexibility and planning. Ross et al. (2014) note that some of the cognitive deficits described in Huntington's disease occur at the intersection between cognitive and psychiatric realms of function. Such cognitive deficits include disinhibition, problems with initiation, and lack of awareness of own deficits (anosognosia). Cognitive deficits can compromise pragmatic communication and will be discussed further in Sect. 18.5.

The common emotional features of Huntington's disease may appear before the motor or cognitive symptoms (Kay et al., 2017; Ross et al., 2014). Depression is common, with symptoms reported in over 50% of individuals with Huntington's disease. Irritability is also frequently present and might be an early symptom along with anxiety. Apathy is another disabling feature. This symptom is present in most individuals by the later stages of the disease and has been correlated with degree of motor and cognitive impairment, while depression and irritability seem to be more variable and independent of disease progression (Thompson et al., 2002).

18.2.4 Progression of the Disease

From the first manifest symptom, Huntington's disease typically progresses over the course of several decades before the death of the affected individual. The development of signs and symptoms of the disease can vary between individuals in terms of onset, severity, and rate of progression. The progress of the functional decline is often specified in different versions of the five-stage scale presented by Shoulson and Fahn (1979). The different stages describe the typical trajectory of decline in the ability to work and the degree of dependence in the performance of basic functions, such as financial management, domestic responsibilities, and activities of daily living.

Since the publication of the Shoulson and Fahn scale, it has become clear that symptoms affecting everyday life may appear before motor symptoms become manifest. Although many variations of the scale exist, progression of the disease is now often described in the following four stages, which will also be used in this chapter: Stage (1) the pre-manifest, asymptomatic, or prodromal stage (including both confirmed gene carriers and first line relatives to persons with Huntington's disease without gene confirmation); Stage (2) an early stage of the diagnosed disease; Stage (3) a stage with moderate disease progression; and Stage (4) an advanced stage of disease progression (Ross et al., 2014).

18.3 Speech and Language in Huntington's Disease

Huntington's disease compromises motor speech production and a range of language skills, including pragmatic language skills. Before examining pragmatic language impairments in Huntington's disease, this section will examine impaired speech production and language in this condition.

18.3.1 Motor Speech Disorders: Dysarthria

> *"I hesitate a lot before I [. . .] talk, because there is no point in talking if you can't, if you don't understand, as I, I only answer and continue to talk and it, it turns out wrong the whole time, the whole time, so there is no point keeping it up the next time either, there is no point."*
>
> (Hartelius et al., 2010, p. 388)

"…it turns out wrong the whole time..." The quote above is an extract from Hartelius et al. (2010). "Effort" and "variability" are recurrent themes in the description of how communication typically changes, particularly concerning the motor speech aspect.

The speech difficulties facing individuals with Huntington's disease reflect their different movement disorders. Early in the disease process, the involuntary, choreatic movements seem to be *superimposed* on the respiratory, phonatory, and articulatory movements of the speech system and do not necessarily disrupt the speech flow. Later, the chorea may *interfere* with the different speech processes, and speech breathing, voice, articulation and prosody are affected in a way that is detrimental to speech intelligibility.

The type of speech disorder is referred to as *hyperkinetic dysarthria* and is generally characterized by variable speech rate, shortened phrase length, and prolongations of inter-word and inter-syllable intervals. Phonation-related changes include harsh and strained-strangled phonation and increased pitch and monotony. Imprecise consonant articulation is also frequently noticed (Duffy, 2013; Hartelius et al., 2003). In a study by Rusz, Klempir, et al. (2014a), it was reported that more than 90% of the 40 consecutively investigated persons with Huntington's disease exhibited some degree of speech impairment. The subsequent acoustic analysis revealed that the speech impairment corresponded to slower articulation rate, decreased number of pauses, imprecise vowel articulation, and excess intensity variations.

If one stops to think about the consequences of the audible speech symptoms again, it is easy to understand their potential effect on everyday communication. Speech is slow, tense and slurred and difficulties are also irregular and unpredictable. These are very challenging conditions, requiring the listener to pay close attention to the speaker in order to understand him or her. The substantial effort that the listener requires precludes simultaneous engagement in other activities, such as everyday household chores. The listener ends up in an either-or situation where conversation is not a natural part of everyday life. It becomes a chore in itself, a task that may require so much effort it may be avoided altogether.

In addition to involuntary, choreatic movements, there are early problems with voluntary movements that affect the planning, initiation, tracing, and termination of movements. These problems are also present in the pre-manifest stage (Kirkwood et al., 2000). The hypothesis that subtle changes in speech motor control may precede the clinical onset of Huntington's disease and that these changes are biomarkers of disease and can be measured acoustically has recurred in the literature over the course of the last three decades. Currently, primarily two lines of investigations are being pursued, one focusing on problems with speech timing (e.g. Skodda et al., 2016; Vogel et al., 2012) and one on voice deficits during sustained vowel phonation (Rusz, Saft, et al., 2014b).

There is large individual variability in type and severity of dysarthria, and symptom development in Huntington's disease. However, speech intelligibility is usually affected in Stage 3 (moderate disease progression) and coincides with incipient swallowing disorders. Later, dysphagia progresses together with respiratory difficulties caused by chest muscle rigidity (Mehanna & Jankovic, 2010).

18.3.2 Language Impairments

Gagnon et al. (2018) conclude that individuals with Huntington's disease may have language deficits in all stages of the disease. In their review of current research in the area, they describe impairments in comprehension and production of sentences and discourse, lexical-semantic deficits, and impaired rule-based linguistic processing. Deficits in narrative discourse, sentence comprehension, semantic processing of action verbs, and verb conjugation have also been reported in individuals who are in the pre-manifest stage.

According to Gagnon et al. (2018), the authors of 24 of the 31 published studies in their review argued that the language deficits described were so-called primary language deficits. The authors of the remaining seven studies instead viewed the language deficits as secondary to motor, perceptual, or general cognitive impairments. Both views may be relevant. However, in an emergent perspective as presented by Perkins (1998, 2005a, 2005b, 2007), the issue of whether an impaired language ability is primary or the result of deficits in other cognitive systems is of less importance. In a genuine communication situation, people depend on their motor and perceptual abilities as well as their cognitive capacity to be able to use language. We will here report findings from research on the effects of Huntington's disease on different core aspects of language as explored in formal test situations and experimental tasks.

18.3.2.1 Impaired Comprehension at Sentence and Discourse Levels

Communication partners report that people with Huntington's disease experience an increasing rate of misunderstandings and impaired comprehension in everyday communication (Hartelius et al., 2010; Saldert, Eriksson, et al., 2010a). This includes reports of less successful inferencing and changes in the understanding of ambiguous meaning. The type and degree of comprehension deficit are often observable in formal test situations. Measures of pragmatic abilities often include tasks where participants' ability to understand metaphors or ambiguous sentences is investigated. Such tasks have also been used in studies of comprehension in Huntington's disease and show reduced abilities compared with matched control groups consisting of individuals without brain damage (Chenery et al., 2002; Saldert, Fors, et al., 2010b).

A reduced ability to comprehend complex logico-grammatical sentences compared with controls matched for gender, age, and education has been reported (García et al., 2018; Saldert, Fors, et al., 2010b). In the study by Garcia and colleagues, impaired grammatical processing was also seen in individuals in the pre-manifest stage. The authors argue that this type of task may be useful as a sensitive bio-marker to detect problems at an early stage.

Comprehension difficulties at a discourse level have also been reported. Compared with controls, individuals with Huntington's disease have been shown to have more problems responding to yes/no questions about both stated and implied information after listening to short stories (Murray & Stout, 1999). Saldert, Fors, et al. (2010b) also reported impaired comprehension of both explicitly stated and inferred information in short narratives. Impaired inference was also seen in the individuals who were in early stages of the disease. Problems with discourse comprehension in Huntington's disease have been linked to cognitive decline as measured by a comprehensive test battery (Murray & Stout, 1999) but not to general cognition as measured by the Montreal Cognitive Assessment (MoCA; Nasreddine et al., 2005), which is a brief screening tool for mild cognitive impairment (García et al., 2018).

People in early stages of Huntington's disease have performed on a par with groups of individuals with left- or right-hemisphere stroke when asked to indicate whether the tone of a voice was happy, sad, or angry in statements and whether sentences were questions, commands, or statements. In their study, Speedie et al. (1990) used audio-recorded sentences where the spoken words were filtered out so that only the emotion could be heard. The authors concluded that this deficit may compromise the ability to understand the more subtle prosodic aspects of communication, which may in turn contribute to social impairment early in the course of the disease. There is additional evidence of impaired ability to process emotional expressions and of effects on social cognition in Huntington's disease (see Sect. 18.5.2).

18.3.2.2 Content and Structure in Narrative Discourse

Short responses and less elaboration of topics have been described as characterizing everyday communication with persons with Huntington's disease. Formal language assessments have also revealed production of shorter and syntactically simple sentences with few embeddings compared with control subjects in analyses of narrative speech elicited in picture description tasks (Gordon & Illes, 1987; Murray, 2000; Murray & Lenz, 2001) and in re-telling in the Cinderella story task (Hinzen et al., 2018). Grammatical errors, including agreement errors, inappropriate use of function words, and missing or inappropriate use of referents have also been reported (Gordon & Illes, 1987; Hinzen et al., 2018; Jensen et al., 2006; Murray, 2000; Murray & Lenz, 2001). Furthermore, Hinzen and colleagues reported more fluency disturbances (e.g. filled and unfilled pauses, prolongations, and repetitions) and impairment in the combination of clauses (e.g. use of coordinate and subordinate clauses) than control subjects in a group of 19 individuals in the early manifest stage of the disease. Nine individuals in the pre-manifest stage were judged to only have difficulties in terms of reference use and combination of clauses.

Due to associations between language measures and measures of cognitive and motor abilities, Jensen et al. (2006), Murray (2000), and Murray and Lenz (2001) suggest that, rather than being a primary language disorder, problems in narrative discourse in individuals with Huntington's disease may be explained by cognitive impairments and speech deficits. The group of individuals in the early manifest stage of the disease in the study by Hinzen et al. (2018) did show impairments in measures of memory and processing speed as well as other aspects of executive functions, while the individuals in the small group of individuals in the pre-manifest stage had scores comparable to those of a control group on the cognitive test battery. Hinzen et al. (2018) also discuss the possible involvement of motor impairments in low results on language tasks. However, they conclude that some linguistic impairments are seen prior to onset of motor symptoms and when standardized neuropsychological test profiles are still normal.

18.3.2.3 Lexical-Semantic Deficits

Word-finding difficulties have been reported in observational studies as well as by persons with Huntington's disease and their communication partners. Disrupted access to semantic knowledge in individuals with Huntington's disease has also been described in experimental studies (Frank et al., 1996; García et al., 2018; Kargieman et al., 2014; Smith et al., 1988). Effects of Huntington's disease on lexical access have been explored in studies using word generation tasks. Ho et al. (2002) studied the performance of 21 individuals, in an early or moderate stage, for an average period of 3.5 years. They displayed a reduction in production of correct words over time, and also a significant increase in word repetitions. The impairment seems to affect both object and verb generation (Lepron et al., 2009; Péran et al., 2004).

There is also some evidence of individuals in the pre-manifest stage of Huntington's disease having a selective impairment in action semantics (Kargieman et al., 2014; García et al., 2018). The deficit has been described as a linguistic deficit, unrelated to measures of executive function or cognition in general.

18.3.2.4 Rule-Based Language Processing

Due to the degeneration of neurons in the striatum in the basal ganglia in Huntington's disease, affected individuals have often been included in studies exploring the role of procedural memory in rule-based language processes. Ullman et al. (1997) and Ullman (2001) describe procedural, or declarative, memory as dependent on fronto-striatal structures in the brain and involved in the rule-based computations of phonemes, morphemes, and words stored in the mental lexicon. Results from experimental studies indicate that individuals with Huntington's disease do present with difficulties in word-rule extraction as well as morphological and syntactic rule application (see Gagnon et al., 2018 for a review). Impaired rule-based language processes have been suggested to underlie some of the symptoms of language impairment seen in Huntington's disease. Effects of morphological impairments on noun inflection have also been demonstrated in individuals in the pre-manifest stage (Nemeth et al., 2012).

18.4 Pragmatic Aspects of Communication in Huntington's Disease

18.4.1 An Emergentist Perspective on Pragmatic Impairment

As Huntington's disease progresses over time, affected individuals may present with social disengagement, low conversational participation, and reduced cognitive processing speed, sometimes overlapped with impulsivity and a lack of awareness of own deficits (Ross et al., 2014). Impaired ability to communicate may be explained by the effects of Huntington's disease on motor and cognitive systems as well as neuropsychiatric changes. Furthermore, the affected structures in the brain include the basal ganglia and the thalamus. These are structures that, along with the cerebral cortex, have been considered to be important for language use (Crosson, 1999; Crosson & Nadeau, 1998; Teichmann et al., 2005, 2006; Ullman, 2001; Ullman et al., 1997). Thus, Huntington's disease may have major impacts on communication and pragmatics.

In an emergentist perspective (Perkins, 1998, 2005a, 2005b, 2007), pragmatic impairment is the result of a restriction on the choice of elements available for encoding or decoding meaning. The term "emergence" describes a process in which a "complex" entity comes out of interactions between "lower-level", more basic, entities. It emphasizes that pragmatics is not a discrete entity that exists independently of other entities, such as speech, language, memory, attention, and inferential reasoning. Instead, pragmatics is described as what emerges when such entities come together in a socio-culturally situated human interaction. This view of pragmatics focuses on the processes within the individual as well as between individuals in interaction in a specific context. The emergentist perspective also claims that there is often no direct link between an underlying deficit and a resulting pragmatic impairment or symptom observed in communication. The symptom may very well be a consequence of more or less intentional compensatory adaptations due to impairments in more basic functions.

Both motor abilities and different cognitive abilities are essential for the construction and sharing of meaning. In everyday social interaction, motor problems related to Huntington's disease in combination with impaired cognitive functions, including verbal and non-verbal language, affect the communicative output. This interferes with the communication partner's ability to understand the message as intended. The chorea and cognitive factors, including language comprehension, also prevent the individual with Huntington's disease from being attentive to and grasping the communication partner's verbal and non-verbal contributions.

18.4.2 Pragmatic Aspects of Everyday Communication

> – *What are your thoughts about the disease today?*
>
> – *Today […] That it's not a good thing, I don't think it is.*
>
> – *Because of the movements? Is that the worst part about it?*
>
> – *That's the worst part about it… the movements. And not being able to talk much. Your thinking doesn't work so well.*
>
> – *Ok, it doesn't? So, the way you move your mouth isn't the only thing that has changed?*
>
> – *No*
>
> – *But how you think, as well?*
>
> – *Also how you think, as well.*
>
> (Conversation between Anne, a 43-year-old woman with advanced Huntington's disease and her support person).

The effects of Huntington's disease on natural everyday communication have been explored using both qualitative, interaction-based methods and quantitative methods (see Table 18.1). The lived experiences of changes in the ability to participate in communicative interaction have been described in interviews with persons with Huntington's disease and their communication partners. Using a triangular perspective, Hartelius et al. (2010) interviewed 11 individuals with Huntington's disease along with seven family members and 10 professional caregivers, individually or in focus groups. One of the participating individuals with Huntington's disease was in the early stage, two were in the early-moderate stage, three were in the moderate stage, and five were in the advanced stage. The family members were partners or parents (one), while the professional caregivers consisted of, for example, support and contact persons, home help service personnel, a rehabilitation assistant, an occupational therapist, and a physiotherapist.

In the interviews, variability in communicative ability within the person with Huntington's disease was reported by all participants. The persons with Huntington's disease reported that they experienced participation in communication as demanding and effortful. Examples of problems mentioned in the interaction include losing their train of thought or forgetting what they had wanted to say. They also described word-finding difficulties, and some of them experienced uttering words that were not intended. Furthermore, they felt that they often needed more time than what was allowed by their conversation partners to be able to participate in conversations. Family members and professional carers mentioned changes in speech, but they had also noticed problems with language comprehension and a lack of initiative to communicative interaction. Moreover, they had noted word-finding difficulties and that misunderstandings could be difficult to solve. They also described how their conversations with the person with Huntington's disease were often on more shallow and tangible topics. Both family members and professional carers reported that they had to adjust their way of speaking to the person with Huntington's disease and use different communicative strategies to support communication.

Communicative interaction in, for example, a conversation is typically structured and regulated by conventional rules regarding turn-taking patterns, management of repair when mutual understanding is compromised, and adaptations of level of

Table 18.1 Overview of reported findings of impairments in everyday communication in Huntington's disease

Problems reported in interviews and questionnaires (Hartelius et al., 2010; Power et al., 2011; Saldert, Eriksson, et al., 2010a)		Neurolinguistic analysis of spontaneous speech (Illes, 1989)	Analysis of communicative interaction (Saldert & Hartelius, 2011)
Persons with Huntington's disease	Communication partners		
Word-finding difficulties. Unintended word choices	Word-finding difficulties	Semantic and phonemic paraphasias and neologisms	Word-finding difficulties (pauses and circumlocutions)
Need for more time to be able to participate	Turn-taking affected with latency or omitted responses	Latencies before responses	Affected turn taking: latency or overlapping speech
Losing the train of thought and memory problems	Attentional (losing the train of thought) and memory problems		
Difficulties continuing conversations and following the topic	Short responses and less elaboration of topics	Short, rapid language segments	
	Lack of initiative to communicate		
Difficulties understanding complex information	Reduced comprehension, especially of metaphors and humor, and less successful inferences		
	Topics tangible, less details	Reduced syntactical complexity	Contributions with low level of content words and detail
	Less successful self-repairs and difficulties solving problems in communication	Self-corrections and aborted phrases	Self-repair attempts and need for other-repair
	Adjustment of partner to support communication		Adjustment of partner to support communication
	Impaired speech (articulation, voice, volume and monotonous speech)		Effects of chorea or motor rigidity and dysarthria on interaction

detail and use of reference in narratives (Schegloff, 2007). These rules guide the participants in their comprehension as well as in their planning of their own contributions in the interaction. Most effects of Huntington's disease compromise the participants' ability to adapt to these conventions and structures.

In a case study including a man with Huntington's disease in the advanced stage, Power et al. (2011) used rating scales to describe pragmatic skills and communicative effectiveness. The authors described limited topic selection, with pauses as well as some interruptions and overlap. They also noted reduced quantity of output (short turns and a lack of initiative and elaboration of topics). However, ability to repair and to adapt communication style to type of topic was also reported. Intelligibility and prosodic ability were described as mildly impaired and body language was affected by chorea. The man himself reported that he had problems "*continuing conversations and following the topic*," "*being part of a fast conversation with other people talking*," and "*understanding complex information*." His reports were consistent with professional carers who knew him, with the exception that they rated his ability to "*describe things in detail*" and "*start conversations with others*" lower than he did himself.

In another study, a parent, a spouse, and a support person rated the degree of change in their everyday conversations with individuals diagnosed with Huntington's disease using a questionnaire (Saldert, Eriksson, et al., 2010a). The persons with Huntington's disease comprised a young man in the moderate stage, a man in in his mid-60s in the advanced stage, and a middle-aged woman, also in the advanced stage. The questionnaire was based on the *Conversation Analysis Profile for People with Cognitive Impairment* (Perkins et al., 1997). It explored different aspects of seven areas within communication, i.e. basic language ability, turn-taking, topic management, repair, complex language comprehension, attention and memory, and voice and speech.

Two of the three communication partners indicated some degree of change on all 27 items included in the questionnaire. All three communication partners reported a large increase in omitted responses and latency in turn-taking. This may reflect what the participants with Huntington's disease in Hartelius et al. (2010) described as a need for more time to be able to participate. The number of participants in Saldert, Eriksson, et al. (2010a) was small, but nevertheless the responses mirror the reports of effects on communication presented in Hartelius et al. (2010) and Power et al. (2011). Furthermore, in Saldert, Eriksson, et al. (2010a), changes in the occurrence and use of circumlocutions due to word-finding troubles, reduced comprehension of metaphors and humor, less successful inferences, more attentional failures (e.g. losing the train of thought) and memory problems were also reported.

The questionnaire used by Saldert, Eriksson, et al. (2010a) was supplemented with analysis of video-recorded conversations in the three dyads. The analysis showed how chorea affected the use of language in the woman who was in the advanced stage of the disease. Although her speech in general was quite monotonous, the involuntary motor activity often resulted in involuntary increases in volume during her speech, which affected her use of intonation and pitch to convey meaning. The chorea also had an impact on her use of facial expressions, although she often also smiled and grimaced intentionally in her contributions in the interactions. In contrast, rigidity of movements caused dysarthria and a decreased use of facial expressions in the young man in the moderate stage and the man in the advanced stage. This variation in means for non-verbal communication reflects the development of chorea and rigidity in the disease (Pagan et al., 2017; Ross et al., 2014).

The observations of the communicative interaction were based on 10-minute-long transcriptions of three different conversations in each of the three dyads. The analysis confirmed the communication partners' reports, as well as findings in other research, of word-finding difficulties with pauses, fillers, cut-off words, circumlocutions, and contributions with low levels of content words and detail (see Extracts 18.1 and 18.2).

Extract 18.1: Example of word-finding difficulties with frequent pauses

Extract from a conversation where a man with Huntington's disease in the moderate stage is explaining for his father what is wrong with the size of his window curtains.

`a bit short (1.8 seconds pause) a bit shoddily short on the side otherwise they are (1.9 seconds pause) otherwise they are like a little wider so much shorter would only go down by the window`

Extract reproduced from Saldert et al. (2010a, p. 177).

Extract 18.2: Example of word-finding difficulties with frequent pauses, fillers and a neologism (*smlis*)

Extract from a conversation where a man with Huntington's disease in the advanced stage is telling his spouse that the roof of their summer house is intact.

`there is nothing wrong with it there no- there are no holes or (1.5 seconds pause) there are no eeh (2.4 seconds pause) nothing smlis open (2.5 seconds pause) or eeh`

Extract reproduced from Saldert et al. (2010a, p. 177).

Turn-taking was also affected, either due to long latency, especially for the older man, or due to overlapping speech, especially for the younger man who sometimes seemed reluctant to let his father speak (see Extract 18.3).

Extract 18.3: Example of turn-taking with frequently overlapping and latched speech

Extract from a conversation between a young man with Huntington's disease in the moderate stage (MwHD) and his father on the topic of the young man's experiences during a trip to Gothenburg. (See Appendix for key to transcription symbols).

```
Father:  what do you do in the evenings when you are there
         (0.5 seconds pause) nothing or are you just staying
         in the room lazing around?

MwHD:    a lot in the room and sleep and so

Father:  but when you [are there]

MwHD:                 [are      ] not (0.5 seconds pause) much
         to do before(0.5 seconds pause) Lise-
         (0.5 seconds pause) berg opens
Father:  en no but you=

MwHD:    =will go there later

Father:  yes (0.5 seconds pause) yes of course=

MwHD:    =is close to Carlanderska
```

Father: hhh yes

MwHD: is close to the Swedish exhibition centre up the hill

Father: yes it is easy hhh=

MwHD: =really close

Extract reproduced from Saldert et al. (2010a, p. 179).

Further, there were several instances in the transcriptions of more or less successful self-repair attempts in cases of word-finding difficulties, but also of initiatives to repair by the communication partners in cases of misunderstanding. Latency, a need for repair, lexical-semantic issues, and short and less elaborated responses have also been noted in a study utilizing a neurolinguistic approach (Illes, 1989). Temporal, syntactic, and lexical impairments in spontaneous speech of persons with Huntington's disease were analyzed in audio-recorded responses to open-ended autobiographical interview questions. The language samples ranged from 2 to 8 min for the participants. Five of the ten participating persons with Huntington's disease were in the early stage, while the other five were in the moderate stage of the disease.

Although the lexical deviations seen in Illes' study were quite small, individuals with Huntington's disease produced more semantic than phonemic paraphasias, and the participants in the moderate stage also produced neologisms. Participants in the study were in the early or moderate stage of the disease. But the author refers to other studies (Gordon, 1985; Gordon & Illes, 1987) that include persons in the advanced stage and that have reported more prominent findings of long pauses before producing a response to an interlocutor. The author suggests that these latencies in the beginning of a phrase are due to problems in the planning of a verbal contribution, but they may, of course, also be the result of pure movement initiation difficulties. Furthermore, a reduction in syntactic complexity was demonstrated in spontaneous speech, and as this was also seen in participants in the early stage, it was argued that it could not be explained by an adaptation to motor speech problems.

Measures of time in relation to number of words produced in Illes (1989) revealed that the individuals in the early stage of the disease tended to produce short, rapid language segments. The author discusses whether this may be related to the co-occurring motor speech disorder or whether it was an adaptation to impaired language and other cognitive abilities. In line with an emergentist perspective of pragmatics (Perkins, 2007), Illes argues that this may be an adaptation to, for example, memory problems and word-finding difficulties. She proposes that the rapid speech may sometimes be a strategy used in an attempt to complete the contribution before losing the train of thought or forgetting the successfully retrieved lexical item. Still, Illes also suggests that a likely explanation is that although the initiation of speech may be a difficult process (just as in the cogwheel phenomenon seen in Parkinson's disease), the speech in itself may be rapid once started.

The analysis in Illes' (1989) study concerned spontaneous speech production in participants with Huntington's disease. Effects on communication have also been studied in video-recorded naturally occurring *interaction* between affected individuals, in the moderate or advanced stage, and their communication partners (Saldert, Eriksson, et al., 2010a; Saldert & Hartelius, 2011). It has been shown that what may at first be perceived as a symptom of the disease may actually work as a resource and a more or less deliberate strategy that enables active participation in communicative interaction. Saldert and Hartelius (2011) presented an analysis of the interaction in natural conversations between a middle-aged woman, Anne, with advanced Huntington's disease and her support person. Although Anne sometimes did produce longer phrases, her speech had been described as occasionally characterized by echolalia in her medical records. Echolalia is a common symptom both in the progression of dementia and in other conditions such as autism (Stengel, 1947; Wetherby et al., 2000). In the field of medicine, echolalia has been defined as a non-communicative and involuntary repetition of words or utterances spoken by another person (Ford, 1989; Schuler, 1979). However, the analysis of Anne's repetitions showed that they were highly functional.

Anne had dysarthria with imprecise articulation along with word-retrieval problems and impaired comprehension. Despite this, she took an active part in conversations. In the analysis, the support person often adapts her contributions to Anne's communicative ability by wording questions or suggestions that can be responded to with a simple "yes" or "no." However, Anne utilizes the words produced by the support person in a way that makes it possible for her to participate and to produce more than minimal responses. Anne uses re-cycling of her communication partner's words to perform repair and produce feedback, but also to extend the topic at hand (see Extract 18.4). In doing this, she also often attempts to vary the prosody to elaborate on the topic and sometimes she modifies the communication partner's utterance by adding an attribute.

Extract 18.4: Example of re-cycling communication partner's words

Extract from a conversation between Anne and her support person (SP) on the topic of effects from working with boat motors. (See Appendix for key to transcription symbols).

```
SP:    did you get your fingers filthy=

Anne:  =yes

SP:    m

Anne:  you did

SP:    yes

Anne:  very dirty
```

Extract reproduced from Saldert and Hartelius (2011, p. 256).

This use of repetition is also common in ordinary conversations between non-brain-damaged individuals (see, for example, Tannen, 2007). However, in this case it illustrates how communication partners may cooperate to compensate for impairments in speech, language, and other cognition seen in Huntington's disease.

In summary, although the number of studies and study participants is small, there are several forms of evidence that Huntington's disease affects pragmatic aspects of everyday communication. In an emergentist perspective, the symptoms noted in communicative interaction may be related to both motor and cognitive problems, including language, as well as to adaptations made by the affected persons and their communication partners. In the next section, we will present results from research on deficits in cognitive abilities and emotional perception in Huntington's disease. These deficits can also compromise pragmatic aspects of communication.

18.5 Cognitive Decline and Emotion Processing

Active participation in functional communicative interaction is dependent on intact cognitive functions, such as memory and executive functions, and an ability to recognize and use verbal and non-verbal expressions of different emotions and other states of mind. Cognitive decline in general neuropsychological functions has been characterized in all stages of Huntington's disease, including before the first motor symptoms appear. Besides emotional problems such as depression, apathy, anxiety, and irritability (see Paulsen et al. (2017) for a review), there is also evidence of an emotion processing deficit in Huntington's disease.

18.5.1 Neuropsychological Symptoms

The cognitive impairment has been described as a cardinal feature of Huntington's disease, although the type of impairments varies (Pagan et al., 2017). Decreased processing speed, attentional deficits, verbal learning and verbal fluency impairments, and memory disorders in terms of short-term memory have been described as early signs of Huntington's disease (Butters et al., 1978; Lemiere et al., 2004; Lundervold et al., 1994; Solomon et al., 2007). The ability to search and retrieve information from long-term memory may be severely impaired even in early stages. Aspects of executive functions, such as planning and cognitive flexibility, are affected during disease progression. Decline of executive functions has been found to be related to the progression of the pathology in the basal ganglia, but the degree of deterioration of executive functions in Huntington's disease varies (Holl et al., 2013). Some aspects of executive processing might be affected in the early stages of the disease, while others are intact.

18.5.2 Social Cognition and Emotion Processing

Social cognition and Theory of Mind (ToM) are concepts that have been utilized in the description of the cognitive impairment seen in Huntington's disease and may explain difficulties in pragmatic aspects of communicative interaction. Problems with ToM tasks have been described in individuals in the pre-manifest stage (Eddy & Rickards, 2015). Individuals diagnosed with Huntington's disease may have apparent difficulties in evaluating negative emotions and understanding statements that are sincere, sarcastic, or "paradoxical sarcastic", that is, an utterance that can only make sense when interpreted as sarcastic (Philpott et al., 2016).

As an aspect of social cognition, the ability to recognize facial expressions has been explored in-depth in Huntington's disease and impairments have been reported in all stages (Bora et al., 2016). Problems recognizing facial expressions of especially negative emotions, i.e. fear, disgust and anger, have been demonstrated in numerous studies in individuals in both pre-manifest and various manifest stages (see, for example, Baez et al., 2015; Ille et al., 2011; Johnson et al., 2007; Kordsachia et al., 2017; Snowden et al., 2008). Impaired recognition of both positive and negative emotional signals has also been reported in studies using non-verbal emotional vocalization stimuli (Robotham et al., 2011). It is not clear to what extent this may be related to impaired cognitive decline in general, although links to measures of various neuropsychological functions have been reported (Bora et al., 2016). Furthermore, in a study of eye movements, individuals who were in a pre-manifest or early stage of Huntington's disease spent less time than controls looking at the regions of the face important for the identification of emotional expressions, e.g. eyes, nose, and mouth (Kordsachia et al., 2018). Based on these results, the authors concluded that the emotion processing deficit may be explained partly by a general disease-related decline in cognition and motor functioning and partly by a social-emotional deficit, which is reflected in reduced eye-viewing.

Problems with emotion recognition have been related to theories of embodied cognition, suggesting that perceiving an emotion involves somato-visceral and motoric re-experiencing (de Gelder et al., 2008; Trinkler et al., 2017). Impaired motor control may thus lead to a decreased recognition and understanding of non-verbally expressed emotion as well as an impaired ability to make oneself understood when trying to express personal feelings. Still, deficits in the production of facial expressions of disgust have been seen in cases of spared ability to imitate the facial movements and the presence of adequate motor control (Hayes et al., 2009). Trinkler (2016) argues that having difficulties in recognizing emotional expressions in another individual and in voluntarily expressing one's own emotions does not mean that the ability to understand the emotions in self and others has to be impaired. The conceptual understanding of emotions does seem to remain relatively intact in Huntington's disease. Although subtle empathy deficits have been reported (Baez et al., 2015), individuals with Huntington's disease usually do perform normally on empathy questionnaires and are able to identify and describe emotions in self, as measured by alexithymia questionnaires (Trinkler, 2016; Trinkler et al., 2017).

Impaired recognition of emotions has been noted across a variety of domains, including facial expressions in isolation, in vocal expressions and in short, verbally based vignettes (Calder et al., 2010). However, numerous studies have reported that *type of stimuli* seems to affect the ability to recognize expressions of feelings. Verbal or contextual clues, e.g. prosody, facial movements, and body language in video-recorded vignettes, have been shown to support emotion recognition in individuals with Huntington's disease, individuals in a pre-manifest stage, and in controls (Aviezer et al., 2009; Baez et al., 2015; Rees et al., 2014; Trinkler, 2016; Trinkler et al., 2013, 2017). This is important to remember in interventions targeting affected individuals and their communication partners.

18.6 Assessment of Communicative Ability in Huntington's Disease

18.6.1 Clinical Guidelines and Frameworks

The European Huntington's Disease Network (EHDN) Standards of Care provides clinical guidelines for the management of speech, language and communication difficulties in Huntington's disease (Hamilton et al., 2012). Early and comprehensive assessments are recommended to obtain baseline measures for the evaluation of progression of symptoms or effects of interventions. Besides assessment of basic areas in speech and language, the EHDN Standards of Care recommend an assessment of the affected individual's communication. Such assessment should include an evaluation of: (1) the strengths and needs in the individual's communication skills; (2) the individual's use of communication in their current environments; (3) the communication partner's communication skills; and (4) the impact of the communication difficulties on the individual and their environment, including emotional, psychological, and psychosocial aspects. It is also suggested that in the future, "communication and social interaction, rather than speech and language as isolated phenomena, will be of main concern in clinical practices" (Hamilton et al., 2012, p. 75).

The World Health Organization's International Classification of Functioning, Disability and Health (ICF; World Health Organization, 2001) is a biopsychosocial framework that may be used as a base for systematic assessments of individuals' health conditions. The ICF framework proposes that, besides a focus on impaired body functions, clinical management of individuals should also include assessment and intervention focused on what tasks and activities the person undertakes and wants to participate in in everyday life. The ICF model is adopted in many clinical guidelines used by speech-language pathologists. Despite this, an online survey of speech-language pathologists working with clients who have progressive dysarthria (including Huntington's disease in 37% of them) showed that a large proportion of respondents reported predominant use of impairment-based methods in their assessments (Collis & Bloch, 2012). According to the respondents, the reason for this was a lack of the necessary tools to target activities like communicative interaction in assessments and interventions. This is also true for the assessment of language and pragmatic abilities in general (Frith et al., 2014; Saldert, 2017).

The EHDN Standards of Care do not provide any concrete suggestions regarding suitable tools and instruments to use in assessment but recommend that assessment should involve both formal standardized tests and informal methods such as checklists and observational protocols. Further, it is stated in the guidelines that methods for measuring changes in speech, language, and communication will need to be developed and evaluated. In the next section, we present a brief review of instruments and methods used in the assessment of pragmatic impairments and functional communication in Huntington's disease.

18.6.2 Assessment of Speech, Language, and Pragmatic Aspects of Everyday Communication

Several instruments exist for the diagnosis and assessment of Huntington's disease. Many include items that directly assess speech, language and communication. The Unified Huntington's Disease Rating Scale (UHDRS; Kieburtz and Huntington Study Group, 1996) is currently the most commonly used clinical and research tool for diagnosis and assessment of progression in Huntington's disease (Ross et al., 2014). This scale comprises motor, cognitive, behavioral, emotional, and functional components. It also includes a rating of speech, but there is no measure of language or communication ability. The motor assessment includes a subscale focused on dysarthria, which is an assessment of speech intelligibility, where the alternatives range from normal to mute and cover "clear, no need to repeat," "must repeat to be understood," and "mostly incomprehensible." The inter- and intra-rater reliability of this subscale has been investigated by Zraick et al. (2004) and has been found to be excellent for both medical experts and speech-language pathologists using the scale. Although valid and reliable, the information gained from the UHDRS is unsatisfactory as a basis for analysis of the motor speech component in Huntington's disease.

Picture description and confrontation naming tasks, such as the *Boston Naming Test* (Kaplan et al., 1983), can be used to examine lexical-semantic ability in Huntington's disease. However, Gagnon et al. (2018) raises methodological issues involved in interpreting results from these types of tasks. Naming pictures involves different processing steps, like visual perception and recognition of the pictured object, as well as activation of the semantic representation and lexical retrieval. Analysis of spontaneous discourse in storytelling, interviews, and conversational interaction is time consuming, but probably provides a more valid measure of pragmatic ability in everyday communication than formal assessments (see Saldert (2017) for a review of assessment of pragmatic ability).

There are numerous established and standardized tests that can be used to assess basic cognitive abilities (see Gagnon et al. (2018) and Paulsen et al. (2017) for reviews of measures used in Huntington's disease). However, Philpott et al. (2016) argued that when it comes to impaired emotion processing and social inference, which may have a very negative impact on interpersonal relationships, it is important to assess it clinically with ecologically valid tools such as *The Awareness of Social Inference Test* (TASIT; McDonald et al., 2003). This instrument provides a measure of ability to recognize emotional expression, for example, in facial expressions and video-recorded vignettes.

Although pragmatic deficits and communication difficulties may be the result of impairment of core cognitive functions, these problems may only become apparent when language is used in conversational interaction or some other more complex language-dependent task. Conversational interaction places high demands on the integration of different cognitive functions, such as memory and executive function. What qualifies as an appropriate utterance or response is highly dependent on the context of the communication and there are also requirements of continuous activation and integration of stored world knowledge, flexibility, and inhibition. More subtle deficits cannot be detected using instruments that measure only core functions in language or other aspects of cognition (Hamilton et al., 2012; Saldert, 2017; Saldert, Fors, et al., 2010b). Instead, test batteries including more complex tasks, in terms of demands on the integration of language abilities and other cognitive functions, have been developed and used.

One example is the *Test of Language Competence* (TLC; Wiig & Secord, 1989). The TLC measures so-called high-level language functions in five subtests. The TLC and tasks from a Swedish test battery with similar subtests, *Bedömningsmaterial för Subtila Språkstörningar* (BESS; Laakso et al., 2000), have been used in the assessment of individuals with Huntington's disease in early to advanced stages (Chenery et al., 2002; Saldert, Fors, et al., 2010b). However, although these types of instruments may detect more subtle language impairments, the normal distribution of scores in a non-brain-damaged population is often large and results need to be interpreted in the light of the examiner's perceptions and experiences of change in language ability.

Pragmatic language skills in Huntington's disease may also be assessed by means of rating scales. Power et al. (2011) assessed the use of language in a 15-min unstructured conversation by a man with advanced Huntington's disease using the *Pragmatic Protocol* (Prutting & Kirchner, 1987). The protocol is a rating scale consisting of a taxonomy of 30 items related to different behavioral aspects of communicative interaction. Each item is rated as "appropriate" or "inappropriate" or noted as "no opportunity to observe". Acceptable levels of inter-rater reliability have been reported (Prutting & Kirchner, 1987). The rating scale was used in both individual and group conversations. Speech intelligibility and pragmatic skills were evaluated. For the latter assessment, the *Communicative Effectiveness Index* (CETI; Lomas et al., 1989) was used. CETI was developed for the assessment of functional communication in aphasia. In this case, the man with Huntington's disease, six nurses and two speech-language pathologists rated his communication effectiveness using a modified 20-item version. Each item was rated on a 5-point scale, ranging from "never able" to "always able." Furthermore, semi-structured guided interviews were conducted with the man and his mother.

The involvement of communication partners is often required in the assessment of pragmatic aspects of communicative ability. The results in an interview study that included individuals with Huntington's disease and their family members and professional carers reflect these different perspectives on the communicative interaction (Hartelius et al., 2010). The *Conversation Analysis Profile for People with Cognitive Impairment* (CAPPCI; Perkins et al., 1997), which was developed for assessment of cognitive impairments in dementia, consists of a structured interview that is conducted with a key communication partner of the person with a communication disorder. The interview is supplemented with a method for the analysis of a sample of the participant's everyday communication. With CAPPCI, the participants' perception of various aspects of communication is explored using 26 questions divided into four main areas: (1) linguistic abilities; (2) repair; (3) initiation and turn taking; and (4) topic management. A questionnaire developed from the questions in CAPPCI has been used in a study of pragmatic aspects of everyday communication in three individuals with Huntington's disease (Saldert, Eriksson, et al., 2010a). However, as with most available instruments used for the assessment of pragmatic ability, these methods are difficult to standardize and validate due to the variable and context-dependent nature of the phenomena they are aiming to measure.

Increasingly, speech-language pathologists are concerned to assess the impact of communication and swallowing problems on the quality of life of people with Huntington's disease. Carlozzi et al. (2016) developed a patient-reported outcome measure in order to assess the impact of difficulties with speech and swallowing on health-related quality of life in Huntington's disease. The outcome measure consists of 47 items and was evaluated in a study in which 507 individuals with Huntington's disease indicated their agreement with statements and items. Based on factor analyses, two different scales were developed, corresponding to difficulties perceived in speech (27 items) and swallowing (16 items). The outcome measure can be used in clinical assessment of difficulties in these areas, both in Huntington's disease and in other populations.

18.7 Intervention

There is no cure for Huntington's disease (Roos, 2010; Cruickshank et al., 2015; Deb et al., 2017) and the effects of medical and nonmedical treatments need further attention (Deb et al., 2017). Yet, many things can be done to support the affected individual and family (Nance, 2012). The individual may live with the disease for many years, and different kinds of support are relevant at different times depending on symptoms, disease progression, and the person's individual needs.

There is little evidence and poor documentation of interventions specifically targeting speech, language, and communication in Huntington's disease (Bachoud-Lévi et al., 2019; Beukelman et al., 2007; Bilney et al., 2003). However, like other types of intervention, speech-language pathology intervention should follow the changes that occur with the progression of the disease and take into consideration the complexity of the disease in terms of how different symptoms are interwoven and interact with personal, social, and environmental factors.

Pharmacological intervention targeting motor and psychiatric functions should be based on the person's functioning in daily life (Roos, 2010) and can have a positive effect on communication. For example, pharmacological intervention targeting behavioral changes such as apathy may have positive effects on the individual's interactions with other people (van Duijn, 2017). Besides pharmacological intervention, music therapy has been thought of as beneficial regarding the quality of life and communication of individuals with Huntington's disease, but there is yet no evidence of favorable effects compared with group recreation therapy (van Bruggen-Rufi, Vink, Achterberg, & Roos, 2017a; van Bruggen-Rufi, Vink, Wolterbeek, et al., 2017b).

Speaking is strenuous for individuals with HD and affects other people's understanding (Hartelius et al., 2003, 2010; Klasner & Yorkston, 2005; Power et al., 2011). Although there are currently no published studies on interventions targeting people with Huntington's disease, interventions targeting speech are similar to those used for other types of hyperkinetic dysarthria. They focus on prosodic aspects of speech including speech rate and phrasing. More relaxed and better coordinated breathing and phonation together with shorter utterances may make speaking easier and have positive effects on articulation, prosody, and intelligibility (Duffy, 2013). Giddens et al. (2010) published an intervention study describing a home training program focusing on respiratory, phonatory, and oral-motor exercises as an adjunct to pharmacological treatment. Both retention and improvement of function were shown after a 2-year period. Even when focusing on specific motor speech functions, the overarching goal should be to achieve optimal communication and increased participation in daily activities (Collis & Bloch, 2012; Killoran & Biglan, 2012; Power et al., 2011; World Health Organization, 2001).

Collis and Bloch's (2012) survey revealed that the degree to which different interventions were given by speech-language pathologists working with people with progressive dysarthria in the UK varied with both the type of underlying disease and the degree of severity of the dysarthria. Communication and social interaction were valued as more important in cases of severe dysarthria, and there was a stronger focus on interaction strategies with patients who had Huntington's disease than with patients who had Parkinson's disease, for example. Development and adaptation of augmentative and alternative communication (AAC) resources is a common speech-language pathologist intervention in cases of dysarthria (see Box 18.1). Outcomes of AAC interventions in Huntington's disease needs to be further explored (Diehl & de Riesthal, 2019). There may be specific challenges related to progressive impairments in visual perception, cognition in general, and the movement disorder, but this is not unique to Huntington's disease.

The use of Talking Mats (Ewing & Murphy, 2017; Murphy & Boa, 2012) has been investigated in Huntington's disease. Talking Mats, which supports both comprehension and expression, may enable people with Huntington's disease and cognitive impairments to share their views and wishes regarding activities, communication, personal care, and accommodation at present and in the future. There is both a low-tech and a digital version of Talking Mats, each involving three sets of pictures representing a topic, a number of different questions/options, and a

Box 18.1 Augmentative and Alternative Communication (AAC)

AAC resources such as (a) pictures and text, (b) schedules, calendars and notebooks, (c) identification cards and communication passports, (d) topic and activity boards and books, as well as (e) software, web applications, and computer access adaptations can supplement personal meetings, conversation and the performance of daily activities for people with communication disorders (Astell et al., 2010; Ekström et al., 2017; Killoran & Biglan, 2012; Klasner & Yorkston, 2001; van Walsem et al., 2016; Yorkston & Beukelman, 2007). It is also important to consider remote communication and the fact that participation in society involves the use of various online services. Efforts to ensure that people with cognitive and communicative disability are not excluded are warranted (Buchholz et al., 2018; MacKenzie et al., 2011; Scholz et al., 2017). Social media, e-mail, text-messaging, etc. are main means of communication with friends and family and need to be assessed and supported, as part of and in addition to AAC resources (Paterson, 2017; van Walsem et al., 2016). The purpose of intervention is to find the aids, methods, and strategies that can support the perceived communication needs in daily life (Power et al., 2011; van Walsem et al., 2016).

visual evaluation scale. Open questions and pictures are used together, e.g. How do you manage cooking? + picture of cooking, and the person is asked to place the picture of cooking under the picture in the visual scale that best matches their opinion.

Talking Mats has been shown to stimulate interaction and increase communicative effectiveness in dyadic as well as multiparty interactions involving people with Huntington's disease (Ferm et al., 2010; Hallberg et al., 2013). It has also been utilized to support self-care and participation in consultations where understanding and remembering the information given by different professionals may be difficult for the individual. For example, oral and dental health is an area where extra support with communication may be needed and where Talking Mats has been satisfactorily used (Ferm et al., 2012). The patients rated their communicative involvement higher when Talking Mats was used, and the dental hygienist felt that the method supported the intervention.

It is important to remember that all parties in the interaction situation are affected and consequently need to be involved in the intervention (Hartelius et al., 2010; Murray & Stout, 1999; Saldert, Eriksson, et al., 2010a). The communication partner may be a resource in the communication. For example, in cases of problems with emotion recognition, research has shown that the synthesis of verbal or contextual clues, e.g. prosody, facial movements, and body language, may offer valuable support (Aviezer et al., 2009; Baez et al., 2015; Rees et al., 2014; Trinkler, 2016; Trinkler et al., 2013, 2017). Thus, communication partners of individuals with Huntington's disease may be trained in supporting communication by being explicit and by using several different channels when expressing themselves. In communication partner training, communication partners learn to adapt and use functional communicative strategies. Training in the management and use of various aids and AAC resources is also important (Ferm & Saldert, 2013; Kent-Walsh & McNaughton, 2005). There is evidence of positive outcomes of communication partner training in dementia and aphasia, although no studies have yet focused on communication partner training in Huntington's disease (Eggenberger et al., 2013; Simmons-Mackie et al., 2016).

Information and education about the disease is central to all intervention in Huntington's disease (Braisch et al., 2016; Duncan et al., 2007; Hartelius et al., 2010; Klager et al., 2008; Murray & Stout, 1999; Piira et al., 2014; Veenhuizen et al., 2011; Williams et al., 2007, 2013). Families approach Huntington's disease in many ways (Brown Moskowitz & Rao, 2017), and the fact that the disease is hereditary and may or may not be known within the family complicates the situation and calls for openness from health care professionals and other experts. Knowledge about the disease is of particular importance considering the need for communication support and caregiving in the home environment and at end of life (Ferm et al., 2018; Klager et al., 2008). Information about speech and communication to the affected individual and family is also often prioritized by speech-language pathologists (Collis & Bloch, 2012).

Due to the complexity of the symptoms and the duration of the disease, the care team around a person with Huntington's disease needs to be multidisciplinary in nature. It typically involves a large number of professionals who collaborate with the individual and the family in order to set functional goals and develop personal treatment and care plans (Brown Moskowitz & Rao, 2017; Killoran & Biglan, 2012; Klager et al., 2008; Phillips et al., 2008; Power et al., 2011; Veenhuizen et al., 2011, 2018; Zinzi et al., 2007).

The standards of care model developed within the European Huntington's Disease Network has been applied in coordinated multidisciplinary team programs (Veenhuizen et al., 2011, 2018), and speech-language therapy is one of the core services (Simpson & Rae, 2012). The patient is offered a multidisciplinary assessment twice a year, including assessments of speech, language, and communication by the speech-language pathologist. Other psychosocial functions where communication plays a significant role, and which are influenced by Huntington's disease, such as parenting (Ferm et al., 2018; Klager et al., 2008), are also assessed. The team assessments serve as a basis for shared decision making on intervention goals and care plans, and patients, caregivers, and professionals appreciate the coordinated multidisciplinary approach (Veenhuizen et al., 2011, 2018). Similar goal-directed and collaborative intervention is exemplified by Power et al. (2011), who used semi-structured interviews and the ICF framework (World Health Organization, 2001) to plan and implement communication support.

Positive effects of multidisciplinary rehabilitation have been reported. Zinzi et al. (2007) carried out an intensive rehabilitation program with 40 patients in the early and middle stages of Huntington's disease. Besides training of oral and written language as well as respiration and speech, the intervention included physical and occupational therapy along with attention and memory exercises. The treatment was given both individually and in groups and had positive effects, as measured by tests, on motor performance and activities in daily life. There was no decline in motor performance over a period of 2 years, which is noteworthy considering the progressive nature of the disease. Patients and caregivers experienced positive effects on speech, swallowing, mood, apathy, and social relationships (Zinzi et al., 2009). Positive comments from the caregivers included, for example, more knowledge of Huntington's disease and increased empowerment and quality of life. The patients reported improved self-esteem and self-worth as well as a decline in depression. Meeting other people was valuable. Multidisciplinary training focusing on verbal, physical, and cognitive functioning has also been shown to have positive effects on grey matter volume and verbal learning and memory (Cruickshank et al., 2015).

Klager et al. (2008) and Brown Moskowitz and Rao (2017) stress the importance of effective communication in the care and support of patients and families experiencing Huntington's disease at end of life. Brown Moskowitz and Rao provide a concrete list of how to behave when entering a room of a person in the advanced stage of Huntington's disease. The list clearly exemplifies the importance of language and body communication. As far as possible, socializing with other people should be encouraged and supported, as should the patient's abilities to share their views and wishes regarding activities, communication, personal care, and accommodation at present and in the future.

Living with a parent who has a neurodegenerative disease such as Huntington's disease may be challenging for a child (Sparbel et al., 2008). It is important for a child to maintain a relationship with the parent, but changes in cognitive and communicative functions make this difficult. It is also a right of the person who is ill to be a parent and care for their child (cf. Power et al., 2011). The children and adolescents in the study by Ferm et al. (2018) understand the parents' need for assistance at home, but communication with the parent and the assistants is a problem, as is communication between the parent and the assistants, according to the children.

18.8 Summary

Huntington's disease is an autosomal-dominant hereditary, neurodegenerative disorder resulting in progressive motor, cognitive, and neuropsychiatric impairments. The disease occurs worldwide but is more common in populations of European lineage. There is as yet no cure for the disease, which usually leads to death within two decades of symptom onset. Evidence from observation studies and reports from people with Huntington's disease and their carers have shown that problems in both motor speech and language use affect pragmatics and everyday communication. In an emergentist perspective, different core functions within the individual and also between communication partners emerges into an individual's pragmatic ability in a specific context. Cognitive impairments impact comprehension of more complex sentences and in discourse. Turn-taking problems along with difficulties in topic management may be the result of lexical-semantic issues and reductions in cognitive processing speed, but the motor speech disorder also causes response latencies. Besides the dysarthria, the movement disorder also affects non-verbal language use.

During progression of the disease everyday communicative interaction may be restricted by less initiative and short, less elaborated utterances, often with an insufficient level of detail, and speech becomes more and more effortful and often difficult to understand. This leads to an increased need for repair in conversations and also avoidance of communicative interaction. The communication partner needs to understand the source of the difficulties and be prepared to take the responsibility for adjustments and adaptations to support the communication. In assessments of the pragmatic aspects of communicative ability in Huntington's disease, the focus should be on language use at a discourse level and in interaction. Still, any inferences regarding communicative ability must also be based on the status of core functions such as language, cognition in general and speech. Existing barriers and resources in environmental factors, for example, the communication partner's skill in supported communication, must also be considered. There is a lack of evidence-based communication interventions in Huntington's disease, although a multidisciplinary approach has been shown to be beneficial. There is a need for more research on the outcome of augmentative and alternative communicative support and training of communication partners of people with Huntington's disease. What we do know, however, is that these types of interventions have been proven successful in cases with similar communication problems.

Appendix: Transcription Key

⌈ ⌉ ⌊ ⌋	Large brackets link an ongoing utterance with an overlapping utterance
=	Latched utterance with no gap between the adjacent utterances
but-	A dash indicates a cut-off
(0.6)	Pauses or gaps in tenths of a second within parentheses

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Chapter 19
Traumatic Brain Injury

Jacinta M. Douglas

19.1 Introduction

This chapter focuses on pragmatic language disorders in the context of traumatic brain injury (TBI). TBI refers to brain injury acquired through a traumatic event such as a motor vehicle crash, assault, or fall resulting in an altered state of consciousness and diffuse damage to the brain (National Institute of Neurological Disorders and Stroke, 2019). The mechanisms whereby damage occurs as a result of blunt trauma to the head are complex and give rise to multiple, interactive, pathological processes in the brain. These processes cause extensive changes including diffuse axonal injury (DAI) and microvascular damage as well as focal effects (contusion and haemorrhage) in the context of widespread neural excitation and metabolic changes (Povlishock & Katz, 2005). Overall, damage occurs most frequently in the frontal and temporal lobes, the hippocampus, corpus callosum and the basal ganglia. Recent diffusion tensor imaging data has also supported the importance of these regions in the context of communication outcomes following TBI by demonstrating that frontotemporal white matter microstructural integrity is associated with social communication abilities in adults with TBI (Rigon et al., 2016).

TBI is the most common cause of disability in young people, with an annual incidence in most Western communities of 150–250 individuals per 100,000 population (Roozenbeek et al., 2013). Globally, it is expected to surpass many diseases as the major cause of disability in 2020 (Hyder, 2007). The complex neurobehavioural effects of TBI result in a range of cognitive, communication, personality, behavioural, psychological and social consequences which disrupt the quality of life of survivors and their families over many years (Roozenbeek et al., 2013). TBI typically affects young people (mostly men aged 18–35 years) at the beginning of their working lives. Those who sustain moderate and severe injuries face the

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challenge of long-lasting deficits necessitating intensive rehabilitation efforts and ongoing support to facilitate community integration. The majority of people with severe TBI require long-term support due to loss of independence, reduced work capacity and relationship breakdown (Access Economics, 2008; Hyder, 2007).

Impaired communication is a well-established consequence of TBI. As many as 70% of those with TBI report difficulties with communication (Ponsford et al., 1995), including motor speech deficits (Jaeger et al., 2000; Wang et al., 2005), word-finding problems (Bittner & Crowe, 2006; Hoofien et al., 2001), comprehension difficulties (Olver et al., 1996; Vas et al., 2015), and impaired pragmatic ability (Bracy & Douglas, 2005; Channon & Watts, 2003; Douglas, 2010a; McDonald, 1993; Snow et al., 1997, 1998; Turkstra et al., 1995). In addition, although longitudinal studies providing detailed communication data post TBI are rare, outcome research has demonstrated that these communication deficits, particularly in the domain of pragmatics, persist into the long term (Douglas et al., 2019; Matsuoka et al., 2015; Snow et al., 1998). In fact, data from one of our early studies showed that pragmatic deficits continued to be evident in 96% of speakers with moderate-severe TBI who were followed up over a 2-year period, despite having had considerable therapy (Snow et al., 1998). Further, increasing evidence demonstrates that deficits associated with negative social outcomes include those that involve changes in the functional use of language (Douglas et al., 2016; Ryan et al., 2013; Struchen et al., 2008; Struchen et al., 2011).

This chapter begins with consideration of the nature of pragmatic impairment, particularly within the complex array of cognitive dysfunction that emerges as a consequence of the typical underlying neuropathology of TBI. The next section addresses the experience of pragmatic language disorders from the perspective of those grappling with the ongoing and far-reaching consequences of TBI in everyday life. From this perspective, the focus moves to the contribution of pragmatic language dysfunction to outcome for those who sustain TBI with an emphasis on social integration. This focus on social outcome then serves as background to consideration of various approaches taken to the challenges associated with assessment of pragmatic language function following TBI sustained in adulthood. This section provides a broad description of an assessment approach that takes into account the impact of contributing individual and contextual factors and provides the foundation for treatment planning. Methods and measures that have been used to evaluate pragmatic language abilities and strategies associated with effective treatment outcomes following TBI are identified and considered. The chapter finishes with a summary of the content covered.

19.2 The Nature of Pragmatic Language Disorders Following TBI

Pragmatic language competence sits in a complex, multifactorial space characterized by interacting associations with cognitive and psychological functions and social and environmental parameters. Pragmatic skills have been variously described as 'the skills underlying competence in contextually determined, functional language use' (Turkstra et al., 1995); 'the emergent consequence of interactions between linguistic, cognitive and sensorimotor processes which take place both within and between individuals' (Perkins, 2005); and 'the wide range of codified but subtle ways in which language use has evolved in a given culture' (Snow & Douglas, 2017). In their recent review, the Joint Committee on Interprofessional Relations between the American Speech-Language-Hearing Association and Division 40: Society for Clinical Neuropsychology of the American Psychological Association described pragmatic competence as including three aspects of communicative function:

> "(1) the use of verbal and non-verbal communication for social functions such as requesting, greeting, or persuading; (2) the ability to adjust language and non-verbal communication according to the listener, physical, and social context (e.g., conversation with a supervisor versus a chat with friends); and (3) the ability to use context cues to understand verbal and non-verbal communication (e.g., to recognize bragging or lies)." (Turkstra et al., 2017, pp. 1872–1873).

Given the multifaceted nature of pragmatic ability, it follows then that the study of 'language meaning in context' (Cummings, 2013) is inherently multidisciplinary, covering scholarly endeavours across philosophy, linguistics, speech-language pathology, cognitive science, and psychology (Cummings, 2005). Figure 19.1 uses the metaphor of 'a cup of competence' (Snow & Douglas, 2017) to illustrate the complexities of pragmatic competence by conceptualizing its various constituent functions (executive, language and social cognition functions) in the context of individual psychological characteristics as well as social-environmental influences. This conceptualization not only depicts the multiple pathways through which pragmatic competence can be compromised but it also captures the enormous interdisciplinary challenge of understanding and managing pragmatic language disorders, particularly those acquired as a result of the diffuse neurological damage associated with TBI.

Research efforts from the 1980s through to the early 2000s provided a detailed characterization of the specific manifestations of TBI-related pragmatic impairment. Deficits described in this literature include difficulties in meeting the informational needs of the listener (McDonald, 1993; Snow et al., 1997, 1998), lack of logical structure and coherence in discourse (Liles et al., 1989; O'Flaherty & Douglas, 1997), difficulty with implied meaning (McDonald, 1992; O'Flaherty & Douglas, 1997), inappropriate choice of conversational content or topic (Togher et al., 1997a; Snow et al., 1997), inappropriate style of interaction (McDonald & van Sommers, 1993; O'Flaherty & Douglas, 1997), inappropriate change in topic or being tangential (Bracy & Douglas, 2005), and impoverished content (Snow et al., 1997, 1998; Stout et al., 2000). The consequences of such deficits result in conversations that are frequently hard to follow, disconcerting and uncomfortable (Bracy & Douglas, 2005; Coelho et al., 2002) and require increased dependence on the communication partner to maintain the flow of conversation (Coelho et al., 2002; Togher et al., 1997b, 2004). Deficits have been identified in comprehension and

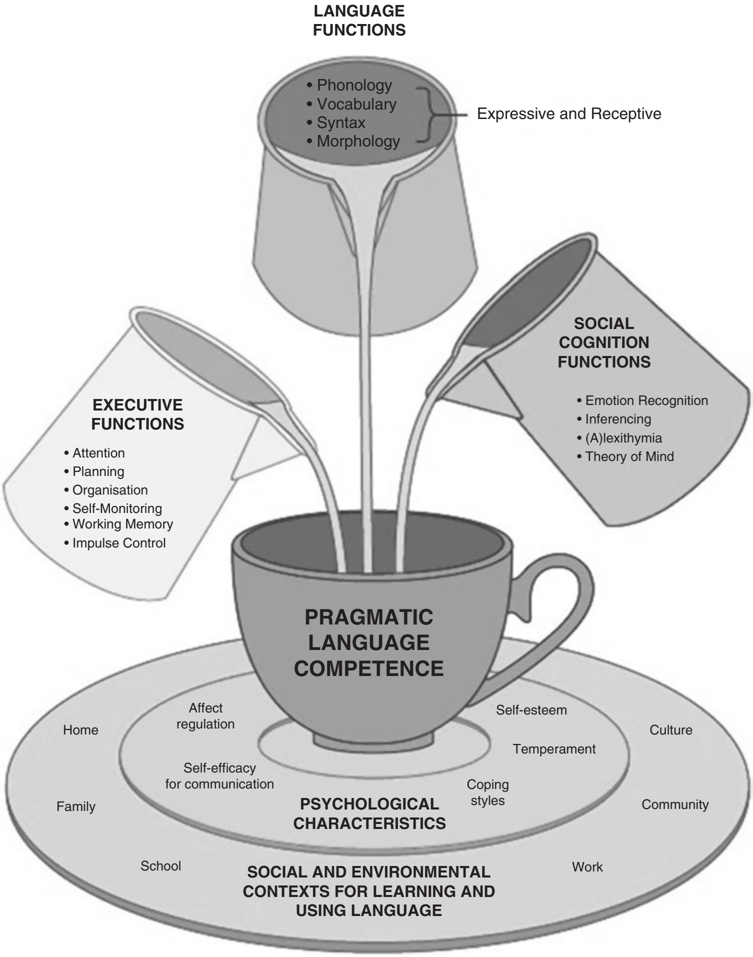

Fig. 19.1 ‘A cup of competence’: constituent functions of pragmatic language competence and psychological and social influences. (From: Snow, P., & Douglas, J. (2017). Psychosocial aspects of pragmatic disorders. In L. Cummings (Ed.), *Research in clinical pragmatics*. Volume 11. Series: Perspectives in Pragmatics, Philosophy & Psychology (pp. 617–649). Cham, Switzerland: Springer International Publishing AG. With permission of Springer Nature)

expression in spoken and written modalities and across types of discourse: conversation, narrative, procedural and expository discourse.

Many researchers conceptualized these pragmatic deficits in TBI as reflecting, at least in part, the impact of cognitive impairments on relatively intact linguistic function (Body et al., 1999; Bracy & Douglas, 2005; Douglas, 2004; Godfrey & Shum, 2000; Hagen, 1984; Hartley, 1995; Martin & McDonald, 2005; McDonald, 1993; Ylvisaker et al., 2001). The cognitive domains most frequently considered by researchers as having significant impact on pragmatic competence are executive function (Bosco et al., 2017, 2018; Byom & Turkstra, 2017; Channon & Watts, 2003; Coelho, 2002; Coelho et al., 1995; Douglas, 2010a; Hinchliffe et al., 1998; LeBlanc et al., 2014a; Martin & McDonald, 2005; McDonald et al., 2014; Matsuoka et al., 2015; McDonald & Pearce, 1998; Pearce et al., 2016; Rowley et al., 2017; Schmitter-Edgecombe & Bales, 2005; Snow et al., 1998; Vas et al., 2015), memory (Hartley & Jensen, 1991; Hinchliffe et al., 1998; McDonald et al., 2014; Schmitter-Edgecombe & Bales, 2005; Youse & Coelho, 2005) and more recently theory of mind (ToM) (Bosco et al., 2017, 2018; Byom & Turkstra, 2017; McDonald et al., 2014; McDonald, Fisher, et al., 2017a; Turkstra et al., 2018). Focus on these domains is logical for several reasons. The frontotemporal regions of the brain and the related circuitry integral to executive, memory and ToM functions are particularly vulnerable to damage as a result of the acceleration-deceleration forces associated with TBI. Accordingly, impairments in these cognitive domains are frequently considered to be among the hallmark deficits encountered by those who sustain TBI. In addition, such deficits clearly have the potential to impact negatively on the functional use of language.

19.3 Relation Between Pragmatic Impairment and Executive Function, Memory and ToM Following TBI

Executive functions can be described as cognitive control processes that include self-regulation, allocation of attention, maintenance and manipulation of information over time, planning and task management (Grafman & Litvan, 1999; Miller, 2000; Rolls, 1999; van Zomeren & van den Burg, 1985). These control processes encompass diverse capacities relevant to pragmatic function, such as initiating and maintaining conversation, inhibiting irrelevant or inappropriate responses, structuring and monitoring task performance, and appreciating multiple perspectives in a situation. Executive functions also enable the efficient deployment of specific skills, such as word retrieval or verbal fluency (Miller, 2000). Deficits may manifest as impaired attention, psychomotor slowing, poor response inhibition, distractibility, initiation difficulties, reduced flexibility, impaired working memory performance (e.g. keeping track of conversations), and difficulties modifying behaviors based on prior experience (Baddeley, 1998; Busch et al., 2005; Muscovitch & Winocur, 2002; Stuss & Alexander, 2005; Stuss & Benson, 1986).

Memory too subsumes multiple functions that impact pragmatic competence (Wilson, 2005). Our interactions with others and across contexts can call upon explicit (conscious, declarative), implicit (unconscious) and emotional knowledge (conscious and unconscious). Our explicit memory includes semantic memory capturing facts and knowledge and episodic memory that supports storage and recall of our personal experiential and autobiographical information. Implicit memory includes our procedural, skill- and habit-based knowledge, while emotional memory can both consciously and unconsciously inform our emotional responses including attraction, fear and avoidance. Memory deficits may present as paucity in content, provision of incorrect information, inability to store new information over time and inappropriate or incomplete contributions to personal exchanges.

ToM is also critical to pragmatic competence. It refers to our ability to understand the mental states of others including their beliefs and desires and to recognize how the mental states of others differ from our own (Siegal & Varley, 2002). It also encompasses the important notion of awareness of false belief, that is, knowledge that another person may have a particular belief that is based on an incorrect assumption. ToM helps us to make sense of the social-relational complexity around us, including acts of deception. It supports our ability to consider how another person is thinking about us and to consider changing our behaviour to improve how we are perceived. Deficits may present as contribution of too little or too much information in a personal exchange, misinterpretation of other people's reactions and inability to recognize the need for conversational repair.

The body of research exploring associations between these cognitive control processes (executive function, memory and ToM) and pragmatic competence following TBI in adults is characterized by substantial variability. Many different neuropsychological assessments and many different pragmatic tasks and measures have been used to index the constructs investigated. As well as diversity in measures, the studies vary considerably on methodological parameters, such as design, sample size and the injury characteristics (severity and chronicity) of participants, which can influence the generalizability of the findings. Overall, 22 studies were reviewed for this section of the chapter. While all included adults with severe injury, some sampled the entire severity continuum from mild to severe (e.g. Channon & Watts, 2003; Youse & Coelho, 2005). Inclusion criteria for injury chronicity also varied markedly within this group of studies. Douglas (2010a) and Snow et al. (1998) assessed participants at least 2 years postinjury in order to sample a relatively stable pragmatic profile. In contrast, participants in Coelho's studies (e.g. Coelho, 2002; Youse & Coelho, 2005) were assessed as early as 1 month to as late as 8.25 years after injury. Finally, small sample size ($n < 30$) with associated compromised power, particularly for the multiple correlational analyses frequently reported, was characteristic of the majority (77%) of these studies.

Notwithstanding these limitations, these studies together provide evidence of significant associations between pragmatic competence following TBI and executive function, memory and ToM, suggesting that deficits in these cognitive domains either co-occur with, or contribute to, pragmatic difficulties in this population. Several studies have used regression or meta-analytic techniques to comparatively investigate correlational patterns. For example, Rowley et al. (2017) focused their exploration on the association between cognitive dysfunction following TBI and pragmatic comprehension or the decoding of implied meaning. They systematically searched the published literature, identifying 10 studies that reported correlations between a range of neuropsychological domains and pragmatic comprehension, and subjected the resultant data to meta-analyses. Results revealed that performance on pragmatic comprehension tasks showed significant correlations with all the cognitive processes included in the review (declarative memory, working memory, attention, executive functions and social cognition[1]). Notably, however, meta-analyses revealed that declarative memory was the strongest correlate ($r = .605$, $p < 0.001$), followed by executive functions ($r = .473$, $p < 0.001$), social cognition ($r = .421$, $p < 0.001$), working memory ($r = .320$, $p < 0.001$) and attention ($r = .291$, $p < 0.001$). The number and magnitude of these correlations support the view that a broad range of cognitive control processes may shape pragmatic competence following TBI.

19.3.1 Executive Function

Douglas (2010a) used standard multiple regression analysis to evaluate the contribution of executive skills to the chronic experience of pragmatic difficulties after TBI. Participants were 43 TBI dyads (adults with TBI and close relatives) and 43 healthy control dyads. All participants with TBI had sustained severe injuries. Their perceptions and those of their relatives were significantly correlated ($r = .63$, $p < 0.001$) and significantly different from those of control dyad participants (F (1, 84) = 37.2, $p < 0.001$). Self and close-other perceptions within the TBI dyads were strikingly congruent with respect to the most frequent pragmatic problems representing violations in three domains of Grice's (1975) Cooperative Principle. TBI participants experienced difficulties with the amount (Quantity) and the relevance (Relation) of information they provided in conversation as well as how they conducted the conversation (Manner). Those with whom they interacted on a regular basis perceived them to have the same difficulties. A tendency for persistent disruption to occur in these aspects of conversational discourse after TBI has also been found based on clinician-ratings using Damico's (1985) clinical discourse analysis (Snow et al., 1997, 1998).

In a study by Douglas (2010a), executive function measures predicted 37% (32% adjusted) of the variance in perceived pragmatic deficits as measured by the La Trobe Communication Questionnaire (LCQ; Douglas et al., 2000, 2007a). Of the three executive function measures used, performance on the FAS verbal fluency task (Spreen & Benton, 1969) was the only one to make a significant, unique contribution to prediction of pragmatic impairment. The FAS task was also the only measure to yield statistically significant associations with perceived difficulties in all four domains reflecting the conversational maxims described by Grice (1975). Snow et al. (1998) also found performance on the FAS task correlated significantly with the number of discourse errors revealed through clinical discourse analysis (Damico, 1985) of conversation samples from speakers with TBI (Snow et al., 1997).

The efficient deployment of specific word retrieval or verbal fluency skills captured by performance on the FAS task clearly taps into a range of executive functions—including self-regulation, allocation of attention, planning, and task

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[1] Social cognition studies used ToM and/or emotion recognition tasks.

management—and it does so in the context of verbal material. Further, phonological fluency tasks, as exemplified by the FAS, are considered to be sensitive to frontal lobe damage (Lezak, 2004), particularly damage to the left dorsolateral prefrontal cortex (Stuss et al., 1998; Szatkowska et al., 2000). In the phonological fluency paradigm of the FAS task, performance also requires inhibition of the more usual meaning-based way of retrieving words and adoption of a retrieval strategy that relies on sound-based lexical representation (Szatkowska et al., 2000).

Other researchers have also found significant associations between inhibitory control measures and pragmatic function after TBI. Channon and Watts (2003) used scores on three nonsocial executive tasks as predictors of pragmatic comprehension. The tasks were the Hayling Test (Burgess & Shallice, 1996), the Six Elements Test from the Behavioural Assessment of the Dysexecutive Syndrome (Wilson et al., 1996), and the Telephone Search While Counting Task from the Test of Everyday Attention (Robertson et al., 1994). These three predictors together gave rise to a significant regression equation accounting for 36% of the variance in pragmatic comprehension. However, the Hayling test error score, measuring inhibitory control, was the only measure that reached significance in the equation. Similarly, McDonald and Pearce (1998) reported a significant association between scores on their disinhibition factor, derived from performance on the Wisconsin Card Sorting Test (Heaton et al., 1993), the FAS verbal fluency task (Spreen & Benton, 1969), and the Rey Auditory Verbal Learning Test (RAVLT; Rey, 1964), and the number of strategies elicited on a request production task. More recently, Pearce et al. (2016) extended these inhibition-related findings when they reported that substantial variance in scores on the LCQ inhibitory control factor (Douglas et al., 2007b) was accounted for by performance on the Hayling Sentence Completion Test and concluded that reduced speed of inhibition may well make a stronger contribution than inhibition failures to prediction of disinhibited communication behaviours following TBI. Overall then, it seems that executive control processes do influence pragmatic competence, but they do not fully account for pragmatic problems after TBI. Indeed, research results to date show that executive function measures leave a substantial proportion of variance (up to two thirds) in pragmatic behaviour unexplained.

19.3.2 Memory

Several researchers have investigated the role of memory in shaping pragmatic competence following TBI. Some studies have focused only on memory as a correlate, while others have included memory in a broader set of correlates examined. Hartley and Jensen (1991) and Youse and Coelho (2005) focused their investigations only on components of memory function using the Wechsler Memory Scale (WMS) (Wechsler, 1945). Hartley and Jensen (1991) reported significant correlations between logical memory and digit span performance with measures of productivity, content and cohesion calculated on story retelling, story generation and procedural description tasks. Youse and Coelho (2005) used the logical memory, digit span and associative learning subtests of the WMS and story retelling and generation tasks. Significant correlations ($r = 0.29$–0.36) were found between digit span and subclauses per T-unit, associative learning and words per T-unit, cohesive adequacy and story grammar (total episodes).

Schmitter-Edgecombe and Bales (2005) found impairment in declarative memory correlated with a reduced ability to infer meaning from textual language. Working memory impairment has also been found to correlate significantly with increasing difficulties in pragmatic comprehension (LeBlanc et al., 2014a). Douglas' (2010a) finding that RAVLT performance was significantly correlated with frequency of conversation difficulties on LCQ items reflecting violations in Grice's (1975) Relation maxim provides further evidence that impaired storage and poorer access to prior linguistic and contextual information contribute to pragmatic problems following TBI. Taken together, empirical results across a number of studies show that declarative as well as working memory deficits make a statistically significant contribution to the prediction of pragmatic problems following TBI. But again, as was the case with executive function measures, memory measures leave a substantial proportion of the variance in pragmatic competence unexplained.

19.3.3 Theory of Mind

A growing body of research has produced evidence that links ToM impairment to TBI and efforts continue to be directed towards understanding the role that ToM plays in pragmatic dysfunction after TBI (Bibby & McDonald, 2005; Channon & Crawford, 2010; Channon et al., 2005; Havet-Thomassin et al., 2006; Martin & McDonald, 2003; McDonald et al., 2003, 2014; McDonald, Fisher, et al., 2017a; Muller et al., 2010; Turkstra et al., 2018). McDonald et al. (2014) examined the role of ToM judgments versus executive function, especially flexibility and inhibition, in producing communication difficulties following TBI. They compared the performance of 25 adults with moderate-severe TBI with that of 28 age-, sex- and education-matched, uninjured control participants on three sets of communication tasks with low executive demands, high flexibility, and high inhibition demands. Within each set, parallel versions of the tasks varied based on low or high ToM requirements. Overall, their findings confirmed that both executive function and ToM made independent contributions to expressive communication skills following TBI. Poor ToM was specifically implicated when there was a strong demand on inhibition. Participants with TBI had differential difficulty with the high ToM version of tasks evidenced through specific problems inhibiting self-referential thoughts in order to accommodate another's perspective. These findings show that taking the perspective of another into account when communicating is a particular challenge following TBI and that this problem is exacerbated when the person with TBI is required to interrupt self-referential thought processes.

In a later study focusing on perception of sincerity in adults with TBI and demographically-matched control participants, McDonald, Fisher, et al. (2017a) found group differences in both non-social and social cognitive processes. Sensitivity to sincerity was moderately correlated with both neuropsychological performance (processing speed and working memory) and social cognitive performance. Subsequent regression analyses further clarified the results. Social cognition performance continued to emerge as a significant predictor of sincerity detection after accounting for variance contributed by age, poor working memory and processing speed and neither mood disorders, group membership nor injury severity made a contribution over and above that of social cognitive performance. Further, when the influence of emotion perception ability was controlled, ToM judgments (the ability of a listener to infer the speaker's communicative intention) predicted sensitivity to insincerity.

Turkstra et al. (2018) also reported evidence of the effect of working memory load on ToM task performance. They compared ToM task performance with varying working memory demands across two groups comprising 58 adults with moderate-severe TBI and 66 age-, sex- and education-matched, uninjured control adults. They found significant main effects for group (higher scores in the uninjured control group) and working memory load (accuracy on ToM questions decreased in both groups with increased working memory demands). These results replicate Turkstra's (2008) previous results. They also reflect those of Bibby and McDonald (2005) that suggested working memory (WM) has a variable influence on ToM task performance that depends on the demands on each in a given task. Turkstra et al. (2018) concluded: "It may be that WM and ToM are so intertwined that we can only observe effects when one or the other is the focus of the task, and cannot truly know the independent contribution of each in everyday social interactions" (Turkstra et al., 2018, p. 120).

As indicated in the preceding review of the role played by executive function, memory and ToM in shaping pragmatic competence following TBI, the number, magnitude and pattern of associations revealed in these studies certainly supports the complex, multifaceted nature of pragmatic language disorders. Research findings also highlight the many ways in which pragmatic function can be compromised as a result of TBI, with its propensity to damage neural systems that are crucial to effective executive, memory and ToM functions. This complex nature of TBI-related pragmatic deficits has not only been revealed in the results of experimental studies within a positivist, quantitative research framework but is also apparent through the insights afforded by constructivist, qualitative inquiries focused on understanding the lived experience of social communication from the perspective of those with TBI.

19.4 The Experience of Pragmatic Language Disorders Following TBI

While incidence and prevalence statistics and quantitative research results effectively shed light on the scope and theoretical nature of the problem, personal description from the insider's perspective powerfully evokes the experience of pragmatic impairment faced by people with TBI as they go about negotiating life in social settings. Over years of working with people with TBI and being privileged to hear their stories, I have listened to many vivid descriptions that expertly capture the challenges of living with pragmatic impairment following TBI. Table 19.1 presents quotes from research participants who outlined their experiences of pragmatic communication behaviours that they perceived as problematic (Bracy & Douglas, 2005; Douglas, 2010c, 2015, 2020; Mackey et al., 2007; Shorland & Douglas, 2010).

These quotes show that people with TBI are frequently well aware of the functional communication challenges they grapple with on a daily basis. Typically, this awareness has grown through repeated social communication failures. Despite knowing that coming up with a topic and starting, participating in, and following a conversation are problems for them, they have had little success in solving these problems. They agree they often say the wrong thing and sometimes go on and on,

Table 19.1 The personal experience of pragmatic deficits following TBI

Personal experience[a]	Pragmatic problems
• *What the hell do I say? I don't know, so I don't speak*	Generating topics
• *I do have problems starting up a conversation … specially when it comes to women*	Initiating conversation
• *I just keep quiet, I have no idea what they're on about* • *I had trouble with continuing a conversation. You say 'hi how are you' and then where do you go from there?* • *Most people can't understand me* • *I'm OK when it's one on one, although sometimes I sort of run out of things to say and then, then sort of the other person doesn't, bring anything new into the conversation, you sort of get stuck*	Following and contributing to conversation
• *I don't say the right thing—I say ridiculous things* • *I say the wrong thing all the time*	Inappropriate comments
• *I go on and on about things, I don't know when enough's enough*	Verbosity
• *I can't seem to pick up even their facial expressions or their voice to know when to say something or even if I should say something*	Reading non-verbal cues
• *I don't seem to show people I'm interested in what they're saying* • *She could tell by what I was saying that I was sincere, but not by the tone of my voice*	Using non-verbal cues
• *You've got to be trying to think about two things, that you're actually doing the actual communication stuff, but then at the same time remember the stuff you're saying* • *I know I've got slow thought patterns, difficulty in word finding sometimes …. being able to comprehend things or assess things quickly in the heat of the moment [is difficult]* • *I get so anxious …. there's so much happening, I never knew* • *…. the relating and talking to people, that's the hardest*	Simultaneous pragmatic, cognitive, emotional and relational demands

[a] Note: Participant quotes from Bracy and Douglas (2005); Douglas (2010c, 2015, 2019); Douglas and Spellacy (2000); Mackey et al. (2007); Shorland and Douglas (2010)

not knowing if, when, and how they should stop talking. Then there is the challenge of the non-verbal domain and feeling both unable to understand and to use facial expression and vocal tone to convey meaning. They clearly capture the complexity of contextually determined, functional language use by highlighting the simultaneous pragmatic, cognitive, emotional and relational demands of social communication in their own words:

> You've got to be trying to think about two things, that you're actually doing the actual communication stuff, but then at the same time remember the stuff you're saying. (Douglas, 2015, p. 206)
>
> I get so anxious …. there's so much happening, I never knew. (Douglas, 2015, p. 207)
>
> …. the relating and talking to people, that's the hardest. (Douglas & Spellacy, 2000, p. 82)

Given the enormous challenges so effectively described in these statements, it is unsurprising that many people with TBI experience social interaction as exhausting, anxiety provoking, and confidence destroying which over time can precipitate a sense of being socially excluded:

> After head injury you know everything just changes and your whole, your whole like social life goes downhill you know, 'cause nobody wants to know you. (Douglas, 2020, p. 13)

19.5 The Impact of Pragmatic Language Disorders on Social Outcome Following TBI

A number of authors have investigated the impact of impaired pragmatic function on social outcome after TBI. Table 19.2 summarises the results of 16 studies that have provided the magnitude of associations between measures indexing pragmatic competence and social outcome. In the most recent of these, Milders (2019) provided an overview of 12 studies and calculated a weighted average correlation across the ten studies that directly investigated the association between perception of social cues and social behaviour and the six studies that directly investigated the association between performance on tasks measuring ability to understand intention and social behaviour. This average correlation essentially represents the effect size of the association between two domains. These analyses revealed the association between social cue perception and social behaviour was consistent with a significant medium effect of 0.35 and the association between understanding intention and social behaviour was consistent with a significant small effect of 0.24.

Of the remaining 15 studies in Table 19.2, five studies (Douglas, 2010c; Galski et al., 1998; Ryan et al., 2013; Struchen et al., 2008, 2011) used regression analysis techniques. Galski et al. (1998) measured pragmatic function with clinician-rated variables derived from narrative, procedural and conversational samples and found 64.5% of the variance in social integration was accounted for by performance on five discourse tasks. The remaining four studies using regression techniques measured pragmatic competence using the LCQ (Douglas et al., 2000, 2007a) and

Table 19.2 Studies investigating the magnitude of association between measures of pragmatic competence and measures of social outcome following TBI

Authors	Year	Sample size (*n*)	Pragmatic function	Social outcome measure	Magnitude of correlation $^{S/NS}$ (shared variance)	Finding
1. Galski et al.	1998	30	Clinician rated discourse variables: narrative, procedural, conversational discourse	CIQ (Social Integration)	Multiple Regression 64.5% ^{S} 5 discourse variables: time; orderings and T-units (narrative); non-repairs and revisions (conversational); words per T-unit (procedural)	Better performance on discourse tasks, better social integration
2. Snow et al.	1998	26	CDA-M: Clinician-rated errors	CHART (Social Integration)	−0.36^{S} (13%)	Better pragmatic function, better social integration
3. Milders et al.	2003	17	Emotion recognition in faces and voices	NBAP (other)	0.2 ^{NS}	Non-significant
4. Dahlberg et al.	2006	60	Self-rated social communication abilities (SCSQ-A): Total Score	CHART-SF (Social Integration and Occupation) CIQ (Social Integration and Productivity)	Social integration—0.43^{S} (18.5%) Occupational—0.13^{NS} Social integration—0.16 ^{NS} Productivity—0.39^{S} (15.2%)	Better social communication abilities, better social integration Better social communication abilities, better productivity
5. Milders et al.	2008	33	Understanding intention	NBAP (other)	0.03 ^{NS}	Non-significant
6. Struchen et al.	2008	121	LCQ (self-rated) Florida Affect Battery	CHART-SF (Social Integration and Occupation)	Hierarchical Regression: Social communication variables ^{S} (8%) Social communication variables ^{S} (5.6%)	Poorer social communication, poorer social integration Increased accuracy of affect perception, better occupational functioning

(continued)

Table 19.2 (continued)

Authors	Year	Sample size (n)	Pragmatic function	Social outcome measure	Magnitude of correlation $^{S/NS}$ (shared variance)	Finding
7. Knox and Douglas	2009	13	Emotion recognition in faces Emotion recognition in social situations	R-CHART—combined Occupation and Social Integration (self-report)	0.65^{S} (43%) 0.69^{S} (48%)	Better interpretation of emotional expression in faces and social situations, better occupation and social integration
8. Douglas	2010	31	LCQ (close other)	R-CHART Social Integration	Multiple Regression: Severity + LCQ 21%^{S}, LCQ unique variance 14%^{S}	Poorer social communication, poorer social integration
9. Struchen et al.	2011	184	LCQ (self-rated)	CHART-SF (Social Integration) CIQ (Social Integration)	Hierarchical Regression: LCQ unique variance 16.3%^{S} LCQ unique variance 11.3%^{S}	Poorer social communication, poorer social integration
10. Ryan et al.	2013	34	LCQ (other) ACS: Emotion recognition in faces and tone of voice	Externalizing Behaviour: ASR (self) ABCL (other)	ASR: 0.51^{S} (26%) ABCL: 0.65^{S} (42.3%) ASR: -0.18^{NS} ABCL: -0.31^{NS}	More frequent social communication difficulty, more frequent externalizing behaviors and poorer emotion perception Reduced social communication mediated the association between poorer emotion perception and more frequent externalizing behaviors
11. Saxton et al.	2013	24	Emotion recognition in faces	KBCI interpersonal difficulties (self)	0.15^{NS}	Non-significant
12. Spikman et al.	2013	51	Emotion recognition in faces	DEX (other)	-0.38^{S} (14.4%)	Better emotion recognition, fewer behavioural problems

13. Ubukata et al.	2014	20	Emotion recognition in faces Understanding intention	R-CHART Social Integration (self)	0.21 ^{NS} 0.10 ^{NS}	Non-significant Non-significant
14. Osborne-Crowley and McDonald	2016	23	Emotion recognition in dynamic faces and body postures	SPRS interpersonal relationships (other)	−0.34 ^{NS}	Non-significant
15. May et al.	2017	40	Emotion recognition in faces Understanding intention	DEX social behaviour (other)	−0.43 ^{S} (18.5%) −0.25 ^{NS}	Better emotion recognition, fewer social behaviour difficulties Non-significant
16. Milders	2019	378 (across 10 studies) 314 (across 6 studies)	Perception of social cues (emotion recognition) Understanding intention	Social behaviour (measures vary across studies) Social behaviour (measures vary across studies)	0.35 ^{S} Weighted average across 10 studies (medium effect) 0.24 ^{S} Weighted average across 6 studies (small effect)	Better emotion recognition, better social behaviour Better understanding of intention, better social behaviour

Note: $^{S/NS}$ designates significant or non-significant finding; *CIQ* Community Integration Questionnaire; *CDA-M* Clinical Discourse Analysis-Modified; *CHART* Craig Hospital Assessment and Reporting Technique; *NBAP* Neuropsychology Behaviour and Affect Profile; *SCSQ-A* Social Communication Skills Questionnaire-Adapted; *CHART-SF* Craig Hospital Assessment and Reporting Technique-Short Form; *LCQ* La Trobe Communication Questionnaire; *R-CHART* Revised Craig Hospital Assessment and Reporting Technique; *ACS* Advanced Clinical Solutions social perception subtest; *ASR* Adult Self Report; *ABCL* Adult Behaviour Checklist; *KBCI* Key Behaviours Change Inventory; *DEX* Dysexecutive Questionnaire; *SPRS* Sydney Psychosocial Reintegration Scale accounted for statistically significant unique variance in social outcome ranging from 5.6 to 42.3%. Ryan et al. (2013) also showed that reduced social communication (LCQ score) mediated the association between poorer emotion perception and more frequent externalizing behaviors in young adults who had sustained TBI during childhood.

The final ten studies used correlation coefficients (*r*) to index the strength of association between measures of pragmatic function and social outcome (Dahlberg et al., 2006; Knox & Douglas, 2009; May et al., 2017; Milders et al., 2003, 2008; Osborne-Crowley & McDonald, 2016; Saxton et al., 2013; Snow et al., 1998; Spikman et al., 2013; Ubukata et al., 2014). Analyses yielded a total of 16 correlation coefficients. Seven revealed significant results: one used clinician-rated discourse assessment; two used a self-rated measure of social communication; and four used performance on tasks of emotion recognition, three in faces and one in social situations. The remaining nine correlation outcomes were non-significant: three measured association between ability to understand intention and social outcome and six evaluated association between emotion recognition (faces, voices, body postures) and social outcome.

Overall, despite some variability across findings, the results of these studies support the proposition that pragmatic function as reflected in self, close-other and clinician ratings has a modest to strong statistically significant association with community integration following TBI and accounts for considerable variance across several measures of social participation and occupational productivity. Further, the statistically significant impact of pragmatic function on social outcome has been reliably demonstrated even after accounting for demographic and injury-related characteristics. While to date, the results demonstrated on specific measures of social cue perception including emotion recognition and understanding another's intention reveal a less reliable finding, evidence suggests that at least a small effect in social outcome is likely to be associated with performance in this domain.

Significant association between pragmatic competence and social participation can be expected given that communication is the means by which we negotiate daily activities and develop and maintain interpersonal relationships. Indeed, ability to use communication effectively in social settings supports a person's sense of connection which in turn underpins psychological wellbeing (Douglas, 2013; Douglas & Spellacy, 2000). Given the pivotal social role of pragmatic competence following TBI, it clearly warrants evidence-based rehabilitation efforts informed by comprehensive and sensitive assessment across the post-injury continuum.

19.6 The Challenge of Pragmatic Language Assessment in Adults with TBI

As we have seen, pragmatic competence is not a unitary static phenomenon. It is an interactive and changing phenomenon reflecting the composite result of the complex interplay between a number of individual factors and multiple contextual parameters. Given that pragmatic performance can be compromised through multiple pathways, it follows that reliable and valid assessment of competence following TBI requires systematic consideration of contributing factors and the extent to which they influence an individual's functional language use (Douglas & Togher, 2017; Snow & Douglas, 2017; Steel & Togher, 2019; Togher et al., 2014; Turkstra et al., 2017). Figure 19.2 depicts individual and contextual factors that warrant attention during the assessment process.

No single tool or procedure is likely to provide an ecologically valid representation of a person's pragmatic competence following TBI (Snow & Douglas, 2000). Indeed, an individualized, collaborative, interdisciplinary approach is essential to capture the social communication profile of an adult with TBI and to document associated activity and participation restrictions for that individual. While formal assessment of pragmatic competence during rehabilitation is generally seen to sit within the professional domain of speech-language pathology (Sander et al., 2009; Wertheimer et al., 2008), it is crucial that health professionals across disciplines are aware of pragmatic language dysfunction and have an understanding of its impact on personal interaction and social activity (Turkstra et al., 2017). Overall, effective assessment begins with the individual. It is important to evaluate systematically how contextual factors shape functional performance and to use measures that have been shown to be both reliable and valid.

19.6.1 Beginning with the Individual

Pragmatic language ability emerges from a melting pot of individual factors. It reflects who we are, where we come from, what we have done, our preferences and style and how we function. Men and women have been found to exhibit differences in pragmatic competence (Douglas et al., 2000), as have young and old adults

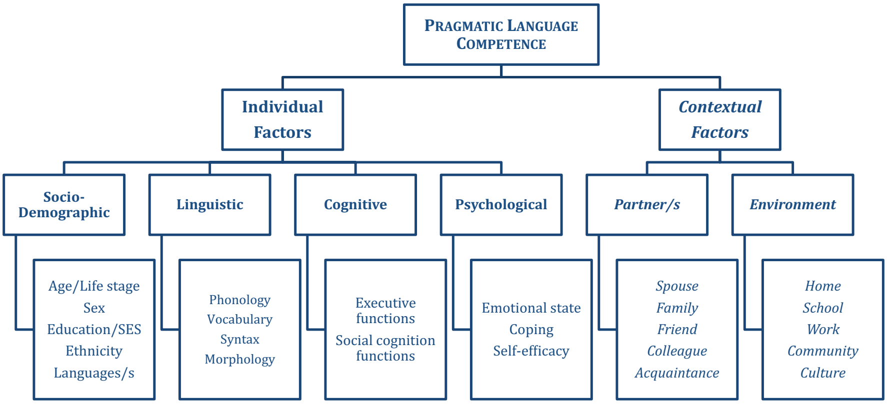

Fig. 19.2 Factors to consider in the assessment of pragmatic language disorders in adults with TBI

(Yorkston et al., 2010) and those with varying levels of education (Snow et al., 1995). Our cultural and ethnic background, socio-economic status and the language/s we speak all shape functional language use across individuals. From an assessment perspective, these personal dimensions need to be considered. Where possible, assessment of individual pragmatic language performance requires comparison with culturally appropriate normative data stratified where appropriate to reflect at least sex and age. Socio-culturally appropriate normative data is also useful for illustrating the extent of deficits and problem behaviours to the person with TBI, their relatives, the clinicians working with them, as well as to those who approve funding for rehabilitation. Unfortunately, appropriate normative data is frequently unavailable, and this lack of normative data is especially the case in developing countries. In these situations systematic individual observation and comparisons with peers and close others can be a useful way of gaining insight into the skills and problems typically demonstrated by the person with TBI.

A large range of standardized assessment measures of specific language and cognitive functions is available, and a number of authors have provided comprehensive descriptions of these measures (e.g. Cummings, 2017; Douglas & Togher, 2017; Honan et al., 2019; Lezak, 2004; Sloan & Ponsford, 2013; Snow, 2013; Strauss et al., 2006; Togher et al., 2013; Turkstra et al., 2005, 2017; Wilson et al., 2017). While standardized assessments provide valuable information about discrete processes, many of these assessment tasks are decontextualized, structured and non-interactive. As a result, reliance on standardized assessment of discrete language and cognitive processes has the risk of failing to identify problems that occur during interpersonal exchanges.

Psychological factors also shape functional communication performance and require attention within the assessment process. Mood state, coping style and self-efficacy can all influence aspects of interpersonal communication including sensitivity to the needs of the conversational partner and the nature of communication-specific coping strategies used in response to communication breakdown (Douglas et al., 2014, 2019). The bidirectional nature of the association between communication problems and psychological function also needs to be recognized in the context of assessment, with evidence suggesting that the presence of acquired communication dysfunction can increase the risk of depression (Davidson & Zhang, 2008; Kauhanen et al., 2000) and operate as a substantial source of stress and anxiety (Douglas et al., 2014, 2019; Karlovits & McColl, 1999).

19.6.2 Taking the Context into Account

Pragmatic competence not only reflects factors specific to the individual, but also varies as a function of context, including the communication partner/s involved and the environment in which the exchange takes place. This contextual variation presents substantial challenges to the assessment process and requires systematic consideration to inform management options. Communication partners have been shown to play a powerful role in shaping pragmatic competence and can scaffold the communication process in ways that can both improve or diminish the effectiveness and equality of the interpersonal exchange (Bogart et al., 2012; Togher et al., 1997a, 1997b, 2013, 2014).

Frequently, communication competence is clinically examined during an interaction with a speech-language pathologist, a relatively unfamiliar communication partner with little shared background and experience to inform the exchange with the person. Information gained from this type of exchange can, therefore, be limited by the contrived nature of the interaction and may not accurately reflect how the person functions in everyday encounters. In contrast, assessment of interaction with a familiar conversation partner, friend or family member more closely approximates not only everyday interactions but also equality of speaker rights and is likely to reflect the person's level of functioning more reliably. In addition, it will enable the clinician to consider partner contributions and responses to the interactive encounter, highlighting potential avenues to treatment that focus on conversation partner training (Togher et al., 2004, 2014). Similarly, the environment in which the interaction takes place will contribute to the demands placed on the person with TBI and the expectations of communication partners. For example, evidence suggests that communication competence in the workplace plays a significant role in influencing return to work outcomes (Douglas et al., 2016; Meulenbroek et al., 2016; Meulenbroek & Turkstra, 2015; Rietdijk et al., 2013) and requires assessment to be tailored to the work role and environment of the individual.

19.6.3 Measuring Pragmatic Competence

As already noted, no single tool provides a comprehensive profile of an individual's pragmatic competence following TBI. A range of assessment tools and procedures that are currently used and have been shown to be effective in delineating pragmatic deficits following TBI are summarized in Table 19.3. These tools cover a variety of methods, including screening checklists, discourse protocols, analysis of transcribed discourse samples, rating scales, self and close-other questionnaires and structured tests. Steel and Togher (2019) have also provided a valuable list of discourse tasks and prompts that have been used with people with TBI to elicit monologic and conversational samples.

Screening checklists, as exemplified by the Cognitive Communication Checklist for Acquired Brain Injury (CCCABI) (MacDonald, 2015), are intended to provide relevant information to support appropriate referral for specialist speech-language assessment. Discourse sampling, transcription and analysis require a considerable time commitment by the clinician. However, such techniques have been shown to be sensitive to the presence of impairment and to treatment-induced change in both primary participants and their everyday communication partners (Mann et al., 2015; Togher et al., 2013; Sim et al., 2013). Discourse protocols support clinicians by compiling standardized elicitation procedures and test batteries and facilitating

Table 19.3 Tools and procedures used to assess pragmatic competence in adults with TBI

Tool/Procedure	Focus	Characteristics
Screening Checklists		
Cognitive Communication Checklist for Acquired Brain Injury (CCCABI) (MacDonald, 2015)	Screening to identify adults requiring assessment of cognitive-communication problems by a speech-language pathologist	• List of concerns in two areas: (1) functional daily communication, and (2) specific functional difficulties • Functional daily communication lists five contexts (family or social; community; workplace; school/academic; and problem solving) • Specific functional difficulties cover auditory and reading comprehension, spoken and written expression, thinking and reasoning
Discourse Protocols		
Mediated Discourse Elicitation Protocol (MDEP) (Hengst & Duff, 2007)	Elicitation of conversation samples in four tasks: conversation; narrative; picture description; and procedural	• Describes sampling procedures to make conversation more balanced and natural (providing active responses, making topics personal and social, and participating in interactional support)
TBIBank (in TalkBank) (http://aphasia.talkbank.org/) (MacWhinney et al., 2018)	Standardized protocol	• Battery of tests and four types of discourse tasks • Website provides guidance for undertaking discourse tasks and includes picture stimuli, instructions and scoring, some norms for a range of analysis types and supporting literature
Conversational Discourse Analysis		
Adapted Kagan Scales (Togher et al., 2010)	Conversational discourse (transcribed) Skills of communication partners providing conversational support	• Two scales: Adapted Measure of Support in Conversation (MSC) and Adapted Measure of Participation in Conversation (MPC) • Intra- and inter-rater reliability established: samples from interactions with adults with TBI • Sensitive to differences in conversational support across partners and intervention-based change (Togher et al., 2013)
Conversation Analysis (Schegloff, 2006)	Conversational discourse (transcribed) Basic organisational structures of interaction (turn-taking, sequence organisation, and repair)	• Detailed analysis of specific communicative patterns in interaction between individuals • Applied to interactions involving people with aphasia, dysarthria, dementia, and TBI (Barnes & Ferguson, 2015) • Sensitive to intervention-based change in TBI population (Mann et al., 2015)

(continued)

Table 19.3 (continued)

Tool/Procedure	Focus	Characteristics
Exchange Structure Analysis (Systemic Functional Linguistics) (Halliday, 1985)	Conversational discourse (transcribed)	• Sensitive to contextual differences (Bogart et al., 2012; Togher et al., 1997a, 1997b) • Sensitive to intervention-based change in TBI population (Sim et al., 2013)
Modified Damico's Clinical Discourse Analysis (Snow et al., 1998)	Conversational discourse (transcribed) Frequency of errors Grices's Cooperative Principle of Conversation (1975)	• Discriminant validity and sensitivity to recovery established (Snow et al., 1997, 1998)
Rating Scales		
Pragmatic Profile of Impairment in Communication (PPIC; formerly PFIC) (Linscott et al., 1997)	Conversation rating: severity of impairment Grices's Cooperative Principle of Conversation (1975)	• 10 feature summary scales • 84 specific behaviour items • Validity examined: TBI population (Dahlberg et al., 2006) • Sensitive to recovery examined (Steel et al., 2017)
Social Performance Survey Schedule (SPSS) (Lowe & Cautela, 1978)	Social competence rating: prosocial and antisocial behaviour	• 100-item questionnaire: 50 prosocial, 50 antisocial behaviours • Discriminant validity examined: established for prosocial behaviours TBI population (Long et al., 2008)
Self and Close-Other Reports		
La Trobe Communication Questionnaire (LCQ) (Douglas et al., 2000, 2007a, 2007b)	Self and close-other report: frequency of conversational problems with optional change comparison (more, same, less) Grices's Cooperative Principle of Conversation (1975)	• 30-item questionnaire • Individual performance can be compared to normative data for self and close-other report and male and female respondents • Comparison of self and other reports provides an index of problem awareness/insight (Bracy & Douglas, 2005; Braden et al., 2010; Douglas et al., 2016; Hoepner & Turkstra, 2013) • Reliability (internal consistency, test-retest) and validity (factor analysis, discriminant) established: normative control (Douglas et al., 2000) and TBI population (Douglas et al., 2007a, 2007b; Struchen et al., 2008) • Sensitive to communication changes in adolescents with TBI (Douglas, 2010b) • Authorised translations completed: Spanish (USA), French (Canada), German, Norwegian, Swedish, Danish, Turkish • Video-based administration developed (Hoepner & Turkstra, 2013)

(continued)

Table 19.3 (continued)

Tool/Procedure	Focus	Characteristics
Structured Tests		
ABaCo (Angeleri et al., 2008, 2012)	Five evaluation scales focusing on separate components of pragmatic competence; uses multimedia and is based on Cognitive Pragmatics theory	• Individual performance can be compared to normative data stratified by age, sex, and years of education, according to Italian National Institute of Statistics • Recently normed on an unimpaired population in English with a focus on politeness markers; regional differences in politeness forms revealed (Davis et al., 2015)
Functional Assessment of Verbal Reasoning and Executive Strategies (FAVRES) (MacDonald & Johnson, 2005); Student version, S-FAVRES (MacDonald, 2016)	Verbal reasoning, complex comprehension, discourse, and executive functioning during performance on real-world functional tasks	• Developed on adults with ABI and a sample of non-injured controls • Individual performance can be compared to normative data for time, accuracy, rationale, and a set of reasoning sub-scores • Reliability and validity: statistically significant differences between adults with ABI and non-injured controls
Montreal Evaluation of Communication (Joanette et al., 2004)	Acute care assessment of non-aphasic communication disorders: prosodic, lexical-semantic, discourse and pragmatic impairments	• Used to examine social communication in adults with TBI in the acute setting (LeBlanc et al., 2014a, 2014b)
The Awareness of Social Inference Test (TASIT) (McDonald et al., 2006; now in third edition)	Social cognition assessment using audiovisual materials	• Three subtests: emotion recognition and two levels of social inferencing • Reliability and validity established: TBI population (McDonald et al., 2006) • TASIT-S Short Form available (McDonald, Fisher, et al., 2017a) with normative data (McDonald, Flanagan, & Honan, 2017b)
Lille Communication Test (LCT) (Rousseaux et al., 2001)	Assessment of verbal and non-verbal communication and gestures	• Systematic evaluation across three domains: participation (greeting, attention and engagement); verbal communication (verbal comprehension, intelligibility, syntax, verbal pragmatics and feedback); and non-verbal communication (understanding and producing gestures, affective expressivity, non-verbal pragmatics and feedback) • Used in rehabilitation and chronic populations of adults with TBI (Rousseaux et al., 2010)
Video Social Inference Test (VSIT) (Turkstra, 2008)	Social inference assessment using video-based materials	• 16 vignettes • Social inferences and explanations of subsequent behaviours

collection of uniform datasets that can increase the efficiency with which research evidence is gathered. Rating scales also produce valuable assessment information, but as already indicated lack of normative data and established sensitivity indices can limit the validity of such scales.

Self and close-other reports provide useful additional perspectives on a person's functioning and have been used effectively as measures of function in the cognitive, neurobehavioural, social and communication domains (Douglas, 2010a; Kreutzer et al., 1996; Prigatano & Altman, 1990). Self-report data are essential because they provide first-hand information about the changes recognized from the person's own perspective. Close others including family members, friends and colleagues/peers from different situations (e.g. work, education/training) also bring important information because they have knowledge of the person's premorbid functioning and spend considerably more time with the person across different and in specific situations than busy professionals. In addition, comparison across self and close-other reports has been shown to be a clinically useful indicator of self-awareness of social communication problems in adults with TBI (Bracy & Douglas, 2005; Braden et al., 2010; Douglas et al., 2016; Hoepner & Turkstra, 2013). Further, comparison of self and close-other ratings of function with appropriate normative datasets can provide valuable information regarding selection of treatment targets.

Structured assessments have a long history of use in rehabilitation and effectively provide important data across the functions that contribute to pragmatic competence. Structured assessments targeting performance on real-world functional tasks (e.g. MacDonald & Johnson, 2005; MacDonald, 2015), social cognition, and social inference using multimedia stimuli (e.g. McDonald et al., 2006; McDonald, Flanagan, & Honan, 2017b) are a relatively recent and valuable addition to the range of tests available to clinicians.

In 2014, Frith et al. (2014) published their review of current assessment practices of speech-language pathologists working with adults with communication impairments following TBI. These authors surveyed clinicians across Australia, Canada, the United Kingdom and the United States of America. They received responses from 265 clinicians describing the areas of communication they frequently assessed and the assessment tools they used. Approximately three quarters of respondents reported assessing the areas of functional communication (79%), receptive (71%) and expressive (70%) language with more than half assessing word-finding skills (63%), high-level language (63%), pragmatic skills (59%) and problem-solving skills (58%). The Functional Assessment of Verbal Reasoning and Executive Strategies (FAVRES) (MacDonald & Johnson, 2005) was the most frequently used tool for assessment of functional performance. The La Trobe Communication Questionnaire (LCQ) (Douglas et al., 2000, 2007a) was the most popular tool in the pragmatic skills assessment category, followed by The Awareness of Social Inference Test (TASIT) (McDonald et al., 2006). While a variety of tools including structured assessments, rating scales, and self and close-other reports were identified in this survey, there was little reported use of conversational discourse analysis in clinical practice. This finding has been previously identified (Coelho, 2007) and may well reflect the combined impact of difficulties in maintaining practitioners' skill levels and prohibitive time demands in clinical practice.

19.6.4 Bringing Assessment Data Together and Planning Intervention

Combining results across multiple data sources is a particularly important principle to apply in the overall assessment of pragmatic competence. The individual is at the centre of the assessment process and acts as the lens through which the assessment results are viewed. Assessment data can be collected from various sources and with a range of tools. Convergence of findings across different data sources strengthens the reliability and validity of results and provides an excellent foundation from which to identify core difficulties, inform treatment planning, measure change and evaluate outcome.

Informed by thorough assessment, the task of working together with the person with TBI to improve pragmatic competence and reduce the impact of functional language deficits begins. This process requires a collaborative approach with the person with TBI at the centre and their close others (family and friends), communication partners from valued life contexts (study, work and leisure), and their health professional team around them (Douglas, 2015). Despite the substantial negative impact of impaired pragmatic competence after TBI, high-quality evidence to inform clinical management of this problem continues to be relatively scarce.

In 2014, the international recommendations for management of cognition following TBI (INCOG guideline) (Bayley et al., 2014) included seven recommendations regarding best practice for the assessment and management of communication disorders following TBI (Togher et al., 2014). Only three are based on evidence from at least one randomised trial with a relevant control group. Evidence currently available supports the effectiveness of context-sensitive interventions embedded in the person's everyday life, communication partner training, and metacognitive strategy training (Douglas & Togher, 2017; Togher et al., 2014). Treatment programs demonstrating application of these factors are exemplified in the work of Togher and colleagues (Behn et al., 2012; Togher et al., 2004, 2013) and our own recent work (Douglas et al., 2014, 2019). This body of work also provides evidence that change in functional outcomes can be facilitated through intervention even for those with severe injury and longstanding problems. Continued research is clearly needed for emerging knowledge to be further developed and translated into practice to make a difference in the lives of people with acquired pragmatic disorders after TBI.

19.7 Summary

Pragmatic language competence makes a crucial contribution to human connection and social wellbeing. Neurological substrates associated with the processes that underpin pragmatic function are particularly vulnerable to the neuropathological mechanisms associated with blunt trauma to the head. Thus, a considerable proportion of people who sustain TBI have substantial and frequently enduring pragmatic language impairments. Pragmatic competence is a complex phenomenon reflecting interaction across cognitive and psychological functions and social and environmental factors. The cognitive domains most frequently seen as contributing to pragmatic competence are executive function, memory and ToM, all of which have been shown to be significantly associated with pragmatic competence following TBI. While these findings highlight the many ways in which pragmatic function can be compromised as a result of TBI, it is important to note that measures of these functions leave a substantial proportion of variance in pragmatic behaviour unexplained.

The complex social interpersonal communication challenges associated with impaired pragmatic competence are well known to people with TBI. Their personal accounts provide powerful renditions of the day-to-day dilemmas they encounter as they navigate conversational exchanges. The impact of these problems is further demonstrated by the findings of studies that have examined the magnitude of associations between measures indexing pragmatic competence and social outcome. The results of these studies show that pragmatic function as reflected in self, close-other and clinician ratings has a modest to strong statistically significant association with community integration following TBI and accounts for considerable variance across several measures of social participation and occupational productivity. Further, this significant impact of pragmatic function on social outcome has been reliably demonstrated even after accounting for demographic and injury-related characteristics.

Given the pivotal social role of pragmatic competence following TBI, it clearly warrants evidence-based rehabilitation efforts informed by comprehensive and sensitive assessment across the post-injury continuum. Reliable and valid assessment of pragmatic competence following TBI requires systematic consideration of contributing factors and the extent to which they influence an individual's functional language use. An individualized, collaborative, interdisciplinary approach is essential to capture the social communication profile of an adult with TBI and to document associated activity and participation restrictions for that individual. No single tool provides a comprehensive profile of an individual's pragmatic competence following TBI and a variety of methods including checklists, discourse protocols and analysis techniques, rating scales, questionnaires and structured tests can be used in the assessment process.

Finally, convergence of findings across data sources strengthens the reliability and validity of results providing a foundation from which to identify core difficulties and inform treatment planning. The task of working together with the person with TBI to improve pragmatic competence and reduce the impact of functional language deficits requires a collaborative approach with the person with TBI at the centre and their close others (family and friends), communication partners from valued life contexts (study, work and leisure), and their health professional team around them. Current evidence supports the effectiveness of context-sensitive interventions embedded in the person's everyday life, communication partner training, and metacognitive strategy training. Intervention has been shown to be effective even for those with severe injury and longstanding problems. A sound foundation exists from which to move forward and continue progress through further quality research.

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Part III
Underserved Populations

Chapter 20
Infants and Children Adopted Internationally

Deborah A. Hwa-Froelich

20.1 Introduction

Children who are adopted internationally (also known as transnational or intercountry adoptions) have different experiences than children who are born to, and reared by their biological parents. These differences can have a profound impact on the children's health and development (for a review, see Rice et al., 2016). For example, many children may have received poor prenatal care, been exposed to substance abuse in utero, and been born premature or small for gestational age (Johnson, 2000). Some children may reside in orphanages run by their country's government, other children may have resided with foster care families, and some children may have experienced a combination of institutional and foster care prior to adoption (Hellerstedt et al., 2008). Dependent upon the country's economic resources, children may experience high child/caregiver ratios and restricted stimulation, social interaction, nurturance, nutrition, and health care (Johnson, 2000; Leiden Conference on the Development and Care of Children without Permanent Parents, 2012). However, children who receive foster care prior to the age of 2 years tend to have better care, nurturance, and stimulation, resulting in better developmental outcomes (Nelson III et al., 2007; Windsor et al., 2011).

Regardless of the type of care children receive prior to being internationally adopted, most begin learning a birth language but are adopted into families that speak a different language. Consequently, they stop listening to, and expressing, their birth language and begin to learn an adopted language (Nicoladis & Grabois, 2002). This interruption in the language acquisition process differs from children who are monolingual because most monolingual speakers do not experience a switch from one language to another. The process also differs from bilingual speakers, because bilingual speakers either receive continuous input of two

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languages from birth or receive continuous input in their birth language and additional input in a second language and are able to use linguistic knowledge of one language to assist in learning a second language. Children adopted internationally (CAI) may not have achieved proficiency in their birth language prior to adoption, their adopted family may not speak the birth language, and there may not be consistent exposure to the birth language following adoption. Consequently, these children stop acquiring their birth language and begin to learn the adopted language at an older age than their monolingual peers. In other words, to catch-up to monolingual peers who did not experience this kind of interruption in monolingual language acquisition, CAI have more language to learn at a faster rate. Thus, the interruption in language learning may have long-lasting effects on the children's language development including social communication and pragmatic language development. The purpose of this chapter is to describe (a) the history of international adoption; (b) the variables influencing post-adoption development; (c) the research on post-adoption hearing, swallowing, speech, language, social communication, and cognitive development; and (d) special clinical considerations for infants and CAI.

20.2 International Adoption History

International adoption is a world-wide phenomenon which has changed significantly over the years. For example, Selman (2015) reported aggregated data from approximately 23 to 27 countries. He reported that over 343,000 children were adopted internationally from 2003 to 2012. However, these numbers of CAI have varied across time. The number of CAI grew steadily from 1950 until 2004 at which time their numbers decreased. In the case of the United States, only 4714 children were adopted internationally to the US in 2017 compared with 22,989 children who were adopted internationally in 2004 (U.S. Department of State—Bureau of Consular Affairs, 2017a). On the other hand, when numbers of CAI to the US are added together from 2000 to 2018, 256,112 children were adopted into U.S. families. Given these numbers, it is likely that education and health professionals will interact with, or serve, some of these children.

Not only has the number of CAI changed, but the countries involved with international adoptions have changed their involvement and adoption policies. For example, intercountry adoptions can be traced back to the 1950s when children were adopted from South Korea following the Korean War (Selman, 2009). From 2001 to 2007, Selman (2009) reported that the countries responsible for the largest number of CAI were China, Russia, Guatemala, and Ethiopia. Currently, the sending countries for the U.S. include Nigeria, Colombia, India, Sierra Leone, Yemen, Zimbabwe, and Laos. In addition, 83 U.S. children were adopted to seven different countries including Austria, Canada, Germany, Ireland, Mexico, Switzerland, and the Netherlands (U.S. Department of State—Bureau of Consular Affairs, 2017b).

The reasons for these changes vary, from countries enacting new adoption laws, promoting domestic adoption of healthy children, having concerns with the abuse, neglect, and exploitation of children by their adoptive parents to using adoption as a political tool. For example, Guatemala stopped all intercountry adoptions in 2007 to develop a process in alignment with The Hague convention issued by the international court for the Protection of Children and Cooperation in Respect of Intercountry Adoption. Haiti and the Democratic Republic of the Congo began new processing procedures which slowed the adoption process, while others like the Ukraine faced political conflict that interrupted the adoption process (U.S. Department of State, 2015). In addition, both China and South Korea decided to promote domestic adoption of healthy children without special needs and international adoption of children with special needs or older children who were not domestically adopted (U.S. Department of State, 2015). China was also concerned that the number of single women who applied to adopt children were not heterosexual and so it added a regulation that only couples that had been married at least 2 years would be allowed to adopt (Selman, 2009). Additionally, after media reports publicized children who were murdered by their adoptive parents, sexually exploited, or abused and neglected, Russia passed legislation in 2013 to stop adoptions to the US (Selman, 2009; Smolin, 2007; U.S. Department of State, 2015). As a result, the number of children available for adoption tend to be older in age and/or have special needs.

20.3 Children Adopted Internationally

20.3.1 An Overserved and Underserved Population

CAI tend to be both an overserved and underserved population. Several researchers have reported that CAI receive more special education and support services than children who did not experience international adoption (Castle et al., 2006; Glennen & Bright, 2005; Glennen & Masters, 2002; Mason & Narad, 2005; Schoenbrodt et al., 2007; Scott et al., 2008). The National Center for Educational Statistics reported that in 2016, the total percentage of children who received special education was 13% (National Center for Educational Statistics, 2016). In contrast, in a longitudinal parent survey involving 130 children who were adopted from Eastern Europe before the age of 30 months, 70% were evaluated by a speech-language pathologist and 35% received speech and/or language services before school-age (Glennen & Masters, 2002). In a subsequent follow-up study of 46 of the 130 participants who were between the ages of 6 and 9 years old, 11.4% were receiving speech and language services, 11.4% had a diagnosis of learning disability, and 25% had a diagnosis of ADD/ADHD, resulting in a total of 54.5% with at least one diagnosis and 59.1% receiving services to support their education (Glennen & Bright, 2005). Scott et al. (2008) also reported that 12.5% of their sample of 24 seven- and eight-year-old girls adopted from China before the age of 2 years, qualified for special education services but 41.6% received tutoring or other kinds of support services other than public education. Thus, both children adopted from Eastern Europe and China had received a high percentage of diagnoses, special education and/or support services. These percentages were significantly higher than the total percentage of children with disabilities (13%) in the US (National Center for Educational Statistics, 2016).

In addition to being overserved, there are clinical reports of children being underserved. Most of the children the author has seen in the International Adoption Clinic at Saint Louis University were either referred or sought services because they were unable to procure services through their school district. The parents of these children have reported several reasons as to why their children do not receive services: (a) school district professionals believed the children were learning English as a Second Language (ESL) instead of learning English as a replacement language and consequently placed the children in ESL services regardless of whether the children demonstrated language learning problems; (b) professionals refused to assess the children, believing they needed more time to learn the language; and (c) professionals recognized the children had developmental delays or problems but conducted limited assessments that did not paint a clear picture of the child's strengths and weaknesses and, consequently, they provided limited and inadequate services. Some case studies have been published and presented (Glennen, 2014a; Hwa-Froelich, 2015a; Hwa-Froelich et al., 2012).

20.3.2 Possible Reasons for Misidentification

One possible reason for misidentification may be misinterpretation of findings from research studies. Researchers have compared CAI with children domestically adopted and with nonadopted peers matched by socioeconomic status (SES) and parent education levels, and compared them to standardized norms. For example, Scott et al. (2011) found that CAI performed better when compared to norms on a standardized test than when compared to a control group, e.g. children who attend the same schools and live in the same neighborhoods. Most parents who adopt internationally have reported incomes of more than 50,000 USD and 70–71% of the parents had a college degree (Hellerstedt et al., 2008). Consequently, their children tend to attend schools with peers from similar socioeconomic backgrounds and their parents tend to have higher academic expectations. When compared to control groups recruited from the same schools and neighborhoods, CAI tend to perform less well than their peers (Cohen et al., 2008; Gauthier & Genesee, 2011; Vinnerljung et al., 2010).

In some research studies, CAI tended to perform similarly to their peers when they were preschool age as opposed to when they were school age (Scott et al., 2011). This change in performance as the children mature may reflect an initial reaction to increased nutrition and stimulation which results in rapid developmental growth. It may also reflect the differences in cognitive, social, and linguistic exposure their peers received because they did not experience the process of international adoption or interrupted language acquisition. Over time, this initial growth spurt may slow or the children may not develop in the same way or at the same speed as their nonadopted peers. In other words, CAI may demonstrate adequate adopted language performance but as the cognitive and linguistic demands increase, they may demonstrate language learning problems necessitating special education services. Future longitudinal studies are needed to determine how to best serve CAI.

Performance may also vary dependent upon the instrument administered to measure the child's performance. In the meta-analysis mentioned above, Scott, Roberts, and Glennen reported that CAI performed better on parent- and teacher-report measures than behavioral measures. Based on the tool used to measure children's development, professionals may unintentionally report that CAI are performing adequately when they may be struggling and may need support. However, the opposite outcome may also be true, e.g. when behavioral measures are administered, the performance of CAI may not reflect performance observed over time.

Another possible reason for misidentification is the complex developmental outcomes that CAI display. Children who have been exposed to adverse early care (poor nutrition, medical care, nurturance, and stimulation), separations from biological parents, relatives, orphanage caregivers, and/or foster care families, and interrupted language acquisition, present a complex picture of strengths and weaknesses that may be interconnected to underlying developmental problems. Foundational neurobiological development may have been limited or over-pruned which in turn may negatively affect later higher cortical development necessary for academic achievement (for a review, see Hwa-Froelich 2012a, 2012b; Ladage & Harris, 2012; Wilson, 2012a). Additionally, without receiving expected and dependent experiences that infants need, the early neurobiological structures and functions may be weakened or missing such that later structures and functions that depend on this foundation may not develop appropriately or adequately for academic achievement (Drury et al., 2011; McLaughlin et al., 2011; Sheridan et al., 2012; Van Ijzendoorn et al., 2005; Vinnerljung et al., 2010).

Children may also have experienced several separations and transitions at early ages (Hellerstedt et al., 2008). During this sensitive period, children do not have the linguistic, cognitive, or social development to understand, explain, and/or cope with these challenges, which can negatively affect attachment with the adoptive parents, future social relationships, emotion regulation, inhibition, and social communication (Hwa-Froelich, 2012a, 2012b, 2012c; Hwa-Froelich, 2015a, 2015b; Wilson, 2012b). McCool describes the effect of maltreatment and trauma on young children in Chap. 22, this volume. In addition, because children must focus their attention and energy on starting to learn a new language, this process may slow their learning in other developmental areas such as cognition, memory, and executive function (Delcenserie & Genesee, 2014; Glennen, 2015; Helder et al., 2016; Juffer & van Ijzendoorn, 2005, 2009). Consequently, children who are internationally adopted often display unusual behaviors and developmental profiles that may not be understood by untrained professionals.

Cultural differences have been reported for children adopted from different geographical areas. Cultural differences exist in terms of the prenatal care mothers receive or do not receive. For example, Johnson (2000) reported differences in prenatal care for mothers who placed children in Eastern European orphanages. In several cases, mothers reported no prenatal care, substance abuse, mental health problems, and multiple previous pregnancies. In China, little to no information was available about the mothers' prenatal care (Miller, 2005). With the One Child Policy in China, parents preferred male infants because sons are expected to care for their parents as the parents grow older. Thus, infants with disabilities and females were placed in public places to be found and taken to an orphanage. However, many of these infants were first-born infants and given to the orphanage so that the parents could try to give birth to a son.

Children may be exposed to different types of care environments, such as institutional care, foster care, or both. These environments may differ in quality of care in terms of nutritional quality and amount, health care, adult-child interaction, and access to developmentally appropriate stimulation for different lengths of time prior to being adopted. South Korea, for example, has one of the oldest, most well-developed systems that employs well-educated staff and provides more foster care for orphan children (Odenstad et al., 2008; Vinnerljung et al., 2010). In a national cohort study comparing IQ and school achievement in adults who were adopted from South Korea, adopted from other countries, and adults who were not adopted, the adults who were adopted from South Korea performed similarly on IQ and school achievement measures as the control group and significantly higher than other adult adoptees (Odenstad et al., 2008; Vinnerljung et al., 2010). Better pre-adoptive care appears to promote better cognitive and academic outcomes.

Linguistic differences in language acquisition patterns may vary. Children adopted from Asian countries may demonstrate different English acquisition patterns than children adopted from Eastern Europe. For example, Hwa-Froelich and Matsuo (2010) hypothesized that English acquisition patterns may differ because of significant linguistic differences between the birth and adopted languages. They compared receptive and expressive language performance between children adopted before age 2 years from Asian countries and children adopted from Eastern European countries. The children were measured at 2 and 6 months following adoption. The two groups did not differ in English language performance 2 months following adoption. After 6 months of English exposure, the Eastern European group had significantly higher expressive English performance, but the Asian children demonstrated more varied receptive language comprehension. The authors suggested that these different performance patterns may reflect either linguistic or cultural differences.

Misidentification of CAI who have special learning needs may be common because few university programs offer educational or clinical training for this unusual population of children and textbooks on child or language development rarely include a chapter on CAI. Speech-language pathologists and other health and education professionals are often unaware that children who are adopted from different countries demonstrate unique developmental outcomes and individual profiles. Consequently, they do not know how to assess or provide intervention services, often missing visual, motor, hearing, swallowing, expressive and receptive language, social communication, verbal memory, discourse, and executive function problems. Although more research is needed to guide professionals who work with infants and CAI, this chapter will present the current research and clinical guidelines for application of this information.

20.4 Hearing, Vision, Speech, and Feeding

20.4.1 Hearing and Vision

Although little research has been done on the hearing and vision of CAI, they are at risk of problems in these areas. Parents and pediatricians often assume that CAI will demonstrate similar behaviors indicative of hearing impairment or ear infection. However, orphanages may not have the funds to provide adequate medical care for respiratory or ear infections (otitis media) so children often do not receive treatment. Thus, the children become accustomed to having infections and demonstrate little to no behavior changes when they have an infection. Consequently, parents are unaware and do not seek medical care, and pediatricians may use ineffective methods to assess hearing (i.e. tuning forks). For example, parents reported that their children who were adopted internationally typically did not have their hearing or vision screened immediately following adoption (Eckerle et al., 2014). Out of 1906 children, 61% had their vision screened and 59% had their hearing screened which means 39% did not receive a vision screening and 41% did not receive a hearing screening. Of the children who did receive screenings, 25–31% had vision problems and 12–13% had hearing problems. The children who had vision problems most often were diagnosed with strabismus, with 36% having multiple abnormalities. The children who failed the hearing screening typically had temporary (otitis media) or correctable hearing loss. Otitis media was reported to occur often during the first year of adoption. These problems have a direct effect on language learning and literacy. Thus, early and consistent hearing and vision screenings are important for this population.

20.4.2 Speech and Feeding

CAI demonstrated little to no problems with articulation of their adopted language in research studies. For example, a large percentage (93%) of 55 children adopted from China performed within 1.25 standard deviations of the mean on an articulation test (Roberts et al., 2005). Their sample was adopted before 26 months of age and was approximately 3 to 6 years old at the time of the study. In studies of children adopted from Eastern Europe, most of the children also performed within an acceptable range and expressed little to no birth language interference on English articulation measures (Glennen, 2007, 2014b). For example, children from Eastern Europe adopted after 2 years of age had standard scores that ranged from 91.64 to 97.82 on an English standardized articulation test after 9 months of English exposure (Glennen, 2014b). Other studies with smaller samples or single cases from other countries, such as Haiti, reported similar findings (for a review, see Hwa-Froelich, 2012c; Hwa-Froelich & Pollock, 2013). CAI appear to learn articulation and phonology of their adopted language quickly and well.

Eating and swallowing problems have also been reported in the literature and in clinical reports (Beckett et al., 2002; Elleseff, 2009; Hwa-Froelich, 2011; Johnson & Dole, 1999). Learning how to eat solid foods is dependent upon infant and toddler feeding experiences. We are all familiar with the cultural variation in types of foods, but little is known about cultural feeding practices. Adoptive parents have described their observations of how children were fed at their children's orphanages. These descriptions included practices of propping bottles of milk in cribs for infants to learn how to feed themselves; extremely hot bottles of milk mixed with gruel given to infants; toddlers fed milk or gruel until they were 2 or 3 years old; and mixing solid chunks of meat in soft food causing some children to gag. The author has evaluated several children at the Saint Louis University International Adoption Clinic with feeding and swallowing problems. These included sensory motor issues with touching certain textures with their hands, lips, tongue, and oral cavity as well as neurological oral motor functions affecting their swallow and velopharyngeal closure.

20.5 Language, Social Communication, and Cognitive Development

There are two common misconceptions about language development in CAI: (a) they are sequential bilingual speakers, and (b) once CAI demonstrate within normal range performance, their development will continue to be within normal ranges of performance like nonadopted children. Both may be false assumptions. From clinical and case study reports, CAI stop speaking their birth language within a few months following their adoption unless parents speak or provide input from other speakers in the child's birth language (Nicoladis & Grabois, 2002; Gindis, 2005). CAI also tend to perform within the average range when compared to standardized test norms or nonadopted peers during their preschool years but lower than average when they are school age (Scott et al., 2011). Other variables to consider when talking about language development of an adopted language include age of adoption and length of adopted language exposure.

20.5.1 Receptive and Expressive Adopted Language Performance

The time it takes for children adopted from different countries to catch-up in their adopted language compared to standardized test norms varies dependent upon the age at which they were adopted. For example, in longitudinal studies following children adopted from Eastern Europe and Asia before the age of 5 years, children performed within average ranges on standardized English assessments in receptive and expressive language after approximately 1 year of exposure (Glennen, 2007; Hwa-Froelich & Matsuo, 2010). Children adopted from Eastern Europe between 3 and 4 years of age, however, had low average scores that fell within the 80–91 standard score range (Glennen, 2014b). It took longer for them to catch-up in terms of mean length of utterance (MLU) configured from language samples. Although standard score equivalents (SSE) of MLUs were within average ranges, these scores were significantly lower than the receptive and expressive standard scores. Glennen (2015) reported that children adopted from Eastern Europe had more difficulty expressing English irregular forms such as irregular plurals and past tense verbs, comparisons, and regular uncontracted plural copula and subject pronouns. Thus, expressive language continued to be delayed after 3 years of English exposure in children adopted from Eastern Europe.

Expressive language samples were collected from children adopted from China before the age of 21 months who were acquiring the French language (Gauthier et al., 2012). The samples were collected during parent-child play sessions when the children were between 42 and 56 months of age. No significant differences were found between the children adopted from China and a control group for MLU, type-token ratio, or total number of clitics. However, they found that the children adopted from China made more complement and direct object clitic errors than the control group and these errors differed from the omission errors that children with language impairment typically make. It is possible that CAI may differ in adopted expressive language performance because of linguistic characteristics between the birth and adopted languages.

Age of adoption was a related variable for receptive and expressive scores at 2.4 months, 9 months, and 1.3 year and months following adoption but not for later measurements (Glennen, 2014b). Age of adoption was not related to MLU SSE. What this evidence tells us is that the adoption age and length of exposure makes a difference in terms of how quickly children can acquire their adopted language to perform at acceptable levels on standardized assessments that are normed on monolingual nonadopted populations, but it does not have enduring effects.

20.5.2 Language Patterns

CAI demonstrate unique patterns of strengths and weaknesses across vocabulary, syntax, MLU, and verbal memory (Delcenserie & Genesee, 2014; Gauthier et al., 2012; Glennen, 2015; Scott et al., 2013). For example, both Roberts et al. (2005) as well as Glennen (2015) have reported that children adopted from China and Eastern Europe attained English receptive and expressive vocabulary scores that were significantly above average test norms. In contrast, however, Gauthier and Genesee (2011) recruited Chinese children adopted before 2 years of age and measured them when they were between 3 and 4 years of age and again approximately 15 months later. They reported above average French receptive vocabulary performance (111.48, SD = 15.52 and 119.22, SD = 17.32) compared with lower than average French expressive vocabulary performance (94.52, SD = 10.69 and 96.87, SD = 11.03). The children from China were significantly different from a French-speaking control group in expressive vocabulary at both time points. Thus, expressive vocabulary, not receptive vocabulary, was an area of weakness for the children adopted from China acquiring French. It may be possible that expressing French vocabulary may be more challenging than expressing English vocabulary for children adopted from China but more research is needed to verify this supposition.

Syntax tends to be an area of weakness for CAI (Delcenserie et al., 2013; Glennen, 2015). In children adopted from Eastern Europe between the ages of 1:0 and 4:11 learning English, they demonstrated lower than average performance on syntactical measures of morphological expression in words and when repeating sentences. These weaknesses were apparent at school age (6- to 7-years-old), but these children were not significantly different at age 5 years (Glennen, 2015). Similar results for recalling sentences and expressive vocabulary performance in children adopted from China learning French before the age of 2 years who were between 7 and 8 years of age were reported by Delcenserie et al. (2013).

Less research is available reporting verbal working memory performance of CAI. Preliminary evidence indicates that short-term verbal memory, which usually involves repetition of words or nonwords, is a possible area of weakness (Delcenserie & Genesee, 2014; Eigsti et al., 2011; Glennen, 2015; Scott et al., 2013). For example, Scott et al. (2013) recruited a group of 6- to 9-year-old children who were adopted from China before the age of 2 years and were learning English and compared their performance to standardized norms. They reported low average phonological memory performance (combined digit span and nonword repetition performance). Eigsti et al. (2011) reported lower word recall performance in a group of 4- to 13-year-old CAI from several different countries between 2 and 86 months of age. Glennen (2015) also reported low average digit span and recalling sentence performance in her 6- to 7-year-old sample of children adopted from Eastern Europe between 1 and 4 years of age but the digit span scores were not significantly different from the standardized norms. In addition, Delcenserie and Genesee (2014) reported findings from a study of 9- to 12-year-old children adopted from China before the age of 2 years who were learning French. Compared with a control group, the Chinese children performed significantly lower on digit span and nonword repetition. Although more research is needed, short-term memory appears to be a weakness for CAI.

In contrast, verbal working memory requires the participant to hold and manipulate information before responding. For example, Delcenserie and Genesee (2014) reported that their sample of children adopted from China performed significantly less well than a control group on backward digit span and a French competing language processing task. However, they also stated that the children's verbal working memory performance was stronger than their short-term verbal memory performance. More longitudinal research is needed to verify these findings and project how these possible areas of weakness might affect later academic outcomes.

Little research has been done with CAI during their adolescence or once they become adults. Preliminary findings indicate that school achievement and language proficiency continue to be weaknesses. In a review of the literature, Norrman et al. (2016) discuss that differences in development can be found in studies of CAI younger than 3 years and CAI between 4 and 11 years of age. They suggest that while CAI demonstrate rapid adopted language learning and acquisition, they do not continue this rapid development during their later school ages. They cited the work of Hene (1988) who reported language outcomes for children adopted prior to age 6 years and measured between 10 and 12 years of age. These children had difficulties with comprehending metaphorical language and mental state vocabulary (e.g. doubt, regard, consider, believe) often found in textbooks.

Norrman and colleagues also described preliminary research comparing four adults who were internationally adopted from Spanish-speaking countries between 1 and 9 years of age and exposed to Swedish for approximately 20 years. They were compared with bilingual Spanish-Swedish speakers who had been exposed to Swedish for about the same length of time as the internationally adopted adults and a control group of monolingual Swedish speakers. Few of the adults adopted internationally performed as well as nonadopted adults on measures of linguistic proficiency. Additionally, speech samples were recorded and played to a panel of judges and only the adult who was adopted at 1 year of age was judged to be native-like. The authors suggest that language proficiency continues to be a weakness into adulthood for CAI. However, given the small sample size, more research on adults who were adopted internationally is needed.

20.5.3 Social Communication

The development of a close social relationship is essential for adequate social and emotional development. It is through consistent, contingent, and collaborative social engagement and interaction that infants learn to trust their caregivers and depend upon their caregivers to guide their interactions with others and the world. Infants develop social communication by associating nonverbal behavior, environmental context, and socio-cultural rules with social understanding and socially competent behavior and communication. Some children receive these expected and dependent social experiences early in life, while others do not which may affect their nonverbal and verbal social communication development.

Nonverbal communication includes interpretation of facial expressions and tone of voice. Several studies provide evidence that CAI have difficulty interpreting emotions from facial expressions and vignettes about emotional experiences (Camras et al., 2006; Glennen & Bright, 2005; Hwa-Froelich et al., 2014; Wismer Fries & Pollak, 2004). For example, in a sample of children adopted from Eastern Europe and Asian countries, Hwa-Froelich et al. (2014) found that regardless of country of origin, the children who were adopted internationally had more difficulty than a nonadopted control group identifying emotions from photographs of facial expressions. They administered the Diagnostic Analysis of Nonverbal Abilities, second version (DANVA-2) which includes subtests of emotion identification using adult and child facial photographs and emotional tone of voice recordings (Nowicki Jr. & Duke, 1994). Although the children had difficulty identifying emotions for both adult and child faces, they had more difficulty identifying emotions of children. The children's structural language performance predicted their emotion identification from photographs.

Hwa-Froelich followed a subset of this sample longitudinally and found that over time, the children improved in their nonverbal abilities. At age 6 years, they did not differ from a group of nonadopted children in emotion identification of adult or child photographs (Hwa-Froelich, 2014). The children's ability to identify emotions by listening to tape-recorded adult and child vocal tones expressed while stating the same sentence in a sad, happy, scared or mad voice was also measured. At age 4 years, the children adopted from Asian and Eastern European countries had more difficulty identifying emotions from tone of voice across both adult and child speakers. By age 6 years, however, CAI had more difficulty identifying emotions from child voices but not adult voices (see Table 20.1 for DANVA-2 error score means and standard deviation). In support of these findings, several parents of children adopted from Asia and Eastern Europe in the same sample as Hwa-Froelich and Matsuo (2019) reported more problems with nonverbal communication when their children were 6 years old than when they were 8 years old (McGownd, 2018). In other words, as CAI are exposed to more social interactions, their social understanding of nonverbal communication seems to improve, but this may be dependent upon their structural language abilities (Hwa-Froelich et al., 2014). More research studying longitudinal social communication in CAI is needed.

CAI may be at risk of weaker pragmatic language skills because they tend to have later developing and weaker expressive language performance when compared to standardized test norms and nonadopted peer groups (Gauthier & Genesee, 2011; Glennen, 2014b; Hwa-Froelich & Matsuo, 2019; Hwa-Froelich et al., 2014). For example, Hwa-Froelich and Matsuo found that 4-year-old CAI performed less well on the pragmatic language subtest of the Comprehensive Assessment of School-age Language (Carrow Woolfolk, 1999) compared to an SES-matched control group of children who were not adopted. The adopted group's standard scores were within the low average range but were significantly lower than the nonadopted group (see

Table 20.1 CASL pragmatic subtest and DANVA-2 means (SDs) by group

Measures	Asian *n* = 17	Eastern European *n* = 10	Adopted group *n* = 27	US Control Group *n* = 27
CASL pragmatic subtest score 4-year-olds	110.60(15.285)	108.40(13.10)	109.657(14.576)	117.394(14.733)
DANVA-2 adult face error score 4-year-olds 6-year-olds	12.294(2.20) 7.353(3.259)	11.40(2.590) 9.01(2.807)	11.963(2.345) 8.0(3.162)	10.778(3.309) 7.148(2.583)
DANVA-2 child face error score 4-year-olds 6-year-olds	11.059(4.458) 4.824(2.834)	11.80(2.670) 6.50(3.689)	11.333(2.922) 5.444(3.215)	9.519(4.042) 4.482(2.486)
DANVA-2 adult tone of voice error score 4-year-olds 6-year-olds	16.941(2.076) 11.942(3.010)	17.00(2.00) 12.80(2.201)	16.963(2.009) 12.259(2.726)	15.741(2.395) 11.667(2.076)
DANVA-2 child tone of voice error score 4-year-olds 6-year-olds	10.059(3.112) 8.370(2.776)	16.90(1.792) 11.30(4.138)	15.963(2.710) 10.519(3.501)	13.519(3.662) 8.370(2.776)

Note: *SD* standard deviation; *n* sample size; *CASL* Comprehensive Assessment of School-age Language (Carrow Woolfolk, 1999); *DANVA-2* Diagnostic Analysis of Nonverbal Abilities, version 2 (Nowicki Jr. & Duke, 1994)

Table 20.1). Although structural language performance explained most of the variance in pragmatic language performance, inhibition performance was significantly correlated with pragmatic language performance. Inhibition, the ability to subdue probable but unsuitable behaviors, was measured by a verbal go/no go task. Because the task was dependent upon following verbal directions of one puppet while inhibiting responses to a different puppet, their verbal abilities may have been a confound in measuring inhibition.

Additional evidence of weaker social communication abilities can be found in parent report data. Parents of CAI have reported lower performance in their children's social communication than parents of children who did not experience the international adoption process (Petranovich et al., 2017). However, Petranovich and colleagues recruited a sample that included children who had received or were receiving speech and/or language services and these children may have performed less well in social communication. In other words, the research on social communication development in this population is emerging. More research is needed to fully understand the effect international adoption has on social communication development.

To provide more context to the kinds of social communication problems children adopted internationally demonstrate, a clinical case study is described. Lena (fictitious name) lived with her biological family and five siblings in Russia until she was 8 years old. At that time, she and two other siblings were removed from their home due to neglect. Lena had attended formal schooling for 2 years before she was removed from her home and school. After she was placed in the orphanage, she no longer received formal education. Instead, she was trained in *life skills* which consisted of learning how to clean and cook. Lena remained in the orphanage for 2.5 years until she was adopted at 11 years of age.

After Lena was adopted, her parents placed her in fourth grade. At school, she received instruction in English as a second language and her parents provided tutoring with a Russian speaker. Lena had difficulty with academic learning and social interactions at school, often preferring to interact with children who were younger than her. Consequently, after fifth grade, her parents decided to teach Lena at home. After working with Lena at home for approximately 2 years, her parents brought her to the International Adoption Clinic (IAC) at Saint Louis University for an evaluation.

Lena was 15 years old at the time of the evaluation. Her parents reported that Lena had difficulty with spelling, word meanings, math, reading, and social skills. The IAC team conducted an evaluation of her hearing, speech, language, literacy, and pragmatic communication skills. Although Lena demonstrated adequate hearing, oral motor structure and function, nonverbal communication, visual memory, expressive language, and phonological awareness, she tended to freeze in social situations, forgetting the names of people she knew, losing the ability to speak, and avoiding social interaction. She also struggled with short-term memory in that she had difficulty remembering simple verbal directions, completing common and simple household tasks, as well as making simple decisions involving choices. Lena's strengths and weaknesses are summarized in Table 20.2.

The intervention plan for Lena was framed around building a secure, trust-based relationship. Clinicians taught Lena strategies to reduce anxiety and to increase a sense of calmness and confidence. Practice having conversations about common topics with her clinician and parents helped to build her confidence. Other clinicians were introduced as well as strangers in the community to increase novelty and stress

Table 20.2 Lena's strengths and weaknesses

Area	Strengths	Weaknesses
Physical function	Hearing WNL Oral Mechanism WNL	
Nonverbal skills	Emotion identification (DANVA2) WNL	
Language	Expressive language	Receptive language/vocabulary Nonliteral, ambiguous, and inferential language
Literacy	Phonological awareness WNL Visual memory WNL L1 influenced spelling errors	Fifth grade level for reading Narrative/expository discourse Drawing inferences
Social communication		Social anxiety affecting memory, flexible thinking for solving problems, and making decisions

Note: *WNL* within normal limits; *DANVA2* Diagnostic Analysis of Nonverbal Abilities, version 2 (Nowicki Jr. & Duke, 1994) and to work on generalizing her skills to other people and contexts. Visual supports such as post-it notes, graphic organizers, and social stories were used to increase short-term and working memory. Lena learned to draw maps visually for common social interactions and create social stories to help her think about what she wanted to say and what other people might expect her to say or do. As Lena gained confidence, her anxiety decreased, allowing her to think more flexibly in social situations.

Other academic areas were included in the intervention plan. For example, Lena worked on auditory discrimination of the English sound system. She was encouraged to compare and contrast these phonemes with Russian phonemes to improve phonological decoding of English words and English spelling. In addition, clinicians worked with Lena's parents on ways to adapt the home school curriculum and added joint book-reading activities to encourage personal enjoyment of reading.

20.5.4 Cognition

Some of the cognitive areas involved in social communication include social cognition and executive function. Social cognition includes awareness, understanding, interpretation, and appropriate reactions to one's own and other's mental and emotional states. Explicit social cognition develops between 4 and 5 years of age (Carpendale & Lewis, 2006; Miller, 2012).

Structural language competence is predictive of social cognition as well as social communication (Carpendale & Lewis, 2006; De Villiers & de Villiers, 2000; Miller, 2012). In support of this, children with language impairment (Farrant et al., 2006) and children who receive less linguistic input such as children who are deaf living with hearing parents (Stanzione & Schick, 2015), tend to perform less well on measures of social cognition such as false belief tasks. On false belief tasks, participants are asked to identify whether they or a fictional character had a mistaken belief about the identity or location of an object. Children who are typically developing and have been exposed to older siblings or families with adults and older children perform better on these tasks (Dunn et al., 1991; Hughes et al., 2005; Hughes & Leekam, 2004).

Because CAI tend to live in smaller families and have weaker structural language than their nonadopted peers, they are at risk of poorer social cognition. For example, children who were exposed to institutional care prior to being adopted demonstrated lower performance on false belief tasks than children who lived in foster care prior to adoption, and children who were not adopted (Tarullo et al., 2007). Children who lived in foster care performed at a level between the other two groups. This indicates that receiving more social interaction within a family improved their social cognition. In a recent study, children adopted from Asia and Eastern Europe performed significantly lower than a nonadopted group on a series of three false belief tasks (Hwa-Froelich et al., 2017). Structural language performance and number of older siblings predicted false belief performance. Problems in social cognition were found to persist in children adopted from Romania before 43 months of age compared with domestically adopted children in the UK at age 11 years of age (Colvert et al., 2008). CAI are at risk for poorer social cognition because of their weaker structural language performance and less exposure to social interactions with older children.

Executive function skills are used when completing a goal, planning or solving a problem. These are behaviors often needed for social interactions (Bernier et al., 2010; Singer & Bashir, 1999). In other words, one must be able to focus one's attention, inhibit distractions, and monitor and adapt responses during social interactions. Several studies have provided evidence that CAI are at risk for inattention and overactivity (Helder et al., 2016), poor inhibition (Levin et al., 2015; Loman et al., 2013; Merz et al., 2013; Tarullo et al., 2011) and weaker executive function (Hostinar et al., 2012). For example, children who were exposed to institutional care demonstrated different event-related potentials (ERP) and electroencephalograms (EEG) when performing a nonverbal inhibition task. These outcomes are an indication of problems with inhibition and sustained attention (Loman et al., 2013; McDermott et al., 2013; Tarullo et al., 2011). In addition, Merz and colleagues asked parents of children adopted from Romania between 5–72 months of age to rate their children's executive function skills when the children were between 6–17 years of age and 2 years later. They found that children who were adopted at younger ages had better reports of executive function than children who were adopted older than 18 months of age, and that these difficulties continued 2 years later (Merz et al., 2013). Executive function weaknesses continued into adolescence (Colvert et al., 2008).

20.6 Special Considerations

There are several considerations speech-language pathologists (SLPs) should consider when assessing or providing intervention for CAI. Considerations include: (a) risk factors; (b) limitations of standardized assessments; (c) appropriate comparison groups; and (d) available resources. From the literature, CAI are at risk of hearing impairment, visual impairment, swallowing or eating problems, language impairment, social communication disorder, weaker verbal working memory, social cognition deficits, executive function weaknesses in attention and inhibition, and problems in visual working memory. Consequently, it is recommended that SLPs should conduct an ethnographic interview with persons most familiar with the child or student to gather information about their concerns and observations. SLPs should include questions about hearing, vision, eating and drinking, understanding and expressing wants and needs, nonverbal and verbal social communication, memory, attention, and inhibition behaviors. This information will guide the assessment and help the SLP determine if observations of the child in particular contexts are needed. Regardless of whether caregivers report normal hearing or vision during the interview, clinical report and research provide evidence that CAI are at risk of hearing and visual problems (Eckerle et al., 2014). SLPs should conduct persistent hearing screenings and recommend that the parents have their child's vision screened if vision has not yet been assessed.

Once the practitioner has determined the areas that need to be assessed, they must select assessment tools carefully. Because CAI are rarely mentioned in samples for standardized assessments, it is recommended to compare CAI to research means and standard deviations reported in the literature. Several researchers have reported means and standard deviations of standard scores from standardized assessments and raw scores or standard score equivalents from informal measures to use for comparison. While it is best to compare children from similar backgrounds, e.g. children adopted from China to children adopted from China, by adoption age, and adopted language, research evidence may not be available for children from all countries of origin or for all adoption ages learning the same adopted language. In these cases, it would be better to compare CAI with other CAI regardless of country-of-origin than to compare them to children who did not experience the process of international adoption. SLPs may have to project performance expectations based on the longitudinal data available on CAI.

In addition, because several studies provide evidence that language and cognitive development slow as these children age (Gauthier & Genesee, 2011; Glennen, 2014b; Norrman et al., 2016; Scott et al., 2011), it is recommended that practitioners continue to follow and retest as these children grow older. They may qualify for special education services during school age when they did not qualify at younger ages.

If CAI meet qualification criteria for special education services, practitioners should consider providing services within a relationship development framework to facilitate parent-child attachment (Hwa-Froelich et al., 2012; Hwa-Froelich, 2015b). It is also recommended that practitioners consider short-term memory aids as intervention strategies for information processing and working memory. These could include graphic organizers, planning sequences, and/or visual reminders. In addition, social communication may be an area of general weakness for CAI. CAI may need assistance with interpretation of nonverbal communication, social understanding, and pragmatic language (Hwa-Froelich et al., 2014, 2017; Hwa-Froelich & Matsuo, 2019). Practitioners should consider strategies for learning to include tiered levels of practice within a social context. In other words, practice communication behaviors one-to-one with the clinician until the client reaches competence. Then practice with a peer who has social communication skills within the client's range of proximal development. Once the client achieves competence with one peer, provide opportunities to practice with a small group, eventually moving practice into large groups. This tiered level of practice increases the chance of social success for CAI.

Finally, it is important to understand that because CAI are adopted into families who represent a higher socioeconomic background and educational achievement, CAI will most likely be compared to peers from similar backgrounds. For this reason, teachers and other professionals may refer CAI more often and many may not qualify for special education services. However, these children may benefit from other support services such as English as a Second Language instruction, tutoring, reading and/or mathematical support, and/or elective speech-language or learning services.

20.7 Summary

Although much research has been done with CAI, more is needed to better understand their development and special learning needs. Longitudinal language development has been published on children adopted between birth and 5 years of age or placed in foster care (Cohen et al., 2008; Gauthier & Genesee, 2011; Glennen, 2014b, 2015; Windsor et al., 2011). These studies have included children adopted into English- or French-speaking families and within country foster care. Little information is available on children who were adopted from countries with different birth languages such as Ethiopia, Haiti, or Syria. There may be differences in adopted language acquisition due to the linguistic contrasts between birth and adopted languages. In addition, little is known about the biological mothers' prenatal care and health or the children's preadoption experiences in these countries. All variables could influence the children's developmental progression and health.

Furthermore, few longitudinal studies have followed CAI into adulthood. Michael Rutter and the English and Romanian Adoption (ERA) Team (1998) is one of the few teams who have followed the longitudinal development of children from the point of their adoption from Romania into the UK to adolescence (Beckett et al., 2002; Beckett et al., 2010; Kreppner et al., 2007; O'Connor et al., 2000). Longitudinal studies need to follow CAI into adulthood to better understand the developmental trajectory of CAI.

Finally, as described earlier in this chapter, international adoption law has changed the process of international adoption. Children are adopted at older ages and may have special learning needs. Little research has been done on how children who are adopted at older ages or who have special health and learning needs develop following adoption. It is also unknown how preadoption care for children with special needs may differ from children who are judged to be healthy. Additional research on these special populations would help practitioners provide improved individualized services.

Until research evidence catches up with the need for services, it is recommended that practitioners think more flexibly about how to provide services. CAI do not follow the developmental trajectory of children who are not adopted or children who are domestically adopted. Dependent upon their early childhood experiences, their learning and social needs may continue to exist across their lifetime (Odenstad et al., 2008). Practitioners should refer to the research data to determine the presence or absence of impairment. But if children do not qualify for special education services, practitioners should recommend other support services such as ESL, Title I reading and/or math support, tutoring, or community-based elective services.

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Chapter 21
Infants and Children Exposed to HIV and Substance Abuse

Dorian Lee-Wilkerson and Shelly S. Chabon

21.1 Introduction

It has been almost 30 years since we first wrote about the complex histories of children exposed to drugs and alcohol (Chabon et al., 1992). Recent reports indicate there has been a rise in opioid use and the recreational use of marijuana has been legalized in many communities. Despite the known physical and neurodevelopmental effects of drug exposure on developing babies, infants, children, and adolescents, there is an increase in the use and misuse of legal and illegal drugs, including opioids. The difficulties caused by early drug exposure are often compounded for children who have HIV infection from birth or who live in families where a parent has HIV/AIDS. This chapter will examine the impact of these early exposures on children's development of language, and particularly their pragmatic language skills. But first, some background on the nature and extent of these problems is necessary.

21.2 The Nature and Extent of Early Drug and HIV Exposure

The opioid crisis has put a new spotlight on the effects of drug exposure on children. Haycraft (2018) reports that disorders of substance use is an international problem and that opioid abuse has reached epidemic proportions. The National Institute of

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Drug Abuse (2019) reports that every 15 minutes a newborn infant is born with symptoms of drug exposure in the United States, and the Canadian Institute for Health Information reported that more than 1800 babies were found to have prenatal drug exposure because of opioid misuse (Quinn, 2018). Some infants with prenatal drug exposure are born in an intoxicated, sleepy, disoriented state and others are born with neonatal abstinence syndrome (NAS) (Oei, 2018).

Neonatal abstinence syndrome refers to signs of drug withdrawal in an infant that occur after birth because of prenatal drug exposure. Signs of NAS appear within a few hours or days after birth. The duration and intensity of NAS will depend on the amount and types of drugs taken by the mother, the infant's genetic susceptibility to drug exposure, and the quality of prenatal care received. Physicians report that symptoms of NAS may last as long as 18 months, and that breastfeeding extends the duration and severity of NAS. NAS is treatable, but failure to diagnose NAS may lead to the development of seizures, 'failure to thrive' or death (Oei, 2018).

Drug exposure may not end at birth. Reports are available showing that children are exposed to drugs during breastfeeding, through accidental ingestion and by recreational use. The Centers for Disease Control and Prevention (CDC, 2018) in the United States reported that 13,052 children were hospitalized for ingestion of prescription opioids between 1997 and 2012. CDC data from 2014 shows that approximately 28,000 adolescents reported using heroin within the past year, 16,000 reported being current heroin users, and 18,000 had a heroin use disorder (Oei, 2018).

The spotlight on the effects of HIV/AIDS has dimmed in many communities because of available prevention methods and treatments. Speech-language pathologists (SLPs) should continue to be mindful, because children living with HIV and/or AIDS are a particularly vulnerable group that may experience a range of physical, social and emotional challenges. The human immunodeficiency virus (HIV) weakens the body's immune system, increasing susceptibility and vulnerability to the effects of infectious diseases. Many children contract the virus from their mother during pregnancy, others during birth. Some children contract the HIV virus during breastfeeding. Signs that a child has the HIV virus include fever, headache, muscle ache and joint pain, rash, sore throat and swollen glands. These initial signs are very mild, are easily ignored, and usually occur one or two months after the child contracts the virus (UNAIDS, 2016).

Not all children will have these initial symptoms. HIV testing has been very effective in identifying most children who have the virus including those who are asymptomatic. As the infection progresses, some children experience frequent illnesses such as ear infections, diarrhea, colds, and upset stomachs. Children may develop oral thrush, cytomegalovirus, yeast infection and lung disease. Children with HIV may also be diagnosed with 'failure to thrive', developmental delay, and seizure disorders. While there is no cure for HIV, medical treatments are available that can control the growth and effects of the virus. Children living in poverty, however, do not have the same access to these treatments. UNICEF reports that only slightly more than half of the 1.2 million children aged 0–14 years living with HIV around the world in 2017 received antiretroviral drug treatment (UNICEF, n.d.). For many of the children the treatment came too late. When children do not receive treatment in a timely manner, their risk of death before their second birthday increases significantly. UNAIDS also reports that only 21.7 million persons received antiretroviral therapy (ART) to treat their HIV in 2017, while an estimated 940,000 died from AIDS-related illnesses (UNAIDS, 2016). Children living with HIV who have access to timely treatment, however, can do well and live healthy and happy lives (Goldberg & Short, 2016).

The pragmatic language disorders (PLD) seen in children with complex histories related to exposure to drugs and HIV are similar to those seen in other children with PLD. Matters of social justice and public health are paramount, however, when evaluating and treating these children. Many of the children may use undesirable behaviors to compensate for or to disguise their PLD. Unfortunately, this group of children are just as likely to receive punishment, as they are to receive support, when they express unusual language behaviors, often because of the stigmas associated with their history of drug exposure and/or HIV/AIDS.

21.3 Complex Histories Related to Drug Exposure

The March of Dimes (2017) reports that the incidence of low birth weight is higher among children born to mothers who smoked marijuana during their pregnancy. Some studies show that children with a history of prenatal exposure to marijuana are more likely to have difficulty paying attention to tasks than children who do not have such a history (Dreher et al., 1994). Children prenatally exposed to hallucinogens and methamphetamines are at an increased risk for premature birth, behavioral difficulties, and poor attention (Singer et al., 2015). Children exposed to cocaine are more likely than their non-exposed peers to experience language delay, executive function impairments, inappropriate play and poor attachment to others (Minnes et al., 2011). Proctor-Williams (2018) observes that children born to mothers who use opioids and other drugs may experience low birth weight, NAS, behavioral disorders and developmental delays. Many reports confirm that children with a history of exposure to drugs frequently experience the consequences listed in Table 21.1.

Children with a history of exposure to drugs also frequently experience environmental and social consequences. And while the physical effects of drug exposure may be relatively direct and clear, the confounding environmental and social effects are less well-defined and may be difficult to measure (Coles & Black, 2006). For example, children with a history of drug exposure have often experienced exposure to multiple drugs and other substances at different times during their development while being concomitantly exposed and susceptible to violence, environmental toxins, unstable living conditions and neglect. Oei (2018) cites the report by Uebel and colleagues presented at the Pediatric Academic Societies (PAS) Annual Meeting in 2015, which stated that children with a history of drug exposure are more likely to be hospitalized for assaults, injuries and maltreatment, and are more likely to die before the age of 12 than are other children. Children with a history of drug

Table 21.1 Effects of drug exposure

Effects of opioids e.g. heroin, morphine, synthetic opioids (oxycodone, hydrocodone, fentanyl)	Smaller brain volume Cognitive delays Motor delays Impulsivity and hyperactivity Attention deficits Depression Anxiety Oppositional behavior and conduct disorder Short stature Low rates of self-esteem Poor social skills
Effects of cocaine	Less age-appropriate play More impulsivity Less secure attachment to their caregivers Delayed language development Lack of tolerance for frustration Easily distracted Difficulty organizing their behavior
Effects of methamphetamines	Attention deficits Hyperactivity Aggressive behaviors Anxiety Depression Sensory integration disorder Learning disabilities Language disorders Executive functioning deficits Sleep disturbances
Effects of amphetamines	Cleft lip Cardiac defects Low birth weight Reduced head circumference Cerebral hemorrhage
Effects of hallucinogens	Learning disabilities Behavior disorders Social/emotional disorders
Effects of marijuana	Attention deficits Hyperactivity Depression Learning disabilities

(continued)

Table 21.1 (continued)

Effects of nicotine	Premature birth Low birth weight Sudden infant death syndrome (SIDS) Conduct disorder Motor delays Cognitive delays

Sources: Macomb Intermediate School District (2010), Cone-Wesson (2005), Oei (2018)

exposure tend to experience more chronic personal stress and vulnerability than other groups of children. Psychologists report that chronic stress and vulnerability is associated with deficits in working memory, general cognitive functioning, attention and executive function in children (Oei, 2018). The identification of these factors in the case histories of these children is challenging and the measurement of their effects and confluence on their development, including language development, is difficult to ascertain.

The way a community views drug addiction and those addicted to drugs also adds to the complex history of a child exposed to drugs. At the time our first article was published in the 1990s (Chabon et al., 1992), a war on drugs was being waged (Drug Policy Alliance, n.d.). This war on drugs criminalized the addict and minimalized the provision of treatment for them. Criminalizing the addict drove those suffering from drug addictions and their families underground. Some mothers who were addicted to drugs would not seek treatment for themselves or for their children who were experiencing drug exposure for fear of incarceration or separation from their child. When a mother was identified as having a drug addiction, she would be separated from her child; sometimes the separation happened immediately after birth.

According to Oei (2018), in Australia more than 50% of babies born to mothers who were addicted to drugs were placed into foster care by 5 years of age. To reduce the possibility of separation, some mothers avoid care for their addiction. Other mothers will refuse prenatal care to avoid detection and potential separation, compounding the effects of the drug exposure for their child. Many children and mothers experience repeated separations from their families, sometimes because of the need for residential drug treatment and sometimes because of incarceration related to drug possession or drug trafficking. The length of separation may vary, but the effect of the separation will be similar, negatively affecting the parent-child bond, creating unstable living situations and interfering with the child's growth and development. Past drug war policies often left the unborn babies, infants and children of those addicted to drugs vulnerable and defenseless.

Views of drug addiction, drug exposure, and drug treatment in the last three decades have changed significantly. *The Comprehensive Addiction and Recovery Act* (CARA) of 2016 no longer uses the term 'illegal' when referring to drug addiction and requires that intervention plans address the needs of the individual addict as well as the needs of the affected family members including infants and children. The CDC (2018) continues to work to develop high-impact approaches that limit the influence of stigma and discrimination when addressing the healthcare needs of individuals who abuse substances because of an addiction. Such shifts in healthcare policy and practice move us away from crime, shame and punishment and towards prevention, reduction of harm and health promotion when planning and implementing effective prevention, assessment and treatment interventions that address the challenges presented by drug addiction and the sequelae that result (Drug Policy Alliance, n.d.).

Nearly every school district in the United States has policies and/or programs in place to address the needs of children exposed to drugs before, during and after birth. These policies help families, teachers, and speech-language pathologists recognize that children with a history of drug exposure are at risk for developing several learning and language disorders, including pragmatic language disorders, because of the physical effects of drug exposure and because of their life experiences related to drug exposure.

21.4 Complex Histories Related to HIV

When left untreated, drug addiction places parents and their children at greater risk for HIV/AIDS. Drug use and addiction have been frequently linked with HIV/AIDS (HIV.gov, 2018). In the 1980s, HIV and AIDS were virtually unknown in the United States of America when a number of cases began appearing and rapidly led to death. The infection and the diseases that soon developed led to fear about and in those impacted by drug abuse. Many of those affected were drug users, especially intravenous drug users, who at times passed the infection on to others through sexual transmission, pregnancy, childbirth or breastfeeding. Little was known about the effects of HIV and AIDS on the development of children during this period, and the public's response to HIV and AIDS was not positive. Those associated with the immunodeficiency were often blamed for having the disease, perhaps because a frequently identified route for the infection was sexual transmission, or perhaps because of other socio-cultural variables.

While there has been a significant decrease in the number of babies born with HIV, and important advances in the identification and treatment of HIV/AIDS, the number of African, African American and Latino babies born with HIV continues to be substantially higher than the general population. Worldwide, there are approximately 36.9 million people living with HIV/AIDS. Of these, 3 million are children, who are less than 15 years old. An estimated 1.8 million individuals worldwide became newly infected with HIV in 2016, including 160,000 children (CDC, 2018). According to UNAIDS (2019), over 13,000 children were newly infected with HIV every month, globally, during 2018.

Some populations may be at greater risk for HIV than others. Sub-Saharan Africa appears to account for approximately 66% of all new HIV infections. Other regions significantly affected by HIV and AIDS include Asia and the Pacific, Latin America and the Caribbean, and Eastern Europe (CDC, 2018). Most children acquire HIV from their HIV-infected mothers during pregnancy, birth or breastfeeding. Treatment reduces the risk of mother-to-child HIV transmission to 2%. Unfortunately, treatment remains widely inaccessible in countries where the burden of HIV is highest (UNICEF, n.d.). The World Health Organization does provide an AIDS Free Tool Kit that is designed to accelerate progress in the testing and treatment of children and adolescents with AIDS. Children with HIV require timely and effective treatment to stay healthy.

McHenry et al. (2018) conducted a systematic review to examine the long-term effects of HIV exposure on children and their neurodevelopment (Table 21.2). McHenry notes that adequate data is not yet available to explain the causal effects of HIV, but the data clearly show that children with HIV are at increased risk of having poorer cognitive and motor outcomes when compared with children who are HIV-negative. Many researchers have reported that the most frequently seen developmental features of pediatric HIV/AIDS are developmental delay and progressive cognitive impairment (Davis-McFarland, 2002; McNeilly, 2005; Sherr et al., 2014).

Davis-McFarland (2002) describes three groups of children with HIV. One group is referred to as 'rapid progressors' because serious signs and symptoms of their disease appear within the first 12–24 months of life due to the rapid loss of the white blood cells that fight infection, known as CD4 cells. Other children known as 'slow progressors' will experience an intermediate progression of the disease. Signs of the inability to fight infection or 'immunosuppression' may not show until 7 or 8 years of age. There is also a small group of children who remain healthy showing minimal or no symptoms of HIV disease through 9–10 years of age (Davis-McFarland, 2002).

Davis-McFarland (2002) poignantly observes that children living with HIV/AIDS have a triple burden of poverty, illness, and family trauma. In terms of family trauma, children may be living with a parent who is also ill, or with parents struggling with drug addiction. The child may not be living with their parent or both parents because of death, incarceration, family violence, or neglect. Davis-McFarland asserts that unlike other diseases and disabilities, HIV/AIDS has had a

Table 21.2 Effects of HIV/AIDS on children

Motor	Cognitive	Hearing	Language and communication
Impaired gross motor skills	Mild intellectual impairment	Severe-to-profound sensorineural hearing loss Conductive hearing loss secondary to otitis media	Language delay
Impaired fine motor skills	Attention deficit disorder	Central auditory processing disorders	Phonological disorders
	Behavioral disorders		Voice disorders

Sources: Davis-McFarland (2002), McNeilly (2005), Sherr et al. (2014)

long history of fear, shame and discrimination, so many families do not want to disclose information about their HIV/AIDS status. She refers to this as the 'burden of secrecy'.

21.5 Working with Children with a History of Drug Exposure and HIV/AIDS

Social and environmental factors and life experiences should be taken into consideration when developing an understanding of a child who has a pragmatic language disorder in addition to exposure to drugs and HIV/AIDS. Knowing the child's risk factors can help SLPs better understand the clinical profiles of children with pragmatic language disorders associated with HIV/AIDS and drug exposure. Considering these factors in the planning of assessment and intervention programs will lead to better clinical outcomes for this group of children. Table 21.3 lists the risk factors associated with HIV/AIDS and drug exposure.

The clinical pictures presented by children with a history of drug exposure and/or HIV/AIDS vary. The variations relate to the amount, type and timing of exposure to different drugs and other substances, the presence or absence of HIV/AIDS, the side effects of treatment for AIDS and/or substance abuse, the effects of genetic and individual factors and associated risk factors.

Below are four cases that will shed light on current assessment and treatment practices used by SLPs when working with children who have PLD related to drug exposure and/or HIV/AIDS. The four cases presented in this chapter show how (1) the knowledge of risk factors is considered when planning for assessment and treatment; (2) the use of culturally-sensitive practices is incorporated to improve clinical outcomes; and (3) the use of a systematic approach is used to guide clinical

Table 21.3 Risk factors for exposure to drugs and HIV/AIDS

Drug exposure	HIV/AIDS
Chronic poverty	Sexually transmitted diseases
Poor nutrition	Drug abuse
Poor prenatal care	Alcohol abuse
Sexually transmitted diseases, including HIV	Discrimination: race, gender or culture
Domestic violence	History of incarceration
Child abuse	High-school dropout
Child neglect	Poor prenatal care
Alcohol and other drug abuse	Chronic poverty
Homelessness or substandard housing	Social stigma
History of incarceration	Family suicide
Unemployment	Child neglect
High-school dropout	
Family suicide	

decision-making. In case one, John's SLP used the International Classification of Functioning, Disability, and Health (ICF) framework (World Health Organization, 2001) to develop the assessment plan and to develop John's clinical profile. John's SLP used the CEEDAR Center's High Leverage Practices for treatment (McLeskey et al., 2017). In case two, Lily's SLP used systems theory (Cordon, 2013) to develop the assessment plan and clinical profile, and positive behavioral interventions and supports (PBIS) (Keller-Bell & Short, 2019) for treatment. In case three, Miranda's SLP used dynamic assessment and for treatment the 'power of play'. In case four, Max's SLP used criterion-referenced assessment and the use of values-driven goals as the approach for treatment.

21.5.1 Case One: John

John is a six-year-old boy who is struggling to meet the demands of the first-grade classroom. His case is a good example of the use of the ICF framework for assessment. Health care professionals use the ICF framework to assess health and health-related conditions that may contribute to a disability such as a pragmatic language disorder. According to the World Health Organization (2001), a disability is an impairment of body functions and/or structures that limits the number and range of activities that an individual can perform and places restrictions on an individual's ability to participate in events. Using the ICF framework to assess a child with a pragmatic language disorder, the SLP would examine the ways body structures and functions may be different because of drug exposure, and how these differences may have affected the child's development of pragmatic language skills. The SLP would also determine if the expressed pragmatic language skills represent a delay, a disorder or a difference. If the child were determined to have a delay or a pragmatic language disorder, the SLP would examine the extent to which the PLD affected the child's participation in life experiences.

John's case history shows that he was diagnosed with drug exposure at birth. His mother did not receive routine prenatal care. She reported that she used heroin, marijuana, alcohol and tobacco, and tried to limit her use of drugs while pregnant. Her heaviest use was during the first trimester, when she was not aware of her pregnancy. At birth, John weighed 4.5 pounds and measured 14 inches in length. His APGAR score was 6 at five minutes which is considered moderately abnormal. He displayed irritability, high-pitch cry and jaundice. His drug screen showed signs of exposure to cocaine and marijuana. He was treated using low light and sound exposure and swaddling. He failed his first hearing screening but passed the second screen before being discharged from the hospital. He was discharged from the hospital after 10 days.

Upon discharge, John was placed in foster care while his mother received residential substance abuse treatment. His father was in jail for illegal distribution of drugs. John's paternal grandparents are deceased. His maternal grandparents live 300 miles away. They assumed custody of John once the agreement was approved by the court. John lived with his grandparents for the first three years of his life. At age three, John's parents regained custody of him and moved him back to his home state. During his time with his parents, John experienced additional exposure to drugs, violence in the home, and placement in and out of foster care as his parents continued to struggle with their addictions. At age six, John returned to live with his grandparents.

John now lives with his grandparents in a stable but relatively unfamiliar home. This is John's first time attending his current school and he does not know any of the children in his class. Many of the children in his class live with their parents and have done so all their lives. All his classmates attended the school the previous year for kindergarten and are very familiar with the classroom routine. John attended kindergarten last year, but a different one and was absent frequently because of the drug-related struggles his parents experienced. His kindergarten teacher did not get to know him well but thought he was a pleasant, easy-going child who did not talk much or cause trouble in the classroom. This year, John's school attendance has been regular, yet he has not learned to navigate the classroom routine or to make friends. Rather than ask for things, he simply takes them. Rather than wait his turn, he pushes. He does not ask for clarification when he misunderstands, nor does he clarify when others misunderstand him. He avoids verbal tasks and is behind his peers in mastering academic tasks. John shows evidence of a pragmatic language disorder exacerbated by his complex physical, environmental, and social history.

John's poor language and communication skills and his complex case history served as the basis for his referral for a speech-language evaluation. John's assessment profile using an ICF checklist shows that his body structures related to speech, language, and hearing are intact. His profile notes that John did not experience many of the frequently-cited conditions reported of children with a history of drug exposure. There were no reports of intellectual impairment, central auditory processing disorder or hearing loss secondary to recurrent otitis media or other impairments of the auditory system. There were no reports of John suffering from fatigue because of cardiovascular and/or respiratory disease.

John passed a routine hearing screening. He discriminated the sounds of speech, recognized rhymes, and identified syllables and was able to blend and segment the sounds of words. John produced all the speech sounds of his language and the utterances that he produced were intelligible. His speech was fluent, and his speech rate was average. John recalled a series of seven unrelated numbers, a series of six unrelated words, and followed three-step directions. The SLP noted some impaired functions. John did not attend to a two-minute story. His average mean length of utterance (MLU) was 3.2, and his vocabulary score was comparable to that of most 4-year-old children. John showed difficulty with comprehension of words with multiple meanings and figurative language. He also showed difficulty understanding utterances with complex clause structures, such as those containing multiple verbs and subordinating conjunctions. John's assessment profile also showed that he is assertive in his interactions with others, but primarily uses nonverbal behaviors to communicate. His verbal interactions with his peers consist primarily of commands, such as ‘gimme’ and ‘that’s mine’. His verbal conversations with adults include 3- to 4-word responses to questions that consist of mostly nouns and verbs.

Using the ICF framework assists SLPs in determining the nature of a child’s pragmatic language disorder, and the degree of disability the language disorder imposes on the child. The ICF framework also assists with identifying the types and levels of support needed for activities of communication, academic learning and social interaction. Additionally, SLPs use the ICF framework to identify the restrictions placed on life participation as a function of pragmatic language disorder, cultural perceptions and social supports. Using the ICF model to assess the degree of disability imposed by PLD, John’s SLP found that his clinical profile was consistent with other children reported to have a pragmatic language disorder. John did not demonstrate the language and communication skills needed to interact with his peers for play and learning. He did not understand indirect requests. He did not clarify or request clarification. John’s participation was restricted as well. He did not actively participate in conversations. He did not engage in social interactions or play. He did not form interpersonal relationships with others or participate in group activities with peers. Classroom learning only occurred with one-to-one instruction with the teacher or instructional assistant (World Health Organization, 2001).

John’s case also provides good examples of ways to use High Leverage Practices to improve pragmatic language skills while addressing classroom and social demands. The Council for Exceptional Children and the Collaboration for Effective Educator Development, Accountability and Reform (CEEDAR) developed 22 High Leverage Practices (HLPs) for teachers and special educators. HLPs support collaboration among teachers, families, allied health providers and support staff to promote student learning, remove barriers to instruction and facilitate the process of securing needed services (McLeskey et al., 2017).

Assessment HLPs use multiple sources of information to develop a comprehensive understanding of the strengths and needs of students. High Leverage Practice #3 states that assessment before planning and implementing treatment leads to achieving learning goals. The ICF model helped to identify the pragmatic language skills John has (activity strengths), those pragmatic language skills he does not have (activity weaknesses), and the restrictions placed on his social and communicative

Table 21.4 John’s ICF profile

	Strengths	Needs
Body functions	Hearing Speech production Memory	Attention Vocabulary Morphology and syntax Pragmatic language skills
Activity	Speech discrimination Speech intelligibility	Play Conversation with peers Response to indirect requests
Participation	One-to-one interactions with adults	Peer engagement Group learning

functions. John's SLP used the HLP of functional behavioral assessment to determine how John uses his pragmatic language skills to participate in activities, including those viewed as problem behaviors. These assessments helped determine if John used certain pragmatic language skills or behaviors to augment his communication and if John used some behaviors to avoid challenging tasks and unwanted interactions, and if some of John's communication behaviors impeded learning.

In John's case, the SLP performed a functional behavioral assessment by asking his family, teachers and childcare providers to participate in short phone interviews and complete a rating scale. The SLP also observed John's behavior in the classroom, in the cafeteria, and during speech therapy sessions. Based on the data, the SLP hypothesized that John used hitting and taking from others to augment his communication and that he used tantrums to avoid challenging tasks. Using these hypotheses, the SLP developed a treatment plan that targeted pragmatic language skills to increase receptive and expressive language and successful social interactions.

Instructional HLPs advance student attainment of learning goals. HLPs for instruction include intensive instruction and adapting curriculum tasks and materials to meet specific learning goals. Instructional HLPs also include teaching explicit cognitive and metacognitive strategies and providing scaffold instruction to encourage active student engagement, to support learning and to promote independence. To foster learning environments in which students feel safe, respected, valued, and motivated to learn new things, social/emotional/behavioral HLPs are used. SLPs provide positive and constructive feedback to guide students' learning and behavior. Teaching students social behaviors, self-monitoring, self-evaluation and self-correction also creates an environment that is conducive to learning.

In John's case, the SLP provided intensive instruction 30 min a day, to model and shape pragmatic language skills. She used explicit instruction to define and demonstrate one pragmatic language skill each week (e.g. verbal requests for puzzle). To deepen John's understanding of social and cognitive concepts, the SLP used comparison and contrast of pragmatic language skills with problem behaviors as a cognitive strategy. The SLP and John's teacher collaborated to provide John with multiple opportunities to practice his new pragmatic language skills in the classroom throughout the week. The SLP, John's teacher and his family agreed to provide continuous reinforcement of John's use of the targeted behavior for the week, via informative feedback. Informative feedback is an HLP that states that children are to be explicitly told what they did well and how they can improve on their behavior as needed. In John's case, his teacher might comment that John did well with asking a question. Then she would state 'Let's practice asking for help by first getting the listener's attention'. Then John and the teacher would practice 'Mrs. Jones, may I get another sheet of paper?' This helped John recognize the ways that his use of language could achieve desired outcomes. Additionally, the SLP worked with John's teacher and family to alter the classroom and home environments to make the pragmatic language skill easier to use than the problem behavior. To do this, the teacher and parents placed certain desired items out of reach to encourage verbal requests and to promote opportunities to practice verbal requests. The teacher, family and the SLP collected data continuously to describe John's response to instruction and used this data to modify John's intervention plan as needed to increase his success.

21.5.2 Case Two: Lily

Lily is a 10-year-old girl. She was diagnosed with pragmatic language disorder, has a history of prenatal drug exposure, and is HIV positive. Her mother died from complications related to AIDS when she was 17 months of age. Her father has been incarcerated since she was six years old. Lily lived in two different foster care homes between the ages of four and seven. At age seven, Lily moved in with her aunt, who was able to find a job, so she could move back to town to care for Lily. The aunt made the move so that Lily could maintain contact with other family members and her father. Lily's aunt is completing the legal process of being named as Lily's guardian. Lily and her aunt live in a two-story townhouse with two cats. Lily's aunt is single and has no children. She works as a car salesperson and is actively working to build her client list to increase the commissions that she earns. She retired from the army at the rank of captain and receives income and benefits from this retirement. Lily now participates in girl scouts, is a member of a community-based soccer team and participates, with her grandmother and two half siblings, in church activities regularly.

Lily is in the 5th grade. She receives speech-language therapy for her pragmatic language disorder. The treatment targets include use of pronouns with shifting references, understanding and using indirect requests, and telling and writing narratives using logical flow and sufficient detail. The use of explicit instruction and numerous opportunities for practice has produced demonstrable progress. Lily continues to perform below grade-level expectations in part because of her failure to complete class assignments. The SLP has observed and the teacher has reported that Lily does well with completing her assignments until snack time. After snack time, Lily does not transition back to work and disrupts the class by repeatedly asking the teacher or classmates to answer off-topic questions. This routinized behavior frustrates both the teacher and her classmates who now frequently respond with "I don't know" or complaints to the teacher and family.

Lily's case demonstrates effective use of systems theory for assessment and positive behavioral interventions and supports (PBIS) for treatment. Systems theory explains how parts of an organization relate to other parts of the organization, and to the organization as a whole. Bowen's family systems theory views families as systems and proposes that we cannot fully understand individuals by viewing them in isolation (Brown, 1999). According to family systems theory, when SLPs assess a child without knowledge of their family and their community and the interactions among the three, they will not develop a full understanding of the child's strengths and needs. In other words, assessment considerations must include the child, their family, community and the ways these 'systems' interact. Viewing the child, the family, and the community as representing many interdependent systems, that respond, change and interact with one another, will help us to appreciate each unique clinical picture that children with complex case histories portray. Table 21.5 shows the different systems that apply to Lily's case.

As frequently reported in the literature, children respond differently to drugs even when the type of drug, amount of drug, and timing of drug exposure are the same. For example, one child may have experienced poly-drug exposure prenatally, live with two parents who are recovering from drug addiction, have sufficient income, private health insurance, and live-in childcare. Whereas a second child may have experienced poly-drug exposure prenatally, live with foster parents, rely on government-supported health care and have parents who struggle with drug addiction and provide inconsistent, erratic parental care. Although the poly-drug exposure appears to be the same, genetic factors, family factors and social supports are clearly different, possibly contributing to how the pragmatic language disorder is expressed and changes over time. Using a systems-theory approach, the SLP can come to understand a child's pragmatic language disorder by understanding the effects of drug exposure, genetics and individual differences, family response and social support.

Using a systems approach can increase the SLP's understanding of the child, the impact of the PLD on the child and their family, an understanding of the ways their complex case history may attenuate opportunities for growth, and ways the SLP and teacher may support the child, family and community to improve outcomes. Using a systems approach helps us better understand how changes in one part of a system bring about changes in other parts of the system or changes to the entire system. With this approach, SLPs can use trial therapy to see if changes made to the child's communication system in therapy bring about favorable or unfavorable interactions within the family system and the community system. SLPs can also use therapeutic trials to determine if changes in the family and community systems favorably or unfavorably affect the child's communication (Luterman, 2017). Systems theory provides a framework for identifying a child's pragmatic language strengths and

Table 21.5 Lily's clinical profile

	Lily's systems	Lily's family systems	Lily's community systems
Strengths	Phonological skills Morphological skills Functional vocabulary Syntax Memory skills	Caregiving, supportive aunt Ties to father and extended family Stable living environment Financial stability	After-school care Girl scouts Soccer team Church activities
Weaknesses	Organizational skills Figurative language Abstract concepts Comprehension and use of indirect requests Narrative skills	Incarceration of father Death of mother	Strict social rules

needs within the context of family and community interactions, classroom engagement and learning and self-regulation. A systems approach provides a strong basis for selecting treatment outcomes that would be supported by the family, and would result in positive changes in the child's, the family's and the community's systems, so that the academic, vocational and social goals of the child can be achieved.

In a 2018 article in the *ASHA Leader*, former ASHA President, Elise Davis-McFarland, shared that her visit to Kenya 'opened her eyes to how other cultures relate to, value and live with people with a disability' (Davis-McFarland, 2018, p. 7). Systems theory provides SLPs with a framework for understanding the effects of drug exposure and HIV/AIDS on the life experiences of the child from the family's cultural perspective. A systems approach also provides SLPs with the opportunity to see the impact of a pragmatic language disorder within the cultural context of the child. In other words, the use of a systems approach for assessment allows a clinician to understand how the pragmatic language disorder of children with complex case histories is seen, understood and responded to by members of their family and community. The systems approach also provides insight into how a child has adapted their communication because of their pragmatic language disorder and the interactions they have had with others as a result.

Children with pragmatic language disorders including those with complex case histories may demonstrate inappropriate behaviors that bring about negative responses from family, peers, teachers and others. The American Psychiatric Association (2013) has noted that many see impaired communication skills such as those related to pragmatic language disorders as inappropriate or problem behaviors. Problem behavioral interventions and supports (PBIS) is a school-based program designed to prevent and/or respond to the negative behaviors displayed by some children (Bopp et al., 2004; Keller-Bell & Short, 2019). One underlying premise of PBIS is that for some children, problem behaviors serve as a form of communication that represent their best attempt to interact with others (Bopp et al., 2004). For these children it is important for SLPs to determine whether problem behaviors are signs of a behavioral disorder, signs of ineffective attempts to communicate, or both. In a case where the problem behaviors represent poor attempts to communicate, further analyses are useful in determining if the child uses the behaviors to express one or multiple meanings, and how the child uses the communication attempts to meet their needs. It is also important to determine the use of the problem behaviors within the child's family and community systems (Bopp et al., 2004; Keller-Bell & Short, 2019).

PBIS provides the foundation for behavioral expectations across school settings including classrooms, cafeterias, playgrounds, gymnasiums, auditoriums, and hallways for all children. Children are taught the rules and routines to follow. The physical environments are arranged to support positive behaviors and engagement (Simonsen et al., 2015; Walker et al., 2005). One PBIS strategy that is frequently used by speech-language pathologists when working with children with communication disorders, including pragmatic language disorders, is functional communication training (Bopp et al., 2004). Functional communication training (FCT) has been shown to reduce the intensity and complexity of problem behaviors and increase communication skills.

FCT is a behavioral approach in which the SLP first identifies those things that trigger a problem behavior. Triggers can include people, activity, time of day, setting, frustration, and intolerance among others. Once triggers are identified, the SLP determines the function of the problem behavior and things that reinforce its use. For example, the function may be to make requests and the reinforcement is having the requests met. Another example of a function may be to stop an activity and the reinforcement comes when all abandon the activity. Once triggers, functions and reinforcers are identified, the SLP would determine a pragmatic language skill that would be an appropriate replacement for the problem behavior. Hegde and Maul (2006) suggest that SLPs select pragmatic language skills that are as easy as or easier than the problem behaviors that are to be replaced.

Functional communication training is often paired with differential reinforcement. Differential reinforcement, as described by Hegde and Maul (2006), can be used to decrease problem behaviors by replacing those problem behaviors with effective communicative behaviors. When SLPs ignore the problem behavior and are generous with their praise and reinforcement of communication attempts, they are using the strategy of differential reinforcement of other behavior (DRO). Differential reinforcement of incompatible behavior (DRI) encourages the child to use pragmatic language skills that physically limit their ability to use the problem behaviors to meet their communication needs. For example, it is difficult to pinch someone to get their attention when the child is using their hands to produce a sign to get someone's attention. It may also be more difficult to have a tantrum to show frustration when the child is using verbal chants to regulate behavior. Differential reinforcement of low rates of responding (DRL) reinforce or reward a child when they perform a problem behavior less often. For example, the clinician might establish a token system so that the child could earn points when not engaging in the problem behaviors. The child could use these points to 'buy' time spent for play. Or the child might be given praise for not engaging in the problem behavior and ignored when the problem behavior(s) are expressed (Hegde & Maul, 2006).

Once the appearance of the problem behaviors decreases significantly, the SLP might use differential reinforcement of alternative behavior (DRA). The desired outcome using this technique would be to replace the problem behavior with an appropriate pragmatic language skill. Using this technique, the SLP would arrange for the child to receive the same 'reinforcing consequence' that they received for the problem behavior. If hitting can be met with attention from someone, so can the utterance of the phrase 'Look at me' (Hegde & Maul, 2006). It is important to note that family and community systems be assessed to determine willingness and ability to implement techniques outside of therapy to increase the positive impact of the behavioral changes made during therapy.

Lily's SLP used functional communication training to address her problem behavior. The SLP noted that Lily's response to the trigger 'transitioning from snack time back to class time' may likely change if she were taught a pragmatic language skill to facilitate the transition. In Lily's case, the SLP taught her how to use the class schedule to find the next task to complete after snack time. She was also taught who to ask and what to ask if she did not understand how to complete the task that followed snack time. The teachers and SLP agreed to make sure that Lily's task after snack was a practice activity of a previously mastered skill. They also agreed that appropriate on-task questions would be quickly addressed and praised, and that off-topic questions would not be answered. When Lily asked two or more classmates off-topic questions during the transition time, she would receive the short verbal reprimand 'Not burning question' time. After Lily completed her practice work, she was immediately seen by the instructional assistant to review the practice work. The review included informative feedback that praised her work and showed the progress she made towards her goal. Following the review, the instructional assistant began the next learning activity with her small group. The plan also included a time at the beginning of school and a time at the end of the school day for each child to ask one 'burning question'. Ongoing data collection showed that the number of off-topic questions asked of others during this transitioning period decreased significantly, and that Lily actively participated in 'burning question' time (Hedge and Maul 2006).

21.5.3 Case Three: Miranda

Miranda is 4 years, 3 months of age. She was identified as at risk for developmental delay at birth because of poly-drug exposure. Miranda's mother is recovering from a heroin addiction. The addiction began when Miranda's mother was prescribed Vicodin, an opioid, for back pain as a result of a car accident that happened when she was 20 years old. At the time of the accident, Miranda's mother did not inform the doctor or her parents that she was struggling with alcohol abuse. Taking Vicodin only added to the struggles with substance abuse faced by Miranda's mother that eventually led to a heroin addiction. A visit to the obstetrician revealed the mother's addiction. Her family immediately placed her in a residential treatment facility for substance abusers who are also pregnant. At that time, Miranda was a 10-week-old fetus.

The treatment plan outlined a medical detox program using buprenorphine. Miranda's mother then received specialized drug and alcohol addiction treatment, which included prenatal care, nutrition services, counseling, supervision and behavioral therapy to manage her addictions. Following Miranda's birth, her mother was provided with follow-up medical care and behavioral therapy, and her family provided financial and emotional support and childcare.

At birth, Miranda tested negative for drug exposure. But she was small for gestational age and her APGAR score at 5 minutes was a 7, which is considered to be at the lower end of normal range. Miranda passed the newborn hearing screening and was discharged from the hospital on the second day into the care of her mother and maternal grandparents. Though not married to her mother, Miranda's father shared custody of Miranda and was an active participant in her care and child rearing. At age two, family concerns were expressed about Miranda not meeting developmental milestones in the areas of speech, language and cognition. By age three, Miranda was enrolled in an early intervention program and had an individualized family service plan (IFSP) that addressed her developmental delays. Miranda made progress and is now in need of a re-evaluation and an update to her IFSP.

Miranda's case illustrates the use of dynamic assessment and the 'power of play'. Dynamic assessment is a language assessment tool used for describing the pragmatic language skills that children have, and their potential for learning new pragmatic language skills. Dynamic assessment helps SLPs determine if children are able to make significant changes in their understanding and use of language with mediated learning experiences. Research supports the idea that those children who are likely to benefit from mediated learning experiences are likely to have a language difference, while those who do not benefit are likely to have a language impairment (American Speech-Language-Hearing Association, n.d.). The goals of dynamic assessment are to describe the strategies children use to learn language and to describe the strategies they use to navigate the communication process (Dockrell, 2001; Feuerstein et al., 2002). Dynamic assessment also provides information about what motivates children to learn and their responsiveness to intervention, which assists with therapy planning, if needed (American Speech-Language-Hearing Association, n.d.). Dynamic assessment incorporates a range of methods for exploring learning potential using prompts, cues and mediation (Peña et al., 2001). It can be used to determine the 'zone of proximal development' or readiness to learn (Gutieérrez-Clellen & Penña, 2001).

Sternberg and Grigorenko (2002) and Campione (1989) describe two ways in which dynamic assessment may be implemented. These include the 'sandwich' method and the 'cake' format. The SLP using the 'sandwich' method gives an initial pretest, followed by a mediated learning experience and ending with a posttest. This method, also known as the test-teach-retest method, is designed to measure improvement achieved during a short-term teaching session. Standardized or non-standardized tests may be used. The SLP using the 'cake' format administers a non-standardized language measure and provides prompts during the testing. This method of assessment, also known as graduated prompting, allows the SLP to assess which forms of support result in improved performance. Both methods assist with identifying those pragmatic language skills that represent language differences associated with life experience and those language behaviors that are disordered, possibly associated with exposure to drugs and/or HIV/AIDS.

Miranda's SLP used the test-teach-retest method of dynamic assessment. She used the Bilinguistics™ Dynamic Assessment Protocol (Bilinguistics, Inc, 2012) to target four pragmatic language skills. The skills included: (1) greetings; (2) making requests; (3) answering questions with a verbal response; and (4) using language for pretend play. The SLP conducted the assessment in the home, with preferred objects and with mom participating. Miranda was given five trials to greet which included opportunities to greet her grandmother, the SLP, the pet fish, the pet cat, and one character in a familiar story book. Miranda was given five trials to answer questions with a verbal response. The questions asked Miranda to name two family members (i.e. 'who is that?') and three familiar objects (i.e. cup, book, and banana). Miranda was given five opportunities to make requests (i.e. up, juice, book, cookie and doll). Miranda was also given five opportunities to engage in pretend play (i.e. drive the car, bathe the baby, comb doll's hair, cook the toy food and eat the toy food).

In the teach phase, the SLP used graduated prompting based on the Bilinguistics™ Dynamic Assessment Protocol and the tasks used for the test phase. The protocol of teaching the behavior included discussing the need for the new skill, modeling the skill, and giving informative feedback about performance level. During the teach phase, the SLP also carefully observed the language and communicative processes used by Miranda and noted which stimuli and therapy strategies increased her motivation and responsiveness (Lidz, 1991). The SLP used this information to identify sources of language differences and pragmatic language disorder. Following the teach phase, the retest phase was completed. Miranda's assessment results may be found in Table 21.6 below.

Miranda's dynamic assessment profile allowed the SLP to consider the effect of information modality on pragmatic language performance. The SLP noted that the therapy strategies that combined auditory with visual information appeared to increase post-treatment outcomes for Miranda. For example, pairing the verbal act of greeting with the actual person or object to be greeted, doubled Miranda's score. Additionally, pairing the visual symbol via a communication board with the desired verbal response increased Miranda's performance as well. Miranda's performance with the communication processes related to pretend play also improved when she was supported by pairing visual and auditory input (i.e. talking about bathing baby using pretend soap and water while pretending to bathe). Miranda's SLP chose play as the context for her language therapy.

Jessica Sinarski talks about the 'healing power of play'. According to Sinarski (2018), children with complex case histories often experience events that serve to strengthen their 'flight or fight' systems and delay the development of their executive functions. These higher-order thinking skills allow adults and children to form attachments with others, have sympathy, tolerate frustration and boredom, and solve problems. In cases where executive functions are delayed or disordered because of brain injury or traumatic life experiences, pragmatic language skills that express politeness, engage in civil debate, seek clarification, and consider the perspectives of others are also delayed or disordered. When pragmatic language skills are delayed or disordered, tantrums may be used to respond to frustration, hitting is used to defend oneself, and the adults in the child's life may respond likewise. Play can help children develop higher-order thinking that fosters problem-solving skills, sympathy, tolerance for frustration and boredom, impulse control, and attachment to others.

Children with complex case histories sometimes hold on to defensive strategies (i.e. fighting, hoarding, withdrawing, etc.) for years, even after their lives have settled into supportive homes. Play can be used to identify the child's defensive strategies, to identify adults' reactions to those behaviors, and identify ways to modify the environment to shape effective communication strategies for both the child and adults. Children with complex case histories may not have a sense of safety or be able to regulate their behaviors. These children may not have the words to express

Table 21.6 Miranda's clinical profile

Language target	Test	Teaching & supports	Retest	Modifiability
Greetings				
1. Greet grandmother 2. Greet SLP 3. Greet fish 4. Greet cat 5. Greet book character	2/5	Modeling- moderate support	4/5	40%→80% High
Answer questions with verbal response				
Who is that? Answer 'mommy' Who is that? Answer 'grandmother' What is that? Answer 'cup' What is that? Answer 'banana' What is that? Answer 'book'	1/5	Direct imitation- maximum support	2/5	20%→40% Moderate
Make requests				
Communication board				
1. Up	2/5	Performs task for child-maximum support	3/5	40% →60%
2. Juice				
3. Book				
4. Cookie				
5. Doll				
Verbal requests				
1. Up 2. Juice 3. Book 4. Cookie 5. Doll	0/5	Direct imitation-maximum support	0/5	0%→0% Low
Pretend play				
1. Drive the car 2. Bathe the baby 3. Comb doll's hair 4. Cook the toy food 5. Eat the toy food	0/5	Direct imitation- maximum support	2/5	0% → 40% High

Notes: Continue with greetings, verbal responses, and pretend play until only minimal support is needed. Continue with nonverbal requests using visual symbols. Suspend making verbal requests until readiness to learn is present

their feelings. They may not have developed the sense that words matter. SLPs can use play to build connections with the child, to help the child build connections with others, and help the child learn the communicative functions of words. SLPs and families can use play to provide moments of laughter and fun that can be used to help the child make connections with people, language, and communication (Sinarski, 2018).

Miranda's SLP used play to strengthen her pragmatic language skills. The SLP used play to facilitate communication between Miranda and her family members.

They played tea party to practice table manners. They played house to practice kitchen and bathroom safety. They played fashion show to practice labeling clothes. They played with dolls to practice routines for getting ready for school and for bed. They played artist to practice labeling colors and objects. Eventually, family members began devising games to practice for family trips and other outings, and as a natural consequence, teaching the words needed to play, practice and participate.

The SLP showed the family and Miranda how to use pretend play to practice the language and communication strategies to use when difficult moments occurred. Family members practiced using language that was on or below Miranda's level of comprehension to acknowledge her feelings and Miranda practiced expressing her feelings and needs in ways to help family members understand (Sinarski, 2018). The family used play to prepare Miranda for the classroom, the playground, and for riding the school bus. The family used play to prepare Miranda for the routines and rituals of church, the routines and rituals for family sleepovers and perhaps most importantly, the routines and rituals for mommy and daddy downtime and grandmother and grandfather downtime, too. Once Miranda showed competence with pretend play, role reversals were practiced, to increase Miranda's skill in understanding the perspectives of others or improve her "theory of mind".

The SLP used nonsense words during play, to teach that some words have multiple meanings. She used scripts to support pretend play and used a wide variety of communicative functions. Play was also used to help Miranda predict the consequences of using incorrect words, incorrect word forms, and not using verbal communication at all. In time, Miranda developed sufficient play skills so that she could play with and learn from her peers. Soon there was no longer a need to teach the use of verbal requests because Miranda learned to use requests to participate in games and to interact with others. The richness of communication between Miranda and her family increased as they learned to play with her and with one another. There were setbacks. Not all play routines were quickly learned, and Miranda was not always able to transfer skills learned through play to other settings. Miranda's family did become less anxious about her and, possibly, more accepting of Miranda as well.

21.5.4 Case Four: Maximillian

Maximillian, a 13-year-old eighth grader, has a diagnosis of pragmatic language disorder secondary to autism spectrum disorder (ASD). He also has a history of prenatal drug exposure. Max was diagnosed at birth with neonatal abstinence syndrome (NAS) that extended his hospital stay for 20 days. While in the hospital, Max received non-pharmacological and pharmacological treatments for his withdrawal symptoms. He also received feeding intervention because of the NAS. Upon discharge, Max was placed on the high-risk register. At age two, he was enrolled in an early intervention program because of developmental delays. At age four, he was diagnosed with pervasive developmental disorder—not otherwise specified (PDD-NOS). In 2013, at age seven, his diagnostic label was changed to ASD.

At the age of 13, Max's academic strengths include computational skills, eye-hand coordination, and visual memory. His language skills, vocabulary, morphology and syntax are average, and his reading skills are on grade level. His social communication skills are poor. He identifies no-one as a friend and none of his classmates considers Max to be a friend. He eats lunch alone and spends his free time on the computer playing games that do not require other players. Because of his challenges with social communication, Max is excused from meeting service-learning requirements and from completing group projects. Max's case is used to illustrate the use of criterion-referenced testing as an assessment tool and values-driven goals as a method of improving pragmatic language skills.

Given Max's reluctance to interact with others, his SLP decided to begin with creating a 'safe space' for Max to promote interaction. Some children have had many unfamiliar adults in their life and as a result have developed a distrust of adults. Some may have had few adults in their life that have shown interest in their well-being and in this case, the child distrusts adults. On the other hand, like Max, a child may have ASD, which makes social interactions difficult. Given any of the above scenarios, it would be important to allow the child time to become comfortable with the examiner. Max's SLP decided to give him the opportunity to meet first in a group situation. The SLP visited Max's classroom and used the visit as the first opportunity to complete criterion-referenced testing by collecting a language sample. The second meeting included Max and several members of his family. This provided the SLP the opportunity to complete a parent and/or family interview and conduct a family observation. Fortunately, Max's mother, aunt, uncle and sister were able to attend the meeting at his school, and his SLP was able to use this session to collect a second sample of language. For some children, it may be necessary to visit the family in their homes if work schedules, family responsibilities or transportation make school visits impractical. If technology were available, video conferencing would serve the same purpose as a face-to-face meeting. Sometimes, phone conferences or the exchange of notes or letters may be the only means of meeting with the family.

Max's SLP began the collection of the third and most formal language sample from him only after he appeared somewhat comfortable with her. The SLP wanted to feel reasonably assured that she could elicit the most 'representative' sample of language as possible. Since Max was 13 years old, the SLP engaged him in a video game to elicit utterances of five words or more with complex syntax by asking 'why', 'how' and 'what if' questions. Once the game ended, the SLP engaged Max in debates about best new recording artists, television series, sports teams, and social media platforms to elicit extended narratives. The SLP also requested recorded samples from Max's family and teachers with Max's permission. If Max's parents and teachers could not have provided a sample, the SLP would have asked them to record a sample and would have given them guidelines for eliciting and recording the sample.

The SLP would analyze these language samples and narratives using C-units. By using C-units, or communication units, as the approach to language analysis, the SLP can identify the grammatical elements Max uses for spoken and/or written language. C-unit analyses would reveal which grammatical rules and vocabulary Max uses to express his ideas in the form of main clauses or ideas with supporting clauses or ideas. This analysis would also show the grammatical errors Max makes, and the types of vocabulary he uses (Miller et al., 2015).

The use of multiple language sampling is effective in assessing how well a child uses language for different activities and with different communication partners. If family and teachers cannot provide data, the child may be willing to record samples and share them on their own. They can also record the child talking with different peers. This may provide important information about their social uses of language and provide good examples of language used with equal partners and sometimes with less dominant partners. Language samples are a source of authentic evidence of grammar, figurative language and social-pragmatic skills.

After collecting multiple language samples, Max's SLP administered a structured criterion-referenced test (CRT) to examine social communication, knowledge and use of figurative language and his ability to appreciate the perspective of others. Max's SLP created this CRT based on reports from teachers and family about the language skills Max needed for home, the classroom and social settings. Examples of skills included in his CRT were to use language to (1) clarify meaning; (2) solve problems; (3) complete group work; (4) regulate his behavior; (5) express his needs and understand the needs of others; and (6) understand terms with shifting reference. Max's SLP administered the CRT in parts over two days with the assistance of his teacher and mother.

In addition to the administration of the CRT, the SLP administered a standardized language test. Direct language assessments or standardized tests are often needed to document eligibility for services. Scores from the formal and informal assessments are often compared. Supportive or contradictory findings from the comparisons may result. In the case of contradictory findings, it is important that the SLP examine factors that may have resulted in increased or decreased performances on the different language measures. The use of surveys can serve this purpose by gathering information about the types of supports provided, the types of peer, family and community interactions and examples of successful and unsuccessful interactions. The survey may also provide information about limitations, if any, the pragmatic language disorder appears to impose on the child's participation in communication, academic, social and other events. Ultimately, the assessment should produce a profile of the child's routine as well as their best language performances on informal and formal measures of pragmatic language (Elleseff, 2015).

Max's profile showed that the longest C-units expressed by him only linked one main clause with one subordinate clause using the conjunctions 'because' or 'that'. No other subordinating conjunctions were used, even when prompted. He used and understood personal deictic terms, but he did not respond appropriately to test items that used terms that shifted reference according to place or time. Max responded well to test items that measured understanding of figurative language using the

Table 21.7 Maximillian's clinical profile

C-units	Deictic terms	Figurative language	Communicative functions
He liked the game	Take this ball; not that one	Recognizes commercial logos	Answers questions
No-one knows that	Put the cup there		Makes statements
No-one came	Put the book here		
Because that's the way it's done	Bring me the pencil		
I don't know that way	Take her the crayon		

recognition of logos, but not to other forms of figurative language such as riddles, jokes, similes or metaphors. The only communicative functions expressed by Max during conversations were statements about his actions and answers to the questions asked by others.

The SLP observed that Max would like to participate in activities with his peers but does not do so unless a peer takes extra effort to include him. Even with peer support, Max's level of participation in the activities is minimal and is mostly limited to observation. The SLP learned from interviews with family members that Max also does not actively participate in family activities. His early life experiences may be described as a state of constant change. He was welcomed and treated well by all family members, but he moved around frequently because of his mother's drug addiction. He lived with his grandparents during his first year of life while his mother was being treated for drug addiction in a residential setting. He lived with his mother for the next two years, but at times lived with his grandparents as his mother struggled to recover from her addiction. By age three, he was back with his mother full time, but they lived in poverty while his mother worked part time and attended school. When Max was seven, his mother married, and Max became a member of a blended family with two younger sisters. When he turned nine, a baby brother was added to the family, and Max's home life was very busy. In the home were two parents who worked full time, scheduled school and after school activities, household chores, and the stresses of a blended family with parents and step parents, grandparents and step grandparents, aunts, uncles and cousins.

Max's family expressed an interest in helping him increase his social communication skills but were not sure what improvements should be made or how to support Max in making such improvements. Family members did not all agree as to which of Max's communication and social behaviors were strengths and which ones were weaknesses or problem behaviors. They all agreed, however, that Max would benefit from intervention.

Adequacy of social communication is determined by how well a child's verbal behaviors align with the values of their family and community. Campe (n.d.) points out that while our personal and community values determine what behaviors are acceptable and which ones are not, values are not readily identifiable or perceived until someone violates them. Max's SLP understood that her values were violated when she viscerally reacted to some of Max's behaviors. To increase her cultural responsiveness to Max, the SLP consulted with his family to increase her knowledge of their views of his behaviors. Family members and the SLP shared their descriptions and interpretations of Max's behaviors, including the perceived meanings and functions of those behaviors, and the appropriateness of the behaviors. Based on these descriptions, the SLP and family considered which of the family, community and school values were being used as the basis for their interpretations.

The SLP started the list of values. She used the six primary values identified by motivational author and speaker, Tony Robbins, as common to most groups (Campe, n.d.). These values include:

1. **Growth**—to develop and become your best
2. **Contribution**—to give to others
3. **Connection**—to have strong relationships with others
4. **Variety**—to appreciate constant, but good change
5. **Certainty**—to have structure and security
6. **Significance**—to contribute in ways that are valued and appreciated

Family members discussed how these values are viewed and expressed in their homes and communities. The SLP shared her views about how the values are viewed and expressed in the school. This discussion led to sharing examples of behaviors that expressed these values and to the addition of values not listed among this core six. Using this discussion, the family and SLP agreed on the top four values that should be used to develop Max's social communication plan. The family and SLP also agreed on the language and social behaviors that they would like for Max to use to express these values, and on the social settings in which these behaviors would be used.

Once the critical decisions of values and ways to express those values were made, decisions about treatment targets and treatment strategies were much easier to make. The SLP developed a treatment plan that included (1) knowing and following the rules of four favorite games (e.g. baseball, basketball, chess, and card games); (2) knowing and understanding the functions of sharing riddles; and (3) completing group projects with two peers. The SLP would teach Max how to play games and practice winning and losing. The SLP would also use explicit instruction to teach how multiple meanings can be used for humor and how graphic organizers can be used to help Max prepare to work on group projects.

Social skills groups were used to assist Max with practicing his newly acquired language and communication skills in a safe and supportive environment. The social skills group included two family members and two peers. The two peers also had a pragmatic language disorder. The children in the group along with Max practiced listening, regulation of behavior, using language and communication to converse and using language and communication to play. In the group, the children shared experiences and Max, over time, became a more active participant.

Making treatment decisions that aligned with the values of Max's family and community reduced the conflicts that may have arisen if Max were taught social communication skills that violated those values. Max's responsiveness to treatment was increased because his new skills were modeled and supported by his family, community, and school. Follow through with treatment plans was increased and communications with the SLP and teacher were more productive. Max's case demonstrates that respecting family and community values will promote positive treatment outcomes for children with pragmatic language disorders, especially those with complex case histories.

21.6 Summary

This chapter illuminated several systematic approaches used by SLPs to guide clinical decision-making when working with children with pragmatic language disorders associated with exposure to drugs and HIV/AIDS. The four cases presented in this chapter illustrated how SLPs critically considered the child's case history, the effects of exposure to drugs and HIV/AIDS, and associated risk factors to plan assessment of the child's pragmatic language skills with a view to creating a comprehensive clinical profile. Each case highlighted how the SLP used the child's clinical profile to plan intervention, making effective use of collaboration, continuous assessment and language therapy strategies.

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Chapter 22
Maltreated and Traumatized Children and Young People

Susan McCool

22.1 Introduction

This chapter is concerned with the effects of childhood trauma and maltreatment on socio-pragmatic development. In Sect. 22.2, we explore key concepts and definitions around childhood adversity, aiming to derive some clarity and consensus from what is a rather confused, and contested landscape. We examine the factors that make it difficult to establish prevalence rates with certainty. We then consider widespread and long-term potential consequences associated with exposure to trauma and maltreatment, particularly when such adversity happens in the critical developmental period of early childhood. Contemporary ecological concepts of resilience are explored, introducing important dimensions of adaptation and functioning that go well beyond the affected individual to include the levels of family, community, and society.

First, it is important to acknowledge some areas not included in this chapter. Globally, the prevalence of children's exposure to potentially traumatic events (PTEs) is influenced by armed conflict, natural disasters and chronic community violence. The level of exposure to such PTEs in affected populations, predominantly in low-income and middle-income countries, is significantly under-researched (Gunaratnam & Alisic, 2017). It cannot, therefore, be the focus of this chapter. However, it is important at the outset to note this gulf in our understanding of children's experience of adversity worldwide.

Additionally, there is increasing recognition of the potentially traumatic impact on children of medical intervention, particularly when they lack understanding of the rationale for procedures. In such circumstances, children may struggle to process what may feel like the contradictory messages of loved ones assenting to them encountering painful and intrusive experiences (Yehuda, 2016). Although this

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emerging area is very relevant to many children with language and communication impairment, owing to the high prevalence of associated medical and disabling conditions, it too is lacking in research activity and shall not be a central focus here. Instead, we concern ourselves primarily in this chapter with trauma in the context of intra-familial violence, abuse and neglect.

22.2 Childhood Adversity, Trauma, Abuse and Neglect

In this section, we will briefly consider the nature and scope of serious childhood adversities, proposing a simple but useful dichotomy of those involving threat and those involving deprivation. Trauma and abuse, it will be suggested, fit into the former category, whereas neglect fits into the latter. We will then review research evidence of the consequences of adversity, particularly when experienced in early childhood at a time when critical developmental foundations are expected to be laid down and when individuals lack the language and cognitive capacities to process adverse experiences adaptively.

22.2.1 What Is Childhood Adversity?

There has been a proliferation of research interest in childhood adversity over recent decades (McLaughlin, 2016). Subsequent to the publication of a seminal study (Felitti et al., 1998), suggesting significant and widespread detrimental health and social effects of adverse childhood experiences (ACEs) lasting into adulthood, this notion has also gained enthusiastic policy uptake, such that it is now recognized as one of the major global public health issues of our time (Landolt et al., 2017). Caution is advised by McLaughlin (2016), p. 3), however, emphasizing that “childhood adversity is a construct in search of a definition”. This author calls for clarity over which experiences surpass common stressors of childhood and argues that the commonplace practice of straightforward arithmetic totaling of adversities is excessively simplistic, neglecting as it does important dimensional and contextual considerations.

McLaughlin (2016), p. 6) proposes the following definition of childhood adversity: “exposure during childhood or adolescence to environmental circumstances that are likely to require significant psychological, social or neurobiological adaptation by an average child and that represent a deviation from the expectable environment”. In agreement with Humphreys and Zeanah (2015), McLaughlin (2016) further postulates that adversities take two distinct forms: those involving *threat* (such as exposure to violence) and those involving *deprivation* from expectable inputs (such as exposure to language). Threat-related adversity encompasses all forms of trauma as well as abusive forms of maltreatment, while deprivation-related adversity relates to circumstances in which there are significant shortfalls in meeting a developing child’s basic needs, such as neglectful forms of maltreatment.

22.2.2 Trauma

The term 'trauma', used in accordance with diagnostic manuals, refers to *exposure* to events involving a significant danger to one's safety, or to witnessing or learning about such events happening to a loved one (American Psychiatric Association, 2013). Trauma has also come to be used to refer to a person's *response* to such exposure, which can lead to some conceptual confusion.

Trauma- and stressor-related disorders (TSRD) are conceptualized in the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) to exist on a spectrum chiefly including, among others, Post-Traumatic Stress Disorder (PTSD) (Goldbeck & Jensen, 2017). The nosology remains problematic, however, with the inclusion in the same category of Attachment Disorders, the origins of which lie in the *absence* of expected inputs rather than the *presence* of threats.

In developed countries, most studies confirm that in excess of 50% of adults will retrospectively report having experienced one or more PTEs before adulthood, such as abuse or witnessing violence. It is important in the context of this chapter to note that the majority of reported exposures take place during adolescence (Landolt et al., 2013). Much less is known about levels of exposure across childhood, particularly in the developmentally sensitive period of infancy, although recent research confirms both the existence of such exposure and its detrimental effects (Osofsky et al., 2017). PTEs tend to co-occur, with around half of respondents indicating exposure to more than one such event (McLaughlin et al., 2013).

Trauma often leads to psychopathology, but not invariably. One helpful way to consider this is that it is adaptive for people encountering an adverse experience to respond in some way. Indeed, it is normal to respond to abnormal situations. When that 'fight/flight/freeze' response persists beyond the need for it, however, we can then begin to deem the individual's response as maladaptive or indicative of psychopathology. Rates of PTSD after exposure to PTEs were calculated at 16% in a meta-analysis (Alisic et al., 2014). Highlighting the importance of determining factors relating to risk and resilience in any set of unique circumstances, resultant psychopathology in this study was found to be more likely among girls, those with prior experience of trauma and where the trauma experienced related to interpersonal violence (particularly from a close caregiver). At present, results of epidemiological research into trauma in childhood vary widely, depending on the scope of events included as potentially traumatizing, demographic differences in the participants sampled, and measurement variables (Gunaratnam & Alisic, 2017).

22.2.3 Maltreatment

Abuse involves acting in such a way as to inflict significant harm. It can involve physical, sexual and/or emotional abuse. Importantly in the context of this chapter, emotional abuse may involve conveying to a child that they are worthless or valued only to the extent that they meet the needs of another person. It may involve conveying inappropriate expectations for the child's age or developmental stage. It includes causing the child to feel fearful, or it may involve exploitation of the child (Scottish Government, 2014).

Neglect is by far the most common form of maltreatment of a child. It involves the persistent failure to meet the child's basic needs, such that there is likely to be serious impairment to the child's health or development (Proctor & Dubowitz, 2014). Neglect can involve the failure to provide for physical needs (e.g. food), the failure to ensure access to required medical care or education, or the failure to protect the child from harm. Importantly in the context of this chapter, neglect can involve inadequate provision to meet the child's emotional and/or developmental needs. Failure to provide sufficient exposure to language and communication models, such that the child's development of these capacities is compromised, would be an example of neglect through absence of expectable inputs.

Child maltreatment is, by consensus, understood to be significantly under-reported (Sedlak & Ellis, 2014). Children who have been maltreated may not be able or willing to disclose this fact. Adults who suspect the maltreatment of children, or who receive disclosures from children, may be reluctant to report it, or indeed may be deterred from doing so for a variety of reasons. Estimated incidence figures vary significantly, depending on who is asked, what they are asked about, when they are asked, and how they are asked. Reports of these figures introduce further variation, with frequent conflation of potentially traumatic events (such as witnessing community violence) with substantiated harm imposed on individuals. So, for example, Lambert et al. (2017), p. 49) cite findings that up to one half of youths in the USA have experienced abuse or have witnessed violence, whereas Sedlak and Ellis (2014) report consistent figures for all forms of maltreatment in the same country at around 40 per 1000, based on officially reported cases and the wider experiences of mandated reporters. Around three quarters of those cases represent neglect and the remainder abuse (Sedlak & Ellis, 2014). Retrospective studies of adults who report having been abused as children indicate much higher figures.

There is no clear answer to the question of why maltreatment of children occurs, and in individual cases the context is likely to involve a complex mix of factors. Persistent areas of investigation include intergenerational transmission across generations of families (Schelbe & Geiger, 2017) and/or links to wider familial violence, such as intimate partner violence (Alhusen et al., 2014). Other investigators examine associations with disadvantageous socio-political environments, such as poverty and discrimination (Drake & Jonson-Reid, 2014). For the purposes of this chapter, it is important to note the significantly increased likelihood that children with disabilities will encounter maltreatment, especially those with language and communication impairments (Crowley, 2016; Giardino et al., 2014).

22.2.4 Consequences of Maltreatment on the Developing Child

Serious and pervasive consequences of early adversity can occur across the lifespan, increasing the likelihood of all forms of physical ill health and psychopathology, as well as detrimentally affecting a wide range of developmental outcomes including communicative and social functioning (Lambert et al., 2017).

Trauma and maltreatment in early childhood are particularly detrimental because they occur at a highly sensitive developmental period considered to be essential to 'laying the foundations' of future development (McLaughlin, 2016). Evidence has accumulated of permanent damaging effects on neurobiology (McCrory et al., 2010), and on general development, mental health and attachment relationships (Vasileva & Petermann, 2018). Likewise, research demonstrates significant hampering of an affected individual's development of key psychosocial capacities such as emotional regulation, linked with organizational and self-regulatory cognitive processes like executive functioning (Ford & Greene, 2017). There is consensus that childhood experience of maltreatment increases risk of poor lifetime outcomes in a host of academic, vocational and health areas (Vasileva & Petermann, 2018).

Any child's response will involve a unique combination of risk and resilience factors (Ungar et al., 2013). Resilience is construed as encompassing a wide range of protective or adaptive factors, importantly not only at the level of the child, but also encompassing factors related to family functioning and context as well as the wider community. In this view, relevant risk factors at societal level include daily stressors for individuals and families arising from struggle, stigma and discrimination (Hyter, 2007).

Aspects such as timing and chronicity of exposure to maltreatment, nature of maltreatment and relationship to the perpetrator are also considered critical variables (Goldbeck & Jensen, 2017). Experiencing interpersonal violence or witnessing it in relation to a key attachment figure is considered particularly likely to lead to complex, pervasive and sustained developmental sequelae (Lambert et al., 2017). Prolonged or repeated exposure to severe stressors, especially without the ability to escape or avoid the maltreatment (such as in the case of abuse of a young child by an attachment figure) heightens risk of multiple morbidity (Osofsky et al., 2017).

Studies have, therefore, focused on developmental sequelae of maltreatment in young children. Indeed, often such investigations focus on highly specific subgroups of maltreated children in order to shed particular light on the nature of developmental associations. Vasileva and Petermann (2018), for instance, reported on a systematic review and meta-analysis of studies investigating aspects of development in children under the age of 7 years and residing in foster care. In all, 41 studies were included. Overall, 39% of the combined sample showed developmental delays, the greatest proportion of which related to cognitive development. In addition, 38% of the sample demonstrated clinically-significant psychological problems, with approximately equal proportions of internalizing and externalizing problems. Levels of both developmental delays and mental health problems, therefore, while far from universal, were found to be significantly higher than levels typically reported in the general population.

McDonald et al. (2013) reported on detailed developmental evaluations of preschool children selected from a specialist family-care facility where all child attendees had substantiated experiences of abuse and/or neglect. Assessments were conducted on 49 children identified by staff as having particular developmental or behavioural concerns, so it is perhaps unsurprising that high levels were found. In 91% of assessments, there was evidence of significant developmental delay and/or significant behavioural problems. Illustrating the high levels of co-morbidity present in the sample, in 63% of assessments both developmental and behavioural concerns were highlighted. In total, 85% of assessments revealed concerns about the preschoolers' emotional or behavioural functioning. The nature of concerns in this respect is illuminative: predominantly the children showed internalizing responses such as being wary or withdrawn (53% of assessments), or a combination comprised of internalizing with externalizing behaviours (a further 15% of assessments). The children were found to lack social skills required for co-operating with their peers, such as sharing and turn-taking. With regard to the nature of developmental problems, by far the most frequent presenting problem was language delay (established in 65% of assessments).

From infancy and across childhood, language and communication develop at the same time as other psychosocial capacities in a highly inter-related manner (Osofsky et al., 2017). A young child experiencing maltreatment, or indeed an older child functioning at an earlier developmental stage, is more likely to lack the emotional and cognitive skills necessary to move towards healthy psychological adjustment. Such a child will also typically lack the necessary language skills to process the experience of maltreatment in a helpful way, either in terms of interior monologue or through expressing it to others (Yehuda, 2016). Relative youth and/or lack of communicative competence may also mean less access to potentially protective relationships in the wider community such as with educators. Therefore, it is likely that there will be heightened longitudinal consequences owing to the compounding influence of early disruptions on later development. Given the centrality of language in learning, impacts on language skills are likely to play a central role in cumulative ongoing developmental cycles of disruption.

22.3 Language and Communication in Traumatized and Maltreated Children

In this section, we first consider the evidence base for the established consensus of an association between trauma and maltreatment on the one hand and disrupted language and communication development on the other. Next, we review the mistaken assumptions, inconsistencies and notable gaps currently characterizing the research landscape in this field, highlighting priority areas for future investigation.

Focusing our attention on particular aspects of language development, we describe first research evidence of impacts on structural language before considering the relatively neglected area of pragmatic language and social communication in this complex and underserved population.

22.3.1 Language and Communication Disruptions in Traumatized and Maltreated Children: What Do We Know for Sure?

Research to date demonstrates close links between childhood exposure to trauma and maltreatment and disrupted language development. The accumulated evidence is drawn together in two meta-analytic systematic reviews (Lum et al., 2015; Sylvestre et al., 2016). The former specifically focused on language (receptive vocabulary, receptive language and expressive language). It reviewed 26 studies, conducted over 4 decades, representing data from 1176 maltreated children and 936 controls. Language skills were consistently shown to be less well developed in maltreated children, as a group, when compared to non-maltreated controls matched for socio-economic status (Lum et al., 2015).

The second review widened its scope to include pragmatics as well as receptive and expressive language. Overall, analyses of data from the 23 independent samples meeting inclusion criteria showed detrimental impacts on assessment performance of maltreated children, as a group, on standardized language tests, compared to children who have not been similarly exposed (Sylvestre et al., 2016). Findings in support of an association between maltreatment and language were not universal (9 out of 23 samples in this meta-analysis did not yield significant effect sizes). However, overall significant effect sizes for receptive language ($g = -0.53$), expressive language ($g = -0.67$) and pragmatics ($g = -0.48$) were taken to confirm links between exposure to childhood maltreatment and less favorable language outcomes.

The consensus that maltreatment and language are linked, however, needs to be tempered with an appreciation of all the important questions to which we do not yet have answers. The cross-sectional nature of studies to date, for example, means that we know little of the nature of the relationship between maltreatment and language development. The temptation to assume a direct causative relationship (as concluded by Sylvestre et al., 2016, for example) must be curbed, for the available evidence does not currently support such a leap. We do not know if the direction of the relationship between maltreatment and language is unidirectional, bidirectional, or whether it is more complex. Indeed, it is not definitely known whether, or to what extent, the relationship is mediated or moderated by one or several other factors either within the child, such as cognitive ability, or external to the child, such as caregiver 'psychological availability', as potentially indicated in Sylvestre and Merette's (2010) comprehensive risk analysis. Longitudinal research is necessary, controlling for a wider range of potentially confounding variables including:

maltreatment differences; child, family and community factors; and variables in how research is conducted.

Detrimental effects of maltreatment on language have been consistently found, but their magnitude should not be over-estimated. When results of studies were statistically combined, on average maltreated children performed between 0.48 and 0.67 standard deviations (SD) below controls on standardized language assessments (Sylvestre et al., 2016). While this confirms language impairment for the maltreated group, some caution is needed because SD scores on formal measures have limited utility in complex populations such as this, where even well-matched control participants from similar socio-economic backgrounds score, on average, below the mean (Eigsti & Cicchetti, 2004).

There is clear consensus that prevalence rates of language disruption in maltreated children are significantly higher than in the non-maltreated population. Importantly, however, serious inconsistency and variability are noted in reported prevalence rates for language impairments in maltreated children, which range from 35% to 73% even within the comparatively well-defined subset of children in foster care (Krier et al., 2018). Discrepancies relate to issues of definition, measurement, and the practical challenges of identification and data recording for this complex population.

Since not all maltreated children experience detrimental effects on language development (Lum et al., 2018), we should not allow group effects to blind us to individual differences. Currently, we know too little about why some children's language and communication appear to be affected and not others. We need to understand more about factors that may predispose, precipitate, and/or perpetuate such effects—and indeed what factors may be protective. Research to date has often failed to account for critical variables such as whether children have remained in environments where maltreatment was experienced and, if their living circumstances did change, the manner in which they changed. Yet, practice determines that important variation exists. In a sample of 82 maltreated children under 12 years (Lum et al., 2018), the total number of out-of-home care placements individual children had encountered ranged enormously from 1 to 185. They also represented a wide range of socio-economic and educational differences known to be associated with language development.

For too long, research in this area has failed to account for important variation in children's experience of trauma and maltreatment. We need to understand much more about differential impacts of the timing, frequency, chronicity and type(s) of exposure to maltreatment in relation to varied language and communication outcomes. For example, we need to determine whether very early exposure to maltreatment carries proportionally more risk, as widely assumed and as tentatively indicated in the review by Sylvestre et al. (2016). Further, the evidence base does not currently support predictions about likely language outcomes based on whether the maltreatment was ongoing and whether the perpetrator was a close contact as opposed to a distant or occasional contact.

Moreover, in important areas, there is recent cause to question some long-established assumptions. One such example is the long-held view that neglect appears to have a greater impact on language development than abuse. Such reports (e.g. Hwa-Froelich, 2015) were based on findings of early studies (Allen & Oliver, 1982; Culp et al., 1991; Fox et al., 1988). Yet, more recent research including a meta-analytic review (Sylvestre et al., 2016) and a statistical investigation of covariance (Lum et al., 2018) indicates that no maltreatment type is more associated with language functioning than any other. In part, discrepancy arises from the difficulty in differentiating children who have been abused as opposed to neglected when, in fact, maltreatment types often co-occur, and one can often overshadow or 'mask' the other (Sylvestre et al., 2016). Clearly, caution is warranted at present, as is further, systematic, multi-factor investigation.

Importantly, current evidence cannot yet explain the nature of the interplay between language and cognition in maltreated children. Specifically, it has not yet been established whether linguistic skills are disproportionately affected relative to other cognitive capacities. There are some indications that this could be the case (Lum et al., 2015), although contradictory findings arise from a detailed risk analysis in which cognitive development emerged as a single, central risk factor for language disruptions (Sylvestre & Merette, 2010). The majority of studies investigating language in maltreated children have failed to report on measures of child IQ (19 of the 26 studies reviewed by Lum et al., 2015). Furthermore, these studies have not typically accounted for other dimensions of cognition that have been shown to be compromised in maltreated children, such as attention, learning and memory (De Bellis et al., 2013) or emotional regulation and executive functioning (Ford & Greene, 2017). If we are to develop optimally effective interventions, then clearly it will be important to elucidate the relative roles of cognition and language development in this context through further investigation.

With a view to developing effective preventions and intervention, future research is needed to determine the mechanism(s) by which language is affected in maltreated children. Krier et al. (2018) propose a complex combination of genetic and environmental risk factors for language disruptions, operating within a context often characterized by heightened neurophysiological stress resulting from maltreatment conditions and diminished caregiver and/or community support for language development. Meanwhile, based on a study of severely neglected children, Sylvestre and Merette (2010) suggest that key challenges include the psychological availability of key caregivers, based on low acceptance of the child and reduced sensitivity towards the child's development needs. Advocating a systemic, ecological approach, they suggest that the caregiver's own experience of adversity, and specifically of abuse and neglect, often underpins the intergenerational transmission of language disruption within the context of maltreatment. They posit a complex interplay of cognitive and linguistic challenges for a maltreated child, from the prelinguistic stage, underpinned by a compromised relational context.

This section has highlighted the consensus view that maltreatment and language development are linked. Research has shown that language development is disrupted at considerably higher rates among maltreated children as a group. More refined approaches to research would elucidate more about the mechanisms by means of which language development is affected in the context of trauma and maltreatment, and about the myriad of factors potentially influencing this. Crucially, we also need to know a good deal more about the specific aspects of language development affected by experiences of maltreatment. We now go on to review what is currently known about impacts on different dimensions of language development, first looking at structural language and then considering social communication and pragmatic language.

22.3.2 Structural Language in Traumatized and Maltreated Children

Groundbreaking studies beginning around two decades ago demonstrated significant disruptions in the structural language development of maltreated children. At an average age of 31 months, syntactic development, as measured via Mean Length of Utterance (MLU), was significantly behind in maltreated toddlers compared to demographically matched controls (Beeghly & Cicchetti, 1994; Coster et al., 1989). Later, Eigsti and Cicchetti (2004) focused specifically on morphosyntactic development in maltreated children by the age of 5 years. While control children matched for socioeconomic status showed significant delays (13 months) against age-expectations on the Index of Productive Syntax, the magnitude of delay was significantly greater in the maltreated group (16 months).

This study demonstrated the exacerbating influence of maltreatment status on syntax, an aspect of development thought to be closely related to cognition (Eigsti & Cicchetti, 2004).

Further, expressive language showed less richness and diversity of lexical items, with a tendency towards general rather than specific terms and greater use of fillers such as ‘oh’ and ‘mmm’ (Coster et al., 1989). Important qualitative differences emerged, particularly in words used to convey the child’s internal state (Beeghly & Cicchetti, 1994). Maltreated toddlers used fewer internal state words overall, and employed a limited range of internal state words, a finding that may be related to the known social and emotional disruptions in the development of maltreated children. They applied internal state words to a reduced range of social agents and contexts, focusing mainly on the here-and-now (Beeghly & Cicchetti, 1994). Moreover, analysis revealed that while the maltreated toddlers produced broadly expected levels of internal state words for ‘task oriented’ aspects such as volition, the paucity in their use was particularly marked for expression of physiological states (e.g. hungry), negative affect (e.g. worried) and moral obligation (e.g. related to permission).

These early studies revealed a promising seam of investigation at the interface of language, cognition and socioemotional development in this vulnerable group, via the innovative use of linguistic analyses and based on systematic observation. Regrettably, for at least the next decade the field failed to capitalize on these solid early foundations, turning instead to a reliance on somewhat restricted and repetitive research using standardized assessments. That research will be reviewed next, before considering promising new avenues for research.

Receptive vocabulary has been by far the most frequently studied aspect of language development in maltreated children (Sylvestre et al., 2016). On average, maltreated children as a group achieve standard scores seven points lower than their well-matched non-maltreated counterparts on standardized tests, where the mean is 100 and the standard deviation 15 (Lum et al., 2015). Caution is urged, however, in extrapolating to wider linguistic attainment from tests of this single component of language (Lum et al., 2015), particularly because receptive vocabulary tests are considered to have relatively low diagnostic accuracy for language problems (Spaulding et al., 2006). Broader receptive language has also been investigated reasonably frequently, yielding overall group averages for language comprehension eight points below controls (Lum et al., 2015). These findings are consistent with results of a meta-analytic review, which reported an effect size of g = −0.53 for receptive language (Sylvestre et al., 2016).

Expressive language has been subject to less examination in robust research, with only 6 of the 26 studies included in the recent systematic review (Lum et al., 2015) focusing on this aspect. Two of the six studies reported specifically on expressive vocabulary, while the remaining four used more comprehensive measures of expressive language. Further investigation of this component is warranted, however, given the comparative magnitude of the impairments indicated: maltreated children scored 13 points lower than comparable peers in expressive language in the meta-analysis by Lum et al. (2015), while the meta-analytic effect size calculated for this aspect of linguistic functioning was g = .67 (Sylvestre et al., 2016)

A small but significant stream of recent research has returned once more to detailed linguistic analysis of samples gained in semi-naturalistic contexts. Knolle et al. (2018) report on data obtained from 32 well-matched child dyads aged 2 to 5 years (each consisting of one maltreated and one not) in peer play sessions facilitated by sensitive adults. Investigation of 'general language sophistication' via measures of vocabulary use, talkativeness and MLU yielded the unexpected finding of equal levels of language competence in maltreated and non-maltreated participants. Authors speculated that this finding might be explained in part by all participants' enrolment in a therapeutic childcare setting which, although not targeting language development per se, was intended to have beneficial effects on a wide range of developmental aspects. They further postulated that the presence of supportive adults rather than the children's parents might explain divergence from the results of previous studies. Nonetheless, this finding is contrary to expectations based on consensus from previous research, so further investigation, and specifically replication, would be in order.

Knolle et al. (2018) also add an interesting new conceptual dimension to study in this field. Research to date has had a deficit-based orientation, based on hypotheses of deficits or delays in language development associated with maltreatment status. Intriguingly, these authors propose a more nuanced consideration of *differences*, rather than deficits, in the language development of maltreated children. Specifically, they highlight their finding that children with prior exposure to maltreatment used twice as many grammatical negations as non-maltreated peers. This finding held true even for participants under the age of 3.5 years. An example of a grammatical negation used by one participant is “I don’t cry when I kick people” (Knolle et al., 2018, p. 454). The authors suggest that these constructions are highly complex in a grammatical sense, involving advanced language skills and representing one aspect of positive adaptation to developmentally adverse circumstances. Alternative explanations, for example that children are merely reflecting their disproportionate experience of parental negative language, are dismissed by these authors because no evidence of heightened negative parental language input to maltreated children currently supports this view. Instead, the researchers associate these language differences with a broader cognitive negativity bias thought to develop in maltreated children, where children’s developing language is central to how negatively they come to view the world and themselves as an agent within it. While interesting, and closely aligned with contemporary asset-based perspectives on children’s development, the results of this study are based on one group of children in one rather specialized therapeutic setting, so caution would need to be exercised in both the extrapolation and interpretation of findings, pending further research.

Results reviewed above have shown that there is broad consensus regarding an association between child maltreatment and structural language disruption, although this in not universally found in empirical studies. Further, important discrepancies between studies exist, depending on the aspect of language measured (for example, receptive or expressive; vocabulary or syntax); the way in which it is measured (standardized test versus linguistic analysis) and the context from which it is sampled (for example, interacting with parents, peers, known sensitive adults or unfamiliar assessors). We go on now to consider what is known about pragmatic language and social communication in maltreated children.

22.3.3 Pragmatic Language and Social Communication in Traumatized and Maltreated Children

The overall intention of this chapter, and of this section in particular, is to consider pragmatics in traumatized and maltreated children. Efforts to this end are somewhat thwarted in a landscape replete with imprecise, inconsistent, and improperly used terms. Linguists have long argued that the notion of pragmatic language has been erroneously conflated with aspects of conversation, and even widely misconstrued as co-terminus with communication itself, to the detriment of both research and clinical practice (Cummings, 2009). That criticism certainly applies within this field. The broader term ‘social communication’ is, therefore, used preferentially in this part of the chapter, with ‘pragmatics’, when used by authors or instruments, appearing in single quotation marks.

Clinical texts stress the impacts of maltreatment on social understanding and communication development (Holosko, 2015; Hwa-Froelich, 2015; Hyter, 2007). A detailed and clinically-rich account of the complex and multi-faceted impact of trauma and maltreatment on communication is provided by Yehuda (2016). She focuses particularly on children's 'pragmatic skills', their narratives, and their ability to interpet ambiguous and symbolic meaning, all within the context of the relationship between those linguistic skills and wider cognitive capacities such as sequencing, cause-and effect and emotion regulation. Given the depth and breadth of clinical discussion on this topic, it is perhaps surprising, then, that in comparison to structural language, aspects of social communication and pragmatic language have been relatively neglected in terms of discrete empirical investigation.

The scope of the review and meta-analysis conducted by Sylvestre et al. (2016), mentioned above, was reported to include 'pragmatics'. Close examination of results indicates that these authors judged 16 of the 23 studies to report aspects they deemed relevant to 'pragmatics'. Importantly, the selected 16 studies are not listed discretely, and nor are there clearly defined criteria by which relevance to 'pragmatics' was judged, so the authors' claims in that respect cannot be independently verified. Nor is it possible to ascertain how many of these studies were among those acknowledged by the authors to have failed to report an effect size ($N = 9$). Overall, reported effect sizes for 'pragmatics' ($g = -0.48$) were broadly comparable with those of receptive and expressive language ($g = -0.53$ and $g = -0.67$ respectively), with 'pragmatics' yielding the smallest effect size. This surprising result may have arisen owing to the review's requirement that studies employ standardized measures (Sylvestre et al., 2016), since a wide range of analytical, observational and caregiver-report measures are often considered more fruitful in exploring pragmatic development (Adams, 2015). The authors themselves attribute the unexpectedly small effect size for 'pragmatics' to the paucity of studies reporting exclusively on neglected children, for whom they hypothesize a greater impact on 'pragmatic' development through a dearth of attuned parental interaction.

Particularly important when considering social communicative function is the environment in which children learn and develop their skills as reciprocal communicators. In this regard, a rich seam of investigation has considered differences in maltreating parents' communication during interactions with their children. To this end, Wilson et al. (2008) conducted a meta-analytic review of 33 observational studies comparing the communication of maltreating (physically abusive or neglectful) vs. non-maltreating parents. Notwithstanding the challenges across this body of evidence, including discrepancies, a host of moderating variables and the characteristic lack of statistical power, the report's authors did find that maltreating parents as a group demonstrate fewer positive communicative behaviors towards their children than comparison parents. Moreover, physically abusive parents were distinguishable by the presence of aversive behaviors whereas neglectful parents engaged in fewer attentive and responsive interactions with their offspring.

Within this context, it is interesting to note that the early work of Coster et al. (1989) and Beeghly and Cicchetti (1994), mentioned above, also gave intriguing insights into differences in communicative development of maltreated toddlers, even at the comparatively young age of 31 months on average. Maltreated preschoolers demonstrated a restricted repertoire of communicative functions, in that they were less likely than non-maltreated comparison children to describe their own experiences, and less likely to seek information from parents during interactions

(Coster et al., 1989). Deficits relative to well-matched controls were evident also at discourse level, in maltreated children's ability to maintain connected 'conversation-related acts' during an interaction with an adult (Beeghly & Cicchetti, 1994; Coster et al., 1989), as measured by Mean Length of Episode (MLE).

In 2010, publication of work by a Spanish research group marked a new stream of interest in social communicative functioning of children and young people relevant to the topic of this chapter, this time in residential care (Moreno et al., 2010). As part of a wider assessment of linguistic functioning, 'pragmatic' functioning of 74 individuals aged between 6 and 18 was assessed by means of a picture-based task. Participants were required to generate statements that would be used by the characters depicted to serve a range of 13 communicative functions. Strikingly, none of the participants were found to be functioning at the level considered to reflect mastery of the requisite skill. Indeed, 87.8% were reported to be functioning at the lowest levels, with 'pragmatic' performance said to be indicative of 'emergency' or 'alarm'.

It should be noted that this study did not involve a comparison group of age and socio-economically matched non-maltreated children. Detail is lacking on important methodological concerns, such as reliability of the instrument. Arguably, the assessment task lacked validity, precision, and comprehensiveness as a sample of purportedly 'pragmatic' competence, testing, as it did in an abstract way, children's responses as to *what other people should say* in certain situations. Nonetheless, this study represented progress in its attempt to report data on some relevant components of social communicative competence in a key sub-population of the maltreated group, in which empirical investigation is challenging. In reporting universal impairment in the sampled population, much of it representing the most significant levels of challenge, this study makes a notable contribution.

In the UK, McCool and Stevens (2011) reported data on perceived communicative functioning of children and young people, also in residential care. Nineteen out of 30 participants showed indications of previously undetected speech, language, and communication impairments on the robustly validated caregiver-completed checklist, the Children's Communication Checklist-2 (CCC-2; Bishop, 2003). Eight of the 19 youngsters who showed impairment had profiles indicative of primarily socio-pragmatic impairment, according to the instrument's originators, with 6 of the 8 at the more severe end of the scale. None of the sample had been referred to speech and language therapy.

Children in residential care, as in this study, represent a special sub-set of the maltreated population. The overwhelming majority of them have experienced multiple adversities, of which maltreatment is just one. They are more likely to be older and to have experienced multiple placements and multiple placement breakdowns (Lum et al., 2018). Furthermore, it has been suggested that in common with many assessments purportedly sampling pragmatics, the instrument used in this study taps into a much broader range of communicative, conversational, and cognitive skills (Cummings, 2009). As such, caution should be exercised in the interpretation of these findings. Nonetheless, the study adds to emerging evidence of substantial levels of social communicative impairment in a significant proportion of maltreated children.

Overall, research into social communicative and pragmatic functioning in maltreated children has not been as plentiful nor as comprehensive as investigation of structural language. The term ‘pragmatics’ has tended to be somewhat liberally applied, when actually exploring wider developmental areas such as social communication, social cognition and conversation. Research into pragmatics for this underserved group has also been beset by the challenges typically encountered for any other population in attempting to find reliable and valid means of measurement, as widely discussed elsewhere in this volume. Despite the gaps and inconsistencies across the body of evidence, there is some evidence from systematic review as well as empirical investigation to support widespread and in-depth clinical accounts, for some children, of significant and pervasive impacts of maltreatment on aspects relevant to socio-pragmatic development. With that in mind, we now turn our attention to the assessment of these aspects in this population.

22.4 Assessment of Pragmatic Language and Social Communication in Traumatized and Maltreated Children

Firstly, it is important to note that there is an initial challenge for speech and language services in ensuring that children who are known to have been traumatized, abused, or neglected are even considered for referral to speech and language services (Frederico et al., 2018). Where maltreated children remain in the care of their parents, the children may not routinely be taken for developmental surveillance or screening appointments of the sort that lead to referral for communication evaluation (Hwa-Froelich, 2015). When children are in out-of-home care, it has been shown that communication impairments often remain unsuspected by care-givers (Frederico et al., 2018; McCool & Stevens, 2011). Furthermore, it has been found that referral may not occur even when impairments are suspected, presumably because other considerations are deemed to take priority (McCool & Stevens, 2011). The chaotic or transient living circumstances of many maltreated children can mean that even if referral is instigated, service policies dictating discharge for failure to attend, or on handling re-referral following discharge, may introduce delays and disruptions to care and preclude meaningful engagement (Byrne et al., 2018).

Frederico et al. (2018) attempted to develop a care-giver completed tool that would identify children in out-of-home care who would benefit from access to detailed speech and language evaluation and intervention. They found that non-clinicians were not able to identify suitable candidates for referral reliably—indeed, it was found that many children who would benefit from referral would be missed. Detailed analysis revealed that while overt speech and fluency concerns were more readily identified, impairments in more complex or subtle aspects such as pragmatic language and comprehension were more likely to be missed. Pending further refinement of such a tool, the authors recommend routine referral of all child protection clients for detailed specialist speech and language pathology evaluation.

For routinely referred children, one significant challenge in assessing pragmatic language may, in fact, be in identifying that the child has indeed been traumatized, abused, or neglected. By their very nature, trauma and maltreatment tend not to be readily disclosed nor openly discussed. It is, therefore, imperative that practitioners in general paediatric practice always remain alert to the possibility, and show vigilance. It is important to be aware that *any* child may potentially be at risk of having been exposed to trauma or maltreatment. Trauma-informed practice determines that speech and language professionals should always be alert to this possibility (Yehuda, 2016).

There are some family circumstances where past or present exposure to trauma or maltreatment are more likely. For trauma, migrants, asylum-seeking families and those with refugee status are more likely to have been affected. For maltreatment, having parents who are living with poor mental health, addiction, and/or domestic abuse (especially in combination) should result in heightened concern and vigilance. Equally, among children there are particular groups where there is greater risk of past or present trauma or maltreatment exposure: children with chronic illness, younger children, disabled children and those with limited verbal communication; children receiving child protection services, whether at home or living in other circumstances (Yehuda, 2016). Where trauma or maltreatment are known or found to have formed part of a child's lived experience, it should be remembered that the effects of trauma on the child's clinical presentation can last for many years beyond the original trauma source, such as in the case of adoption (Yehuda, 2016).

The results of research by McDonald et al. (2013) provide some indications of how to differentiate children with primary developmental disabilities from children whose developmental delays are secondary to maltreatment. For the latter group, it is the presence of behaviours indicative of anxiety in addition to developmental delays that is said to mark out their history of maltreatment. Indicators of note are reported to be hypervigilance, startle responses (for example in response to sudden or loud noises), marked shyness, separation anxiety, and dissociative responses such as momentary 'blanking' or staring. Although helpful, these pointers do not take account of the fact that children with primary developmental disabilities are not mutually exclusive from maltreated children. In fact, disabled children are at significantly increased risk of maltreatment (Crowley, 2016). Children whose developmental profiles include challenges with emotional regulation, such as children with autism or Attention Deficit Hyperactivity Disorder, are considered to be at increased risk not only from maltreatment but also from a lasting propensity to become overwhelmed in response, leading to complex clinical presentations (Yehuda, 2016).

Several authors recommend the use of an ethnographic interview in case-history taking for traumatized or maltreated children (Hwa-Froelich, 2015; Hyter et al., 2001; Westby et al., 2003). By this, they mean a process that considers multiple constructions of the child's reality, incorporating and respecting different perspectives and contextual influences. Yehuda (2016) details the areas a trauma-informed case history will explore, including questions about particular prenatal, postnatal, and developmental stressors, and experiences of disruptions, grief and loss. Helpfully, she lists specific additional questions that should be incorporated into case histories in the case of children living in out-of-home care or post-adoption.

There are practical implications for the clinician in aiming to meet such recommendations. Yehuda (2016) advocates open conversations about such topics, several of which it is noted should not be discussed in the presence of a child, and some of which can best be explored at different levels over time which may necessitate alterations in how service delivery is arranged. Probing into some of those areas will be unfamiliar territory for many speech and language practitioners, so it may offer reassurance to note that clear professional boundaries are recommended and that, when indicated, onward referral for specialist trauma assessment is expected (Yehuda, 2016). She cautions that in cases of trauma and maltreatment case history information may be fragmented, reflecting the child's disrupted experience. This can prove detrimental to the process of assessment of pragmatic language and social communication, which often relies on care-giver completed checklists. Indeed, McCool and Stevens (2011) found that residential care workers were sometimes unable to complete the Children's Communication Checklist-2 (Bishop, 2003) because the child had not been known to them for the required 4 months. Further, Yehuda (2016) describes how the child's lived experience of a lack of cohesion and coherence can be reflected in their communicative style. This compounds the clinical challenge of trying to determine the origins of current clinical manifestations.

For the assessment process itself, Hwa-Froelich (2015) recommends comprehensive assessment, including taking wide perspectives on development in pragmatic language and social communication, social-emotional aspects and different dimensions of cognition. Observations of play-based, interactive engagements with parents/caregivers and siblings/peers are advocated for young children. For school-aged children, Hwa-Froelich (2015) recommends gathering a spread of authentic indicators and measures of functional language and communication performance, including samples of spoken language and writing from school, and teacher observation. Teacher observation may be supported by the use of instruments such as the CCC-2, as mentioned above. Alternatives include the Observational Rating Scale component of the Clinical Evaluation of Language Fundamentals 5th Edition (CELF-5 UK; Semel et al., 2017) or the Metalinguistics Profile of the CELF-5 Metalinguistics (Wiig et al., 2014). However, as found by Frederico et al. (2018), caution must be exercised in relying on reports of non-specialists regarding pragmatic language skills in traumatized and maltreated children, as this area is particularly prone to misinterpretation.

Observation is often proposed, therefore, as a means of obtaining information about pragmatic performance of traumatized and maltreated children in everyday contexts. Using observation to sample pragmatic and social communication skill for any child, however, is fraught with challenge to minimize and account for sampling error and bias (Cummings, 2009). For the traumatized and maltreated child, the challenge is compounded by a key consequence of their disrupted developmental pathway, and that is variability in how they function. Yehuda (2016) stresses how assessment of traumatized children can be particularly challenging because of marked and often unpredictable inconsistencies, from one occasion to another or indeed from one moment to another. She highlights not only that traumatized and maltreated children have overall raised stress levels, but that all manner of seemingly innocuous stimuli in the observational or test situation may trigger trauma reactions which impact on performance. Moreover, trauma reactions can vary a great deal, ranging from withdrawal, through dissociation, to hyperarousal. For the trauma-informed clinician, dynamic and sensitive observation involves careful noting of the antecedents of such reactions, the behaviours themselves, and the child's response to support.

Formal assessment of pragmatic and social communication functioning may be particularly vulnerable to the 'flight/fight/freeze' effects of triggered trauma reactions. Such assessments often present 'staged' situations involving ambiguity, humour, sarcasm, or dissonance. These experiences in particular can trigger the child to re-experience trauma-related emotions. In the past, the child may have had to contend with overwhelming ambiguity and dissonance, particularly when experiencing inconsistent maltreatment from an otherwise needed, loved, or trusted person. Being caught up in a cascade of intense emotion, being intensely vigilant to perceived threats, or indeed being 'shut down', all these responses will lessen the child's chances of attending to subtle social signals and processing ambiguous or contradictory communicative cues in the assessment of pragmatics. Traumatized and maltreated children, therefore, may exhibit heightened sensitivity to testing. Flexibility may need to be exercised in how the assessment process proceeds, including the decision to avoid or alter test items or procedures likely to trigger responses in the individual, even if this invalidates the instrument's administration and means it cannot be interpreted via standard scores (Yehuda, 2016).

Traumatized and maltreated children are likely to be already primed for negative responses in test conditions. Paradoxically, while they may crave praise, they may have hostile reactions when it is provided (Yehuda, 2016). They can have heightened reactions to perceived difficulty or failure. Any perception of confusion, disorientation, or failure during assessment of pragmatic language or social communication may prompt additional distress and anxiety, further impeding test performance. So, while there is general critique of standardized tests purportedly assessing pragmatic language (Cummings, 2009), there are additional reasons to question the validity of such instruments for use with traumatized or maltreated children. Such children are prone to significant fluctuations in their performance, which can be precipitated by anxieties provoked by the test situation and the 'pragmatic' stimuli involved.

As noted above, an ethnographic approach to case history taking is widely recommended for this population (Hwa-Froelich, 2015; Hyter et al., 2001; Westby et al., 2003). It has been argued in this section that traditional approaches to assessing pragmatic language and social communication via checklists, observations and tests—already recognized as generally flawed—are especially limited in the case of traumatized and maltreated children. For this reason, it is suggested here that the ethnographic approach has merit beyond the case history, lasting throughout the ongoing process of assessment and intervention. Recommended methods include naturalistic observation and narrative description of observed events, leading to rich and nuanced interpretations. For speech and language practitioners aware of the challenges and complexities of deriving a valid appraisal of pragmatic language (Cummings, 2009), the adoption of a truly ethnographic approach will be welcome. Such an approach brings opportunities for detailed and systematic consideration of contextual influences on traumatized and maltreated children's pragmatic functioning in dynamic reciprocal interactions, with a range of their typical communication partners, across a representative range of ecologically valid settings and situations. Such an approach invites the use of time-consuming but potentially productive techniques such as conversation analysis and discourse analysis.

Having examined special issues in the assessment of pragmatic language and social communication in traumatized and maltreated children, we turn in the next section to consideration of recommendations for intervention.

22.5 Intervention for Pragmatic Language and Social Communication in Traumatized and Maltreated Children

There is, unfortunately, a significant gap in the literature with regard to empirical intervention studies of specialized communication interventions for traumatized, abused, or neglected children. In fact, a recent systematic review by Byrne (2017) found no original research studies from the past two decades reporting effects of direct speech and language pathology intervention for children in out-of-home care. The author concluded that there is currently no reliable evidence base on which practitioners can base decisions about management options or treatment methods.

There are complex reasons for the dearth in intervention research within this population. The transience of the population can disrupt engagement with services (McCool & Stevens, 2011), a problem compounded by inflexible discharge and re-referral policies in services (Byrne & Lyddiard, 2013). Conducting research in this area is fraught with methodological challenges, such as how to control for variables such as socio-economic status and differences in children's experiences (Lum et al., 2015), and ethical issues, for example the matter of who provides consent and/or assent for participation (McCool & Stevens, 2011) and the extent to which case-study data has to be homogenized to protect anonymity (Byrne et al., 2018).

Published work on intervention, therefore, is largely at the descriptive level. Yehuda (2005) gave rich clinical accounts of three cases from her work with traumatized and dissociated children in an inner city elementary public school. This was followed up in her book (Yehuda, 2016) with extensive clinical examples and detailed principles of intervention in the context of childhood trauma and adversity. Likewise, Hwa-Froelich (2015) outlines useful general guiding principles and suggestions for social communication intervention for children exposed to maltreatment. Byrne et al. (2018) set out to describe the case studies of eight children in out-of-home care accessing speech and language services, using a retrospective case note review design. Restrictions in reporting permissions limited the richness of detail regarding individual cases. Nonetheless, the authors extracted relevant themes at the level of service delivery, thereby making an original, if still anecdotal, contribution to the nascent literature. Meanwhile, Moreno and colleagues took a different approach, describing instead the design of an intervention focusing on language and pragmatics for children in residential care (Moreno et al., 2011) and reporting on its outcomes (Moreno et al., 2012).

Taken together, these accounts highlight potential intervention targets for speech and language practitioners with this population. These include the language of body states and emotions, listening and conversation skills, linguistic structures to support narrative, knowledge of cause and effect, and finally, sequencing (Yehuda, 2005, 2016). Yehuda gives suggestions for recognizing and responding to moments when children become triggered by reminders of trauma, and consequently become activated or dissociated. She describes specific techniques, known as grounding, in which the therapist helps re-orient the child to the present, and reassures them of their safety and security within the therapy environment. In a similar vein, Byrne et al. (2018) advise special awareness of startle and alarm responses to seemingly innocuous sensory stimuli such as environmental noise. Overall, the play and language stimulation techniques they suggest are reassuringly familiar to any speech and language practitioner familiar with young children, with modelling and expansion playing a central role.

For individual young children exposed to maltreatment, a social-pragmatic developmental play-based approach to therapy is advocated (Hwa-Froelich, 2015). Caregiver-mediated approaches designed originally for young children with autism are recommended by this author, for their emphasis on relationship-based intervention, targeting aspects such as joint attention, emotion regulation and intersubjectivity. It is further suggested that adaptations can be made in order to tailor such approaches for older children who have been maltreated. It follows that interventions designed for children with primary pragmatic impairments, such as the individually-delivered Social Communication Intervention Project (SCIP; Adams, 2015), are similarly likely to offer useful frameworks.

Moreno et al. (2011) take a different approach, outlining a group intervention designed for children aged 5 to 12 years who have been exposed to abuse and are currently in residential care. The model proposed is for weekly 2-hour sessions, focusing on morphology, syntax, semantics, and pragmatics, as well as wider cognitive and affective areas considered by the authors to be of relevance. The aim is for goals to be functional and therapy to be interactive. Collaboration with children's parents and school staff is considered key. The intention is that each child will have individually selected targets within the intervention, although the challenges in achieving that are acknowledged as a limitation.

Outcome data from this Spanish research group appear in a related paper (Moreno et al., 2012). It is not clear why this study was not included in Byrne's (2017) systematic review, as it seems to meet the criteria of reporting on the outcomes of speech and language pathology intervention for this client group. Participants were 21 children (notably, 7 male and 14 female) in residential care. Important differences exist compared to the intervention originally described in Moreno et al. (2011), in that the intervention had a much more central focus on socio-pragmatic functioning, it ran for 25 sessions rather than 20, and children in the intervention study represent a sub-set of the original intended age range (8 to 12 years). The primary outcome measure is a picture-based task in which the child is expected to put themselves in the situation of a character and suggest what they would say in pursuit of particular communicative ends.

Caution should be exercised in interpreting the results of this study, given the involvement of the research group in the design of this instrument and in its limited usefulness as a ‘pure’ measure of the child’s linguistic pragmatic functioning since cognitive skills such as theory of mind are required to perform well (Cummings, 2009). Additionally, there is significant risk of bias from several sources including the lack of a control group and non-blinding of assessors. Nonetheless, this study appears to be the only one of its kind reporting original data on intervention for maltreated children. Moreover, the intervention focuses primarily on areas of communication and pragmatics, so its report of significant progress for all child participants appears promising.

Direct work with traumatized and maltreated children is only one management option available to speech and language professionals. Advising and consulting with others, such as case workers, is advocated by Byrne et al. (2018). Facilitative roles are recommended, such as equipping foster carers and residential care workers with the skills to promote language and communication development (Byrne et al., 2018; Vasileva & Petermann, 2018). Direct approaches to upskilling and empowering foster carers and others are possible, potentially with the incorporation of video feedback techniques and group social support common to many parent-mediated approaches to language development. Equally, innovative use of digital and online technologies would seem to offer intriguing possibilities, with demonstrated appeal to busy foster carers seeking easy access to guidance on language development in particular (Golding et al., 2011). Flexibility and accessibility may be particularly important here, where primary concerns for child safety and permanency of placement often, not unreasonably, take precedence over engagement with speech and language services (Chambers et al., 2010; Sylvestre et al., 2016).

This section has shown that there is almost no empirical evidence base to guide decision-making around pragmatic language intervention for traumatized and maltreated children, although there are detailed clinical accounts and useful recommendations. Future research should seek to establish the comparative appeal, effectiveness, and efficiency of different models of direct and indirect speech and language therapy intervention. The final section of this chapter now seeks to draw together suggestions for clearer understanding and investigation of pragmatic language in this population.

22.6 Future Directions: Towards a Greater Understanding of Pragmatic Language in Traumatized and Maltreated Children

This chapter set out to explore pragmatics in an important but underserved population, namely, traumatized and maltreated children. This effort was hampered by basic definitional inconsistencies, both in relation to childhood adversity and in relation to pragmatic language. It was further impeded by notable gaps in the literature concerning both the nature of pragmatic functioning in this population and in the evidence base for intervention. In this final section, we examine potential ways to develop a more nuanced understanding of pragmatics as applied to this complex population.

Terminology within the wider clinical pragmatic arena is contested and in flux. The term 'Pragmatic Language Impairment' (PLI) evolved relatively recently out of a landscape that has historically lacked agreement (Ketelaars et al., 2017). As such, its definition remains unclear, and consensus regarding its underlying mechanisms remains elusive (Ketelaars et al., 2017). Attempts over the past decade to coalesce professional opinion in related areas around an agreed nosology have provoked intense debate (Norbury, 2014) about relationships between, on the one hand, Developmental Language Disorder (DLD) and, on the other hand, Autism. As explained by Adams (2015), for some theorists PLI is considered to lie at an intermediate point between DLD and Autism, whereas for others it represents a complex co-existing condition. Moreover, there is discord (Brukner-Wertman et al., 2016) over newer categorical conceptualizations such as Social (Pragmatic) Communication Disorder (SPCD). It remains to be seen what the field will make of terms such as the proposed 'DLD with impairment of mainly pragmatic language' within the International Classification of Diseases 11 (World Health Organization, 2018). The relevance of such terms to the patterns of social communicative difference associated with maltreated children is as yet unclear.

As a further challenge, important questions remain about the origins, nature, and course of pragmatic difference in the maltreated population (McCool & Stevens, 2011). Can we assume, for instance, that atypical pragmatic development can be regarded in the same way when it arises primarily from disrupted socio-developmental relationships and contexts rather than from neurogenetic differences within the child? Are such assumptions supportable, given that disruptions occur at such an early stage in development as to invoke similar atypical patterns in development? Or should such assumptions be avoided, because enhancing the maltreated child's socio-developmental inputs *could* substantially alter their pragmatic developmental trajectory? This is especially important given the significant upward trend towards children being removed from homes in which abuse or neglect has occurred (Byrne et al., 2018).

Illuminating in this respect is recent research by Lum et al. (2018) in Australia. Consistent with previous research, these investigators found that while, as a group, maltreated children performed below developmental expectations on standardized measures of language and social skills, significant variability was evident for both domains. Some of the children performed within the expected range for their age, indeed some performed at the highest possible levels (+3 standard deviations). The researchers set out to investigate which of a range of variables might account for this variation. Like previous researchers, they were interested in maltreatment history (type and child's age when experienced) but, adding a novel perspective, they also examined the influence of different aspects of the children's current living arrangements (all were residing in out-of-home-care, for example in kinship care, foster care or group homes).

Intriguingly, different patterns were found for language skills as opposed to social skills. For language, maltreatment history was found not to be an influence on the child's current level of functioning but instead the educational level of the current care-giver was related. The converse was true for social skills. Here, maltreatment history had a bearing on current functioning but not out-of-home setting characteristics. Specifically, for social skills, but not for language, a history of neglect was associated with current performance. Children thus affected did not show *more problem behaviors* compared to maltreated children overall. Rather, they were distinguishable by a *lack of prosocial behaviors*. The authors concluded that while language might be amenable to facilitation simply through placement with appropriate care-givers (and that training could assist care-givers in learning language-enrichment techniques where necessary) social skills, however, might require more in the way of targeted intervention to improve. The study offers enticing new insights, but since it was a cross-sectional study of only 82 children, replication and extension are necessary before firm conclusions can be drawn.

Although preliminary in nature, this study lends support to an ecological orientation to the development of language, pragmatics, and social communication skills in traumatized and maltreated children. Eschewing an impairment-based, child-focused model in favour of a bio-social-ecological approach as suggested by Ungar et al. (2013), we can begin to situate the child's pragmatic development within a complex multi-layered set of experiences and influences, many of which are extrinsic to the child. The challenge for the future is how best to understand the socio-pragmatic development of individuals who may have encountered complex and unique patterns of intergenerational, genetic, and epigenetic influences; prenatal exposures to stress hormones and/or potentially neuro-teratogenic toxins; inter-personal relationships affected by trauma, abuse and/or neglect; together with family and/or community contexts characterized by disadvantage, disruption and/or stigma. Additional group studies of intra-child impairment on standardized assessments are unlikely to be particularly illuminating, whereas discourse-based, ecological approaches may well prove more so.

Furthermore, findings of differential post-maltreatment environmental impacts on language and social skills (Lum et al., 2018) shed light on a further critical area for future clarification: the delineation between structural language, pragmatic language and social communication. Norbury (2014) argues for a move towards precisely this dissociation. Moreover, she cites new thinking that pragmatic language is significantly underpinned by both structural language skills and social cognitive competence, highlighting the inter-relatedness of these areas. With regard to social cognition for traumatized and maltreated children, it is important to note the mounting evidence of significantly compromised emotional regulation and executive functioning in this underserved population (Ford & Greene, 2017). With this in mind, it is interesting that in the above study by Lum et al. (2018), the key social skills found to be lacking in the maltreated sample were described as “prosocial communication, cooperation, assertiveness, responsibility, empathy, engagement and self-control” (p. 167). Social communication is undoubtedly an important part of this profile, but nonetheless only a part, with social cognition also a key component. Future research, therefore, needs to be much clearer not only about maltreatment and subsequent history, but also about precisely the developmental skill or skills being investigated.

22.7 Summary

This chapter adopted a dichotomous framework whereby serious childhood adversities can be classified as those exposing the child to threat and those involving deprivation of expectable inputs. Both can lead to calamitous, lifelong consequences on physical and mental health and on educational, vocational and social outcomes. A substantial body of research has demonstrated clear associations between early exposure to trauma and maltreatment and developmental language disruptions. There is evidence of significant and pervasive potential effects on structural language, pragmatic language, and social communication as well as important related areas such as attachment, executive functioning and, increasingly, social cognition. This chapter reviewed specific challenges in clinical assessment and intervention of pragmatic language and social communication for this population, such as unpredictable and variable responses. Indeed, trauma responses such as dissociation can be unwittingly triggered by the staged pragmatic ‘disruption’ often employed as a therapeutic device in clinical settings. Additional or alternative assessment was proposed, in the form of naturalistic sampling, conversation analysis, and discourse analysis. Likewise, recommendations were made for an ecological approach to intervention, with an emphasis on facilitating functional reciprocal language and communication in the context of relationships. Resilience is construed at the levels of child, family, community, and society. Pragmatic language competence, it is argued here, should be understood and promoted in the same way.

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Chapter 23
African American Children and Adolescents

Yvette D. Hyter, Glenda DeJarnette, and Kenyatta O. Rivers

23.1 Introduction

A social (pragmatic) communication disorder (SCD) is characterized by persistent difficulty with verbal (spoken and written) and nonverbal (gestures, facial expressions, spatial distances) communication for social purposes in the absence of medical or intellectual disabilities (American Psychiatric Association, 2013; American Speech-Language-Hearing Association, 2019). SCD is a more recent iteration of pragmatic language impairment (see Adams et al., 2005, 2012; Bishop, 2000), which "restricted pragmatic deficits" to verbal communication (Ketelaars & Embrechts, 2017, p. 31). Children with SCD are likely to have problems with understanding social cues, with language structure, limited vocabulary knowledge, as well as impaired discourse production, comprehension and management (Adams, 2013; Adams et al., 2015, 2018; Bishop, 2000; Timler, 2008).

SCD is frequently associated with a diagnosis of autism spectrum disorder (ASD). However, several other populations also exhibit disorders of pragmatics, including children with neurodevelopmental syndromes resulting from maltreatment and/or prenatal alcohol exposure (Coggins et al., 2003; Hyter, 2003, 2017; Timler et al., 2005), and those with developmental language disorders (DLD) (Adams, 2013; Landa, 2005). There is some overlap in language and communication profiles of children with ASD, SCD and DLD (Adams, 2013; Adams et al., 2015, p. 295), as illustrated in Fig. 23.1. Social pragmatic communication is

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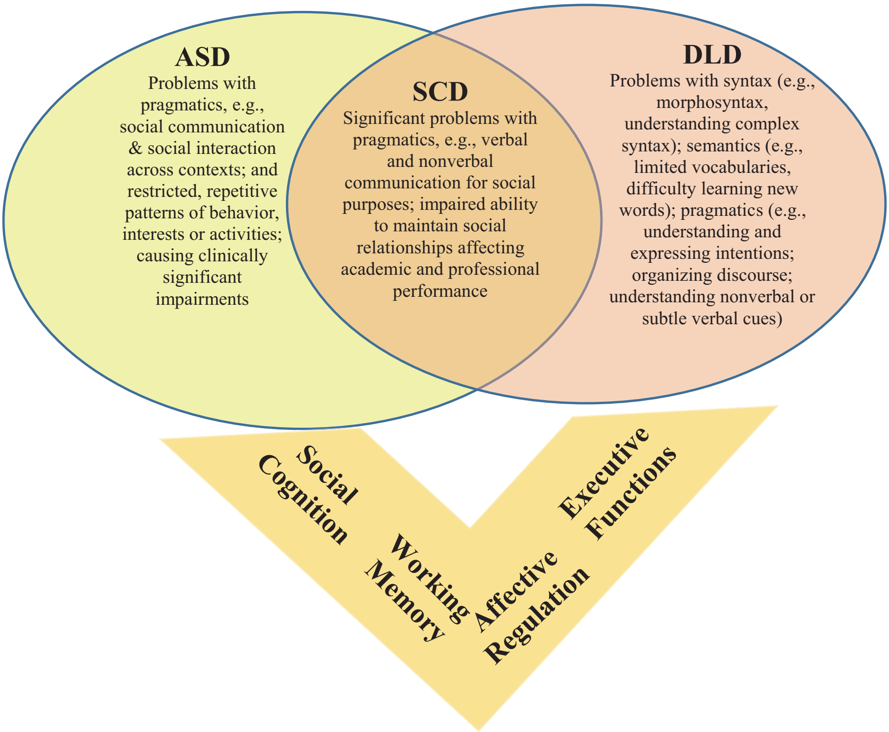

Fig. 23.1 Graphic depicting the relationship among ASD, SCD, and DLD

supported by cognitive skills such as social cognition, executive functions, working memory, and affective regulation, and also by pragmatic language (Hyter, 2012, 2017).

This chapter focuses on pragmatic language disorders in African American children and adolescents who have been demographically identified in health disparity studies as being underrepresented and/or not adequately served by health professions (Ellis et al., 2016; Flores and the Committee on Pediatric Research, 2010; Mandell et al., 2009; Mehta et al., 2013). The language variety spoken by many (but not all) African Americans in the U.S. provides a unique cultural lens through which to examine pragmatic abilities and can be used as a model by which other Black languages (Makoni et al., 2003) and language varieties around the world can be analyzed. This language variety has been referred to by several names over the years, including Black English (BE), African American vernacular English (AAvE), African American English (AAE), and other terms that have changed in relationship to the way the speakers of that language variety self-identified (Green, 2002). Throughout this chapter we will refer to this language variety as AAE.

The origins of AAE are contested. There are four different views about the origins of AAE. The first view is that AAE has structural similarities to Niger-Congo languages—a substratist view. This perspective argues that “West African or substrate languages influenced the sentence and sound structures of AAE” (Green, 2002, p. 9). The second view is the creolist hypothesis, which asserts that AAE is a creole because it shares some features of other creoles of English such as Jamaican Creole or Gullah (Rickford, 2015; Winford, 2015). The third perspective is in the tradition of an Anglicist or dialectologist framework, which indicates that features of AAE are found in other varieties of English (Van Herk, 2015). McWhorter (2017a), who takes an Anglicist perspective, states that the connection of AAE with West African languages is minimal. He argues that there was a mix of dialects to which those who were enslaved in the U.S. were exposed. McWhorter asserts that this mix of dialects was spoken by British indentured servants “who black slaves worked alongside in early America” (McWhorter, 2009). A fourth view is that the development of language produced by enslaved Africans in the U.S. and Caribbean was influenced by ecological factors, such as racial segregation (Mufwene, 2015). Linguists do agree, however, that AAE is a complex language variety—more complex than the language variety referred to as White American English, (WAE), or as McWhorter has written, Black English is a “larger English” (McWhorter, 2016).

Examining the pragmatic aspects of AAE allows us to inspect the development and divergence of cultural linguistic traits shaped by historic social, political and economic forces (Hyter, 2007). The resilience of these cultural linguistic traits over time suggest that they are markers of pragmatics (DeJarnette et al., 2015; Hyter, 2007). We will discuss the relationship between pragmatics and culture, and then examine four domains of social pragmatic communication as they are manifested in the language of African American children and adolescents. These four domains are theory of mind, speech acts or communication functions, use of prosody and voice, and discourse.

23.2 Pragmatics and Culture

Pragmatics is comprised of linguistic, nonlinguistic, cognitive, and contextual domains of communication as listed in Table 23.1. These domains influence communicative behaviors (Hyter, 2017; Hyter et al., 2015, p. 9; Huang, 2015; Perkins, 2007).

Pragmatic skills are influenced by culture—the underlying beliefs, values and worldviews of groups of people with a shared history of problem-solving practices passed from one generation to another (DeJarnette et al., 2015; Hyter, 2007; Hyter et al., 2015; Hyter & Salas-Provance, 2019; Stockman, 2010; Ting-Toomey & Chung, 2012; Wierzbicka, 1991). Having effective pragmatic skills is part of becoming a culturally competent communicator in one’s speech community (Curenton, 2015, p. 57). When thinking about development and disorders, it is important to acknowledge the role of culture because communication and language exist within a sociocultural context - the social, cultural, economic, political and ecological situations that shape development (Hyter & Salas-Provance, 2019; Imai et al., 2016;

Table 23.1 Examples of the domains of pragmatics

Pragmatic domains	Example
Linguistic	Use of phonology, morphology, syntax, semantics used to communicate intentions and regulate discourse (de Villiers, 2004; DeJarnette et al., 2015; Hyter et al., 2015)
Nonlinguistic	Use of gestures, facial expressions, prosody and voice to facilitate communicative interactions (Hyter et al., 2015)
Cognitive	Use of implicature, perspective taking, and theory of mind to interpret and comprehend communicative interactions (Carston, 2004; DeJarnette et al., 2015; Horn, 2004; Hyter et al., 2015; Perkins, 2007)
Contextual	The influence of socialization and cultural history (how historical experiences are interpreted), and of social activities in which interlocutors are engaging on communicative interactions (DeJarnette et al., 2015; Hyter, 2007; Hyter et al., 2015; Rivers et al., 2012)

Kidd et al., 2016; Makoni et al., 2003; Otto, 2014). This need to recognize the role of culture in communication seems to increase exponentially when considering pragmatic language and communication skills and impairments. This is because culture not only provides the lens through which communicative contexts and social interactions are interpreted, but can serve as a basis for differentiating typical social pragmatic communication development from disordered development (DeJarnette et al., 2015; Hyter, 2007; Hyter et al., 2015; Rivers et al., 2012).

Kecskes (2017) identifies two different schools of thought regarding the relationship between pragmatics and culture. One school focuses on the similarities and differences of pragmatics among different cultural groups—a cross-cultural view. This cross-cultural view examines communication mismatches (Kecskes, 2017). Examples of these mismatches are how some polite language forms may work in one language and culture but not in another (see Brown, 1995; Brown & Levinson, 1987) and how different uses of language may contribute to employment discrimination (see Gumperz, 1982).

The other school, referred to as intercultural pragmatics, emerges from a socio-cognitive theoretical perspective (Kecskes, 2017, p. 401). Intercultural pragmatics focuses on the previous experiences of groups of people and the current communicative context, and how those past experiences and current situations *both* shape the way groups construct or understand communicative interactions. According to Kecskes (2017), this intercultural approach to pragmatics "adopts a dialectical perspective by considering communication a dynamic process in which individuals are not only constrained by societal conditions but also shape them at the same time" (p. 406). The main driving force of these intercultural engagements is the act of blending prior experiences with the current situational experience. It is this intercultural view of pragmatics that we employ in this chapter to explicate the pragmatics of many African American children and adolescents.

23.3 Etic Versus Emic Views of Pragmatics

Throughout this chapter we will discuss pragmatics using both an etic perspective and an emic perspective (DeJarnette et al., 2015; Pike, 1967). Etic describes the examination of pragmatics from outside of the cultural group of interest. An etic analysis of speech acts, for example, would compare African American children's production of speech acts to *a priori* taxonomies of speech acts developed on other cultural groups, such as speakers of White American English (DeJarnette et al., 2015). An emic analysis is intracultural in that it focuses on the "unique characteristics and behaviors of a particular cultural group" (DeJarnette et al., 2015, p. 67). It is also a necessary addition to etic perspectives because it is through the emic lens one can begin to understand cultural behaviors. An emic analysis, for example, would compare data from a particular cultural group, e.g. African American English speakers, to speakers from that same cultural group. Consistent with a socio-cognitive perspective, humans have the capacity to create collective reality, which informs cultural values (DeJarnette et al., 2015; Moll & Tomasello, 2007) that drive daily interactions (Hyter, 2007; Hyter & Salas-Provance, 2019).

23.4 Pragmatic Language Disorder

The fifth edition of the *Diagnostic and Statistical Manual of Mental Disorders* (DSM-5; American Psychiatric Association, 2013) defines a language disorder as "persistent difficulties in the acquisition and use of language across modalities (i.e. spoken, written, sign language, or other) due to deficits in comprehension or production" that include language structure, meaning, and function; and that "language abilities are substantially and quantifiably below age expectations resulting in functional limitations in effective communication, social participation, academic achievement or occupational performance" (American Psychiatric Association, 2013). Some disorders can cause a disability, but a disability must be viewed within not only a biological or psychological context, but also within social and cultural contexts.

The World Health Organization (WHO) uses a biopsychosocial model to conceptualize disability. The biopsychosocial model acknowledges the relationship between social contexts and health (e.g. health behavior, communication and/or language abilities, health outcomes, access to health care, health disparities) (Engel, 1980; Harris et al., 2012; Hyter & Salas-Provance, 2019). Using this model, WHO defines disability as the interaction between a person's abilities (e.g. linguistic, cognitive) and the environmental and personal contexts in which that person exists (WHO and The World Bank, 2011). Environmental contexts refer to the situations in which a person lives such as one's home, work, or school, and includes societal attitudes about the person's abilities. It can also include health care policies that affect services that a person is able to access. Personal contexts include individual characteristics that do not result from the person's abilities such as gender, race, ethnicity, or personality (Howe, 2008; WHO and The World Bank, 2011). These contextual factors serve to facilitate or hinder a person's ability to participate actively in his or her day-to-day life (Howe, 2008; WHO, 2001; WHO and The World Bank, 2011).

Sometimes, the pragmatic language of African American children and adolescents has been socially constructed as a disability by viewing these behaviors from solely an etic perspective; that is, examining pragmatic language from outside of the African American cultural perspective by using pragmatic taxonomies (and assessments) developed on cultures other than those for whom they were developed and on people of African descent (DeJarnette et al., 2015; Hyter et al., 2018). In this part of the chapter, we review components of pragmatics that are typical for AAE child and adolescent speakers.

23.5 Social Pragmatic Communication Skills of African American Children and Adolescents

Four types of social pragmatic communication behaviors will be used as examples to illustrate typical and atypical development of these skills for AAE speakers. These four areas are theory of mind, speech acts (or communicative intentions), voice as a pragmatic tool, and discourse management, which includes conversations, narrations, and expositions. We will highlight the importance of utilizing an emic (intracultural) analysis rather than only an etic (intercultural) analysis when differentiating typical from disordered social pragmatic communication abilities for African American speakers.

23.5.1 Theory of Mind

Theory of mind (ToM) is a social cognitive skill that includes intention reading, false belief and mental state awareness. ToM allows one to understand that their perspectives, intentions, and mental states may be the same or differ from others' perspectives, intentions and mental states (de Villiers, 2004; Hyter et al., 2018). Intention reading is the ability to utilize landscape of consciousness[1] (Bruner, 1986), which refers to the subjective internal states or consciousness experienced by characters in a narrative, for example. False belief refers to the ability of a person to separate or suspend their knowledge and/or beliefs from

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[1] Both the Landscape of Consciousness and Landscape of Action (the actions/behaviors of the characters or others) are important for narrative development, requiring "a metarepresentational ability to adopt the meaning attributed to the situation by others." (Ligeza, 1998, p. 80).

what others *might* believe (Wimmer & Perner, 1983). For example, if a child sees a crayon box[2], opens it and discovers that cotton balls are inside the box, they know that (1) crayons are not inside the crayon box, and (2) they thought there were crayons inside the crayon box before opening it. This same child demonstrating awareness of false beliefs would infer that someone else who had not looked inside the crayon box, might *believe* that there were crayons inside the box. This ability to infer others' cognitive and emotional states or intentions predicts emotional knowledge[3] development (Curenton, 2015; Seidenfeld et al., 2014); is instrumental in children understanding and predicting their own as well as others' behaviors (Westby & Robinson, 2017); supports children in initiating and maintaining social relations with peers (Curenton, 2015; Espelage et al., 2018; Hofmann et al., 2016); and plays a role in reading comprehension (Filiatrault-Veilleux et al., 2016; Hyter et al., 2018; Peskin et al., 2016; van Kleeck, 2008).

There are currently very few studies investigating ToM in child AAE speakers (Curenton, 2015; Mills & Fox, 2016). Using an emic approach, Curenton and Gardner-Neblett (2014) state that African-based cultures emphasize social emotional knowledge in children because it is believed that in order for children to become linguistically competent, they must learn to convey their own emotions and recognize the emotions of those with whom they are conversing. Curenton (2015) found that some 3- to 5-year-old African American children who were able to explain emotions also exhibited prosocial abilities. ToM is used by African American school-age children when describing internal states of characters in stories and when telling fictional narratives, particularly when there are no visual stimuli (Mills, 2015a; Mills et al., 2013).

Failure to understand others' intentions, emotions, or mental states may indicate an impairment in ToM. Consider the following scenario. A group of African American adolescents are telling stories about what good cooks they have in the family. Each story teller within this group tries to embellish more than the previous story teller. One adolescent who was listening to all the other stories, shares a story about how the cooks in his family "suck." This adolescent might be exhibiting a ToM impairment in that he was not able to pick up on the cultural nuance that everyone was trying to make the cooks in their family seem better than anyone else's in this group discussion. He was not able to participate in the "one-upmanship" that was in line with the thinking and stories of the other members of the group.

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[2] The Crayon Box content task, originally called the "smarties" test, was developed by Perner et al. (1987) and augmented by Gopnik and Astington (1988). It is a widely used false belief task.

[3] Emotional knowledge or competence is "the ability to understand and describe emotions, the ability to understand that emotions are the consequences of interpersonal interactions and situational events, and the ability to demonstrate socially appropriate and age-appropriate emotions" (Curenton, 2015, p. 46).

23.5.2 *White American English Speech Acts in AAE Speaking Children: An Etic View*

Speech acts (SAs) are an important component of pragmatic language behavior. They are "the speaker's use of utterances with certain intentions in mind and the effect the utterance has on a listener in a given context" (Rivers et al., 2012, p. 2). The study of SAs in young African American children is sparse (Hyter et al., 2015). The few studies that have been conducted have used an "etic" analysis where speech acts present in the pragmatic behaviors of WAE speakers are imposed on the speech act behavior of AAE speakers (DeJarnette et al., 2015). Using this "outside in" approach, researchers examining specific SA behavior in AAE speaking children have noted that, to some degree, young speakers of AAE use SAs known to be present in young speakers of WAE (Hwa-Froelich et al., 2007; Stockman, 1996; Stockman et al., 2008).

Employing an etic approach to explore more broad representation of speech acts in young AAE speakers, DeJarnette (2006) examined SA behavior of nine African American children, aged 2 to 4 years old, in their home environment. The children interacted with caregivers and siblings in non-structured play. This study found that SAs that have been noted in the literature regarding WAE child speakers (Dore, 1974, 1975, 1978; Halliday, 1975; Moerk, 1975; Tough, 1977) were observed in the home environments of these young AAE speakers. The WAE SAs performed by young AAE speakers included agreeing with proposal, calling attention, elicited imitation, imitation, perform activity, performatives, refuse to answer, request/propose, statement, transfer object, wh-question, yes/no question (see Table 23.2). These SAs represent, consecutively, the following intentions: confirm another's proposition; direct attention to an object or event; direct another to imitate; utter again what has just been said; describe a specific event while enacting it; protest/tease/warn; not respond when obliged to do so; request action or permission; declare/share facts; indirect or direct request for exchange of an object; inquiry about something; and request affirmation or denial (see Table 23.2). DeJarnette (2006) found that there were trends for using certain SAs across age groups such that agree with proposal (confirm another's proposition), calling attention (directing attention to object or event), statement (declare/share facts) and wh-question (inquiry about something) were more frequently used by young AAE speakers in the home environment.

In a study to explore further the presentation of WAE SAs in young AAE child speakers and to check the pattern of use, DeJarnette (2006) analyzed video samples of ninety-five African American children, aged 2 to 4 years old, living in an urban Midwest setting in the United States. Videotaped samples were transcribed for child utterances, context and/or adult-interlocutor utterances as they occurred in a day care setting and across interaction sets (play, classroom activities, lunch, naptime and snack). Transcriptions were examined for instances of the twelve SAs identified by DeJarnette (2006). High interrater reliability was determined as two SLPs experienced in language sampling analysis rated a random sampling of the data for representative SAs. Parents used acknowledged features of AAE, such as semantics/

Table 23.2 White American English (WAE) speech acts (Dore, 1977; Moerk, 1975) noted in young African American English child speakers (DeJarnette, 2006; DeJarnette et al., 2015) and frequency comparison with WAE findings

Speech acts	Description	Frequency of SAs	
		AAE	WAE
Agree with proposal	Confirm another's proposition	7.88	17.00
Calling attention	Direct attention to an entity or event	7.88	5.80
Elicited imitation	Direct another to imitate	0.33	Not reported
Imitation	Utter again what has just been said	4.66	5.54
Perform activity	Describe a specific event while enacting it	4.00	22.30
Performatives	Protest, tease, warn	4.11	10.80
Refuse to answer	Not to respond when obliged to do so	0.33	Not reported
Request/propose	Request action or permission	0.66	18.50
Statement	Declare, share facts and information	60.55	13.80
Transfer of object	Direct or indirect request for exchange of an object	0.33	Not reported
Wh question	Specify inquiry regarding subjects, entities, time or process	11.70	9.62-11.70
Yes/no question	Requesting agreement or disagreement from another	11.70	9.62-11.70

lexicon (e.g. "man" for reference to son and "little mamma" as reference to daughter), phonology (e.g. ð /d/ [them ; dem]; /æsk/;/æks/), morphology (e.g. deletion of plural marker when number is stated; "ain't" as auxiliary), syntax (e.g. double negatives "not no"; zero copula "She a girl"; regularized reflexive "He hurt hisself") (Washington & Craig, 1994; Williams & Wolfram, 1977).

Except for elicited imitation, all speech acts displayed by WAE child speakers were also detected for young AAE child speakers in this study. Findings included some predictable age-related distribution for use of SAs:

(1) statement (sharing/declaring fact or information) was the most frequently used SA by all age groups although it was used less at 2-year-olds when compared to 3- and 4-year-olds;
(2) 2-year-olds used wh-questions (inquiry about things) more than 3- and 4-year-olds, but 3- and 4-year-olds used yes-no questions (confirmation/denial) more often than 2-year-olds;
(3) imitation decreases with age and function tends to change from practicing and repeating at 2-years-old to repeating at 3-years-old and finally it is used only to emphasize points at 4-years-old;
(4) refuse to answer occurred for 2-year-olds only; transfer of object (request for exchange of object) occurred only at 4-years-old.

In both home and school contexts it appears that young AAE speaking children displayed SAs commonly found in WAE. Additionally, there seems to be a minimum "core" of pragmatic SAs. Stockman (1994), cited in Stockman (1996), found a common core, "minimum competency core" [MCC], in seven children between 33 and 36 months of age. This MCC was identified using an etic analysis process and is defined as "the set of linguistic features that the least competent normal child should demonstrate" (Stockman, 1996, p. 359). The SAs or communicative functions that comprised the MCC were:

- Comments
- Requests for information
- Requests for objects/acts
- Unobligated responses
- Obligated responses
- Spontaneous imitations
- Initiated repairs
- Solicited repairs (Stockman, 1996, p. 361)

Using an emic analysis, DeJarnette (2006) found that a "core" of most frequently occurring SAs emerged across age groups for AAE speakers and this core included the following speech acts:

- agree with proposal (confirm another's proposition)
- calling attention (directing attention to object or event)
- statement (declare/share facts)
- wh-question (inquiry about something)

This "core" pattern has not been reported in the literature regarding SAs in young speakers of WAE (Dore, 1977; Keenan et al., 1977; Moerk, 1975) and, therefore, this core may reflect sociocultural nuances for AAE children's use of WAE SAs. While there are some etic-based data regarding SA behavior in young AAE speaking children, such data is lacking for older children and adolescents. Thus, further study of non-contrastive SAs and their pattern of use in AAE speaking children and adolescents is warranted.

23.5.3 *African American English Speech Acts: An Emic View*

Although it has been noted that AAE speakers use some SAs found in WAE, the pattern of verbal and/or nonverbal performance is distinctively different from WAE performance of SAs. This demands that AAE SAs be examined from an "emic" perspective where the SAs are identified from their origin in the AAE speaking community. As indicated by Hyter et al. (2018), "speech acts are historically transmitted social communication behaviors that allow sociocultural identity" (p. 135). Indeed, pragmatic language behavior for AAE speakers reflects sociocultural influences of this speech community (a group of people who share linguistic culture—the underlying beliefs, values, and uses of language). It can be said that pragmatics is what AAE does. That is, AAE is always about how language is used in context.

Table 23.3 AAE communicative functions and intentions expressed

AAE communicative function	Intention expressed
Ideological identity (self and collective affiliation)	To express thoughts that demonstrate individual or group connection to historical, economic, political, social cultural heritage
Portraying emotions (keeping ‘cool’ or acting ‘fool’)	To demonstrate loss or maintenance of emotional control; to display emotional state, commitment or attachment
Perspective taking/tools for thinking (use of wit all the while assessing effect on receivers); social exchange (interacting)	To demonstrate mental flexibility in devising or responding in snide or witty manner
Logging facts (factual information sharing)	To directly or indirectly share verifiable information
Controlling one’s world (overcoming odds/ oppression)	To exert self or group ability to rise above adversity
Use of sound for power (vocal projection for emphasis)	To exude rhythm/cadence in voice to personify concept expressed
Use of gesture for power (gesticulation for emphasis)	To use body and limbs to project attitude and force of message
Engaging in social exchanges	To use verbal and nonverbal behaviors to keep interaction vibrant

Speech acts in AAE have been identified over the last four decades (DeJarnette et al., 2015; Goodwin, 1991; Green, 2002; Hyter, 2000, unpublished results; Hyter et al., 2018; Mitchell-Kernan, 1972, 1973; Morgan, 1996, 2002; Rickford & Rickford, 1976; Rivers et al., 2012; Smitherman, 1975, 1977, 1994, 2000). In an attempt to capture the sociocultural characteristics of AAE pragmatic language behavior, and SAs in particular, DeJarnette et al. (2015) identified a conceptual framework for the communicative functions of SAs found in AAE speaking communities. The AAE communicative functions and the intentions that these functions express are presented in Table 23.3. Each communicative function emanates from an intuitive foundation of cultural intelligence and reflects how an AAE speaker uses verbal and nonverbal resources (speech acts) to express intentions. Intentions of the communicative functions expressed through SAs include: intention to engage with others (perspective taking/tools for thinking; social exchange; logging facts); and intention to exert control (controlling one’s world; use of sound for power; use of gesture for power) (DeJarnette et al., 2015). Extrapolating from the existing literature, DeJarnette et al. (2015) have identified several AAE SAs that can reflect these AAE communicative functions singularly or in combination (see Table 23.4).

Sample 1: Emic View of Speech Acts for Young AAE Child Speakers

The transcript below is of a YouTube video (2015) that illustrates several AAE SAs performed by a three-year-old preschooler as she interacts with her adolescent brother. Some lines of the transcript were excluded due to space limitations. This three-year-old demonstrates the cultural transmission of SA behavior in AAE including:

Table 23.4 African American English (AAE) speech acts and intention of communicative function

	Intention of the communicative function		
Speech act	1. Self-regulate	2. Engage others	3. Exert control
"Call & response" *Speaker proposes something and looks to audience for affirmation*		X	
"Givin' skin; Givin' dap" *Gesture used to affirm or signify unity*		X	
"He said-she said" *Checking facts by tracing the rumor mill*		X	X
"Ignorin'" *Treating others as non-existent/invisible*	X	X	X
"Instigatin'" *Starting a battle between others*		X	
"Loud Talkin'" *Pumping up the voice for attention and effect*	X	X	X
"Markin'" *Mimicking*		X	X
"Neck roll; cut eye; finger waggin/snappin" *Exaggerated movements to punctuate message*	X	X	X
"Playin' the dozens" *Dishing out insults about mother or family for one- upmanship*		X	X
"Rappin'" *Rhythmically making the facts plain*		X	X
"Readin'" *Verbal put down of someone who veils the truth*		X	
"Sermonizin' (testifyin'; preachin')" *Verbal tones like a preacher's style*	X	X	X
"Signifyin' (joanin', cappin', soundin', dissin', bustin', blazin', snappin')" *Insult that sends a message*		X	X
"Suck teeth" *Articulation depicting disgust or incredulity*	X		X
"Wolfin (braggin')" *Braggadocio, having bragging rights*		X	X

- neck-rolling (e.g. line 2: non-verbal gestures for emphasis)
- readin' (e.g. line 8: "Yes, you was," where she admonishes her brother to tell the truth)
- rappin' (e.g. line 15: "An' you heard me", with a rhythmical way of 'tellin' it like it is')
- signifyin' (e.g. line 37: "You luh' [love] yo [your] ownself", throwing an indirect insult at her brother)

[General context: Brianna (a typically developing child) and her adolescent brother are in a bedroom as the brother was making the video.]

	Speaker	Transcript	Speech act
1	**Brother**	"You idn't tell me nothin' Brianna. Whachu tell me?	
2	**Brianna**	[Lookin up at her brother, squinting her eyes, wrinkling her nose and neck-rolling her head] "I tol'you fo'times to hea me and you was in nat room."	*Neck-rolling*
3	**Brother**	"What room?"	
		Lines 4–6 are excluded	
7	**Brother**	"No, I was not."	
8	**Brianna**	[turns to face brother while shutting the door behind her] "Yes, you was."	*Readin'*
9	**Brother**	"Nooo"	
10	**Brianna**	[walking toward brother, then away moving her hands in up and down gesticulations that end with the arms bent at the waist and two hands facing upward looking with definitive expression in her brother's direction]	*Waggin'*
		"Yes you was—You cut duh light on or nothin' you cain' do"	*Readin'*
		Lines 11–14 excluded	
15	**Brianna**	[expressively gesticulates hands around and then in a repetitive downward direction]	*Waggin'*
		"Even now, you can' do nothin' what I say."	*Signifyin'*
		[hands on her upper thighs, upper body moving back and forth and neck-rolling side to side]	*Waggin'*
		[rhythmic phrasing] "an' you heard me" [rhythmic phrasing] "an' you ta:k to me you don' listen"	*Rappin'*
		[keeping same body posture, but exaggerating neck roll for emphasis]	*Waggin'*
		"Ta:k bout I wanna go in dat room"	*Readin'*
		"I say you not goin' nowhere"	
		Lines 16–36 were excluded	
37	**Brianna**	[looking at her brother and using neck roll] "I'on't know you 'on't luh' me." [moving arm across her chest and then in the direction of floor] "You 'on't luh' nobody-you luh' yo ownself."	*Signifyin'*

Sample 2: Emic View of Speech Acts for Adolescent AAE Child Speakers
The transcript below presents AAE adolescents in a classroom setting (DeJarnette et al., 2016). One of the speakers, Jerrell, has an interaction pattern that is "different" from that of his cultural linguistic peers, although he does use AAE SAs. The adolescents in this interaction demonstrate several AAE SAs including:

- dozens (line 5: "Yo momma!")
- givin' dap (line 15: Jayden gives pound in air to Jerrell)

- ignorin' (line 19: Jerrell's peers act as though he is invisible)
- signifyin' (line 24: "You lookin' los' ovuh dere.")

[Context: Classroom sample of Jerrell (adolescent demonstrating pragmatic language and social communication difficulties) and four classmates who are assigned to work on a science project by classroom teacher, Ms. Chase. Jerrell positions his desk, so it is spread a bit away from the group. No one tells him to come closer.]

	Speaker	Transcript	Speech act
1	**Aniyah**	"Alright y'all les get dis thang started!"	
2	**Xavier**	"I think we oughtta give the whole thing to Jerrell to do" (laughs).	
3	**Jerrell**	Stares in the teacher's direction	
4	**Xavier**	"Whatcho say to dat, Jerrell?	*Intigatin'*
5	**Jerrell**	"Yo momma!"	*Dozens*
6	**Tiana**	"Leave him 'lone an les do dis."	
7	**Jerrell**	Wads a piece of paper and throws it hard at Xavier	
8	**Xavier**	"Hey, you betta' chill man or I'll sic yo momma on you!"	*Dozens*
9	**Teacher**	From the other side of the room Ms. chase tells Jerrell and Xavier to stop clowning around and get to work.	
10	**Jerrell**	(seeming to direct his comment just into the air) "get off me!"	
11	**Teacher**	Ms. chase raises her eyebrow and is ready to reprimand Jerrell when Jayden speaks.	
12	**Jayden**	"It's alright, Ms. chase, we got dis."	
13	**Jerrell**	Jerrell turns to Jayden	
14	**Jerrell**	"Thanks for coverin' me homie."	
15	**Jayden**	Jayden gives a pound sign in the air to Jerrell and Jerrell reciprocates.	*Givin' dap*
16	**Jayden**	"Okay, look, Uhm checkin' out duh book and it say we got to follow steps one to five to dis right. So, Tiana and Aniyah, y'all check out steps one and two and my man Xavier gone look at three to five wit me. Alright?"	
17	**Tiana**	"Soun' good to me."	
18	**Aniyah**	"Yeah"	
19		Tiana and Aniyah start talking to each other about their part while Xavier and Jayden start talking about theirs. Neither pair invites Jerrell to join.	*Ignorin'*
20		Jerrell sits quietly doodling for several minutes.	
21		Suddenly, Jerrell leaves his seat and stands awkwardly in front of Tiana and Aniyah but says nothing as the girls continue to discuss what needs to be done.	
22		Both girls look at each other intently trying not to pay attention to Jerrell at all.	*Ignorin'*
23		Xavier sees the girls' discomfort with the situation.	

Table 23.5 Grice's maxims in African American English

Grice's Maxim	Explanation	Culturally responsive gloss of Grice's maxims appropriate for AAE
Quality	Be truthful and accurate about information	There is a time to be truthful, and a time to exaggerate; exaggeration or embellishment is valued, or in instances of racism there is a time to hide the truth
Quantity	To give the right amount of information in a given situation	There is a time to talk around a subject or to use indirectness
Relevance	Information shared is directly connected to the topic at hand	There is a time to go off topic for a purpose
Manner	To be clear and unambiguous	There is a time when double-speak or ambiguity protects from oppression

	Speaker	Transcript	Speech act
24	**Xavier**	"Jerrell, man, whatcho doin'? You lookin' los' ovuh dere."	*Signifyin'*

Adolescents and young adult AAE speakers often relate to SAs presented in rap and hip-hop forms. Rap and hip hop allow the expression of intentions in direct and indirect ways that may or may not agree with Grice's (1975) conversational maxims (quality, quantity, relevance and manner) and thus require a culturally sensitive way of examining SAs in AAE. The intentions often expressed in rap and hip hop include to:

- insult (signify, play the dozens, rap, snap or cap)
- mock (markin' or loud talking); brag (woofin' or toasts)
- call-response (putting out a request for affirmation and having a response)

In the AAE oral tradition these intentions often defy Grice's maxims as exemplified in Table 23.5.

In an excerpt from the documentary "Talking Black in America" (YouTube, 2017), hip hop jousting is explained by Keith Cross, literacy and arts educator from Stanford University as follows, "It's as much about the language itself and the connections that are being made as it is about like how the language is being delivered." In this same YouTube excerpt John Baugh, a linguist at Washington University in St. Louis, reiterates the importance of this genre by suggesting that, "An individual's ability to speak spontaneously, authoritatively in the vernacular is not only highly prized, but is literally used in verbal combat." Hip hop and rap epitomize SAs, that is, how to do things with words (Austin, 1962).

23.5.4 Speech Act Impairment in AAE Speakers

Any African American child or adolescent who is appropriately diagnosed with a language disorder (rather than a language difference) has the potential to display a pragmatic language or social communication impairment. Given the reduced receptive and/or expressive language skill set consistent with the diagnosis of impairment, such individuals might exhibit a significantly reduced ability to understand or produce socially coded intentions in a given context. Specifically, the AAE child or adolescent with a diagnosed language impairment may have a very limited number and range of SAs that are known to be unique to the AAE speaking population, that is, contrastive with WAE. Or, they may have a very limited range of SAs that are known to be shared with the WAE speaking population, that is, non-contrastive with WAE. In sample 2 above where Jerrell and his classmates interact, Jerrell demonstrates use of several AAE SAs including *playin' the dozens* (line 5), givin' dap (line 15) and signifying (line 24). However, Jerrell does not display the range of quick-witted retorts displayed by his peers. Additionally, Jerrell does not read well the nonverbal cues given by his peers (lines 22 and 24) and his own nonverbal behavior is awkward for the cultural context (lines 7 and 21). Jerrell's mismatched pragmatic language and social communication behaviors are noticed by his peers who respond in ways to move the class activity along, sometimes trying to include Jerrell and at other times excluding him.

Children who have difficulties or disordered pragmatics may not adjust their speech register to match the communicative context. An example of this limited change in register is talking to one's classroom teacher using the same register used to talk with peers, as demonstrated in the script below:

- Child: (to the teacher) Hey . . . Hey. . . I said, HEY [use of colloquial term to get teacher's attention]
- Child: I'm tryin'na get'cho attention
- Teacher: I am not your friend or a horse

Another characteristic of disordered pragmatics is making unrelated topic shifts during a conversation. The example below is of a child [child 2] making such a shift:

- Child 1: I was tryin'na tell the teacher sum'in
- Child 2: We was goin'na go to the mall [*unrelated topic shift*]
- Child 1: What'cho talkin' bout?

The following interaction of a group of 4-year-old AAE speaking children at free play in the construction center illustrates how the group uses both AAE and WAE SA behaviors. The typical AAE language users in the scenario show a good range of use for both AAE SAs (Call and Response, Cut eye, Loud Talkin', Markin', Signifyin, Wolfin') and WAE SAs (Calling Attention, Imitation, Performative, Statement, Yes/No question). However, Portia who demonstrates impaired language output has one conventional WAE speech act (Statement) and no AAE SAs and her style of interaction does not follow cultural norms. Just as was the case with the adolescent sample, peers in the young child group recognize the mismatch of social cultural abilities in Portia and they respond using AAE SAs to address them.

[Context: Four-year-old AAE speaking children at free play in the construction center with blocks, tools and plenty of space. One child, Portia, demonstrates impairment of pragmatic language and social communication behaviors.]

	Speaker	Transcript	Speech act
1	**Felicia**	Les go buil' a tower y'all.	*WAE: Request or Propose*
2	**Trey**	I'mma buil' de bigges' one.	*AAE: Wolfin'; WAE: Statement*
3	**Marvelle**	Uhn-uhn, mine go be de bes'. Now who else comin'?	*AAE: Wolfin';* *WAE: Statement, Request/Propose*
4	**Portia**	[Looking all around, but not directing her speech to anyone in particular] Me go a sink.	*WAE: Statement*
5		[The other children look at Portia and then at each other]	*AAE: Cut eye*
6	**Percel**	[in a whisper to the others]. Y'all hear that girl?	*AAE: Signifying;* *WAE: Calling Attention*
7	**Felicia**	[In a loud voice for Portia to hear, but while looking at Percel] She not buildin' wit' us noway, so come on.	*AAE: Loud Talkin';* *WAE: Statement*
8		[Felicia, Trey, Marvelle and Percel start collecting blocks and tools to build as Portia moves back and forth between them sometimes in the way, but not seeming to realize it]	
9		[Portia crosses in front of Felicia who has her hands full of blocks]	
10	**Felicia**	Watchit gi::l!	*AAE: Signifying (bustin')* *WAE: Performative*
11		[Portia does not move any faster to get out of Felicia's path and Trey notices what happened]	
12	**Trey**	Could say sorry!	*AAE: Readin';* *WAE: Statement*
13		[The children put their blocks and tools down around the middle of the building area]	
14	**Felicia**	Mine dope, y'all!	*AAE: Wolfin'* *WAE: Statement*
15	**Marvelle**	Yoz like chicken lil, but mine is taller den a house!	*AAE: Signifying, Wolfin*
16	**Felicia**	His wall gone fall!	*AAE: Signifyin'; WAE: Statement*

	Speaker	Transcript	Speech act
17	**Trey**	Man, mine bedda dan all y'alls'!	*AAE: Wolfin;* *WAE: Statement*
18	**Percel**	We know how to buil'!	*AAE: Call-Response;* *WAE: Performative*
19	**Felicia**	You right!	*AAE: Call-Response;* *WAE: Agree with proposal*
20	**Marvelle**	Yep!	*AAE: Call-Response;* *WAE: Agree with proposal*
21	**Trey**	Sho' nuff!	*AAE: Call-Response;* *WAE: Agree with proposal*
22		[Portia sits on the floor next to where the others are building their towers and she starts to sing her ABCs ending with "now, me ABC, tell me, me, me, me]	
23	**Felicia**	[sucks her teeth and mocks Portia] ABC, tell me, me, me	*AAE: Suck-teeth, Markin'* *WAE: Imitation*

In sum, pragmatic language and social communication impairment are suspected when the African American child or adolescent withdraws from social interaction and/or does not appropriately use culturally relevant SAs to express intentionality. Additionally, a pragmatic language and social communication impairment may be suspected when peers withdraw from interaction with a target child or adolescent. Finally, pragmatic language and social communication impairment may be present when the child or adolescent displays inordinate dependence on adults to convey their intended message to peers as noted in Jerrell's case.

23.5.5 African American Voice and Prosody as a Pragmatic Tool

McWhorter (2017b) has argued that characteristic patterns of AAE have a voice presence that is captured by nuances in speech sound (mostly vowels) and co-articulated productions. These productions include prosodic variations. He uses the term "blaccent" to represent black voice production that can be detected even when the speaker uses white standard English grammar:

> "ALTHOUGH WE DON'T PUT IT THIS WAY, in relation to white American English, black people tend to have an accent…. This issue can be viewed as having two levels. The surface level…the "classic", obvious differences in sound between black and white American speech. The deeper level is the aspect of the sound that leads one to wonder why even if a person doesn't sound black in an immediately obvious way—for example, like the typical rapper—we can still somehow detect "blackness" in the voice." (p. 66) "Most important, the blaccent is normal. *What would be strange is if it didn't exist.*" (p. 82)

McWhorter (2017b) suggests that "blaccent" is not slang, and includes *more* than AAE grammatical patterns, deeper pitch of voice, and southern accenting. Moreover, he conjectures that not all African Americans speak with blaccent though most have this cultural trait in their voice. This subconscious trait reflects cultural exposure that when all else does not identify one as black, the blaccent remains as a vestige of the fact that one belongs to the African American experience.

McWhorter sees blaccent as a culturally heritable trait rather than a trait of genetics or biological predisposition. Some studies, however, have found evidence that differences between black voice and white voice exist at acoustical, physiological and perceptual levels for adults and at the acoustical level for prepubescent and pubescent children. At the acoustical level, fundamental frequency (F_0) has been studied for adults, prepubescent and pubescent children. For adults, both African American males and females were found to have statistically significantly lower F_0 compared to a WAE voice (Ducote, 1983; Xue et al., 2001). Adult African American voice has also been shown to demonstrate greater variability (pitch sigma) and range (Hudson & Holbrook, 1982; Jones & Mayo, 2001; Mayo et al., 2001; Mayo & Manning, 1994; Tarone, 1973; Walton & Orlikoff, 1994). Similar to findings for adult African American voice, the voice of prepubescent and pubescent African American males and females has lower F_0 compared to WAE, with some studies suggesting a statistically significant difference (Wheat & Hudson, 1988) and others noting differences but not at a statistically significant level (Awan & Mueller, 1996; Gelfer & Denor, 2014). Regarding pubescent African American male voice, Hollien and Malcik (1962, 1967) found that F_0 was lower and these adolescents showed greater pitch range and early voice change than noted in adolescent WAE speaking males.

Research of the physiology and perception of African American voice has focused on adult voice rather than on prepubescent and pubescent voice. When examining vocal tract physiology, formant frequency analysis shows that adult African American males and females show lower formant ranges for F_1 (tongue height and jaw opening), F_2 (tongue body movement front to back), and F_3 (lip round to spread) (Mayo & Manning, 1994), with some variability depending on the vowel and formant examined (Andrianopoulos et al., 2001a, 2001b). Perceptual study of African American voice has taken the form of rating favorable versus less favorable voice. It has been found that African American voice is rated less favorably or differently when compared to ratings of WAE voice (Larimer et al., 1988; Saniga et al., 1984). Overall, the acoustic, physiological and perceptual data tend to support McWhorter's speculation that blaccent is a real phenomenon with identifiable as well as subconscious characteristics that are products of cultural exposure, such that voice timbre is retained to identify African American voice (blaccent) even for African Americans who have primary exposure to WAE productions.

Loudness is one feature of African American voice that is not mentioned by McWhorter (2017a, 2017b), but is often anecdotally noted when AAE voice is rated as being less favorable (Larimer et al., 1988; Saniga et al., 1984). Being "boisterous", often expressed by increased volume of the voice, is a characteristic of several SAs as noted above. The loudness characteristic is used in AAE voice to assign illocutionary force (commitment of the speaker and intention) to the utterance. The use of loudness does not mean that the speaker is angry, upset or out of control. An often-misinterpreted voice feature of AAE voice is prosody.

Prosodic variations are one of the hallmarks of AAE (Hyter et al., 2018; Nielsen, 2012, unpublished results; Thomas et al., 2010), and often mark speech acts such as those described earlier in Table 23.4. Prosody is the interface of loudness with variations in pitch (intonation), rate (rhythm), and stress and has been identified in a phenomenon known as "forestressing" for AAE speakers (Baugh, 1983). Baugh (1983) has described forestressing as the tendency to put primary stress on word syllabication where WAE usually places secondary stress. Words such as 'police', pronounced as /po➜lice/ by a speaker of WAE, would be pronounced /➜police/ in AAE and used for emphatic intent.

Another prosodic characteristic of AAE is falsetto, an unusually high pitch. This is also used to communicate indignation, make a challenge in an argument, emphasize a point, and is often employed during emotionally (or racially) charged interactions (Alim, 2004; Nielsen, 2010; Podesva, 2016). Vowel elongation also has been found to be used for emphasis and differentiating new (i.e. unknown) from given (i.e. known) references in narrative (Hyter, 1994, unpublished results; Hyter et al., 2018).

23.5.6 African American Voice ("Blaccent") as a Tool for Pragmatic Performance

While McWhorter's discussion of blaccent does not consider the use of voice for pragmatic and social communication purposes *per se*, blaccent cuts across geographic, socioeconomic and political divides within the African American speech communities and has the practical, that is, pragmatic function of connecting a person of African American descent to her or his speech community or communities. Indeed, AAE voice, and thus blaccent, uses suprasegmental features of speech such as loudness, intonation and falsetto pitch to capture or emphasize intention in pragmatic language and social communication (Alim, 2004; Hyter et al., 2018; Nielsen, 2010; Tarone, 1973; Thomas, 2015; Wolfram & Schilling-Estes, 2006). In fact, blaccent, and loudness as a part of it, is very apparent in using *sound for power* as a function of SAs (Table 23.3) and for specific SAs that capitalize on prosodic rhythm and voice volume to send an intended message. This includes markin' (Mitchell-Kernan, 1989; Morgan, 2002; Nielsen, 2012, unpublished results), sermonizin'/preachin' (Green, 2002; Smitherman, 1975, 2000), rappin and signifyin' (Green, 2002; Mitchell-Kernan, 1989; Sistrunk, 2008; Smitherman, 1977), and call and response (Green, 2002; Smitherman, 1975, 2000) (see Table 23.5). In the following sermonette, blaccent is used in various SAs:

Speaker	Statement	Speech act
Preacher	One day I was stanin' at de bus stop and I hu:d dis chil' talkin' ovuh her momma	
	—'Momma you thank you white?'	*Loud talkin'*
	God maidjew black and beautiful, so don' ack like you white when we out heyuh.'	*Signifyin'*
	Outta de mouf uh babes!	*Readin'*
	Chu::Ch can I get a witness!!!	*Call*
Congregation	Amen!	*Response*
Preacher	We gotta stop ackin' like we white when weez black like God bless us to be Chu::Ch	*Readin'*
	I say gi' God de glooory	*Sermonizing/ starting to wind up voice for whoopin*[4]
	Tuhuh	*(vocal play)*
	Gi God de glooooory,	*(Sermonizing/start whoopin)*
	Tuhuh	*(vocal play)*
	Gi' God de glooooooooory!	*(bigger whoopin)*
	Be who you iz cause He made you dat way fo His glooooooooooooooory!	*(Sermonizing /full whoopin')*
	Non' be ackin' like you all stuck up,	*Signifyin'*
	[in falsetto voice] whichya noz so fa in de aiuh you be breevin' the staws an trippin ovuh you own feets.	*Signifying*
	[in a moaning tone] Uhmmm, Can I get a witness??	*Call*
Congregation:	Say it preachuh!!	*Response*

"Sermonizing" is a SA that eloquently shows AAE voice, blaccent, in its cascading use of vowels and volume (loudness). It is typical for real sermons by AAE preachers to be characterized by an increased use of AAE features (Green, 2002). A similar description can be made of sermonizing that emerges from conversations or interactions among AAE speakers. In African American culture the voice is used in both sacred and secular spaces to verbally express intentions. One can use a sermonizing style of voice on the street, or a child can use it during play as much as in the church. In a one-minute YouTube (2013) video clip, a young child called Riah, of about 3 or 4 years of age, is playing preacher. She uses many of the vocal, prosodic and physical mannerisms used by preachers in some African American churches:

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[4] Whoopin' is a "rhythmically stylized presentation" where the preacher uses body movement, and intonational changes (e.g. vowel elongations, pitch variation) to make a point (Debose, 2015; Pinn, 2002, p. 63).

Riah	I decla dat da Lor i' gooooood	*Vowel elongation*
	Jesus saiiiiiid dat todaaaay dat da x xx	*Vowel elongation;*
	Huh	*Vocal play*
	Haaaaa ya luuuuu: Ya	*Vowel elongation;* *Whoopin'*
	It's ovah, it's ovah, it's oooooovah	*Repetition for emphasis; whoopin'*

23.5.7 Prosodic and Voice Impairment in AAE Speakers

The extension of vowels in musical trains help to punctuate the performance of the word in African American oral tradition. African American children and adolescents who have voice impairments that impact their use of suprasegmental features will have difficulty appropriately infusing their SAs and oral communicative discourse with voice nuances to express various culturally based linguistic intent. Those African American children who have pragmatic language and social communication impairments will have difficulty using features of voice to accurately reflect communicative intentions, or they may have difficulty understanding the intention of another's SAs because of their inability to appropriately read suprasegmental cues. Voice impairment can affect psychological or physiological abilities. For example, the child or adolescent who is a loud talker all of the time (vocal hyperfunction) is not able to use the voice for subtle intentional cues or prosodic emphasis. Additionally, the child or adolescent who has a pragmatic language disorder may not be able to interpret or use loudness, prosodic patterns, or pitch patterns that have cultural significance.

23.5.8 Conversational Discourse

African American English speakers use extended extracts of language (discourse) to connect with and relate to each other out of shared historical, sociopolitical and sociocultural experiences. Discourse patterns in the African American speech community reflect oral traditions that have been passed on across generations by a people whose history as descendants of people who were enslaved marks them as "involuntary immigrants" (Ogbu, 1991; Ogbu & Simmons, 1998) or people who were forced to migrate, for whom assimilation is often unobtainable due to inhumane treatment and persistent discrimination. Rickford and Rickford (2000) discuss the concept of "spoken soul" that aptly captures African American discourse. Spoken soul is practiced to varying degrees by most African Americans including those for whom WAE is characteristic of their professional and personal language use (Rickford & Rickford, 2000). Indeed, as Rickford and Rickford suggest, African Americans have historically used spoken soul to engage in various speech acts such as to express ethnic identities, to confide in friends, and to joke or tell stories (Rickford & Rickford, 2000, p. 4).

"Spoken soul" captures the essence of African American discourse and is the term used for the remainder of this discussion of African American discourse. The essence of spoken soul reflects the will of African Americans to preserve cultural habits, traditions and values through language performance, and is one of the reasons that AAE pragmatic expressions persist and are resilient. Spoken soul's method/style of interacting through talking, or to "conversate" (Rickford & Rickford, 2000), fits Hymes' (1974) "SPEAKING" model for discourse, which refers to **S**etting or scene; **P**articipants; **E**nds or intentions; **A**cts (speech acts); **K**ey (tone or rhythm); **I**nstrumentality (style or register); **N**orms (rules of engagement); and **G**enre (sacred or secular). A review of the first sample in section 23.5.3 illustrates that young African American children are acculturated with spoken soul in conversational discourse. There, in one of the bedrooms of their home (**S**etting), preschooler Brianna hosts a conversation with her brother (**P**articipants). Brianna manages the conversation using communicative functions (**E**nds) and speech acts (**A**cts). Her topic management strategies include repetitiveness (telling her brother not to say he's sorry) and using dramatic gestures and voice tone to emphasize her message and keep her brother's attention. She uses verbal rhythm and non-verbal gestures (**K**ey) as part of the conversatin' to punctuate her points. Her conversatin' is an informal style (**I**nstrumentality) as she uses verbal and non-verbal behaviors that are culturally based to engage with her brother (**N**orms). Her conversatin' is secular (**G**enre).

Spoken soul allows African Americans to express cultural values and symbols through doing things with words. van Keulen et al. (1998) attempt to identify communication styles (spoken soul) that reflect African American cultural values and symbols. van Keulen et al. include the following in their list: sharing (collectivist orientation to life); touching (tactile behaviors that connect speaker and listener); distance (proximity with closeness representing connectedness); relationship intimacy (speaker and listeners are relating on more than a superficial level and exhibit a closeness in doing so); rituals (verbal and nonverbal habits that affirm affinity to the culture); jiving (intentional trickstering that is non-malicious); boasting (bragging for the intended purpose of exaggerating desired attributes); uniqueness (self-affirming and cultivating authentic traits); and assertiveness (behaviors that promote a proposition strongly).

Sharing some similarities with the communicative styles that capture values and symbols as indicated by van Keulen et al., Vetter (2013) notes seven discourse patterns for spoken soul and marks them as follows:

1. "Exaggerated language"—where unique words and expressions are generated to emphasize a point being made by the speaker
2. "Mimicry"—an intentionally direct imitation of another's speech or gesture
3. "Improvisation"—spontaneous witticism or remark that capitalizes on some change in the context of the conversation
4. "Braggadocio"—expression suggesting bragging rights by the speaker
5. "Tonal semantics"—use of intonation and prosody to convey meaning

6. “Repetition and rhythm”—use of sound repetition (alliteration) and song-like beat to the verbal expression
7. “Signification”—artful insult sending a message

In addition to discourse patterns, discourse management in spoken soul shows cultural traits. Discourse management “…is the ability to construct and comprehend text connected in the form of a conversation, narrative or exposition/explanation” (Hyter et al., 2018, p. 137). Discourse management is important for discourse coherence and includes: topic selection (which must consider the mental state/interest of the audience); method of introducing the topic into the conversation (i.e. skillful integration into the conversation); development and maintenance of the topic in an interesting and relevant way for the audience; and terminating a topic (avoiding abrupt ending). Conversational discourse (spoken soul) in African American child and adolescent speakers reflect social cultural practices for topic initiation and maintenance strategies (Connor & Craig, 2006; Craig et al., 2003; Craig et al., 2014; Craig & Washington, 2004; Flowers & Flowers, 2008; Hyter et al., 2018; Terry et al., 2012).

Speech-language pathologists in charge of assessing, diagnosing and implementing intervention for pragmatic language disorder must be cognizant that features of spoken soul in AAE speaking children and adolescents are not disorders. Indeed, “conversatin’” is a legitimate style of conversational discourse in the African American speech community that uses culture-specific speech acts (see section 23.5.3) to influence SPEAKING, i.e. Hymes’ (1974) model of discourse as noted above. Professionals, therefore, must be aware that AAE speaking children and adolescents who are conversatin’ (like Brianna, for example, in the first sample of section 23.5.3) will tend to use words vivaciously, “tell it like it is”, cleverly retort or respond, gesticulate to emphasize a point, strongly and loudly assert, and poignantly reiterate as they engage in discourse in any given setting and with various participants (van Keulen et al., 1998). Again, these are typical productions of pragmatics within a language variety, not disorders (Hyter et al., 2018).

23.5.9 Narrative and Expository Discourse

Spoken conversational discourse is the oral pattern for discourse that has been shown to influence narrative and expository forms in African American child and adolescent speakers (Charity et al., 2004; Connor & Craig, 2006; Craig & Washington, 2004, 2006). Thus, the cultural linguistic patterns of spoken soul used in conversational discourse are reflected in narrative and expository forms of spoken soul. Both Vetter (2013) and van Keulen et al. (1998) identify the liveliness of spoken soul as a core concept that captures cultural nuances for conversational discourse and this same spirited core concept is found in narrative and expository discourse. Also, the social cultural patterns of repetitiveness, talking around a topic (circumlocution), using communicative devices to connect with the communicative partner or audience, and dramatized style of delivery influence the presentation of coherence found in spoken soul narratives and expository explanations produced by African American children and adolescents. Thus, in projecting cultural and social influences, the presentation of narrative and expository forms of spoken soul appear counter to etic considerations for these dialogical expressions.

In an etic way of thinking about narrative and expository text coherence, relevance and felicity must be explicit. Etic tradition denotes that in the construction of forms, coherence (making sense as a whole) and how cohesive the elements of a narrative or explanation are, are critical. Additionally, events and elements in discourse are said to need to be connected in relevant and felicitous ways and that there must be a logical sequence that makes sense. However, for spoken soul other factors may be more important such as repeating, talking around a topic, or embellishing a topic. These characteristics can appear to impact fluency and coherence for those outside of the cultural linguistic community while those who use spoken soul are unbothered by digression or exaggeration.

There is a sizeable literature that describes the narrative skills of AAE speakers (see the systematic literature review by Hyter et al., 2015). Gorman et al. (2011) found that African American children produced more stylistic embellishments (e.g. fantasy stories, suspense mechanisms) in fictional narratives than did their Latinx[5] and Euro American counterparts (p. 176). These embellishments served to increase narrative quality and listener engagement (Gorman et al., 2011). Mills (2015b) found that fictional narratives of school-age African American children included such features as “interesting modifiers, repetition, and internal state words” (p. 39) more often than personal narratives, which led Mills to conclude that fictional narratives may be more “informative and culturally fair” (p. 39) for assessing the language skills of African American children and adolescents. Mills found that typical narrative analysis measures—narrative length, lexical diversity, average sentence length—did not differ between African American children in general education and those in gifted education. She also reported that African American children (Mills, 2015b) performed similarly to other WAE speaking children reported in other studies on measures of syntactic complexity—total number of C-units (Norbury & Bishop, 2003; Strekas et al., 2013), number of different words (Cole, 2001), and mean length of utterance in words (Hoffman & Gillam, 2004).

In separate observations, Champion (2003) noted themes in narrative production of African American speakers such as use of sound in narratives (tonality, ideophone); ways of connecting elements in a narrative (repetition, piling and association, parallelism); ways of moving back and forth among topics (digression); ways of painting a picture of ideas (allusion, symbolism and imagery). Kersting et al. (2015) used Champion’s (2003) codes along with others, such as cultural reference, to see to what degree narrative forms attributed to AAE oral traditions are differentially used by African American adolescents and WAE speakers. As anticipated, they found that African American adolescents used AAE discourse features significantly more than WAE speakers and that parallelism (“playing one set of words or

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[5] Latinx is a gender-neutral reference for Latino, Latina and Latin@ (Salinas & Lozano, 2017).

images against another without changing the theme of the narrative") and cultural reference ("used for cultural vocabulary and idioms") were two AAE discourse codes that most distinguished the groups of adolescents (Kersting et al., 2015, pp. 96–97).

There is not as much literature about expository discourse used by AAE speakers (Hyter et al., 2015). Characteristic social cultural patterns that have been noted in research regarding discourse management include repetitiveness, talking around a topic (circumlocution), using communicative devices to connect with the communicative partner or audience, and dramatized style of delivery (Ball, 1992, 1996, 2002). Koonce (2015) found that third- and fourth-grade speakers of AAE provided rules, details, and strategies related to explaining how to play their favorite game or sport. The mean length of T-Units were similar for this group as were reported for WAE speakers of the same age. Also, Koonce stated that the participants in her study "demonstrated the ability to stay on topic throughout their explanations, produce extended language free of excessive revisions, fillers, or pauses" (p. 85). Ball (1992, 1996) observed that African American adolescents had patterns of text structure that would conflict with coherence as prescribed for WAE users. The patterns fit with AAE oral traditions and include interspersion where narrative is placed within an expository text, circumlocution where explanations have elements that are thematically linked, and a recursive pattern where repetition is used to emphasize or reiterate.

Smitherman (2000) found that in the expository writing of African American adolescents, repeated themes that pointed to the African American community, tradition and culture were important (e.g. use of culture specific terms and idiomatic expressions, folklore and proverbs). Smitherman also found that cultural communicative devices that captured tone, rhythm and drama were also important to the African American adolescents' explanations (e.g. evocative language and sermonic tone). Overall, what emerges from observations of discourse in African American child and adolescent speakers is that cultural identity and cultural reference are themes that are staples in narrative and expository forms of spoken soul. Whether through uniqueness of the words, idioms or proverbs used, sound/tonal characteristics expressed, or images developed, cultural identity and cultural reference are expressed in discourse of African American children and adolescents.

23.5.10 Discourse Impairment in AAE Speakers

Discourse impairment must be viewed from the lens of what is culturally acceptable for the African American child or adolescent. The African American child or adolescent who is neither able to use age appropriate, culturally acceptable patterns or standard school patterns for organizing, formulating, producing or comprehending text in conversation, narrative or exposition might be identified as presenting a discourse management impairment. Yet, the African American child or adolescent who uses cultural patterns for conversational, narrative or expository discourse but is not able to code switch/code shift to meet standards set by the school must not be considered "impaired".

23.6 Assessment and Intervention

Culturally responsive assessment and intervention processes should accommodate language varieties (Hyter & Salas-Provance, 2019). For social pragmatic language, utilizing assessments that focus on the use of language in context is essential. Often these types of assessments are informal assessments such as observational measures (e.g. *Pragmatic Observational Measure*, Cordier et al., 2014; *Targeted Observations of Pragmatics in Children's Conversations*, Adams et al., 2011) and narrative and expository text analysis (e.g. *Index of Narrative Macrostructure*, Justice et al., 2016; *Narrative Scoring Scheme*, Heilmann et al., 2010; *Expository Scoring Scheme*, Heilmann & Malone, 2014; *Test of Narrative Language-Second edition*, Gillam & Pearson, 2017). Some of these measures are described in more detail in Hyter et al. (2017). Given these guidelines, we suggest that conversational interactions among peers, and oral and written narrative and expository samples be used as tools of pragmatic assessment, and that a dynamic assessment method be employed. Employing an emic approach and using data from studies on theory of mind (e.g. Curenton, 2015; Curenton & Gardner-Neblett, 2014; Hyter et al., 2018; Mills, 2015a; Mills et al., 2013), speech acts (e.g. DeJarnette et al., 2015), narrative macro and microstructures (e.g. Champion, 2003; Gorman et al., 2011; Kersting et al., 2015; Mills, 2015a, 2015b; Mills et al., 2013), and expository text (e.g. Ball, 1992, 1996, 2002; Koonce, 2015) can be used to support interpretation of the results of collected samples.

Dynamic assessment is a process where clinicians first teach strategies that children will need to use on an assessment task. In this manner the clinician is serving as a mediating agent, providing mediated learning experiences (Feuerstein et al., 2006). The clinician then observes and takes notes of how quickly the child is able to learn the strategies (modifiability), and finally measures the child's ability to demonstrate the learned skill(s). This assessment method can provide information that is culturally and linguistically responsive (Peña et al., 2001, 2006). Peña et al. (2006) used two wordless picture books to collect narratives from first- and second-grade children. After collecting the narratives, they taught strategies to the children and then collected their narratives again. They found that children who received the mediated learning (strategy teaching) produced greater changes in narrative macrostructure (i.e. complete and complex narratives) than did children who did not receive mediated learning, although the narratives of children with language impairments also improved after mediated learning. They also concluded that observations of how readily modifiable a child was (i.e. the effort of the clinician to teach the child a strategy and the child's responsiveness to the instruction) was "the strongest and best predictor of language ability" (p. 15).

With regard to intervention, it is important to engage culturally responsive processes. An essential component of culturally responsive intervention is for clinicians to engage in continual reflective practice in order to recognize cultural nuances in communication and to move closer to culturally responsive care. Additionally, it is important to collaborate on intervention goals and approaches with the individual and/or family members in need of communication support (Hyter & Salas-Provance, 2019). There are articles that address pragmatic intervention and at least three systematic reviews of pragmatic treatment (Gerber et al., 2012; Law et al., 2011; Petersen, 2011), but few of the studies examined included African American children, adolescents or AAE speakers as participants. Interventions with evidence for supporting the social pragmatic communication of AAE speaking children and adolescents are minimal. Nevertheless, general principles of culturally and linguistically responsive interventions must be incorporated into the practices of speech-language pathologists. These general principles include:

1. Engaging in *critical self-reflection* by becoming aware of and by calling into question your own cultural perspectives, assumptions, biases and beliefs. Those who are critically self-reflective are aware of the impact the identities and long-held assumptions they bring into the intervention setting may have on the intervention outcome (Hyter & Salas-Provance, 2019).
2. Exercising *cultural humility* (Ortega & Faller, 2011), which requires continual critical self-reflection and the willingness to learn from the individuals and families with whom we are working.
3. *Learning* about the political and economic history and the world views of others (Hyter et al., 2017; Hyter & Salas-Provance, 2019; Pillay & Kathard, 2018), which will allow us to situate our speech-language practice into culturally relevant contexts for those with whom we are working.
4. Engaging in *cultural reciprocity and intentionality*. Cultural reciprocity is the ability to convey the values that underlie the clinical practices in which you are engaged, and to be able to make connections with the values that underlie the clinical decisions/needs expressed by the individuals and families with whom we work (Hyter, 2014; Hyter & Salas-Provance, 2019; Kalyanpur & Harry, 2012). Cultural intentionality is being able to generate solutions to clinical problems from various perspectives (Jones, 2014).
5. Being willing to strive for *equity* and *social justice* through your clinical practice. Engaging in practice that promotes equity (i.e. everyone having what they need to be successful in their daily life) and social justice (i.e. working for all persons to have equal access to all resources and services) is based on the premise that communication is a human right (Hyter et al., 2017; Jogerst et al., 2015).

23.7 Summary

In this chapter, we focused on explaining the social pragmatic communication skills of African American English-speaking children and adolescents. We provided examples of typical pragmatic skills and those skills that might be considered impaired. Additionally, we strongly suggested that adding emic (intracultural) analyses to the etic (intercultural) analyses we typically perform in the area of pragmatics, is important for capturing a more wholistic picture of African American children's and adolescents' social pragmatic strengths and possible challenges. The chapter concluded with assessment suggestions, and strategies for engaging in culturally and linguistically responsive intervention.

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Chapter 24
Children and Young People with Written Language Disorders

Gary A. Troia

24.1 Introduction

Literacy proficiency is important for a variety of reasons: (a) it is both a tool for learning and for demonstrating learning (Cunningham & Stanovich, 1991; Graham & Perin, 2007a, 2007b; Keys, 2000; Shanahan, 2009; Sparks et al., 2014; Sperling & Freedman, 2001); (b) it is a significant predictor of performance on assessments used for school accountability purposes (Jenkins et al., 2004; Reeves, 2000; Silver & Saunders, 2008); (c) it serves as a gateway for employment and promotion and is expected for matriculation into and completion of college (e.g. Hernandez, 2012; Jackson, 2005; National Commission on Writing for America's Families, Schools, and Colleges 2004; Rychen & Salganik, 2003; Smith, 2000); and (d) it is essential for participation in a global information and technology society (e.g. Anderson et al., 1985). Alas, a majority of students in the United States do not demonstrate grade-level mastery of reading and writing skills (National Center for Education Statistics, 2012, 2018). This is likely due in part to weak K-12[1] classroom literacy instruction that fails to address the needs of a diverse student population (e.g. Applebee & Langer, 2011; Cutler & Graham, 2008; Fresch, 2003; Gillespie et al., 2014; McCarthey & Ro, 2011; Moats et al., 2006; Pressley & Wharton-McDonald, 1998; Rowan et al., 2004; Stringfield et al., 1997; Swanson, 2008; Vaughn et al., 2002). However, the focus of this chapter is on those children and young people who display written language disorders and the intersectionality of written language and pragmatics in such individuals.

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[1] Kindergarten, the earliest year of compulsory education in the United States, which most students enter the year in which they turn age 5, through grade 12 secondary school, called high school, at which point successful completion grants students a diploma or certificate of completion.

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24.2 Defining Written Language Disorder

Written language disorder is a broad term encompassing both deficits in reading and writing texts. On the one hand, reading disability may be characterized by: (1) difficulties in recognizing words in isolation and in text (depending on the severity of word recognition difficulty, comprehension may or may not be significantly impacted), often referred to as dyslexia and presumed to be caused by underlying problems with processing phonological information accurately and rapidly (e.g. Catts & Kamhi, 2005; Moats, 2008; Vellutino et al., 2004); (2) difficulties with comprehending text despite word recognition skills that are fairly intact, often presumed to be associated with an underlying spoken language impairment (e.g. Kamhi & Catts, 2012); or (3) a combination of both word recognition and reading comprehension difficulties, which may be caused by a variety of conditions including severe dyslexia, severe oral language impairment, and other developmental disorders.

Writing disability, on the other hand, may be characterized by: (1) difficulties with handwriting and keyboarding, often referred to as dysgraphia and which may be due to motor coordination difficulties, fine motor weakness, or severe spelling difficulties (spelling and handwriting are strongly related; e.g. Berninger & Wolf, 2009; Berninger & O'Malley, 2011); (2) difficulties with spelling in the absence of handwriting difficulties, likely the result of dyslexia given that word recognition and word spelling rely on similar cognitive and linguistic operations and shared knowledge sources (e.g. Berninger et al., 2008; Bourassa & Treiman, 2001; Ehri, 2000; Masterson & Apel, 2007; Moats, 1995); (3) combined spelling and handwriting/keyboarding difficulties, a disorder associated with writing mechanics/conventions; (4) difficulties with expressing ideas in writing without accompanying deficits in writing mechanics, most probably due to an underlying spoken language impairment; and (5) difficulty with all aspects of writing.

Written language disorder is considered a type of neurodevelopmental disorder and, more precisely, a specific learning disorder, according to the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association, 2013). It has a reported prevalence of between 4 and 9% in DSM-5. However, prevalence estimates vary substantially depending on how the condition is operationalized, whether reading or writing disability is the focus of concern, and the gender of the affected individuals, with males generally diagnosed with a specific learning disorder more frequently than females (e.g. Badian, 1999; Katusic et al., 2009; Shaywitz et al., 1990). Specific learning disorders such as written language disorder are characterized by unexpectedly low academic achievement considering the child's age, instructional history, and general intellectual and sensory capacities (Alt et al., 2017; Lyon et al., 2003; Pennington & Bishop, 2009).

Specific learning disorders appear to originate from neurobiological differences marked by structural and functional deviations in the brain and putative genetic susceptibilities (e.g. Catts et al., 2012). The underlying cause of a specific learning disorder is most likely multifactorial. Importantly, environmental variables such as exposure to oral language and early literacy experiences can interact with intrapersonal genetic and neurological differences to manifest a specific learning disorder in reading or writing (e.g. Hoff, 2013; Stothard et al., 1998). While many individuals who struggle with comprehending spoken and written language (e.g. those who display reading comprehension problems) often also struggle with expressing language through oral and written modes of communication, there is nevertheless dissociation of receptive and expressive difficulties in that some individuals possess adequate comprehension but poor expressive abilities. That is to say, underlying language difficulties can lead to problems in both reading and writing, but it is entirely possible for a person to possess intact reading (and receptive oral language) abilities but exhibit poor writing (and expressive oral language) abilities (Conti-Ramsden & Botting, 1999).

24.3 Pragmatic Deficits in Children and Young People with Written Language Disorder

It is important to note that most evidence regarding the pragmatic abilities of school-age individuals with deficits in reading and/or writing comes from studies in which participants were identified as having spoken language impairment and not necessarily dyslexia or dysgraphia. Thus, it is not clear if children and adolescents who just struggle with word reading, spelling, and/or handwriting also display pragmatic language difficulties. An additional relevant point to consider is that, in some cases, children and adolescents with oral language impairment may exhibit difficulties with pragmatics that appear unrelated to their deficits in language content and form, though they would not be considered to have co-occurring autism spectrum disorder (Bishop et al., 2000; Brinton et al., 1997a; Conti-Ramsden et al., 1997).

Conversely, youth with spoken language impairment do not consistently demonstrate pragmatic deficits when evaluated using theory of mind tasks (Farmer, 2000; Gillott et al., 2004; Miller, 2001, 2004; Shields et al., 1996), performance on which has a significant relationship with linguistic competence (e.g. Milligan et al., 2007). Theory of mind, a constituent of social cognition, refers to the understanding of others' mental states, beliefs, knowledge, and desires. It typically develops prior to kindergarten entry as the child slowly decouples from egocentric thought (see Eyuboglu et al., 2018), with cognitive and language precursors including the conception of intentionality, recognition that perspective is not constant or universal, and the comprehension and use of mental state words (e.g. “believe”, “know”, “think”).

Researchers have identified an array of pragmatic deficits displayed by children with spoken language impairment. These deficits include trouble with (a) joining in others' social interactions (Brinton et al., 1997b; Craig & Washington, 1993; Liiva & Cleave, 2005), (b) participating in cooperative activities (Brinton et al., 1998a), and (c) conflict resolution (Brinton et al., 1998b; Horowitz et al., 2006; Timler, 2008). Additionally, their attempts to influence others are frequently rebuffed and they often exhibit inappropriate comments and questions and inflexible communicative patterns (Craig, 1993; Craig & Washington, 1993; Guralnick et al., 1996; Hadley & Rice, 1991; Vallance et al., 1999).

Among children with reading disability, scholars have observed notable deficits in pragmatics. For instance, an early study by Bryan et al. (1981) compared the conversational skills of 20 male children with reading disabilities and 20 male children without reading disabilities in grades 2 and 4 (approximately 7- to 8-years-old in grade 2 and 9- to 10-years-old in grade 4). Each participant was videotaped as he played the role of a talk-show host interviewing another child. Children with reading disability exhibited relative weaknesses with initiating and maintaining a conversation with their "guest" and were less assertive than their peers without reading problems. Toro et al. (1990) examined how well elementary school children with and without reading disability solved a series of presented problem situations involving social interactions. The children were asked to generate a satisfactory solution to the presented problem situation. Those with reading disability struggled substantially more than their peers without reading problems to identify suitable solutions to the social problems. Similarly, Carlson (1987, cited in Toro et al., 1990) compared the social problem-solving strategies of 30 secondary students with a reading disability and 30 students without a reading disability. The students with reading problems performed more poorly when trying to identify a social problem, generate alternative solutions to that problem, and identify consequences of each proposed solution to the problem.

More recent work has established that children with specific learning disorder display poorer performance than their unaffected peers on theory of mind tasks that are thought to be related to pragmatic abilities (e.g. Caillies & Sourn-Bissaoui, 2008; Cardillo et al., 2018; Carruthers & Smith, 1996; Eyuboglu et al., 2018; Martin & McDonald, 2003; McTear & Conti-Ramsden, 1991; Norbury, 2005). As noted previously, the possibility of co-occurring spoken language impairment was not adequately ruled out in these studies, so we do not know if these were children and adolescents who exhibited dyslexia, reading comprehension disorder, or a combination of the two.

A few studies have investigated the pragmatic language abilities of students with purported dyslexia, though these studies did not clearly identify the participants as having a core phonological deficit resulting in word recognition and spelling difficulties in the absence of broader spoken language impairment (i.e. true dyslexia), so their results must be interpreted cautiously. For instance, Kasirer and Mashal (2017) noted that the children and adolescents with purported dyslexia in their study exhibited weaker comprehension of conventional metaphors (e.g. *a sharp tongue*) using a multiple-choice task than typical peers, though they performed similarly when interpreting novel metaphors (e.g. *a pure hand*) or creating metaphoric expressions using a stem-completion task.

Lam and Ho (2014) used a caregiver checklist of pragmatic skills to compare the performance of a small group ($n = 22$) of 8- to 12-year-old Chinese students with purported dyslexia to a control peer group ($n = 24$). The students with dyslexia had significantly lower scores on items measuring communicative initiation and use of social context for communication and a composite of pragmatic language skills (with additional items measuring the areas of stereotypic language and nonverbal communication). Their findings comport with survey data reported by Riddick et al. (1997), in which students with purported dyslexia exhibited incoherent and disorganized utterances and inappropriate topic initiation skills, which ultimately led to their communicative partners failing to make sense of their communicative attempts.

Cardillo et al. (2018) investigated the comparative performance on pragmatic language and related theory of mind tasks of a small group of 8- to 10-year-old Italian children with and without dyslexia ($n = 21$ in each group). To assess pragmatics, participants were asked to explain a metaphor or to select an illustration from an array that represented the non-literal meaning of a target metaphor. They were also asked to respond to questions about a story that required inferencing and to answer questions about emotions and behaviours of individuals in social situations described using figurative expressions. To assess theory of mind, verbal and pictorial social perception tasks were used in which the children had to respond to questions about the described individual's point of view or identify with pictures the emotion associated with a social situation shown in a photograph. The students with purported dyslexia performed more poorly on the metaphors and story inferencing tasks, but not the social situations task. While the students with dyslexia performed more poorly than their typical peers on the point of view task, they did not do so on the emotion identification task. However, most of these initial significant differences between the groups were rendered nonsignificant when vocabulary knowledge (specifically synonym recognition) was controlled, suggesting that many of the pragmatic and theory of mind weaknesses observed in children with dyslexia may be explained by vocabulary differences.

Research studies examining the pragmatic skills of children with writing disability are virtually non-existent. There is preliminary evidence that measures related with pragmatic language skills, such as first- and second-order false belief measures of theory of mind, have an indirect influence (through oral discourse abilities) on writing quality in at least young, mostly typically-developing writers (Kim & Schatschneider, 2017). Moreover, writing tasks that place a premium on perspective-taking, such as persuasion-oriented school writing assignments, tend to be challenging for younger students and those with specific learning disorder given the lack of contextual cues and feedback from a present communicative partner to gauge one's persuasiveness (Erftmier & Dyson, 1986; Frank, 1992; McCann, 1989; Rubin & Piche, 1979). Specifically, children with reading and writing disabilities typically have trouble producing persuasive papers that display clarity, logic, convincing arguments, and effective reconciliation of opposing viewpoints (e.g. Ferretti et al., 2000; Wong et al., 1996). Unfortunately, we do not know if their difficulties with persuasion are related clearly to pragmatics (because pragmatic abilities were not directly evaluated) or are perhaps associated with broader limitations in background knowledge (McCutchen, 1986) or cognitive development (e.g. Gallagher & Noppe, 1976). Additionally, these studies do not isolate writing disability from other conditions or differentiate types of writing disorders.

24.4 The Effect of Pragmatic Difficulties on Reading and Writing Performance

It is likely that pragmatic difficulties have a pronounced impact on writing performance. This is especially true when perspective taking is at a premium, such as when crafting a written text for a specific audience unfamiliar to the writer (Langer, 1986; Rubin & Piche, 1979). Perspective taking relies on one particular aspect of pragmatic functioning—presupposition—which is directly related to social cognition and theory of mind. After all, how much a student presupposes about a reader's prior knowledge in a written text is predicated on his or her inferences about what the reader believes, knows, and wants.

Children with spoken and/or written language impairments tend to presuppose too much shared knowledge between themselves and their readers. This renders communication ineffective because their reader must reconstruct a situation model for the text without sufficient detail (see Kintsch (1988) and van Dijk and Kintsch (1983) for discussion of mental representations of discourse structures). A situation model is an integrated understanding of the meaning of a text formed from its constituent propositions that are surface coded via linguistic units and other knowledge residing outside the text. Sometimes, children with such impairments presuppose too little shared knowledge, rendering a text cumbersome to read and laden with irrelevant or tangential information (Troia, 2011).

Perspective taking is relevant for all aspects of the writing process. For instance, difficulty taking a reader's perspective obscures the need to revise and edit. Thus, when faced with the task of polishing one's own paper to meet the needs of a particular audience, weaker writers tend to have difficulty detecting problems and correcting their errors or missteps (Bartlett, 1982). Likewise, insensitivity to the reader's perspective makes planning and drafting text more difficult, because without a clear conception of plausible audience experiences, knowledge, motivations, and values, generating and organizing ideas for a paper will be misguided.

For reading too, perspective taking is important, as the child with a written language disorder must infer the author's communicative intentions behind what is presented, how it is presented, and why so—the author's stance. If the child reading the text presupposes shared knowledge inappropriately, she may regard at least some information presented in the text being read as superfluous or redundant with what she assumes she already knows about the topic. Major points of the text the author felt were important to share in a particular manner are likely to be disregarded or skimmed with little contemplation. The child also might fail to activate relevant and sufficient prior knowledge to assist with comprehending the text because the purpose for reading may be unclear (e.g. Paris et al., 1983). In either case, reading comprehension, especially inferences about hidden relationships between ideas in text, will suffer (e.g. Leu et al., 1986). More fundamental linguistic errors due to poor presupposition skills may involve incorrect interpretation (and use) of referential deictic terms (e.g. Bishop, 1997) in texts that mark noun relationships (e.g. *I* vs. *you*, *a* vs. *the*), spatial relationships (e.g. *here* vs. *there*, *this* vs. *that*), and temporal relationships (e.g. *before* vs. *after*, *now* vs. *then*).

Children and adolescents with spoken and/or written language disorders frequently exhibit problems with discourse regulation, another aspect of pragmatics that is a prominent contributor to successful reading and writing. Effective discourse regulation is possible when the reader/writer possesses a strong understanding of the structure of the discourse form used in a given social context for specific social purposes. This includes deep knowledge of genres, the socially-situated flexible language configurations used to meaningfully communicate ideas (e.g. Chapman, 1999; Roth, 1986; Roth et al., 1995; Roth & Spekman, 1984), as well as a firm grasp of the topic being addressed (e.g. McCutchen, 1986; Westby, 2002).

Research has shown that students with language disorders are relatively less competent in discourse regulation when writing, particularly (a) topic organization and maintenance (e.g. Botting, 2002; Norbury & Bishop, 2003), (b) successful use of strategies to avoid misunderstanding by the reader such as paraphrasing to simplify information, repeating important ideas for emphasis, and elaborating on novel or controversial points (e.g. Adams & Bishop, 1989; Brinton & Fujiki, 1982), (c) grammatical cohesion via appropriate use of anaphoric and cataphoric reference, substitution, ellipsis, and clausal structures characteristic of the genre (e.g. Lapadat, 1991), and (d) lexical cohesion created through use of synonyms, antonyms, hyponyms, repetition, and collocation. Consequently, and because the reader's response to a written text often is not available during the production of that text, students with language disorders are likely to produce short pieces of writing bereft of detail and lacking strong organization that fail to attend to genre conventions and the reader's needs (e.g. Englert & Raphael, 1988; Graham et al., 1993; Graham & Harris, 1989, 1997; Nodine et al., 1985).

While reading, the child with a language disorder who possesses weak discourse regulation skills can be expected to exhibit poor comprehension because of limited genre and topic knowledge (beyond whatever problems the student has with word reading). Research clearly shows that reading comprehension suffers when schema activation—the mental representation of textual propositions, images, and their ordering (see Anderson, 1983)—is inadequate due to limited genre and/or topic knowledge (e.g. Afflerbach, 1986; Anderson & Pearson, 1984; Best et al., 2008; Langer, 1986; Rupley & Wilson, 1996). Readers with low background knowledge also rely more heavily on textual cohesion devices such as those mentioned above to aid their comprehension because they lack the information needed to make inferences otherwise; that is, in texts with few cohesive devices, poor readers often cannot fall back on their knowledge to fill in gaps (McNamara et al., 1996, 2017). Reading comprehension strategy use, such as periodically summarizing the gist of a portion of text, rereading or momentarily skipping especially difficult sections or words, and identifying the main theme of a piece, why it is important, and how it relates to other texts and one's knowledge and experiences, is aligned with discourse regulation. As would be anticipated, students with language disorder tend to be less aware of such strategies and use them less frequently (e.g. Cain & Oakhill, 2007; Gersten et al., 2001; Paris et al., 1983; Schumaker & Deshler, 1992).

A final aspect of pragmatics with which children with language disorders might struggle is figurative language. Figurative language comprehension and use are important for reading and writing success because nearly two-thirds of English is non-literal (Arnold & Hornett, 1990). In addition, approximately a third of teachers' utterances contain words with multiple meanings (the foundation of figurative expressions) or idiomatic expressions, and about 7% of reading materials used in elementary schools contain idioms (Lazar et al., 1989). Children are extensively immersed in figurative language in school. Figurative language is critical to academic success via comprehension of instructional and conversational language expressed orally or in writing and effective use of non-literal language when expressing oneself in social or academic endeavors (Low, 1988; Palmer & Brooks, 2004; Qualls & Harris, 1999). Research findings indicate children and adolescents with language disorders demonstrate significant problems interpreting and using conventional figurative expressions (e.g. idioms, proverbs, metaphors, similes, humour, hyperbole, indirect requests) and slang (Abrahamsen & Sprouse, 1995; Lee & Kamhi, 1990; Nippold, 2007; Rice, 1993; Rice et al., 1991), as well as literate figurative expressions such as personification, allusion, and symbolism (e.g. Nippold, 2007).

Students with language disorders who struggle with figurative language would be expected to comprehend less well while reading most texts in which such non-literal expressions are embedded (e.g. Ortony et al., 1978). Conversely, poor readers are exposed to fewer sophisticated vocabulary words that serve as building blocks of figurative expressions, so there is likely a reciprocal relationship between reading and figurative language competencies (e.g. Cain et al., 2005; Levorato et al., 2004). For example, a study of Polish students in grades two and three (ages 8 to 9 years) found that poorer readers recognized and interpreted metaphors less well than better readers, often using a literal frame of reference (Wiejak, 2014). Both text comprehension and figurative language understanding involve metalinguistic skills (treating language as an entity for conscious examination; see Pinto et al., 2011), which helps explain why the relationship between reading and figurative language comprehension may be bidirectional.

When writing, figurative language helps authors craft texts that creatively illustrate complex relationships between ideas, people, and things in novel or authentic ways. Use of figurative expressions in writing permits students to participate fully in their social worlds via credible means and is essential to some genres, such as poetry. Children and adolescents with language disorders who cannot incorporate non-literal language into their writing will not be able to display the full range of communicative functions of writing. Likewise, when these students are expected to fulfill multiple communicative functions with a single composition (e.g. informative, heuristic or learning, and imaginative functions for a report on novel approaches to combating climate change), they may fall short because research has demonstrated that they use the full array of language functions less frequently than their typical peers (e.g. Lapadat, 1991; Spekman, 1984).

24.5 Illustration of Pragmatic Difficulties in Written Expression

To illustrate some of the pragmatic difficulties that may be experienced by children with written language challenges, two contrastive examples—one using informational papers and the other persuasive papers—are presented below. These papers, each written by a different child during a timed 15-minute period on a desktop or laptop computer via keyboard, were produced by students in grade four or five from the general education population. The papers in which pragmatic difficulties are evident were written by students who scored low average or below average in reading and writing on associated norm-referenced tests, while those papers in which pragmatic abilities are relatively strong were written by students who scored high average or above average on the same norm-referenced tests. However, only one student with poor pragmatics in their writing was identified with a written language disorder.

The students wrote in response to a prompt and, for the informational papers, used a source text for reference. For reader ease, the typewritten papers below have been corrected for errors in writing mechanics including spelling, capitalization, and punctuation. It should be noted that, because students were allotted only 15 minutes to write, it is evident some did not finish. For the informational papers, one student responded to a prompt using a source text that described the tale of Calvin Graham, a 13-year-old World War II veteran who joined the Navy and helped save fellow enlisted personnel. But he was injured while doing so and won a Bronze Star and Purple Heart for his efforts, though it was later found out he lied about his age and consequently he was stripped of his medals and discharged from the Navy. The other student responded to a prompt using a source text that described how China, facing severe automobile traffic congestion in major cities, has begun experimenting with roadway elevated high-capacity transit buses to ease congestion. For the persuasive papers, one student responded to the question, “Should cell phones be allowed in classrooms?”. The other student responded to the question, “Should families be able to pick who their children’s friends are?”.

	Evidence of relative pragmatic strength	Evidence of relative pragmatic weakness
Informational papers	Calvin Graham was in sixth grade and lived with a very rude stepfather. He heard that his cousins joined the Navy team, and that they recently kicked the bucket. Now he wanted to join the Navy team more than anything. One day he lined up with some of his buddies, but there was a dentist to check and see if the person is old enough to join the Navy. The dentist said he was not qualified and that he needed to go back home. But he fought back. Calvin Graham said he wouldn't go anywhere without joining the Navy. Then, finally, the dentist gave up and let him go. He finally was a fighter! Before the South Dakota made it to the Pacific with Graham in the backseat, the battleship had become part of a task force. One day, a bomb hit the South Dakota and 50 men or so got hurt. The South Dakota was fixed in Pearl Harbour. Then, on November 6, Graham turned 13! Then the Navy got to know that Graham was not old enough to be a Navy officer, and they sent him back with a very bad attitude. The South Dakota had disappeared into smoke. Graham had joined school but then got kicked out. In 1994, Graham…	One word: no, nope, wrong on so many levels. To me, it's most likely deadly. People and cars are going to drop like flies. I am not going to China anymore, so don't text me. Don't call me and try to persuade me. It is not happening. Sorry, just no. I am not going to risk my life to ride on this weird bus thing. I just think it will collapse on a car and kill somebody, and I could break my neck. That is just not right.

	Evidence of relative pragmatic strength	Evidence of relative pragmatic weakness
Persuasive papers	I strongly believe that cell phones should not be allowed in classrooms. Here are three reasons why I strongly believe this. One reason why I believe this is that phones can be a distraction for some students. For example, kids may be tempted to play or text on them, which is distracting them. This also causes disruptions in the class. Loud noises and sounds can cause disruption in the class. One last reason why I strongly believe this is that all these distractions can lead to not getting the education you need. Education is very important for everyone, and everyone has to have a good education if they want to be successful in life. You may believe that phones should be allowed in class. You might think this because phones would come in handy just in case there was an emergency, and yes this is true, but there are phones in all classrooms for teachers to use, and phones should always be kept in lockers or backpacks. This way they will not get lost or broken. You may also think that phones would come in handy for school work, but I disagree. There are many resources in a classroom from books to multiplication charts and calculators; having phones in classrooms might also lead to students cheating on tests. Cheating on tests is really bad because you don't get the education you need, and your teacher won't know what you might need to work on, so your teachers won't know to help you. These are all the reasons why I strongly believe that cell phones should not be allowed in classrooms, and that phones are better to be used outside of school	No, they should not because I think kids should be able to pick their own friend because if the family picks the friend for them, then they might not like that friend. So I think they should be able to pick their own friend, and if that friend…

One of the most noticeable differences between the papers written by relatively strong versus weak writers is text length. In the same period of time, stronger writers wrote about double or triple the number of words of their weaker counterparts. Of course, the weaker writers may have written much less because these students often struggle with the mechanics of writing, which can overwhelm the child and divert precious cognitive and motivational resources from generating ideas and translating them into appropriate language to the necessities of typing, spelling, etc. Nevertheless, more specific pragmatic difficulties are evident in the papers written by the weaker writers.

The child who wrote the informational paper about transit elevated buses in China clearly has an undeveloped sense of informative genre structure. He wrote more of an opinion piece rather than shared what was learned about the topic with an unfamiliar reader, even though he was explicitly directed to do just that. It is also obvious that this student presupposes too much shared knowledge, as there is not a clear topic introduction. The reader is left to infer that the "weird bus thing" is elevated above traffic (and the student appears to confuse the fact that the bus is elevated, not cars). There is no mention of why the bus exists and that it is, in fact, used in China. The paper also exhibits an informality that is at odds with the desired purpose. Cohesion is weak in this paper. The student does not employ lexical cohesive devices such as synonyms adequately, resulting in unnecessary repetitiveness, or grammatical cohesion in the form of clausal structures commonly observed in informational text to link ideas together (and the text even appears rather jumbled, like stream of consciousness).

The student who wrote about Calvin Graham, on the other hand, understands that the reader likely does not know anything about the subject and attempts to provide enough information to give the reader a sense of Calvin's life story (though there are a couple minor inaccuracies regarding the sequential ordering of events and the student did not complete relaying the account). The writer of this paper even uses figurative language ("kicked the bucket") not found in the source text. Though the stated purpose of the paper is left implicit for the reader (e.g. "Let me tell you what I learned about an amazing young man, Calvin Graham, and his adventures in the Navy"), this is not unusual for students in late elementary school when writing informative texts.

The student who responded to the question regarding families' input on one's choice of friends states their stance on the topic but the reader must infer the topic itself by reading the first sentence carefully. Also, poor control over referential cohesion is evident by the confusing use of "they" in the paper. The writer of this piece does give a single reason to support the stance taken ("they might not like that friend"), displaying some awareness of the required text structure, but that is all. There is not even any elaboration of that single reason (presumably the student did not have time to complete the paper). In contrast, the student who wrote about cell phones in classrooms introduces the topic effectively and clearly states an opinion. This writer provides multiple reasons for the stated opinion, elaborates on those reasons, and even logically rebuts potential counterarguments, which suggests strong presupposition skills. And for the most part, this student uses cohesive devices appropriately and effectively (e.g. conditional and adversative adverbials). These papers written by strong and weak writers make it apparent that students with weaker written language skills often struggle with multiple aspects of pragmatics, including presupposition and discourse regulation, regardless of text genre.

24.6 Summary

School-age individuals who struggle with literacy are at heightened risk for difficulties with pragmatics (because written language disorder is associated often with oral language impairments), and those children and young people with deficits in pragmatic language abilities are likely to experience trouble engaging in many reading and writing tasks. Both reading and writing require a keen sense of the author's (when involved in a reading activity) or reader's (when planning, drafting, revising, or editing a text) thoughts, intentions, and views to communicate successfully through written language. This is made that much more challenging by the mere fact that there is usually not another interlocutor when performing literacy tasks to help the student gauge how well they are interpreting the text or meeting the needs of their audience.

Pragmatic skills including (a) presupposition, (b) the regulation of discourse through effective organization, strategy use, and cohesion, all of which depend to some extent on genre and topic knowledge, and (c) figurative language comprehension and use each appear to be highly relevant to written language. Students with deficits in these particular skill sets also often display weak literacy achievement. However, it is unclear at this point which lies first in the causal chain—poor pragmatic abilities or underdeveloped literacy competencies—or even if they are truly causally related, though there is adequate theoretical grounding for assuming so. Future research efforts should attempt to address this issue head on by using longitudinal observational investigations and experimental (single case or group) treatment studies.

Another issue that requires further research to be resolved is whether students with written language disorders at the lexical and sub-lexical levels of language (i.e. dyslexia and dysgraphia) actually demonstrate pragmatic difficulties. There are only a handful of studies that have examined pragmatic language skills in children with dyslexia (none could be located with samples of children diagnosed with dysgraphia). All these studies are plagued by poorly defined samples, making it unclear if the students involved were correctly identified as a distinct group from other children with other kinds of reading and/or spoken language problems.

Based on the available work, it appears that students with purported dyslexia do indeed display weak pragmatic skills, at least in discourse regulation and non-literal language. However, it is noteworthy that in one of these studies (Cardillo et al., 2018), most of the statistically significant differences in pragmatic abilities between children with dyslexia and their unaffected peers were rendered non-significant when vocabulary knowledge was controlled. Thus, it is conceivable that some pragmatic difficulties observed in these students (and perhaps even children and young people with other kinds of written language disorders) may be due to limited vocabulary, and this possibility deserves further attention. Lastly, no studies have directly examined the pragmatic abilities of children with writing disabilities of any kind, though Kim and Schatschneider (2017) did find theory of mind task performance was related to oral language skills, which were in turn related to writing performance. So, there is reason to believe that students with writing disabilities may face difficulties with pragmatic language skills.

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Chapter 25
Children, Young People and Adults Who Use AAC

Jill E. Senner

25.1 Introduction

The American Speech-Language-Hearing Association (2019) states that "augmentative and alternative communication (AAC) is an area of clinical practice that addresses the needs of individuals with significant and complex communication disorders characterized by impairments in speech-language production and/or comprehension, including spoken and written modes of communication." AAC can be *augmentative* when it enhances or supplements residual natural speech or *alternative* when it replaces natural speech. AAC approaches often aid in language production (i.e. expressive communication) as well as in comprehension (i.e. understanding language).

Beukelman and Mirenda (2013) remark that "there is no typical person who relies on AAC. They come from all age groups, socioeconomic groups, and ethnic and racial backgrounds. Their only unifying characteristic is the fact that they require adaptive assistance" for communicating (p. 4). According to the National Joint Committee for the Communication Needs of Persons with Severe Disabilities (2019), recent estimates suggest that there are over 2 million individuals using AAC in the United States. A variety of developmental and acquired conditions can result in severe communication impairments in both adults and children. Many of these disorders include, but are not limited to, those covered in this volume such as autism spectrum disorder, Down syndrome, Fragile X syndrome, Parkinson's disease, amyotrophic lateral sclerosis, and traumatic brain injury.

The need for AAC services may be temporary, such as in cases of young children who may improve speech and language skills with development or in individuals recovering from an accident, an illness, a stroke, or who are intubated and unable to talk. Most individuals who use AAC, however, do so throughout their lifetimes.

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This includes individuals with chronic communication impairments, resulting from developmental or acquired disorders, as well as those with degenerative disorders that preclude their use of natural speech. AAC systems and strategies can vary widely and depend on the needs of the individual over time. A person's AAC system often involves the use of the body, such as gestures and sign language (i.e. unaided AAC), as well as using aids external to the body (i.e. aided AAC) which include simple picture boards and books as well as sophisticated speech-generating devices (SGDs) and mainstream technologies. Individuals using AAC often employ multiple modes of communication such as vocalizations, facial expression, and body language as well as a range of aided AAC systems to convey messages (Blackstone & Berg, 2012). People who use AAC systems may choose to use different communication modes and methods in different settings and with different communication partners (Warrick, 1988).

The ability to use AAC approaches effectively requires a growing understanding of pragmatics. The following sections will summarize the importance of pragmatic skills, variables affecting pragmatic skills, and characteristics of pragmatic skills in individuals who use AAC. They will also introduce assessment strategies and discuss appropriate interventions.

25.2 The Importance of Pragmatic Skills in Individuals Using AAC

To achieve communicative competence (i.e. communicate effectively), individuals using AAC must have knowledge and skills in (1) operating a communication system; (2) using the language of a system; (3) understanding ways to compensate for the limitations of a system; and (4) using the social rules of communication, also known as *social competence* (Light, 1989). Light (1989) suggests that social competence includes both sociolinguistic and sociorelational aspects of interaction. Included in sociolinguistic skills is an understanding of discourse strategies such as initiating, maintaining, and terminating interactions, turn taking, and the cohesion and coherence of conversation. Sociolinguistic skills also include interaction functions (e.g. expression of wants and needs, social closeness, information transfer) and specific communicative functions such as requests for information, protest, and self-expression. Each of these skills is context dependent, that is, depends on partner, setting, and task demands, and is evaluated in terms of how appropriate and effective they are (Light, 1989). Effective interpersonal communication skills, otherwise known as sociorelational skills (Light et al., 2007), include having a positive self-image, showing an interest in others and a desire to communicate, being an active participant in conversations, being responsive to communication partners, and putting others at ease (Light, 1989).

It has been suggested that regularly interacting and developing friendships with peers is necessary for learning social competence (Lilienfeld & Alant, 2005a).

Cooper et al. (2009) remark that “friendships are the building blocks of strong social networks” (p. 154). Young adults interviewed attributed some difficulties with loneliness and friendship formation to poor communication. Young children learn to perceive themselves through their interactions with communication partners. However, children with disabilities may experience rejection and isolation. The impact of negative social relationships experienced by many children with disabilities may be exaggerated for children with complex communication needs due to their communicative disadvantages in interacting with peers (Clarke & Kirton, 2003). Individuals who require AAC often have particularly limited social networks (Lilienfeld & Alant, 2005a), often consisting only of close family members and paid professionals. Social networks, including family and friends, have been found to be important in decreasing feelings of loneliness in young adults with cerebral palsy using AAC (Cooper et al., 2009) and in aiding adults who use AAC to find and maintain employment (Bryen, 2006).

25.3 Factors Affecting Pragmatic Skills in Individuals Using AAC

There are several variables that may interact with one another to affect social competence in children and adults using AAC (Calculator, 1999). These variables are summarized in Table 25.1 and explained further here.

(1) *Characteristics of the individual using AAC*. Individuals using AAC may differ in their language skills, cognitive abilities, motor skills, motivation to communicate, personality, and social experiences. Each of these characteristics can influence pragmatic skills. This point will be expanded in Sect. 25.4.

(2) *AAC system characteristics*. Each AAC system has its own unique features such as language organization, access methods, and type of output which can affect social skills. For example, poor quality voice output on a speech-generating device may be a barrier to successful use of the device (Crisp et al., 2014) and result in communication breakdowns (Clarke & Wilkinson, 2007). Language organization and access methods can affect rate of message production and pre-utterance pause length which may affect partner perceptions of competence. Also, an idiosyncratic gesture will not be useful unless communication partners understand what it means.

(3) *Communication partner characteristics*. Communication partners may have differing expectations of and attitudes towards the individual using AAC, thus affecting social interactions. For example, communication partners with previous experience with individuals with disabilities may have more positive attitudes about individuals using AAC (McCarthy & Light, 2005) and confidence in initiating interactions with people who use AAC (Ostvik et al., 2018). This point will be expanded in Sect. 25.5.

Table 25.1 Variables affecting interaction

1. Characteristics of individuals using AAC
    - a. Personality
    - b. Language abilities
        - i. Message Length (Hoag et al., 1994)
    - c. Cognitive abilities
    - d. Motor skills
        - i. Rate of Message Production (Farrier et al., 1985)
    - e. Social skills
    - f. Motivation
    - g. Socialization (Lilienfeld & Alant, 2005b)
2. AAC system characteristics
    - a. Access method
        - i. Rate of Message Production (Farrier et al., 1985)
    - b. Language organization
        - i. Message Length (Hoag et al., 1994)
        - ii. Rate of Message Production (Farrier et al., 1985)
        - iii. Pauses Preceding Utterances (Todman & Rzepecka, 2003)
    - c. Quality of Speech Output (Clarke & Wilkinson, 2007; Crisp et al., 2014)
3. Communication partner characteristics
    - a. Attitudes & Expectations (McCarthy & Light, 2005; Ostvik et al., 2018)
    - b. Confidence (Ostvik et al., 2018)
4. AAC instruction
    - a. Direct Instruction (Glennen & Calculator, 1985)
    - b. Communication Partner Instruction (Kent-Walsh & McNaughton, 2005; Light et al., 2002)
5. Other variables
    - a. Communicative Opportunities (Andzik et al., 2016)
    - b. Communicative Purposes (Light, 1988; Light, 1996)

(4) *AAC instruction*. Direct instruction in the use of a device can affect the communicative functions used by the individual using AAC. However, communication partner knowledge and skills regarding AAC can also heavily influence interactions. For example, caregivers of pre-symbolic children with severe disabilities may misinterpret atypical attempts to initiate social closeness and benefit from instruction to recognize subtle cues (Light et al., 2002). This point will be expanded in Sect. 25.7.

(5) *Other variables*. Other variables such as the frequency with which one has opportunities to communicate and the purpose of communication can also affect interactions. Light (1988) suggests that there are four purposes of communicative interactions including communicating wants and needs, information transfer, social closeness, and social etiquette. The goal of social closeness communications is to establish, maintain, and develop relationships with others. In social closeness interactions, the focus is simply on being together. Thus, unaided forms of communication may predominate, whereas communication to express wants and needs requires different types of interactions (Light, 1997).

Given the complex nature of interactions between these variables, it is difficult to draw generalizations about characteristics of pragmatic skills in individuals using AAC. However, the following sections summarize the available evidence.

25.4 Characteristics of Pragmatic Skills in Individuals Using AAC

Despite the importance of social competence in effective communication, relatively little is known about pragmatic skills in individuals who use AAC. In children with severe speech and physical impairments without any accompanying cognitive deficits, general patterns have been observed when communicating with caregivers. These include (1) taking fewer conversational turns; (2) responding more frequently than initiating communication; and (3) exhibiting a restricted range of communicative functions (Light et al., 1985). Partner-dominated interactions are also seen between school-aged children in academic environments (Andzik et al., 2016; Clarke & Wilkinson, 2007), with children using AAC serving as respondents in as high as 91% of communicative opportunities. These asymmetries in initiation have been reported in instructor interactions (Andzik et al., 2016), in experimentally arranged peer interactions (Clarke & Wilkinson, 2007), with speaking peers with disabilities at school (Clarke & Kirton, 2003), and in naturally occurring interactions with peers in inclusive classrooms (Chung et al., 2012). Furthermore, during naturally occurring events at school, students tend to interact primarily with adults rather than peers in greater than 90% of communicative opportunities (Andzik et al., 2016; Chung et al., 2012). Even in inclusive classrooms where peers were in frequent proximity, students using AAC interacted primarily with an assigned staff member, most frequently instructional assistants or special educators (Chung et al., 2012; Ostvik et al., 2018).

Senner (2011) interviewed 21 parents of teens and young adults with developmental disabilities who use AAC. These parents indicated deficits in multiple skill areas in their children on the Pragmatics Profile from the *Clinical Evaluation of Language Fundamentals* (Wiig et al., 2013). Many parents indicated their children exhibited difficulty using specific pragmatic functions such as introducing appropriate topics of conversation and maintaining conversations. However, considerable individual differences were found. Many parents identified the need for direct social skills instruction for their children. However, only a third of parents reported their children having participated in pragmatic intervention. Parent comments from this study also suggested that educating communication partners may also be beneficial in supporting pragmatic skills in teens and young adults using AAC. Finally, parents highlighted that AAC systems must provide adequate vocabulary for social participation (Senner, 2011).

The employment rate of adults using AAC has been estimated at around 15%, about half the rate of employment for persons with disabilities without complex communication needs (Bryen et al., 2007). In a survey of employers, 91% indicated that an understandable, standard voice was a job requirement, and 61% indicated that intermediate or advanced communication skills were necessary (Bryen et al., 2007). The vast majority of employers required in-person or telephone interviews. Access to and use of generic communication technologies allowing remote communication such as email or phone have been listed as a means to expand social networks in individuals using AAC (Bryen, 2006) and are also required for many occupations (Bryen et al., 2007). However, access to these technologies may be challenging for some individuals using AAC and thus limit opportunities for friendships and employment (Bryen, 2006; Bryen et al., 2007; Cooper et al., 2009). Overall, adult focus group participants who use AAC have rated making and keeping new friends, dating, and finding jobs as research priorities (O'Keefe et al., 2007). Beginning and maintaining friendships, dating, and seeking and maintaining employment all require social competence.

The factors that affect use of AAC by adults are numerous and complex. Conversational control, "the manner and extent to which an individual directs and restrains communicative interaction" (Farrier et al., 1985, p. 65), was found to be markedly lower in an experiment in which neurotypical subjects used an AAC system as compared to their communication using speech. The slower rate of message production seen in individuals using AAC is one factor thought to be responsible for reduced conversational control (Farrier et al., 1985). Reduced message length also affects perceptions of participation, management of partner attention, and degree of social ease (Hoag et al., 1994) in neurotypical subjects using AAC to communicate. Finally, equality in conversational turns can affect perceptions of communicative competence. In one study, greater equality was accomplished by teaching individuals using AAC to fulfill non-obligatory in addition to obligatory turns (Light & Binger, 1998). Obligatory turns are those that typically require an answer (e.g. those that follow a partner's question such as "How are you?"). However, non-obligatory turns are those that follow a partner's comment or statement (e.g. commenting "too bad" after a partner indicates failing a test). Increased use of non-obligatory turns by an efficient communicator resulted in observers' judgments of increased communicative competency. However, increasing the frequency of non-obligatory turns was not effective in improving judgments about slower communicators' competency.

Communication breakdowns are also frequently documented in individuals using AAC. Adults with developmental disabilities and intellectual impairment have been observed to attempt to respond to partner's requests for clarification by simply repeating the message instead of revising the message to make it more easily understood (Calculator & Delaney, 1986). Effective repair of communication breakdowns typically involves a hierarchical approach, moving from less to more complex resolution strategies. For example, the first strategy an individual might use could involve repeating the message. However, if that fails, the individual should include additional information by adding gestures, cuing the communication partner to the topic (Dowden, 1997; Hustad et al., 2002), or adding more words to the message.

25.5 Communication Partner Skills and Attitudes

The development of communicative competence "is inseparable from socialization and partner interaction" (Lilienfeld & Alant, 2005b). Success of a communication interaction between a person using AAC and a communication partner depends heavily on the skills of the communication partner (Kent-Walsh & McNaughton, 2005). Blackstone (2006) remarks that "being an effective communication partner or AAC facilitator is not intuitive. It often requires one to change long-established, unconscious ways of communicating" (p. 12). Interaction patterns of communication observed in parents of children using AAC often include controlling the topic, dominating conversational turns, and being more directive by requiring specific responses (Light et al., 1985; Pennington & McConachie, 1999). These behaviors may negatively affect communication development in children with complex communication needs (Yoder & Warren, 1998).

Communication partners range from more to less familiar and can include (a) life partners (such as a spouse for an adult or parents and siblings for young children); (b) good friends; (c) acquaintances (e.g. neighbors, classmates, co-workers); (d) paid professionals (e.g. personal care attendants, therapists, teachers); and (e) unfamiliar partners such as servers in a restaurant, store clerks, and other people with whom an individual using AAC might interact within the community (Blackstone & Berg, 2012). Over time and across a person's lifespan, communication partners may change circles (e.g. a relationship with a friend may evolve into a marriage). As a person's communication partners evolve, so will their communication needs. Often, an individual who uses AAC may have a restricted range of communication partners, consisting primarily of close family members and paid professionals (Lilienfeld & Alant, 2005a).

Communication partner attitudes can also affect social interactions with individuals using AAC. In school-aged children, positive peer attitudes have been viewed as facilitators of relationships with students who use AAC (Ostvik et al., 2018). Gender has been found to correlate with attitudes, with girls demonstrating more positive attitudes towards peers using AAC than boys. In addition, children exposed to individuals using AAC (e.g. those who attended integrated schools) had more positive attitudes than those with little experience of children with disabilities (Beck & Dennis, 1996). In general, factors that affect attitudes towards individuals using AAC are consistent with those in the general disability literature. For example, McCarthy and Light (2005) found that males generally had more negative attitudes towards individuals using AAC than females and that those who had previous experience with individuals with disabilities generally had more positive attitudes about people who used AAC.

25.6 Assessment

Given the wide range of ages, diagnoses, and types of AAC used by individuals with complex communication needs, individuals who use AAC are a heterogenous group. Accordingly, each individual's strengths and areas of need must be assessed to determine appropriate intervention targets and strategies. Language sample analysis, writing down and analyzing what an individual who uses AAC says, can provide rich information about pragmatic language use in children and adults who use AAC (Van Tatenhove, 2014). Many devices have built-in keystroke recording to help with the process. However, videotaping is still highly recommended because built-in logging does not record an individual's use of unaided modes of communication or the communication partner's responses, which are critical to interpreting the function of each utterance. A number of commercially available language sample analysis tools are available to make interpretation faster and easier.

The Pragmatics Profile in the *Clinical Evaluation of Language Fundamentals—5th Edition* (CELF-5; Wiig et al., 2013) is a 50-item checklist that provides normed scores on verbal and nonverbal social communication skills. The profile is completed by an informant familiar with the student such as a teacher or parent and each item on the questionnaire is rated on a 4-point Likert-type scale. The number checked is related to the frequency of occurrence of each skill: *Never or Almost Never* (1)*, Sometimes* (2)*, Often* (3), and *Always or Almost Always* (4). Despite the fact that the Pragmatics Profile was not specifically designed for individuals who use AAC, as per the CELF-5 Examiner's Manual, an item analysis can be done to identify student strengths and needs. For example, an item receiving 4-point rating indicates appropriate development and use of the specified skill, an item receiving a 3-point rating might only require monitoring, and a 1 or 2-point rating could indicate that the skill requires direct or indirect intervention. The descriptive information obtained from using the ratings on the Pragmatics Profile has been used to evaluate social skills in teens and young adults who use AAC (Senner, 2011).

One instrument that was specifically developed for use with individuals with complex communication needs is *Social Networks* (Blackstone & Berg, 2012). This instrument is unique in that not only does it document current communication behaviors of an individual using AAC, but it also helps to identify family members and others who might benefit from communication partner training. Parents, peers, and caregivers can improve the quality and quantity of their interactions with adults and children who rely on AAC following communication partner instruction. Furthermore, this instrument captures the multi-modal nature of communication by cataloging an individual's modes of expression.

25.7 Intervention

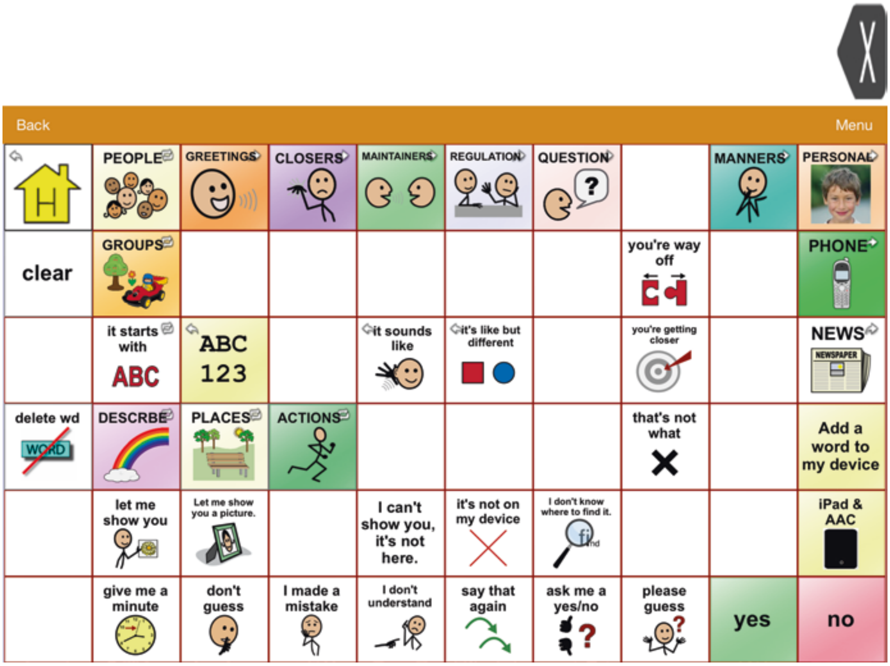
Historically, AAC intervention has emphasized communication for basic needs such as requesting. Even a more recent survey of speech-language pathologists (SLPs) revealed that 95% of respondents ranked communicating wants and needs as the most important communicative purpose for young children using AAC (Finke & Quinn, 2012). Although expressing wants and needs is important, "individuals must concurrently be taught skills that allow them to fulfill their need to be socially connected with others" (Chung et al., 2012, p. 363). Fortunately, there are a number of strategies that are effective in improving social skills in individuals who use AAC and in increasing communicative participation of AAC users (Thomas-Stonell et al., 2015).

25.7.1 Vocabulary

Light and Binger (1998) remark that "providing access to the right vocabulary is critical to ensuring the success of communication" (p. 13). Some AAC systems may not include adequate vocabulary to allow an individual to perform a specific pragmatic function such as maintaining a conversation. If this is the case, customization of messages may be appropriate. For example, inclusion of control or regulatory phrases on an AAC system (e.g. "Wait, I have something to tell you." "That's not what I meant.") and teaching the individual using AAC to use these phrases may be appropriate in helping an individual to gain conversational control (Buzolich & Lunger, 1995; Farrier et al., 1985) or repair a communication breakdown. These may be stored as whole sentences or phrases (rather than generated word by word or letter by letter) to improve efficiency due to the time-dependent nature of many social interactions. A sample communication display for repairing a communication breakdown can be found in Fig. 25.1.

25.7.2 Modeling

Partner-augmented input (PAI), also referred to as natural aided language, aided language modeling, or aided language stimulation, is a modeling strategy whereby communication partners use the child's AAC system themselves by pointing to the symbols on the child's speech-generating device while simultaneously talking. PAI can and should be provided in the natural environment to model when and how to use targeted pragmatic skills such as greeting or maintaining a conversation. Use of this strategy has been associated with gains in pragmatics, semantics, syntax, and morphology and is effective in individuals of varying ages, disabilities, and language skills (Biggs et al., 2018; O'Neill et al., 2018; Sennott et al., 2016).



Back Menu

	PEOPLE	GREETINGS	CLOSERS	MAINTAINERS	REGULATION	QUESTION		MANNERS	PERSONAL
clear	GROUPS						you're way off		PHONE
	it starts with ABC	ABC 123		it sounds like	it's like but different		you're getting closer		NEWS
delete wd	DESCRBE	PLACES	ACTIONS				that's not what		Add a word to my device
	let me show you	Let me show you a picture.		I can't show you, it's not here.	it's not on my device	I don't know where to find it.			iPad & AAC
	give me a minute	don't guess	I made a mistake	I don't understand	say that again	ask me a yes/no	please guess	yes	no

Fig. 25.1 This is the Repairs (i.e. Repair Strategy page) from the Baud and Senner (2013) Add-On Social Pages for use with Core Vocabularies. Note the use of regulatory phrases such as "That's not what I meant." These pages can be downloaded for free from talcaac.com

25.7.3 Direct Interventions

Direct interventions have been effective in teaching children and adults who use AAC to perform a variety of communicative functions including requesting (Glennen & Calculator, 1985), increasing turn-taking, and asking partner-focused questions (Light & Binger, 1998; Light et al., 1999). Training techniques have included: (1) Explanation of the goal and importance of the strategy being taught (using appropriate language); (2) Modeling appropriate use of the strategy by an instructor or individual using AAC; (3) Providing multiple opportunities for the individual to practice the target skill, in either role playing scenarios with an instructor or in functional contexts with coaching from an instructor; (4) The use of appropriate prompts such as an expectant pause, direct imitation or pointing cue; and (5) Performance feedback (Buzolich & Lunger, 1995; Calculator, 1999; Glennen & Calculator, 1985; Light & Binger, 1998; Light et al., 1999; Lilienfeld & Alant, 2005a).

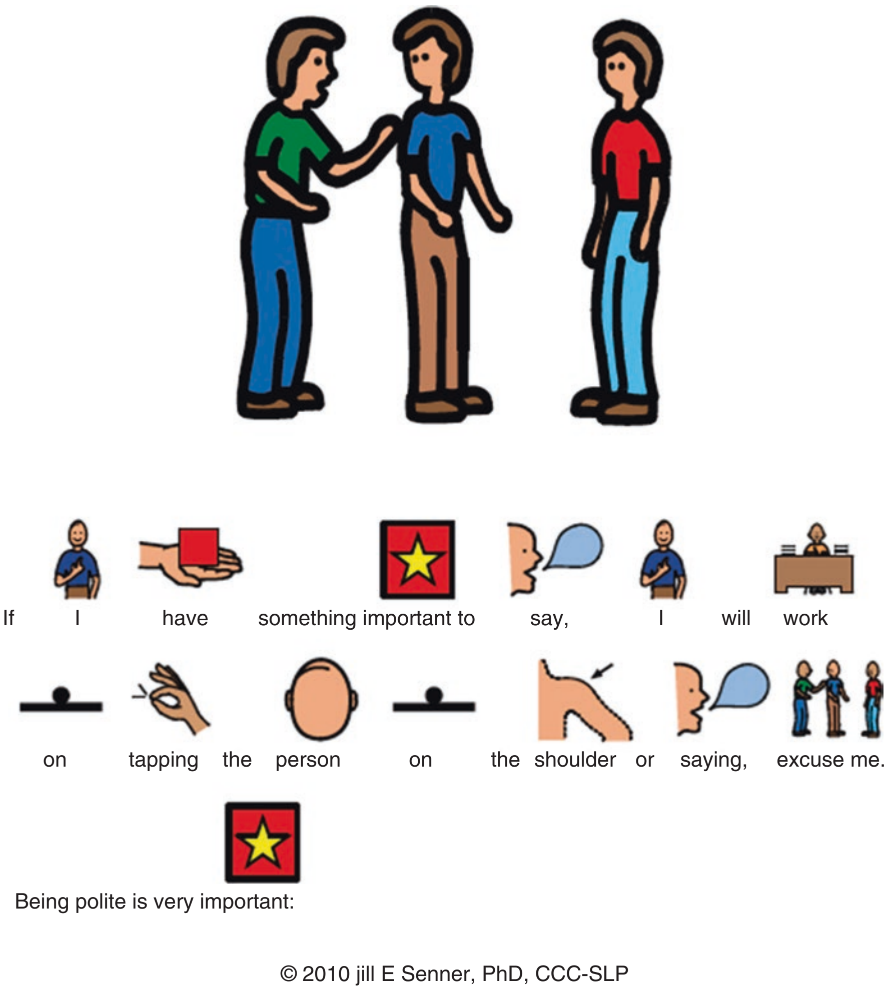
Despite the effectiveness of these interventions, data suggest some individuals with disabilities may not generalize AAC use for communicative functions beyond those for which they received direct instruction. For example, in a study in which two children with quadriplegia and age-level receptive language skills were taught to request objects, no generalization to other communicative initiation skills was observed (Calculator, 1999; Glennen & Calculator, 1985). It has been suggested that interventions that occur in natural settings and in the context of meaningful activities are associated with greater communicative competence (Lilienfeld & Alant, 2005a; Warrick, 1988).

Finke and Quinn (2012) surveyed SLPs about strategies they used to promote or maintain more active communication in young children (under the age of 5) who use AAC. They noted the importance of including appropriate vocabulary for initiating on an AAC system as well as modeling initiation using the child's system (i.e. providing PAI). However, additional strategies utilized included acknowledging all communication attempts from the child, using activities that are meaningful and motivating, and creating communicative temptations (i.e. structuring the environment to entice a child to communicate).

The use of social stories, video models, and scripts have also shown promising results in teens and young adults who use AAC. Social stories are individualized short stories written from an individual's perspective that explain difficult social situations through visual supports and text (Gray & Garand, 1993). Video modeling interventions involve an individual watching videotapes of positive examples of adults, peers, or him- or herself engaging in a pragmatic skill (Delano, 2007). Script training may be used to teach a variety of social interactions (Terpstra et al., 2002). Scripts are visual or auditory supports that include roles for all who participate, and statements or questions related to a specific communicative purpose such as social closeness. Scripts can be pictures, audio files, written words, phrases, or sentences that enable the individual to perform a targeted skill such as starting or continuing conversations (Krantz & McClannahan, 1998; Stevenson et al., 2000).

In research reported by Senner and Baud (2017a), nine participants (four males and five females) ranging in age from 15 years 3 months to 22 years 1 month ($M = 19.44$, $SD = 1.95$) participated in a four-week online social skills class involving reading social stories, watching video models, and using scripts (Fig. 25.2). All nine participants used dynamic display speech-generating devices. Participants were taught how to interrupt appropriately, such as waiting patiently in proximity of others, tapping the communication partner on the shoulder and/or saying, "excuse me" if the message was urgent. Eight participants had higher interrupting post-intervention test scores than pre-intervention test scores. One participant showed no improvement. Results were statistically significant ($Z = -2.588$, $P = 0.010$) using a Wilcoxon Signed Ranks test. On the Pragmatics Profile (Wiig et al., 2013) question pertaining to interrupting, five participants showed improvement and four remained the same. The difference was also statistically significant ($Z = -2.121$, $P = 0.034$).

Another packaged intervention combining video models and scripted conversation in conjunction with least-to-most prompting was investigated with adolescents with autism who used AAC (Thirumanickam et al., 2018). A least-to-most prompting hierarchy (also known as system of least prompts) is a prompting hierarchy in which the least amount of prompting is provided at the beginning with additional cues provided within a specified interval and order as needed (i.e. with increasing assistance). After up to nine intervention sessions, three of the four study



If I have something important to say, I will work on tapping the person on the shoulder or saying, excuse me.

Being polite is very important:

© 2010 jill E Senner, PhD, CCC-SLP

Fig. 25.2 Sample page from a social story about interrupting. Note the inclusion of a directive sentence, one designed to suggest a response and gently direct behavior

participants demonstrated improved performance in conversational turn taking (e.g. responding to a communication partner's question and asking a follow-up question).

The use of mentors, older, more experienced adults who use AAC, has also demonstrated promising results. Adolescents and young adults who used AAC reported enjoying the social support of interacting with an older, successful communicator who also used AAC (Light et al., 2007). Many of the participants also felt they benefitted from talking to someone who could help them set goals and problem-solve.

Adults with cerebral palsy who demonstrated good communication skills, literacy skills, and leadership potential were taught to improve sociorelational skills via an online training program in an effort to become mentors. One targeted skill, being other-oriented, defined as demonstrating respect for and interest in a communication partner, was taught using the acronym LAF: (a) L = Listen to the partner and communicate respect; (b) A = Ask the partner questions to find out more about his or her interests and concerns; and (c) F = Focus on what the partner is saying (Light et al., 2007). The participants were also taught behaviors to avoid such as criticizing, reacting hastily, and talking too much about oneself. All the participants learned to become more other-oriented as a result of the training and felt satisfied with the training received.

25.7.4 Communication Partner Instruction

Live and online parent training in AAC has been linked to positive changes in children's communication (Bruno & Dribbon, 1998; Douglas et al., 2017; Romski et al., 2010). Parent-implemented naturalistic behavioral interventions such as Joint Attention Symbolic Play Engagement and Regulation (JASPER), combined with use of a speech-generating device, resulted in improvements in joint attention, spontaneous communicative utterances, novel words, and comments in children with autism (Kasari et al., 2014). Parent training in AAC has also been shown to increase parent provided communication opportunities, child communication, and parent responses to child communication (Douglas et al., 2017).

Shared experiences and proximity are important for the development of social relationships between students using AAC and their peers. Children must have opportunities to interact with each other around common interests and activities. In addition, teaching peers strategies and skills to promote interaction have been linked to positive effects on interactions with individuals using AAC (Chung & Douglas, 2015; Lilienfeld & Alant, 2005b; Therrien et al., 2016). A recent systematic review of interventions found that frequency of interactions between children who use AAC and their peers could increase throughout the school day with appropriate supports (Therrien et al., 2016). Interventions may or may not also involve the individual using AAC. However, interventions that included multiple training elements were more effective than single-component interventions. Instruction of group home and adult day program staff has also been found to increase communication opportunities and active communication in an adult using AAC (McNaughton & Light, 1989).

Effective communication partner traning programs should include the following elements: (1) Theory/strategy description; (2) Demonstration and modeling; (3) Practice; (4) Feedback; and (5) Coaching (Joyce & Showers, 1980; Senner & Baud, 2017b). In *theory/strategy description*, instructors provide a verbal description as well as information regarding the theoretical base for the strategy being taught. *Demonstration and modeling* may include live or videotaped use of the target strategy. Participants should then *practice* the target skill or strategy in a controlled environment. *Feedback* can be provided by peers, coaches, or self-administered and involves observation and reflection on use of the target strategy or skill. Finally, *coaching* involves a live observation and feedback cycle in the natural environment. The Kent-Walsh and McNaughton (2005) eight-step instruction model for use with communication partners of people who use AAC contains all these training elements and can be used to teach communication partners to use a variety of targeted strategies that encourage rather than inhibit communication.

Partner attitudes can also be influenced by intervention. For example, providing additional information regarding the individual using AAC has been associated with formation of more favorable attitudes (Gorenflo & Gorenflo, 1991). Some classroom-based strategies for increasing peers' understanding of AAC include reading and discussing books about people who use AAC (e.g. Sarah's Surprise by Nan Holcomb (1990)), providing hands-on experiences with a variety of AAC systems, and having students engage in role-playing activities in which they are not able to use natural speech and/or need to use a communication board or device (King & Fahsl, 2012).

25.8 Case Study

Amelia Brown is a 20-year-old female with diagnoses of spastic quadriplegic cerebral palsy and dysarthria of speech. Amelia uses direct selection to access an Accent 1000 speech-generating device. Direct selection is the ability to physically touch an item, point to, or press a button. She uses her right index or middle finger to access devices and requires a keyguard to improve accuracy. A keyguard is a shield with a set of holes that fits over the display to prevent her fingers from touching unintended words. Results from the Receptive One-Word Picture Vocabulary Test, 4th Edition (ROWVT-4; Martin & Brownell, 2011) place her approximate language age around 6 ½ years. Her hearing and vision are functional for her needs.

Results of the Social Networks Assessment revealed that Amelia produces three sign approximations including holding her fist near her chin for "mom," holding her fist near her forehead for "dad," and placing her right fist to her right ear for "sleep" or "loud." Generally understood gestures included nodding/shaking her head for yes and no and pointing towards desired objects with an open hand. She intentionally vocalized to gain attention (e.g. when a caregiver is in another room), laughed to express humor or pleasure, and cried when upset. Amelia's signs and gestures are best understood by familiar communication partners who know her well. Most of Amelia's communication partners are family members and paid professionals.

Amelia's communication was efficient, however, results of the Pragmatics Profile indicated particular difficulty initiating and maintaining conversational topics. Her mother rated Rituals and Conversational Skills item #5 "introducing appropriate topics of conversation" as *Never or Almost Never* and item #6 "maintaining topics using typical responses" as *Sometimes*. Language sampling revealed the ability to navigate her speech-generating device well and to generate a number of multi-word utterances (e.g. "I watch TV with Mom", "I listen to music Christmas"), with occasional errors in word order and verb tense noted. She took approximately 80% of obligatory turns in conversation, however, she only took 15% of non-obligatory turns. No partner-focused questions were observed during the language sample.

Following assessment, the following pragmatic goals were included in Amelia's implementation plan: (1) Amelia will take non-obligatory turns during conversation in four of five charted opportunities when provided with an expectant pause; and (2) Amelia will ask partner-focused questions during conversation in four of five charted opportunities when provided with an expectant pause. She attended once-weekly therapy sessions and direct intervention included the use of social stories, video models, and script training from the Chat with Me series (Technology & Language Center, Inc, 2017a, 2017b) for each of the skills above. She was also coached in using these strategies during conversation with a familiar communication partner (her mother). Within three months, Amelia had met criterion for both of the goals listed above. At about the same time, Amelia had attended a family wedding and as per parent report, several friends and distant family members had remarked about Amelia's "improved communication."

25.9 Conclusion

Given the diversity of individuals who use AAC, it is difficult to draw generalizations about pragmatic skills in this population. However, one thing is clear. Individuals who use AAC have tendencies to be more passive communicators. This may affect their ability to form friendships and in turn impact their quality of life. Pragmatic skills should not be sacrificed while working on expression of wants and needs, learning how to operate a device, or select symbols. Rather, pragmatic skills can and should be taught concurrently with other communicative competencies beginning in childhood. Furthermore, partner training in strategies to support communication are as important as direct intervention with individuals using AAC.

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Chapter 26
Adults in the Prison Population

Karen Bryan

26.1 Introduction

Rates of incarceration and prison policies vary in the western world. People are imprisoned partly to remove those deemed dangerous to protect others, and partly to punish them for breaking the law. The numbers of people incarcerated have grown significantly in countries such as the United States (US) and the United Kingdom (UK). Against these increased rates of incarceration, re-offending rates remain high. Around 39% of young offenders in the UK re-offend and enter adult prisons (Ministry of Justice, 2019). The experience of being in prison is associated with increased physical and healthcare needs. This chapter will examine these needs, with special consideration of the language and communication challenges faced by adults in prison. The discussion will address the social disadvantage that places an individual at risk of incarceration, the developmental and psychiatric disorders that are found in adults in the prison population, and the unique needs of different categories of offenders. It will also highlight the importance of speech and language therapy within the prison setting, and the role of pragmatic language skills in verbally mediated interventions designed to reduce re-offending rates.

26.2 International Incarceration Statistics for Adults

Using the World Prison Population List (Walmsley, 2018), the US has the highest prisoner rate, with 655 prisoners per 100,000 of the national population. The US is by far the leader among large industrialized nations in terms of the number of people it incarcerates. Russia comes closest at 451 prisoners per 100,000 of population,

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although there is no available data regarding China's incarceration rate. The US also has the largest prison population, with roughly 2.2 million people incarcerated in the country in 2014. China's estimated prison population totalled 1.7 million people that year. Other nations with population sizes comparable to the US have far fewer prisoners.

The majority of US prisoners in federal correctional facilities are of black or African-American origin (US Department of Justice, 2018). As of 2011, there were about half a million male and about 26 thousand female black, non-Hispanic prisoners. They made up 40% of all incarcerated persons in the US but accounted for only 13% of the total US population. About 237 thousand prisoners in state facilities were sentenced for drug-related offences, accounting for roughly 17.4% of all state prisoners. In the US, drug-related offences, such as trafficking and possession, were the most common cause of imprisonment in state prisons. The next most common causes were crimes such as robbery and murder, at 13.6 and 12.2%, respectively.

In other OECD countries rates are lower. Scotland, England and Wales have the highest imprisonment rates in western Europe, with 143 people per 100,000 of population in Scotland and 141 per 100,000 in England and Wales. This compares to only 59 and 51 per 100,000 in prison in Sweden and Finland, respectively (Ministry of Justice, 2018a). The prison population in the UK has risen over the last 30 years but has reduced in the last two years. Internationally, analysis shows that there is no link between the prison population and levels of crime (Lappi-Seppälä, 2015). Possibly, this may relate to political reasons for imprisonment in different countries, with the US having a focus on protection of the public by removing people who threaten safety, the UK having an equal emphasis on punishment and rehabilitation, and countries with the lowest levels of incarceration focussed more on rehabilitation.

26.3 Adult Prisoners and Disadvantage: A UK Perspective

In December 2018, the adult prisoner population in England, Wales and Northern Ireland was 82,384 (HM Prison Service, 2018). Taking the adult population in England and Wales as an exemplar, we can examine the characteristics of the prison population. These characteristics are analysed in detail annually in the Bromley Briefings Prison Factfile which are produced annually by the Prison Reform Trust (2017, 2018). Based on the adult prison population in England and Wales in 2012 (Ministry of Justice, 2012), we can state that:

- 24% of prisoners were taken into care as a child (31% for women and 27% for men)
- 29% experienced abuse as a child (53% for women and 27% for men)
- 41% observed violence regularly in the home (50% for women and 40% for men)
- 59% regularly truanted from school
- 42% were expelled or permanently excluded from school

- 47% have no educational qualifications
- 68% were unemployed in the four weeks before custody (81% for women and 67% for men)
- 13% have never had a job
- 15% were homeless before entering custody
- 54% have children below the age of 18
- 16% have symptoms indicative of psychosis (25% for women and 15% for men)
- 25% identified as suffering from both anxiety and depression (49% for women and 23% for men)
- 46% of women and 21% of men have attempted suicide
- 64% have used class A drugs
- 22% drank alcohol every day in the four weeks before custody

Thus, it is clear that a very large proportion of the adult prison population have a troubled background with significant markers of disadvantage, including nearly half having no educational qualifications and a high proportion unemployed prior to entering custody.

The re-offending rates suggest that prison is unsuccessful in rehabilitating prisoners. 48% of adult prisoners re-offend within one year of release (Ministry of Justice, 2018b). This includes 58% of women, rising to 73% of women with a sentence of less than 12 months and 83% of women who have more than eleven previous convictions (Ministry of Justice, 2018c).

Women are a minority within the prison population in England and Wales, accounting for 5% of the adult prison population. The reasons for their offending differ from men and they often have multiple and complex needs (Ministry of Justice, 2018d). On 30th November 2018, 3807 women were in prison in England and Wales. 60% of women were remanded into custody by a magistrate's court. 41% remanded by a crown court did not receive a custodial sentence. 83% of women committed a non-violent offence, with theft being the most common reason for incarceration. 62% of women received a sentence of less than six months (Ministry of Justice, 2018d).

48% of adult prisoners in the UK re-offend so there is a significant proportion of the population revolving in and out of prison. Given the adverse impact of communication difficulties on accessing healthcare and on employment outcomes, we might hypothesise that adult prisoners with communication difficulties are much less likely to benefit from verbally mediated interventions to prevent re-offending and are less likely to gain employment.

26.4 Young Offenders

Most of the research into offenders with language and communication difficulties has been conducted with young offenders in community settings—see Snow (2019) and Snow and Bryan (2018) for recent reviews. These reviews outline the high proportion of young people in contact with criminal justice services who have speech, language and communication difficulties. In the UK, we must take care not to criminalise automatically such people, as Youth Justice Services take prevention referrals as well as deal with those who have offended. The relatively small amount of research that has been conducted shows that:

- Speech and language therapy services can be delivered effectively in criminal justice settings (Bryan et al., 2007; Snow & Woodward, 2017).
- Improvements in language functioning are detectable on standardised tests when speech and language therapy provision is added to the support package available to young people (Gregory & Bryan, 2011).
- Staff perceive a benefit to the wider delivery of justice services for young people when they have access to training and support to manage communication difficulties supplied by a speech and language therapist (Bryan & Gregory, 2013).

In December 2018, there were 839 young people under the age of eighteen in custody and 924 eighteen years or over in youth custody in the UK (Youth Custody Service, 2018). Data from Bryan (2004) suggests that at least 60% of these offenders will have speech, language and communication difficulties that will affect normal everyday functioning, education and engagement in verbally mediated interventions. This would be 1058 offenders using the December 2018 population. The youth re-offending rate in the UK stands at 39.3% (Ministry of Justice, 2019), suggesting that around 415 young offenders with significant speech and language difficulties will enter the adult estate each year. While this is an extrapolation from current figures, it does have resonance with other characteristics of the adult prison population as discussed in Sect. 26.3. Snow (2019) has set out a compelling research agenda for this field. There is a need for significantly more research into the nature of communication difficulties, their impact on the young person's rehabilitation, and the potential for improved communication to support access to preventative measures such as engagement in education or meaningful work and re-engagement with families.

26.5 Physical and Mental Health Problems in Adult Prisoners

Adults in prison experience a wide array of physical and mental health problems, often at an increased prevalence over the general population. In many of these conditions, language and communication difficulties arise. Although there is a dearth of studies of these difficulties in the adult population, they are well documented in young people who are in custody, especially in relation to mental health, neurodevelopmental disorders, and social and cognitive difficulties (Hughes et al., 2017). There is a further complication in the adult population in that developmental conditions presenting in adults may also be accompanied by conditions associated with ageing. This issue is addressed in Sect. 26.9.

26.5.1 Physical Health Problems

Problems that affect physical health are commonplace in adults in prisons. A study conducted in Italy attempted to assess health conditions of all inmates in six Italian regions. The study captured 92.2% of the adult prison population in these regions and represented 28% of the entire Italian prison population (Voller et al., 2016). A total of 15,751 inmates were enrolled in the study. The mean age was 39.6 years with an age range of 18 to 60 plus. All inmates were examined by a doctor using a standard set of tests based on the Clinical Modification of the International Classification of Diseases (ICD-9-CM) criteria (World Health Organization, 2011a). On average, the inmates presented with 2.2 disorders each. 32.5% did not present with any disorders. The most common disorder was psychiatric disorder (41.3%), followed by digestive (14.5%), infectious (11.5%), cardiovascular (11.4%), endocrine, metabolic and immune (8.6%) and respiratory (5.4%) conditions. Diseases of the nervous system accounted for 4% of disorders. The authors commented particularly on the over-representation of chronic diseases associated with lifestyle in a relatively young population (Voller et al., 2016).

26.5.2 Psychiatric Disorders

Fazel et al. (2016) conducted a systematic review of the prevalence of psychiatric disorders in prisoners worldwide. They note that differences in methods of identification and use of characteristics that are highly correlated to criminogenic factors (such as disregarding norms and rules, low threshold for violence and inability to profit from experience) lead to variations in prevalence figures. However, a systematic review of 33,000 prisoners and over 100 studies showed a consistent finding of one in seven prisoners having a major depression or psychosis (Fazel & Seewald, 2012). Another consistent theme is the high rate of substance abuse among prisoners. Butler et al. (2011) showed that there is a high rate of comorbidity between mental illness and substance misuse. Such co-morbidity is detrimental to the prognosis for the individual with a psychiatric disorder and increases the likelihood of re-offending and premature mortality following release (Chang et al., 2015).

Studies also consistently show higher rates of psychiatric disorders, particularly depression and drug dependence in female prisoners (Binswanger et al., 2010). A recent problem is novel psychoactive agents. The Inspectorate of Prisons in England and Wales stated that these substances, particularly synthetic cannabinoids, have led to increased violence in prisons as a direct result of drug intoxication or increased bullying due to drug debts (HM Inspectorate of Prisons, 2015). However, reliable detection of these substances is difficult partly due to disincentives to self-report. Longitudinal studies are needed to fully understand whether prisoners bring psychiatric disorders to prison with them, or whether these develop in the prison environment (Fazel et al., 2014).

There are significant adverse outcomes for prisoners from psychiatric disorders. Suicide rates are difficult to validate given variation in methodologies for reporting deaths and reluctance in some cultures to record deaths as self-inflicted. Fazel et al. (2011) showed that in Western Europe, most countries report around 100-150 suicides per 100,000 prisoners, but France is an outlier with 179 per 100,000 (Duthe et al., 2013). Suicide rates are also lower in the US with 41 per 100,000 in local jails and 16 per 100,100 in state prisons. This lower rate is thought to reflect the high proportion of African American and Hispanic prisoners who have lower suicide rates (Bureau of Justice Statistics, 2015). Self-harm is also an adverse outcome of psychiatric problems. Hawton et al. (2014) showed that 5–6% of men and 20–24% of women in prison in England and Wales self-harmed, with risk factors being younger age and short sentences. Guidelines for suicide prevention include early screening of prisoners, actions taken in response to early screening, and on-going risk monitoring (Konrad et al., 2007). Given that screening is likely to be verbally mediated, any prisoner with communication difficulties may not have their level of risk recognised or fully recognised.

Many interventions aimed at improving prisoner mental health have been evaluated but most studies are small scale. In addition, heterogeneity of the prison population and practical difficulties such as obtaining permissions and running interventions over time result in limited research. Barker et al. (2014) conducted a systematic review of evidence-based activities and concluded that multi-factored suicide prevention programmes appear more effective. They also suggest that using trained inmates to provide social support, and positive staff attitudes towards prisoners may also be influential factors in suicide prevention. There are no studies on the outcomes for prisoners with communication difficulties in relation to such programmes. However, we might hypothesise that if social support is a positive factor in suicide prevention, this will be more difficult to access for prisoners with communication difficulties.

26.5.3 Prisoners with Learning Disabilities

A significant number of adults in prison have intellectual or learning disabilities. Jones and Talbot (2010) showed that adults with intellectual difficulties including learning difficulties or disabilities were over-represented in prison populations, with estimates varying from 20 to 30% depending on definitions and methodologies for identification. Jones and Talbot also demonstrated that despite the Disability Discrimination Act 2005 in the UK placing a statutory responsibility on public bodies to identify and make reasonable adjustment for the needs of people with disabilities, intellectual disabilities were largely unrecognised in the UK prison system.

Murphy et al. (2017) systematically screened nearly 3000 new prisoners entering three category B male prisons in city locations. (Convicted criminals are generally placed in category B prisons if they are not deemed to be the highest level of security threat. However, they are still recognised as being ‘high risk’ and require significant security measures to ensure they do not escape.) The Learning Disability Screening Questionnaire (LDSQ; McKenzie & Paxton, 2006) was used. Prison staff with experience of working with people who have intellectual difficulties were trained to administer the LDSQ within 7 days of admission to the prison. Of the 3778 entering prison, 2429 were screened. But 396 refused, 216 were non-English speaking and the remainder were unable to give consent or were suffering from serious mental health problems. 169 (or 7%) were identified as having an intellectual disability on the LDSQ. This study demonstrates the feasibility of screening for intellectual disabilities within an adult prison environment. It is important that people with intellectual disabilities are identified because they are known to find it difficult to understand written information such as prison rules and to have difficulty using systems such as booking doctors’ appointments. They are also more likely to be depressed, anxious, and bullied (Talbot, 2008).

Although the Bradley Review in 2009 made over 90 recommendations for the care of people with learning disabilities in the criminal justice system in England and Wales (Bradley, 2009), including screening for intellectual difficulties, there is still no systematic screening and support for these prisoners.

26.5.4 Prisoners with Deafness

People with deafness are over-represented in the prison population in both the UK and in the US (Williamson & Grubb, 2015). A survey of prisons and young offender institutions in England and Wales identified 135 deaf or hard of hearing prisoners (Gahir et al., 2011). The US Bureau of Justice Statistics stated that 7% of prisoners rising to 13% for over-45s had a severe hearing loss or deafness based on a 2004 survey of inmates (Maruschak, 2008). Several studies have also shown increased occurrence of sexual offending amongst deaf offenders (Young et al., 2000). Miller and Vernon (2003) reported the rate of sexual offending by deaf prisoners to be four times the rate of hearing offenders. Williamson and Grubb (2015) conducted a systematic review of the literature on the reasons for this bias. They examined the characteristics of deaf offenders in relation to their personalities, language and brain development. They found a dearth of literature but some evidence for sexual offending linked to sexual abuse experienced by the deaf person in childhood, given that there are increased rates of child sexual abuse in the deaf population (Miller et al., 2005). However, Williamson and Grubb (2015) suggest that it is only when deaf children who have been abused experience further unfortunate circumstances such as social isolation or limited sexual education that they become perpetrators of abuse.

Language barriers were also noted to affect the social and psychological development of deaf adolescents. Vernon and Rich (1997) reported that in a sample of twenty deaf offenders, eighteen could not speak and six had minimal use of sign language, reducing their ability to communicate with others. Deaf people are also noted to experience a higher rate of learning difficulties and disabilities than the hearing population. Vernon and Greenberg (1999) highlight this prevalence and suggest that developmental disorders are linked to a propensity towards use of violence. Young et al. (2001) propose a direct link between developmental disorders and sexual offending. Their study of 204 deaf offenders showed that 84 had communication difficulties that masked an underlying disorder such as Asperger's syndrome. Research suggests that if these disorders are unrecognised and therefore not managed, they can result in deviant behaviour such as sexual offending (Allen et al., 2008). A large, longitudinal study of children with developmental language difficulties suggests that language difficulties may be associated with sexual offending, although the results are preliminary (Mouridsen & Hauschild, 2009). More research is needed to understand fully why deaf people are more likely to commit sexual crimes and to prevent this from happening.

Problems with mental health may also contribute to offending behaviour in deaf individuals. However, the literature is very limited. Also, Williamson and Grubb (2015) note that mental illness is difficult to assess and identify where the deaf individual's use of sign language, facial expression and alternative ways of communication can be mistaken as a mental impairment, particularly where staff are not experienced signers with knowledge of the deaf community and its culture. A study of deaf people in prison who use sign language to communicate concluded that the needs of this group of offenders are not fully recognised or met (O'Rourke & Grewer, 2005). While the mental health needs of this group of prisoners appears to be different from that of the hearing population, more research is needed to understand these needs and the pattern of offending behaviour in deaf individuals in prison (Young et al., 2000). Hearing impairment is considered further in Sect. 26.10 on older female prisoners.

26.6 Pragmatic Language Difficulties in Adults in Prison

Prisoners with pragmatic communication difficulties are immediately disadvantaged. Prisons are complex rule-governed institutions with significant restrictions on personal freedom imposed by those rules. Learning the rules and applying the rules requires significant pragmatic language understanding. While assessing language skills in a random sample of prisoners, I had to see one inmate in the segregation unit as he had attempted to smoke in the education unit. He reported checking carefully and there being an absence of 'no smoking' signs. He had not understood the implications of the prison being a no smoking establishment with smoking only allowed in certain outdoor areas.

Negotiating relationships with other prisoners and staff is necessary to establish a place within a wing and to build a small network of trusted people who will 'look out' for you. This is difficult for someone with speech difficulties, someone who finds it difficult to convey information, or for someone who finds it difficult to understand aspects of communication such as sarcasm, tone of voice, implication or warning.

Processes such as booking a phone call and booking a visit for a friend or family member require completion of official forms. If prisoners need help with such processes, they can be deemed to have a weakness that other prisoners may choose to exploit. This can lead to exploitation or to prisoners opting out of such processes to save face, thus increasing their isolation.

Completing the regular 'canteen', which is a regular order of food or personal items that can be ordered using a limited allowance of cash and any earnings from paid employment within the prison, involves completing a complex form. Prisoners with communication or literacy problems often rely on other prisoners to complete this form for them. Asking another prisoner to help can result in a requirement to order something for the helper willingly or otherwise.

Communication difficulties can also lead to staff being unable to ascertain what is troubling a prisoner. I recall a prisoner showing frustration with an officer who was trying to obtain additional phone credit for him as he wanted to call his mother. The officer was aware of the prisoner's frustration but could not ascertain the cause and asked me to intervene. The prisoner found it difficult to convey factual information and also to express his feelings. Using structured questioning, scaffolding his responses and checking back meaning, slowly revealed that he was grateful for the additional credit and recognised that the officer was trying to help. However, the key issue was that his mother had not answered his calls for over a week and he was concerned that she might be ill. Understanding his concern accurately required time and skills that the officer might not have. Such mis-communications are common in the prison environment and may lead to prisoners lashing out when their needs are not understood. This is referred to as 'kicking off' in the prison environment.

Although the adult prison regime provides rehabilitation to prevent re-offending and provision to address issues such as mental health and illegal drug taking, as well as more specialist provision to prevent sex offending, these interventions are all verbally mediated, and most are offered in a group therapy context which makes very significant demands on the person's language skills (Bryan et al., 2015). As yet, there is no systematic assessment of speech, language and communication difficulties for adult prisoners in the UK, either routinely or before embarking upon a verbally-mediated intervention. There is also almost no access to speech and language therapy services for adult prisoners. This means that where a prisoner is struggling to communicate with staff, there is no support for the prisoner or for the staff to address communication difficulties. Failure of a therapeutic intervention will usually count badly against a prisoner who, for example, does not complete a groupwork intervention. Yet, the influence of communication difficulties is not considered.

The pragmatic language difficulties examined in this section can be most acute in certain prison settings and contexts (e.g. entering prison) and for specific categories of prisoners with particularly challenging needs. The chapter explores these prisoners and their experiences in the following sections.

26.7 Experience of Entering Prison

Entry into prison can be a time when the consequences of poor verbal communication skills can be particularly keenly experienced. Williams et al. (2013) examined prisoners' subjective experience of early imprisonment. 170 men were interviewed as they entered custody and three weeks later. 75% were re-offenders. 168/170 gave some free narrative about their life, with prompts used if the prisoner appeared to have difficulty in responding. The authors noted that the free narratives of their experiences were often very brief and that this was not a very verbally fluent cohort of men. In addition, the authors noted a bias in that the sample included those willing to take part in an interview. As well as concerns about adverse aspects of imprisonment such as depression, loneliness, missing relatives and friends and worries about personal safety, good relationships with staff and staff who were able to facilitate good relationships between prisoners were perceived as making a life-saving difference. It is possible to speculate from this study that many of the men had a restricted ability to convey information via verbal language, and that those with language difficulties would be less likely to engage positively with staff. Williams et al. (2013) also stated that few prison staff are trained to create a therapeutic milieu in a prison.

26.8 Foreign National Prisoners

More awareness appears to be given to language issues in foreign nationals who have minimal or no ability to communicate in English. We might hazard a guess that this is because (a) speaking another language is an obvious issue, and (b) finding (and funding) an interpreter is an obvious solution. Sen et al. (2014) showed that the foreign national prisoner (FNP) group within UK prisons has risen rapidly. Ministry of Justice (2018a) state that 11% of the UK prison population are foreign nationals, with 9047 foreign nationals in prison in the UK in September 2018. The FNP group has a higher incidence of self-harm and suicide (Borrill & Taylor, 2009), which is associated with uncertainty about their ongoing fate. Only a quarter of FNPs report that their main language is English (HM Inspectorate of Prisons, 2018).

Language difficulties are further exacerbated by recurring problems with the quality of translated materials and interpreting facilities (HM Inspectorate of Prisons, 2010). Information packs are unavailable in 31 of the 160 languages within the FNP group, which may exacerbate feelings of social isolation (Barnoux &

Wood, 2013). Bhui (2009) suggest that communication problems for FNPs may reduce the ability to express mental health issues, reduce access to services, exacerbate difficulties with understanding legal documents, inhibit wider understanding of the environment and context, and enhance feelings of hopelessness and vulnerability. We might also note that these issues would apply to an English-speaking prisoner who has difficulty with communication.

Graf et al. (2013) showed that one in three of male and one in two of female FNPs are clinically depressed. Sen et al. (2014) concluded that diagnosing mental health difficulties in FNPs is problematic and that diagnostic and treatment tools specific to FNPs are needed. They also call for a systematic study into the prevalence of mental health problems in FNPs.

A more recent study of culturally and linguistically diverse women in prison in Australia—where 20% of female prisoners in New South Wales speak a language other than English—showed that limited English proficiency was a barrier to care. The use of fellow prisoners as interpreters was valuable but significant challenges to their use emerged, such as vulnerability to coercion, loss of confidentiality, and errors in interpretation of information (Watt et al., 2018).

Psychiatric assessment and risk assessment largely rely on verbally-mediated tests and discussion. Therefore, all prisoners with communication difficulties are vulnerable to their risk status and their mental health difficulties not being fully recognised. Incomplete understanding of a prisoner's risk status can potentially endanger staff and fellow prisoners who are in proximity to the prisoner.

26.9 Older Prisoners

As already discussed, the worldwide prison population is increasing. In countries including the UK, the USA, Australia, Canada, and New Zealand, the fastest rise in prisoners is in the number of older prisoners (Maschi et al., 2012; Stevens et al., 2017). One of the reasons for this increase is the growing number of men convicted of historical sexual offences (Prison Reform Trust, 2017). The term 'older prisoners' usually refers to prisoners over 50 years of age. This reflects the well-documented fact that prison accelerates physiological ageing (Hayes et al., 2012), although there is some variation in studies.

26.9.1 Multi-Morbidity in Older Prisoners

Gates et al. (2018) examined multi-morbidity patterns in the entire over 50s male prison population in the Department of Corrections in the South East Central region of the United States. Multi-morbidity and functional limitations are associated with populations with limited economic resources which includes most imprisoned individuals (McLean et al., 2014). There were 2010 men aged 50 years or older with a sentence of over six months. Data was extracted from their medical records using ICD-9 codes to identify all chronic diseases, mental health problems, substance use disorders, and functional limitations. 56% of the older population had multiple morbidities and 10% of the group had functional limitations. Three patterns of comorbidity were identified. These were associated with chronic diseases, geriatric conditions (e.g. joint problems and dementia), and the third category of substance misuse and mental health disorders. 54% of the older prisoners appeared in two or three of the categories. Communication difficulties were not included as a potential functional limitation, but problems with oral health, such as tooth loss having the potential to affect speech, and hearing impairment were recognised.

A study of factors related to quality of life in older prisoners showed that physical health, psychological health, social relationships and environmental factors all contributed to reduced quality of life (De Smet et al., 2017). The prisoners were in sixteen prisons in a Dutch-speaking region of Belgium. There were 93 cases which represented 45% of the entire population of older prisoners in Flanders. Their mean age was 65.2 years. The prisoners were examined on a series of tests such as the Mini International Neuropsychiatric Interview (Sheehan & Lecrubier, 2006), the Forensic Camberwell Assessment of Need (CANFOR; Thomas et al., 2008), the Tilburg Frailty Scale (Gobbens et al., 2013), and the WHOQUAL BREF which is a quality-of-life scale that can be applied irrespective of context and which has been validated for use with prisoners (Saloppé and Pham 2006).

The results showed that 24.2% of the older prisoners had at least one psychiatric disorder and 15% graded themselves as vulnerable people. All respondents had at least one physical health problem. 63.4% did not engage in personal conversations with prison staff, which is suggested as the reason for ‘social relationships’ being the lowest scoring of the four scales of the WHOQUAL. This has been reported in other studies which suggest that social isolation in older prisoners may be a response to increased perceptions of potential rejection and victimisation (Ireland & Qualter, 2008). De Smet et al. (2017) suggest that special attention should be given to psychiatric and age-related symptoms of older prisoners as these are less likely to be noted by staff, and older prisoners appear to be less effective self-advocates than their younger peers.

A study of distress in older prisoners in the USA used a cross-sectional approach to examine 125 participants aged 55 years or over (Bolano et al., 2016). They used a ‘teach to goal’ methodology to obtain informed consent, which has been shown to be effective for older adults with low literacy levels (Sudore et al., 2006). The main distress instrument used was the Memorial Symptom Assessment Scale (MSAS; Portenoy et al., 1994) which has been used to measure physical distress in vulnerable adult populations (Ritchie et al., 2014). Measures of physical and psychological distress, social distress and existential distress are given. The results showed that 44% of the older prisoners had at least one symptom of physical distress, with pain being the most common symptom. 54% reported existential distress, with missing out on things in life due to substance abuse (30%) and having unfinished business (27%) being the most common symptoms. 56% reported psychological distress, with depression (26%) and anxiety (30%) the most common symptoms reported.

45% reported social distress on the Three Item Loneliness Scale (Hughes et al., 2004), with previous studies showing that loneliness is a predictor of functional decline and death (Perissinotto et al., 2012). 49% of the participants experienced three or more forms of distress.

26.9.2 Mental Health in Older Prisoners

The mental health of older prisoners can be particularly challenging. Murdoch et al. (2008) examined depression in older prisoners who had a life sentence or an indeterminate sentence. The prisoners were from two category B prisons in the UK. The Geriatric Depression Scale (Yesavage et al., 1983) and the Mini Mental State Examination (MMSE; Folstein et al., 1975) were used to assess 121 prisoners who represented two thirds of the older (over 55 years) prisoner population in the two prisons. The results showed that 48% of the prisoners scored in the mild depression range and 3% scored in the severe depression range. Of the 49% who scored below the threshold for depression, the majority were in the borderline depression range. Prisoners with reduced cognitive functioning as measured via the MMSE demonstrated higher depression scores. Contrary to expectation, higher Geriatric Depression Scale scores were not related to the effects of imprisonment or the length of sentence. Rather, the association was with the imported burden of chronic ill health.

Fazel et al. (2001) found that 32% of older prisoners had a diagnosable psychiatric illness and the most common diagnosis was depression. Despite the high levels of those diagnosed with a depressive illness, only 12% were on antidepressant medication. It has been acknowledged that mental health services in prisons are aimed at the younger, more vocal, prison population and older prisoners may be in danger of being ignored (HM Inspectorate of Prisons, 2004).

Di Lorito et al. (2017) conducted a systematic review of psychiatric disorders in older prisoners as compared to rates in older people in the community. They reviewed nine studies and found that dementia (3.3%) and alcohol abuse (15.9%) levels were comparable to the community sample and all other psychiatric disorders have a higher prevalence in older prisoners. The prevalence rates in the older prisoners were: any psychiatric disorder 38.4%; depression 28.3%; schizophrenia or psychosis 5.5%; bipolar disorder 4.5%; personality disorder 22.9%; posttraumatic stress disorder 6.2%; and anxiety disorder 14.2%. In addition, 11.8% of the older prisoners were found to have cognitive impairment. Dealing with high levels of psychiatric illness in older prisoners is clearly a challenge for policy and practice in prison healthcare.

26.9.3 Cognitive Impairment in Older Prisoners

There is a significant burden of cognitive impairment and dementia in older prisoners. Combalbert et al. (2018) recruited 138 male prisoners who were over the age of 50 and who had been in prison for at least one year from seven prisons in France. They also recruited a control group in the community. The participants were assessed with the Mini Mental State Examination (MMSE; Folstein et al., 1975), the Frontal Assessment Battery (FAB; Dubois et al., 2000), which is used for rapid evaluation of an individual's executive functioning, the French version of the Nottingham Health Profile (Bucquet et al., 1990), and the World Health Organization Quality of Life Questionnaire (The WHOQAL Group 1993), which questions four aspects of quality of life: physical health; mental health; social relationships; and environment. 18.8% of the prisoners had an MMSE score suggestive of dementia and 89% had a FAB profile suggestive of executive functioning difficulties. Also, the prisoners rated their health and quality of life significantly more negatively than did the comparison men.

Risk factors such as substance abuse, post-traumatic stress disorder (PTSD) and a history of traumatic brain injury have been implicated in cognitive deterioration among prisoners (Loeb & Abudagga, 2006; Mallik-Kane & Visher, 2008). An alternative explanation for cognitive deterioration may be a lack of interpersonal interactions and cognitive stimulation. Studies have shown increased levels of engagement in social, physical or intellectual activities and higher cognitive ability scores in older people aged 65 years and above (Sposito et al., 2015), and loneliness has been shown to be a marker of cognitive decline (Cacioppo & Hawkley, 2009). Difficulties with memory, spatial orientation, and language would be particularly restricting in terms of adapting to prison life, and in building relationships with staff and other inmates. Sposito et al. (2015) suggest that as well as preventing reporting of problems, communication difficulties may prevent prisoners' participation in prison activities which could slow or halt cognitive decline. They suggest that all prisoners over the age of 50 should be routinely screened for cognitive disorders.

Flatt et al. (2017) examined PTSD in 238 older age (55 years and above) prisoners in a county jail in the USA. The Primary Care PTSD screen (PC-PTSD; Prins et al., 2003) was used. Nearly 40% of the older inmates had a positive screen for post-traumatic stress and they were significantly more likely to have an impairment in two or more activities of daily living, traumatic brain injury, pain in the last week, and poor self-rated health. The authors suggest that screening for PTSD in prisons may help to identify older prisoners who would benefit from additional mental health assessment and treatment.

Gaston (2018) reviewed the impact of dementia on older prisoners in Australia. She suggests that prisons are not designed for older people or for people with dementia which causes problems for physical and psychological health. Gaston calls for early identification of dementia so that support can be provided along with measures to slow progression. However, Brown (2014) suggests that cognitive impairment may not be recognised until a prisoner's behaviour begins to clash with expectations of the prison environment. As dementia progresses, the prisoner will experience problems with comprehending instructions, socialising, and completing activities of daily living such as performing hygiene-related tasks. These failures may lead to reprimands and disciplinary actions which may adversely impact on the prisoner's physical and mental health.

26.10 Older Female Prisoners

Most of the research that informed the discussion in Sect. 26.9 is based on older male prisoners. But women form a growing minority in the prison population and elderly female prisoners are a smaller subgroup of the female prison population. A study by Handtke et al. (2015) examined the experience of 13 out of a total of 19 elderly (50 years and over) female prisoners with long-term sentences in Switzerland. As well as medical information from their records, the prisoners had a semi-structured interview about their experiences of ageing in prison. The women felt disadvantaged by being female, being in prison and by ageing. These were described as three layers of vulnerability. The authors made a set of recommendations to improve the experience of elderly female prisoners. First, given the significance of social relationships (Reviere & Young, 2004), prisons should have a stronger emphasis on social support networks for elderly female prisoners, which may require revision of rules around visits. Secondly, security and medical staff should be educated about gender and age-specific needs of prisoners. Thirdly, consistent use of handbooks for prison staff and policy makers that are gender sensitive and built on a human rights approach should be made. An example would be the World Health Organization, Penal Reform International (World Health Organization, 2011b). In addition, prison health care should provide good quality care for older women and access to specialist services outside of prison.

A study of functional impairments and the experiences of older female prisoners in the US identified 353 women prisoners aged 55 or older in California (Williams et al., 2006). 120 questionnaires were completed with a 59% response rate. In addition, ten older women prisoners or former prisoners were interviewed. The mean age of the women was 62 years, 12% were aged over 70 and 68% were white. 33% reported three or more co-morbid conditions and 78% took five or more medications. 58% reported visual impairment, 52% reported hearing impairment with 27% reporting difficulty in hearing orders from staff, 28% had experienced memory loss and 22% reported incontinence. In addition, 4% reported difficulties with eating. The authors noted that the questionnaire may not have been completed by older women with literacy difficulties, and that literacy difficulties are associated with chronic disease and poor disease self-management (Williams et al., 2006).

26.11 Dying Prisoners

Turner et al. (2018) propose that older prisoners face a ‘double burden’ when incarcerated in that they are deprived of their liberty and their health and wellbeing needs are not met. In addition, for those of advanced age, a sentence may effectively be a life sentence given that they are likely to die in prison. Hanson (2017) suggests that sentencing reforms and compassionate release programmes in the US have failed to reduce the numbers of elderly or seriously ill prisoners. It is clearly a significant challenge to provide good quality, multi-disciplinary, end-of-life care in a prison environment. Depner et al. (2018) suggest that palliative care can be provided in prison and describes a peer care model in the US, where healthy inmates are trained to provide intimate care. The benefits include improved care for the dying prisoner, reduced workload for staff, and rehabilitation benefits for the caregivers. More research is needed to establish the viability of such schemes.

26.12 Access to Healthcare for Older Prisoners

With a growing population of older prisoners, access to healthcare is an increasingly pressing issue. Heidari et al. (2017) explored access to healthcare for older prisoners in Switzerland. Thirty-five older prisoners were interviewed from twelve prisons, with interviews conducted in the relevant native language. The study showed psychological and environmental barriers to accessing healthcare. Psychological factors included anticipated negative consequences of healthcare-seeking behaviours, such as having to mix with a new group of prisoners and limited experience of applying healthy practices to their lifestyle, previously reported by Loeb et al. (2008). In addition, fear of increased isolation by being in a health unit or centre further discouraged healthcare-seeking behaviour. The environmental factors reported related to no health service provision at night and at weekends and non-availability of services such as physiotherapy and dentistry.

Sullivan et al. (2016) showed that 85% of older prisoners (aged 60 and over) were in receipt of prescribed medication when they were committed to prison. Older prisoners experienced delays in confirming medication, changes to medication, communication difficulties, and enforced helplessness. This study shows that there is a need to increase awareness of prescribing issues specific to older prisoners who are likely to have greater and more complex medication needs than their younger peers.

Sumner (2012) has developed a healthcare assessment protocol for all prisoners as part of a prison healthcare department’s work to ensure that older prisoners’ needs are recognised and managed. The healthcare department is run by nurses with general practitioner input at three surgeries per week. All prisoners are interviewed by a member of healthcare staff on arrival at the prison to take initial baseline observations such as weight and blood pressure. Any medical conditions are recorded, and medical and surgical history, current and past mental health, use of illicit substances such as alcohol and drugs and prescribed and non-prescribed medication usage are all explored. This assessment relies on self-report and may, therefore, be compromised by an inability to communicate fully. Bryan et al. (2007) have demonstrated that self-report is limited in young offenders.

Where prisoners are over 65 years of age, an Elderly and Disabled Assessment (EDA; Sumner, 2012) is conducted. The EDA consists of six computerised evaluations to explore areas where conditions associated with ageing are known to potentially impact on health. The six areas are: continence; mental health; skin condition; mobility and respiration; nutrition; and vision, hearing and speech. The latter are tested as these are known to reduce a person's ability to adapt to prison life by reducing the likelihood of involvement in activities such as education and interaction with fellow prisoners. Where appropriate, referrals to an optician, a hearing aid clinic, or a speech and language therapist can be made. Each prisoner has a care plan with the regime being adjusted where possible, e.g. longer mealtimes when a prisoner has eating difficulties. The care plans are shared with key workers and are regularly monitored by the healthcare team.

This is an example of integrated healthcare provision that is extending to accommodate the needs of older prisoners. Sumner's study shows that this can be accomplished, and there is a need for prisons to adapt systematically baseline health screening to accommodate the needs of older prisoners.

26.13 Policy and Research Agenda for Older Prisoners

In 2012, a meeting of 29 experts was convened to establish the priorities for improving the management of older prisoners. Williams et al. (2012) state that as well as the legal and moral arguments for attention to the healthcare needs of older prisoners, there is also a wider benefit to society in that more than 95% of prisoners are eventually released to the community in the US. Many of these people will have chronic health conditions and will rely on expensive emergency services or are hospitalised after release (Mallik-Kane & Visher, 2008). Therefore, earlier identification and management of age-related conditions and chronic illnesses could enable independent functioning in the community through use of community healthcare resources. The consensus recommendations of the meeting were:

- To define older prisoners as 55 or over, with arrangements to include younger prisoners who have cognitive or functional impairments in activities of daily living
- To train staff in prisons, probation and health in the care of older people
- To define the functional requirements that are necessary to live in prison and to use that list to screen for impairment annually in prisoners over the age of 55 years

- To screen for dementia annually by developing an optimal tool, and to use the results to guide decisions about housing, care programming, medical treatment and discharge planning
- To identify the needs of older women prisoners
- To create uniform policies for housing older prisoners with provision of a continuum of care, including assisted living and 24-hour nursing care
- To ensure release provision includes linking older prisoners to post-release healthcare, close supervision of people with cognitive impairment after release, and available support for health literacy and self-efficacy in the community
- To create national medical eligibility criteria for early release and to remove barriers that could prevent some prisoners from accessing the application process
- To enhance prison palliative care services and to ensure that all healthcare providers are trained in pain management and communicating with patients

Further research is needed into the health and wellbeing of adult prisoners. This could be justified from a moral and ethical stance, but it would also enable prisoners to achieve better outcomes, i.e. lead healthier lives and reduce re-offending. The taxpayer has a vested interest in society achieving this goal. Specifically, research is needed to understand how young offenders with speech, language and communication difficulties navigate entry to adult prison and their outcomes. Studies are needed to show what proportion of the adult prison population has speech, language and communication difficulties and how these impact on their ability to benefit from the regime and from interventions to prevent re-offending. Research is needed to understand how prisoners with communication difficulties impact on the work of prison officers and other staff, and to establish what support staff need in managing these prisoners. There is some research to suggest that positive social relationships with staff and with other inmates constitute a positive coping strategy for adults who are in prison. The impact of communication difficulties on social relationships in the context of prison also needs to be better understood.

Assessment of risk in prisoners with speech, language and communication difficulties also needs to be investigated. The use of non-verbal techniques to supplement verbally-mediated assessments should also be explored. The needs of specific groups of adult prisoners, who are likely to have communication problems, also needs further research; particularly for deaf prisoners, those with developmental disorders, those with mental health problems, and older prisoners who may have health conditions associated with ageing.

26.14 Summary

Adults in prison experience many forms of disadvantage which impact negatively on their health. Many conditions have negative consequences for speech, language and communication functioning, but these difficulties are rarely identified in adults in prison, despite effective communication with both staff and other prisoners being recognised as a positive coping factor for prisoners. Older prisoners are the fastest-growing sector of the prison population in the western world. There is increasing recognition of the problems of caring for older people in a prison environment, with access to healthcare being a further issue. It is important that more research is conducted into the speech, language and communication needs of adult prisoners and how these impact on access to both healthcare and to provision aimed at preventing re-offending. Research is also needed to understand how best to support prison staff in managing people with a range of communication difficulties.

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