

Literacy Studies: Perspectives from Cognitive Neurosciences,
Linguistics, Psychology and Education

Thomas Lachmann · Tina Weis *Editors*

Reading and Dyslexia

From Basic Functions to Higher Order
Cognition

 Springer

Literacy Studies

Perspectives from Cognitive Neurosciences, Linguistics,
Psychology and Education

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While language defines humanity, literacy defines civilization. Understandably, illiteracy or difficulties in acquiring literacy skills have become a major concern of our technological society. A conservative estimate of the prevalence of literacy problems would put the figure at more than a billion people in the world. Because of the seriousness of the problem, research in literacy acquisition and its breakdown is pursued with enormous vigor and persistence by experts from diverse backgrounds such as cognitive psychology, neuroscience, linguistics and education. This, of course, has resulted in a plethora of data, and consequently it has become difficult to integrate this abundance of information into a coherent body because of the artificial barriers that exist among different professional specialties.

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Chapter 1

Introduction to Reading and Dyslexia



Thomas Lachmann

Abstract The objective of this chapter is to introduce the topic of Reading and Dyslexia. A more complete understanding of the processes behind literacy acquisition, of skilled reading and of the underlying neural mechanisms, is required to fully understand dyslexia. The process of literacy acquisition is described as a form of procedural learning. During this learning process, pre-existing functions from different domains are recruited and coordinated, which are relevant but not specific for literacy acquisition. Further, it is argued that there is not merely one cause for dyslexia. Instead, everything that disturbs the fine-tuned coordination between the functions involved in learning to read and write is a potential cause, which is not restricted to a certain domain.

Keywords Literacy acquisition · Definition of dyslexia · Diagnosis · Brain development · Automatization · Learning disability · Phonological processing · Visual processing · Temporal processing · Procedural learning · Functional coordination · Training and remediation

1.1 Literacy Acquisition and Skilled Reading in Different Orthographies

To most of us reading and writing are everyday activities, secondary to spoken language. Usually, we do not estimate these activities as something extraordinary. However, reading and writing are in fact extremely complex skills, involving reams of cognitive functions, none of which is specific for literacy. Reading and writing are

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human cultural inventions. Therefore, reading and writing skills have to be taught and learned, and thus, successful literacy acquisition depends on the interaction of both the learning environment and individual factors. The learning environment encompasses macro- and meso-level factors of the society, such as teaching methods in institutions and families, orthographic systems, culture, and politics (Hung, Frost, & Pugh 2018; Joshi 2018; Morais 2018; Wimmer & Ludersdorfer 2018). The individual factors include general learning ability and motivation, individual brain development, possible specific processing deficits, etc. (Hung et al. 2018; Wimmer & Ludersdorfer 2018, see Part II). Yet, literacy functions to change both the society and the individual's mind in a feedback process (Morais 2018).

After several years of schooling, preexisting functions (including basic visual and auditory abilities, phonological processing and speech perception, oculomotor control, attentional and executive functions, short- and long-term memory, higher order language processing, and many others) become coordinated to create literacy specific cognitive procedures. These become automatized after intensive training (Lachmann 2018; Nicolson & Fawcett 2018). The process of automatization is accompanied by structural and functional changes in the brain when neural reading networks become functionally specialized (Hung et al. 2018; Wimmer & Ludersdorfer 2018). Thus, reading and writing are undoubtedly, truly extraordinary achievements of the human brain.

Part I of the present book addresses different aspects of literacy acquisition and skilled reading. Chapter 2 focuses on the impact of socio-cultural-political factors on literacy development and the feedback process of literacy on the individual and society (Morais 2018). Chapters 3 and 4 focus on brain structures and functions involved in literacy acquisition (such as the Visual Word Form area; Hung et al. 2018; Wimmer & Ludersdorfer 2018), and Chap. 5 outlines the relationship between linguistic comprehension and reading comprehension, reviewing evidence from monolinguals, second-language learners, and various orthographies (Joshi 2018).

1.2 Developmental Dyslexia: From Basic Functions to Higher Order Cognition

Even though literacy acquisition is quite a challenge for the brain, with the appropriate learning conditions and instructions almost all individuals will eventually become fluent in reading and writing, no matter which orthographic system is involved. However, some individuals have serious problems to acquire these skills adequately. If these problems are restricted to literacy development (i.e., no general cognitive deficit) and are not justified by insufficient learning conditions, then this specific learning disability is defined as developmental dyslexia. In Part II of the present book, a number of renowned theories on developmental dyslexia are introduced and discussed.

After more than a century of intensive research on this topic, there is still a debate about the cause of dyslexia (see Nicolson & Fawcett 2018, for a review). For this reason, most definitions (including the clinical definition from the World Health Organization) are still descriptive. These definitions require that for the diagnosis of dyslexia, there should be a significant discrepancy between individual performance scores from standardized reading and spelling tests when compared to (a) those of a reference population, and (b) indicators of the individual's general cognitive development (usually IQ). In other words, although there is common consensus that dyslexia is a neurodevelopmental disorder, when it comes to the diagnostic criteria in practice, it is currently still all about statistics rather than etiology. It means that the prevalence rate is preassigned and whether or not a person will be diagnosed as dyslexic depends on the distribution of certain test scores in a specified reference population. Since such statistical data can only be considered meaningful when some proficiency has been acquired, a diagnosis cannot be given before the middle or end of Grade 2. As a result, intervention usually starts later than it should (Lachmann 2018).

Is it really the case that we have that little knowledge about the cause of dyslexia? At the outset of systematic research on dyslexia, the focus was primarily on visual deficits. However, since the late 1970s overwhelming evidence was collected on the essential role of phonological processing for literacy development (see Nicolson & Fawcett 2018, for a short review). Specifically, phonological awareness was identified as a strong early predictor for subsequent reading and spelling performance. Yet, phonological awareness deficits do not always lead to problems in performance and not all dyslexics display such deficits before literacy acquisition. Nonetheless, the majority of children with developmental dyslexia show deficits in overall phonological processing. Hence, deficit in phonological processing was used to generalize the universal cause for dyslexia. However, phonological processing abilities do not only function as an important prerequisite for learning to read and spell, but are also enhanced and further developed when these skills are acquired. This means that the quality and quantity of reading experience has an impact on phonological processing abilities. Consequently, it cannot be said to what extent phonological processing deficits are the cause for or the effect of dyslexia. Nevertheless, the importance of phonological processing abilities for successful literacy acquisition is irrefutable even if the impact is considered as bidirectional. If phonological processing deficits are considered to be causal, then they still reflect symptoms at the cognitive level, which are secondary to information processing deficits at a more basic level (Calcus, Hoonhorst, Colin, Deltenre, & Kolinsky 2018; Galaburda 2018; Jaffe-Dax, Daikhin, & Ahissar 2018; Lallier et al. 2018; Nicolson & Fawcett 2018; Witton & Talcott 2018). These basic deficits may be the result of anomalies in brain development (Galaburda 2018; Jaffe-Dax et al. 2018; Lallier et al. 2018; Stein 2018; Witton & Talcott 2018). Importantly, anomalies in brain development may not only cause problems in basic auditory processing but also in the visual domain (Lachmann 2018; Stein 2018), in cross-modal integration (Lachmann 2018), in general temporal processing (Galaburda 2018; Jaffe-Dax et al. 2018; Stein 2018), or in automatization (Nicolson & Fawcett 2018). These may then

lead to secondary symptoms at the cognitive level (e.g., speech perception deficits; see Calcutt et al. 2018), which are not restricted to phonological processing (see Nicolson & Fawcett 2018, for a review).

If anything can be concluded from the immense body of research findings collected over the past decades, it would be that there is not merely one cause of dyslexia (see Burgess, Witton, Shapiro, & Talcott 2018). In fact everything that disturbs the fine-tuned coordination between the functions involved in learning to read and write is a potential cause (Lachmann 2018), which is not limited to a certain domain. Therefore, understanding dyslexia requires a better understanding of the processes behind learning to read and write (Lachmann 2018; Nicolson & Fawcett 2018), of skilled reading, and of the underlying neural mechanisms (see chapters of Part I).

The Functional Coordination approach (Lachmann 2018) describes literacy acquisition as a form of procedural learning (Nicolson & Fawcett 2018). During this learning process, preexisting functions from different domains are recruited, which are relevant but not specifically designed for the task of literacy acquisition. By guided instruction, these functions are then optimized for the task and subsequently coordinated to create specific procedures for reading and writing. The coordination requires fine-tuning of complex functions to warrant cross-modal integration. This leads to a novel synthesis of functions that finally become automatized, as a package, over several years of intensive practice (Nicolson & Fawcett 2018). Consequently, developmental dyslexia is described as a Functional Coordination Deficit (Lachmann 2018), since a failure in coordination is most liable to manifest deficiencies in alphabetic and orthographic coding and decoding. This means, developmental dyslexia is actually not a consequence of a deficient automatization per se but of the automatization of suboptimal functional coordination. Various anomalies in brain development and the ensuing basic and secondary cognitive deficits can hamper this functional coordination. Part II of the present book delves into this subject.

1.3 Training and Remediation for Children with Dyslexia

A remediation program must be based on theoretical and causal assumptions and empirical findings. It was argued that developmental dyslexia can be caused by different anomalies in brain development (Galaburda 2018; Nicolson & Fawcett 2018; Stein 2018) that result in basic and secondary cognitive deficits. However, these deficits are not restricted to a certain domain (auditory, visual, cross-modal, executive, etc.; see chapters of Part II). At the behavioral level, these deficits all have the potential to disturb the process of learning to read and write. Therefore, it is impractical to create a training program that is restricted to solely one isolated function on the basic or cognitive level. Instead, it is more useful to train the coordination of functions (Lachmann 2018). This would involve implementing a multifunctional, cross-modal, hierarchical, adaptive program that is individualized

for each participant (Klatte, Bergström, Steinbrink, Konerding, & Lachmann 2018). Such a training program should be administered at the earliest opportunity with respect to the process of literacy acquisition and not necessarily the age of the participant. However, the design implemented should be age specific, taking into consideration motivation, feedback and cognitive development.

Furthermore, any remediation program implemented should be evaluated using a longitudinal control group design (see Klatte et al. 2018; Tallal & Jenkins 2018). This evaluation should not simply test the effect on the functions and sub-skill(s) that were trained in the program (e.g., phonological awareness) but rather on how it transfers to reading and writing performance in the long run.

However, it cannot be expected that any established program would impact all dyslexic individuals to the same extent, given the multi-causality assumption of dyslexia. On the other hand, the differential effects of a training program, i.e., the response to intervention of an individual or a group, do also provide insight to the causal factors of their reading and writing problems. If a substantial number of participants in a remediation program demonstrate no (or only a short-term) transfer to the behavioral level, then it is rather unlikely that the trained function(s) and sub-skills would play a major role for the learning process in the general population.

Hence, Part III of the book presents two remediation programs, which provide data from evaluation studies. These were developed for two different orthographies. *Fast ForWord*[®] was developed for nontransparent English (Tallal & Jenkins 2018) while *Lautarium* was developed for the German phonetic system and a transparent orthography (Klatte et al. 2018).

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Part I
Literacy Acquisition and Skilled Reading
in Different Orthographies

Chapter 2

The Methods Issue Revisited: From a Developmental and a Socio-Cultural-Political Perspective



José Morais

Abstract This chapter aims at discussing several aspects of the “methods issue” rather than at offering a systematic review of the specialized literature. It will: (1) distinguish, in terms of cognitive processing, between the notions of learning to read and write and of literacy development; (2) consider learning to read and write in the context of previous and subsequent developmental stages; (3) recall the history of the methods used to teach reading and writing abilities, in connection with social and cultural contexts; (4) contemplate the training of parents and teachers for, respectively, educating their children for literacy, and teaching their pupils to read and write.

Keywords Literacy · Illiteracy · Levels of literacy · Data on world literacy · Learning to read processes · History of teaching-to-read methods · Phonics and whole-language methods · Teaching illiterate adults · Literacy in socio-political context · Literacy and democracy

2.1 Introduction

At the last Society for the Scientific Study of Reading (SSSR) meeting held in Porto, Portugal (June 13–16, 2016), no oral or written presentation targeted the learning to read methods issue mainly and explicitly. This might indicate that the issue is over, or that the people involved, for one reason or another in this issue, are tired of the aggressive polemics that for so many decades have shaken them. However, the issue is not over; it is over from the theoretical scientific perspective, but not entirely over from the perspective of applied research. It is most certainly not over at all from the perspective of actual pedagogical practices.

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After analyzing the concept of literacy and offering a portrait of the situation of literacy in the world (Sect. 2.2), and after restating the theoretical psycholinguistic ground as synthetically and as clearly as possible (Sect. 2.3), this chapter deals in Sect. 2.4 with the learning to read methods, and attempts to draw a historical and political account. In Sect. 2.5, it addresses parents' and teachers' preparation.

2.2 Learning to Read/Write, and Literacy Development

“Learning to read and write” is an underspecified expression, in much the same way as “learning to play” is underspecified. Indeed, learning to play football is very different from learning to play tennis, and both very different from learning to play chess, and chess from poker, and these from playing the piano. The game or ability at issue must be specified. Likewise, learning to read and write depends on the writing system. Although there are very general traits common to, and similar constraints on learning whatever the writing system, learning to read and write in an alphabet does not help much in learning an ideographic or a morphographic system, or even a syllabic one. As it would be impractical to specify the writing system each time I speak of learning to read and write, I invite the reader to take into account that here I am only concerned with the alphabetic writing system, more specifically the Latin alphabet.

Learning to read and write must be specified on a further dimension. To read is to transform written words into spoken ones (more exactly, to find for the former(s) the correspondent(s) among the latter), and to write is the reverse. In principle, it is possible to acquire these abilities without knowing the oral language that the written words represents, more precisely without knowing the meaning of those spoken words. In a classical example, Milton's daughters learned to read in languages they were unable to understand (in both their written and oral form). Obviously, except in such an unusual situation and in most of the present print-verbal transformation devices, one reads (writes) to access (code) meaning from (into) print. Yet, the precise definition of reading and writing (or spelling) does not imply understanding nor intentional meaning communication under a written form.

It is this precise definition of reading and writing that creates the necessity of distinguishing the ability to read and write from literacy. Literacy was introduced in English from Latin via Middle Age French, and became the usual word for referring to the ability to read and write. Although it is a recent word (it appeared in the last quarter of the nineteenth century), some non-English-speaking countries or provinces have imported it. This is the case in Portugal (but not in Brazil), Spain and Québec (more recently, also in the French-speaking cantons of Switzerland, and Belgium). Official texts still resist adopting “littératie,” partly because it is wrongly believed to be an Anglicism, and partly because “lettré,” which corresponds to literate, is not merely a person able to read and write but an erudite or someone who possesses culture and knowledge.

I proposed (Morais 2016) to use, in French, the word “*alphabétisé*” to designate a person who is able to read and write in the alphabetic system in the strict sense considered above; and to use “*lettré*,” to designate someone who reads and writes skillfully and uses productively her/his ability of reading and writing. The idea of productive use implies that these abilities are purposively employed for operating on meaning. Below, I distinguish between terms such as illiterate (“*illettré*”), functionally illiterate, and literate, including between types of literacy.

Illiterate, according to the UN and UNESCO (1978), is the individual of at least 15 years of age, who is unable to read and write a single short sentence characteristic of or consistent with her/his daily life. This is more demanding than the criterion used in the Modern Age of being unable to put a signature, other than a cross, in a contract (typically the contract of marriage). This is clearly insufficient to enable an individual to take a significant part in the social, economic, political and cultural life of the modern world (although there are exceptions, such as the case of one member of the present Brazilian parliament who, although being completely illiterate, voted on important matters, including the presidential impeachment). By definition, all those who are not illiterates in that sense should be considered literates, but the term of functional illiterate designates those who have learned to read and write to some extent at school but did not practice enough and suffer tremendous difficulties. There are no criteria for establishing functional illiteracy and there is no systematic evaluation of these people. Non-governmental associations and foundations have reported there are two million and a half people aged from 18 to 65 years who had been schooled in France (ANLCI 2013) and around seven million adults in the UK (World Literacy Foundation 2015, p. 12).

In Morais (2017), I called *productive literate* the person who can read and write in a predominantly automatic way all words (isolated or in text) that are consistent with her or his level of spoken language and knowledge. In the case of alphabetic writing, at least, automaticity in reading and writing refers to the immediate and direct access, through complex but non-conscious processing, to the phonological and orthographic word forms, respectively. The person is called productive literate to the extent that (s)he uses those abilities in a productive way, i.e. to acquire knowledge (through reading) and to communicate it (through writing). Most of these literate people read frequently and skillfully, but write only occasionally, thus they are only partially productive. We do not actually know to what extent and how long skillful reading and writing can be maintained as a result of scarcity or absence of practice.

I called *critical/argumentative literate* the person who uses reading in a critical perspective, organizes his/her thoughts about the matter in an argumentative way, and uses writing to communicate such critical evaluations and arguments. This is crucial for not only personal involvement but also cooperative participation in the democratization of institutions and communities, and for allowing socio-cultural changes and the progress of knowledge. People who go further the critical/argumentative stance and use reading and writing in a creative or innovative may be also called *creative literates* (it is the case in principle of scientific researchers). Orthogonally to all these three literacy types, it is usual to consider domains of literacy, for example, in addition to scientific literacy, philosophical literacy, literary literacy, media literacy, etc.

How are those types of literacy distributed among the population? Concerning adults (from 25 to 65 years old), we may refer to the OECD (2016b)'s analysis. For literacy, and like the PISA's studies on adolescents, considered below, this study on adults only examined reading comprehension. It distinguished six levels of proficiency, including below level 1, this level requiring the person "to read brief texts on familiar topics to locate a single piece of information (...) identical in form". On the average over the 34 countries involved, almost all developed ones, 4.5% of the people scored below level 1; and 14.4% reached level 1, which requires "to locate information that is identical to or synonymous with the information given in the question or directive", "simple cycling through more than one piece of information", and recognition of "basic vocabulary". It thus appears that roughly one among five adults displays very rudimentary reading ability. In the absence of tests on word reading and spelling abilities, it is difficult to match the OECD's levels scale with the notion proposed above of productive literacy. Tentatively, it might be reasonable to match it with level 3, which requires to derive "meaning across larger chunks of text": 35.4% would thus reach the status of productive literate without going beyond it. Finally, both level 4, which involves evaluation of persuasive discourse, and a fortiori level 5, which requires evaluation of logical and conceptual models of ideas, seem appropriate to the criteria of critical/argumentative literate: they were reached, averaged over countries, by 10% and 0.7% of the people. On the whole, and noting again that these features are not estimated on a person basis independent of country, slightly less than 50% of the adults would be literate, of whom only slightly more than 10% would be critical literates.

The PISA data for 2015 (OECD 2016a) about schooled 15-year old adolescents in 69 countries can serve as predictor for adult literacy within a few decades. The approach is basically the same as for the adults: only reading comprehension was tested, although eight levels were distinguished (level 1 was subdivided into 1a and 1b, and 6 was the upper level). According to a longitudinal inquiry, further schooling and professional study would be seriously compromised if the adolescent fell below level 2, which was the case for 20% of them (still averaging over countries). Level 3, requiring the reader "to locate, and in some cases recognize the relationship between, several pieces of information (...), integrate several parts of a text in order to identify a main idea, understand a relationship or construe the meaning of a word or phrase", might indicate productive literacy, although probably overestimated: it was attributed to 28%. For Level 4, it was necessary to interpret "the meaning of nuances of language" and "hypothesize about or critically evaluate a text"; however, the tasks only required a modest critical capacity: for example, in one task, confronted with two letters, the participant was expected to indicate the one that included a discussion rather than mere statements. It was attributed to 20%. Finally, excellence (levels 5 and 6) was reached by only 8%. Thus, 28% may be an overestimation of critical/argumentative literacy.

Obviously, the differences between countries are huge. Among the European countries, the highest mean score was obtained by Ireland (only 10% schooled adolescents below level 2, and 70% presumed to be productive literates, among whom 37% would be critical literates), and the lowest by Kosovo (78% of poor

readers, and 4%, presumed productive readers, including very few critical ones). Out of Europe and OECD, Brazil, despite being a big and rich country, presented 52% poor readers, and only 24% presumed productive literates, of whom only 9% would be critical readers. Thus, given this oligoliteracy (Goody & Watt 1963), i.e. limitation of the literacy capacity to a small proportion of the 15-year-old adolescent population, Brazil risks to remain governed by an oligarchy for many years, unless an educational revolution takes place.

To understand better what literacy acquisition involves in terms of reading processes, it is necessary to consider their normal development, assuming that teaching to read and write respects the main contributions of scientific research in this domain.

2.3 The Developmental Conditions of Learning to Read and Write in the Alphabetic System

For many generations through three millenarities, many individuals, mostly children, have been taught, and learned, to read (less so to write), this with very little knowledge or reflective and analytic awareness of what they were doing. Since many different ways of teaching were used (see Sect. 2.4), they also learned in different ways with more or less success, needing more or less time, and developing different degrees of awareness of how alphabetic writing represents language. Probably, most of them only experienced a very tiny intuition of the alphabetic principle.

This large variety of learning roads may give the impression, shared by many people, that one learns to read whatever the method, and that methods do not matter too much. However, this is not true because we must distinguish word reading and object recognition. Word reading requires developing a specific neural basis. The perceptual system that learners (children or adult) develop to read words is not the same as the one they use to recognize objects. In the latter case, they do not acquire a new system; they only use a preexistent system on new exemplars, with great memory limitations. Thus, the fact that there may be many learning roads does not imply they lead to the same outcome, nor to the same proficiency.

Furthermore, we are aware neither of our reading processes nor, to a great extent, of what we do to acquire them. The idea that the letters of the alphabet, in a more or less transparent way due to the consistencies or inconsistencies of the orthographic code (which makes necessary the notion of grapheme), represent phonemes is a recent one. In the classic Huey (1908)'s book, "The psychology and pedagogy of reading," letters represent sounds, or phones. The author also uses "phonetic," the quality of phones, and "phonic," a learning to read method, but neither "phonology," "phonological," nor "phoneme". Indeed, the notion of phoneme already proposed by a linguist, Baudouin de Courtenay, in the last quarter of the nineteenth century, remained for a long time in the linguistic domain, and its full admission only occurred in the 1940s when Troubetsky and Jakobson clearly distinguished between phonetics and phonology.

The fact that the phoneme has no acoustic invariance, and therefore is neither a sound nor a perceptual “unit,” and that it actually corresponds to a dynamic relation between the speech articulators, was introduced only in the 1960s by Alvin Liberman and the Haskins group (A. M. Liberman, Cooper, Shankweiler, & Studdert-Kennedy 1967). This work immediately impacted the way of conceiving reading and learning to read. This was expressed in many chapters of two influential books, edited respectively by Kavanagh and Mattingly (1972) and by Reber and Scarborough (1977). At the same time, the development during reading acquisition of phonological and, as a special case, phoneme awareness, was theorized by Isabelle Liberman (I. Y. Liberman 1973) and became the object of experimental investigation (I. Y. Liberman, Shankweiler, Fischer, & Carter 1974).

Below I briefly describe what I have called the developmental *conditions* of learning to read and write in the alphabetic system (Morais 1994, in French; 1996 and 1998, for respectively the Brazilian Portuguese and Spanish editions). There are no successive stages or phases in learning to read. Instead, three specific acquisitions must take place, each being a condition for the subsequent, but developing in large part simultaneously, in reciprocal interaction. This progression is valid whatever the learning method, but can be much more facilitated by some methods than by others. With the less efficient methods, the learner also eventually grasped the alphabetic principle, became able to decode and, hopefully, to automatically address lexical orthographic representations, but these skills are intermingled with non-specific ways of recognizing written words (cf., for a different but not incompatible perspective, Lachmann & Weis 2018).

Acquiring at least some intuition of the alphabetic principle, i.e. that letters (more exactly, graphemes) correspond to phonemes, is the first condition to read words and any sequence of letters that could potentially be a word (being able to read new, unknown words is a necessary attribute of being a reader). It is acquired by being presented with alphabetic strings and led to mentally represent the corresponding phonemes and how these can blend. Blending phonemes is not blending sounds, so that to abstract the phoneme from a sound it is necessary to combine two kinds of abilities, perceptual (speech sound recognition, for example /fa/) and inferential (abstracting /f/ by being visually exposed to syllables like “fa”, “fi” and others sharing the letter “f”). This is facilitated by playing initially with sounds in which the phoneme-target is more apparent in the sound and in its articulation. Phoneme awareness is not phonological awareness, although it may be considered as a special case of phonological awareness (but for practical purposes, one would better treat them as distinct). Phoneme awareness is not an instantaneous acquisition, although there is probably some initial insight. It develops with decoding until the learner uses it in an efficient way. For that reason, tasks evaluating phoneme awareness (not used as such in reading) are the main predictors of individual differences in decoding skill, and reciprocally, but only for some time (mostly in the first year). Later, as for many other skills, it ceases to be necessary for reading, and becomes an important matter only for psycholinguists and (it should, at least) learning-to-read teachers. Most skilled adult readers may show, in appropriate tasks, that they remain aware of phonemes, but they find extremely difficult, and usually are unable, to explain correctly what it is.

Developing decoding skill is the second condition. It is highly dependent on phoneme awareness and skills at the beginning and this dependency decreases as decoding becomes based on larger and more complex structures (complex onsets, rhymes, syllables, morpheme-related phonograms . . .). Decoding is conscious, intentional, controlled, but it is not the only mechanism involved in sequential reading. There is also an implicit learning, based on frequency, of how parts of words tend to be spelled and pronounced. This learning of statistical regularities can be observed quite early in the first year (e.g., Martinet, Valdois, & Fayol 2004). It coexists with, and may occasionally dispense, decoding (see Hung, Frost, & Pugh 2018).

As these two mechanisms become dominant and involve larger word structures, they begin to be overcome by the automatic access to stored representations of the words in a specialized long-term memory. This is the third, final and decisive acquisition. It is the repeated successful decoding, and possibly also the use of implicit knowledge of spelling-sound word parts, that allows this storage (Share 2004). However, more exactly, its acquisition does not finish with learning to read; one might even say that learning to read never finishes, as, by being exposed to new written words (in our language or in others we may learn in adult age), we continue to acquire orthographic representations. In my view, it seems worth saying that learning to read is completed when the mechanisms of skilled reading (automatic access to stored word representations) are in place.

Plausibly, this lexical long-term storage may impoverish, as a consequence of either a long lack of exposure to some words or, more generally, of aging. Additionally, such a lexical store is never fixed, as it must increase with the diversification of reading. Many theoretical questions can be raised, and are addressed experimentally, concerning this store and the access to it. They concern two main issues: one is how it is structured: the other, what types of information are represented in it. This mental lexicon must be at least orthographic, i.e., abstract, not visual even if dependent on vision in sighted people (or on the tactile sense in the blind). But it may be also both orthographic and phonological or, alternatively, activation of the stored word orthography immediately propagates to a corresponding phonological store. Finally, to what extent and how semantic information is automatically accessed is an important question, about which there still is little knowledge.

2.4 A Historical and Sociocultural Account of Learning to Read and Write Methods

Huey (1908) offered, in his Chapter XIII – The history of reading methods and texts, a detailed account of how reading and writing had been taught in the alphabetic system since the Greeks and Romans until his time. His first remark is important: “Among the early peoples who used an alphabet each letter was used for a definite purpose,” which was, according to him and certainly to those peoples, “to represent a definite sound.” We know that each letter, even in a one-to-one correspondence, does not represent a sound but a phoneme, and it requires an additional mental effort

for converting a sequence of sounds into the correct word. An imperfect but still interesting analogy in the strict speech domain is the name of Brazil, pronounced “Braziu” by the Brazilians but “Burajiru” by the Japanese. After hearing other examples of Japanese conversion of an Indo-European language, it would not be difficult to overcome the conversion and understand the words. If I read cat as “keate” and rat as “reate,” I will understand that “beate” should be bat (A. M. Liberman 1995, p.24). Something similar may have happened with the Greek and the Roman children. Huey was right saying that univocal letter-sound correspondence “made the letters of much greater importance at present” (he was probably thinking of English, not of Finnish). As it is well known (cf. Seymour, Aro, & Erskine 2003), it makes a great difference to learn to read English, French and European Portuguese than to read Spanish, German or Italian. We should therefore not be surprised, or feel pity for the Greek and Roman children upon hearing that “the ABC method of learning to read became general (. . .) and persisted to recent times in the Western world” (. . .) The Greeks and Romans, in teaching the child letters, taught their combination into syllables and words, and then of words into sentences” (p. 240).

The implementation of the method was a matter of imagination, good will or prepotency. Huey mentioned that one Greek “purchased twenty-four slaves as playmates for his stupid boy, giving to each the name of a letter” and that Quintilian “advised giving the young child blocks and tablets containing the letters (. . .) and that he should be allowed to trace with a pen the forms of the letters as engraved on ivory tablets.” In modern times, imagination is more appropriate than mere prepotency, and for English the gingerbread method was invented: “To Master John the English maid/A horn book gives of gingerbread,/And that the child may learn the better, As he can name he eats the letter.” One of its enthusiastic advocates was Basedow (1723–1770), who proposed to give the school a baker to prepare gingerbread each morning and assured the cost would be modest, as “it is not necessary for any child to eat the alphabet more than three weeks.”

In the Middle and the Modern Ages, reading teaching was mostly organized by the Church, whatever the confession, or directly influenced by it, and was associated with religious instruction. In the Abecedarian of the nineteenth century there was the Credo and the Paternoster, later the Ave Maria, etc. The Reform did not change this. “The German word for primer, Fibel, appeared in 1419, and signifies a little Bible” (Huey 1908, p. 269). To America the Puritans brought an ABC Catechism, which was only replaced by the “New England Primer” in ~1690. One says of this little book, present in every home (three million copies were sold), that it accompanied John Adams through his life¹. It was a Church book, containing the alphabet, lists of syllables such as ab, ib, ib, etc., and of words with increasing number of syllables, rhymes, moral injunctions, prayers, etc.

¹John Adams, third president of the USA, wrote that direct democracy, i.e. by the people, is arbitrary, tyrannical, cruel, that the people cannot judge, act or think, and that the destiny of the poor is the work, whereas the rich are qualified for the superior functions given their education, independence and leisure.

The spelling-books, necessary for English, appeared in the middle of the eighteenth century. In USA, the most famous, the Webster's Spelling Book, written by Noah Webster,² was first published in 1783. Used in all the country, five million copies had been sold until 1818, and 47 million until 1847; in 1900 it was still sold at the annual rate of hundred of thousands. Huey estimated that the Webster's Book was "artificial in its arrangement of words, thought, and vocabulary, most ill-adapted to the needs of its users and to the various ages of the children." Yet, he recognized that, through its universal use, it contributed to the homogeneity of language and of pronunciation across the USA. It most probably helped to avoid the situation of Brazil, where Portuguese is highly diversified within the country and very different from the European Portuguese in terms of lexicon, syntax, pronunciation, and, to a smaller extent, orthography. Regarding the learning method, the book did not contain anything particularly relevant or new.

The first American primer to advocate the whole-word method for reading was probably the Worcester's Primer of 1828. According to its author, the child "may learn first to read words by seeing them, hearing them pronounced, and having their meanings illustrated; and afterward (. . .) to analyze them or name the letters of which they are composed." In the USA, the whole-word method was more clearly presented in the 1840s, however it was still claimed in the "Word Builder" of 1860 to be new and original. It is only by 1870 that the whole-word method "began to be adopted by progressive teachers in various parts of the country," (Huey 1908, p. 259) and the literature only entered reading-books by ~1880.

Huey noted that the alphabet method was almost universally used until ~1880 and had not been discarded yet by the time of his book. This was in contrast to Europe, in particular Germany, where imitative pictures of the sounds of the letters appeared as early as 1534, for example *r* or *m* placed near, respectively, the picture of a dog and of a cow. Thus, the idea was to associate the visual form of the letter to the "sound" of its name. As Huey wrote: "Germany much earlier than America began to realize that spelling was not the only or the best approach to reading, but the spelling method held its ground there until well into the nineteenth century" (p. 256). Perhaps the spelling inconsistency of English has masked for a longer time the pertinence of teaching the alphabet phonetically. As Huey writes, the whole-word method also appeared earlier in Europe. It could have been the case of the Comenius' book of 1657 or 1658, which, translated into ten European (plus four Asiatic) languages remained the most popular textbook in Europe for more than one hundred years. It seems, anyway, that the book was little used as a method.

²Noah Webster, who, before Lincoln, defined democracy as the government of the people for and by the people – although later on he considered people, equality, and democracy as "metaphysical abstractions" –, was formerly a free mind, abolitionist, federalist, who eventually converted to Calvinism, wanted language to serve and to fear God necessary to social order, and wrote his own version of the Bible. Editor and journalist, he is called the "father of American education and schooling." Indeed, he also wrote dictionaries, attempted to normalize the pronunciation of English words, highly diversified by the many regional dialects, and reformed the orthography (colour becoming color, centre center, etc.).

According to Huey, in Europe the ABC practice remained “until Jacotot (1770–1840) advocated the word-method as a part of his system, and set forth clearly the arguments for it” (p. 285). This is not entirely exact for two reasons. First, it was the teacher Nicolas Adam, who used his own whole-word method and proposed it in a book published in 1787. Second, there was *stricto sensu* no Jacotot’s method. However, he had a much greater influence than Adam, and it is justified to dedicate here some comments to his life and ideas.

Joseph Jacotot proposed, more generally, that learning should proceed in an analytic way, which does not necessarily imply a whole-word method. The authors of several learning books claimed to have been inspired by Jacotot’s system, but among them there was also, for example, an ABC-book, by M. Rousset. Anyway, as Jacotot was a humanist whose explicit aim was to contribute to the emancipation of the people, his presumed involvement in the whole-word method for learning to read may help understand the association of this method with humanism. As a young man, he had a main role in the organization of several revolutionary federations. A doctor in humanities, law and mathematics, he became professor of literature at the Catholic University of Louvain where he conceived a system of “universal teaching” (as, for him, all intelligences were equal) based on the idea that everyone, child or adult, is capable of self-instruction (its main principles being repetition, immediate application of everything that is learned, and research of relations with everything else), leaving to the teacher the task of orienting and supporting the student’s attention. This would be valid for all domains, scientific, literary and artistic. Recently, referring to Jacotot’s system, the Marxian philosopher Jacques Ranci re criticized the “myth of pedagogy,” i.e. that the teacher’s explanations are crucial. Later, I will come back to this idea.

The alphabetic method began to face two main competitors at more or less the same time (since the seventeenth century), namely the whole-word method and, somewhat earlier, the phonic method. Based on the myth that the child can embrace the world and give order and meaning to it, the whole-word method led subsequently to the sentence and text methods, i.e. to the whole-language method. This is also presented as approach or philosophy, rather than method, as, indeed, it is very difficult to specify its procedures and to evaluate them through experimental testing. Concerning the phonic method, it started with the Jansenists from Port-Royal, who opened small schools where the teachers had classes for five or six children and presented the consonants followed by a neutral “e” so that “be” and “a” would make “ba.” The difference between the alphabetic and this phonic method is tiny for the plosives, but the difference may help. Huey wrote that, in the USA, this phonic method was introduced in several regions in the nineteenth century, apparently with success. Note that the Jansenists’ phonics was still based on sound and was overtly synthetic, as it was based on making a syllable from two sounds. Thus, it is completely different from what we call phonics today, based on mentally abstracting from the consonant something that we call phoneme and synthesizing it with the vowel.

In the old phonic method, the children combine sounds given by the teacher. It is the teacher who changes the sound of the official letter name and gives this

new sound to the children, hoping that it will be more accessible for their correct reading of the CV syllable. Today, many teachers still use these phonics. However, the present phonics teacher, even when they use the more accessible sound names of the consonants, does not explain that be+a makes /ba/. The teacher offers the children different combinations of C and V, with identical or with different C or V, while pronouncing each syllable, and thus just shows the reality, but an arranged reality, and it is the children who analyze the material, compare what is the same and what is different in the sound and visual form of the syllables, and from this mental analysis extract an intuition that enables them to read. This is their intuition of the phoneme, without knowing what is the phoneme, and perhaps without immediately giving it a sound as we do. This process is thus the exact opposite of the Jansenist's phonic: let the children do it! But, before, show them the written language as an enigma and let them find the answer! They will!

In the USA, from the old version of the phonic method developed quite soon what has been called the "phonetic" method. Its most well-known version was the "Pronouncing Orthography," published by Edwin Leigh in 1864. "In this system the letters were given various special forms to represent their different sounds, these forms being slight modifications of the ordinary form. Silent letters were printed, but in hairlines" (Huey 1908, p. 260). The reason justifying these letter manipulations was the larger number of sounds (44) than letters (26). Indeed, due to the numerous inconsistencies of English orthography, even adult readers of English find it difficult to segment speech phonemically. Adolescents 16 to 18 years old do it worse than young readers from grades two to four (Calfee, Lindamood, & Lindamood 1973), undergraduates in linguistics are at pains to segment three and four phoneme words correctly (Scholes 1993), and the performance of psychology students still remains far from perfect after a short instruction on graphophonemic segmentation (Connelly 2002).

With Leigh's method, the children could immediately find the way the letters are pronounced in a particular word and read it correctly. However, according to Huey, the method did not survive, despite its initial great success, because it was hard on the eyes, caused confusion in reading, and made trouble for the printer and the scripter. Yet, there might be better ways of modifying the visual form of the letters according to their phonological role in the word context. In Morais (2016), I presented an illustration of such a code, which, created for French by a visual artist, Sarah Cleeremans, and named *Phono* by her, allows us to display for each written word both its usual spelling (as all the letters are present in some form and/or position) and its graphemes identified by joining the letters corresponding to a phoneme (see Fig. 2.1). This system, which might also be called a "Graphemic or graphophonemic alphabet" (the grapheme being actually defined by the phoneme), is not necessary for the skilled reader. However, as for the beginning reader of Hebrew, who can take benefit from diacritics to read correctly words and sentences, the beginning reader of French (or English) could learn to read more accurately and faster with this system than with the current presentation. It is not a problem of principle, but of finding the more appropriate design. *Phono* should be tested and, if necessary, improved.

Fig. 2.1 Pierre Burney in
“L’Orthographe”, 1970

La langue française
a un certain caractère
esthétique, puisque
les mots ne sont pas
seulement le calque
du son, mais présentent
une sorte de
physiognomie graphique
où les lettres
superflues font figure
de « signes particuliers »
ou « d'ornements ».

The learning to read and write methods issue is more than a technical one. It has been considered by the educational communities as basic for the definition of a literacy policy. In many countries, it even became a component of national politics. In the USA, in particular, “the transformation of literacy from an educational concern to a national political issue has been swift and significant”, being hotly debated in state legislatures (first in California, then in Texas under the initiative of Governor George W. Bush), Congress (which blocked President Clinton’s literacy legislation) and federal courts (Davenport & Jones 2005). According to these authors, President Bush, his “reading czar” Reid Lyon of the NIH, and the congressional Republicans “hammered out the historical No Child Left Behind legislation”, which was a “back to basics” or “old phonics” move (I call “old phonics” the teaching of grapheme-phoneme correspondences without appropriate activities to develop phoneme awareness and related metaphonological skills). As Marilyn Adams wrote in her report to the Congress, “the question of how best to teach beginning reading may be the most politicized topic in the field of education” (Adams 1994). In France and Brazil, similar debates also occurred and are still alive (cf. specific chapters, in Morais 2014, 2016, respectively).

In each of the three countries mentioned above and also in others, the confrontation is assumed as opposing a method (endorsed by the “skills” or phonics side) to an approach or philosophy (the whole-language). This partly accounts for their impermeability to the opponent’s argumentation. Actually, in my view, the opposition has also sociopolitical roots. The whole-language approach is related to a humanist and liberal conception of education typical of the intellectual elites belonging to the medium-high social class. Even though the phonics method is the most efficient for all children, the children’s precocious cognitive and linguistic development promoted in these families can compensate to some extent the lack of a

systematic skills approach at school, as at home the understanding of the alphabetic code is usually stimulated. On the contrary, the dominant neoliberal high class, whose main values are the competition for developing technological expertise and top administration competences, thus fearing humanism and the associated human rights, is happy with phonics. Ignored by this dispute, the children of the low-working classes are indeed best served if they are taught in school according to a phonics approach including both phonemic skills and systematic spelling teaching (as demonstrated by Roberts & Meiring 2006). In France it was recently shown that the advantage, measured on word reading fluency, word spelling, reading comprehension and written production, of the phonic method over the whole-word method is more than twice larger for the children whose none of the parents had finished secondary school than for those with at least one parent graduated at this level (Deauvieu, Reichstadt, & Terrail 2015).

First, literacy, including alphabetic literacy, is from its onset an instrument in the hands of the elite and exploited by the powerful to their advantage. Alphabetic writing was invented from other writings across successive changes and adaptations to the languages, without conscious knowledge of what this instrument involves in terms of mental capacities. The alphabetic or ABC method was the most superficial one could imagine: writing is made of letters, so letters and their names must be taught. The old phonic method, developed in Port-Royal, among people who were studying logic and language, was a progress, but did not go further into the mental mechanisms, because, for that, a much more comprehensive analysis of phonetic variations (which was done much later at the University of Kazan, in Russia) and an experimental investigation of speech perception and production (which is the enormous legacy of Alvin Liberman and his colleagues) would have been necessary.

Interestingly, the discovery of the phoneme posed more difficulties to science than the discovery of the atoms. Even discounting the fact that Greek philosophers had postulated the existence of the atom, soon after they invented the alphabet, the atom had been inferred by an English chemist, Joseph Dalton, in 1808, and the relations between atoms by another English scientist, this time physicist, Michael Faraday, in 1832. No English scientist discovered the phoneme in that century: certainly, all of them were frequent readers, but English orthography seems to have contributed to the concealment of the phoneme to their mind's ears. Eventually, by the end of the century, assailed from all sides, the atom began to display its internal constitution, and the (also British) physicist Joseph Thomson discovered one of its particles, the electron. In much the same way, after the conceptual formulation of the phoneme, it has been necessary to show how the modulations of the consonantal formant transitions, due to the action of the speech articulators, and depending on their vocalic context, correspond to particular phones perceived as categories through phonetically irrelevant variations and influenced by literacy itself. Penetrating the atoms of language as well as those of matter required a long analytic process. The fact is there: it has been more difficult to penetrate the atoms of language than the atoms of matter.

During all this time, the battlefield of learning-to-read methods was open for the confrontation of the apparently technical approach, the phonics, that require patience and effort, and the global approach that would reveal immediately both language and meaning (with all their promises). Today, the science of literacy has clearly backed the former approach of teaching reading and writing in the alphabetic system. Unfortunately, as described in detail by Morais (2014) but it was also the case in other countries, the defenders of the global approach have been strongly influencing the educational authorities of many countries, including by preventing the future teachers of reading and writing to be correctly informed about the teaching of the science of literacy.

In the title of this section I referred to methods of learning to read and write, but I limited its content to the description of learning to read. This is a consequence of the social discrimination against writing, even greater than the one against reading. It was believed that children should be able to know and recite the religious texts (Bible, Koran, etc.), but that teaching them to write would be foolish; it would give them a powerful instrument of action and intervention. Writing was therefore limited to the oligocrats and their courts. Writing is still socially discriminated against. The reasons why PISA assesses reading but not writing are unclear: it may be because expertise in writing is not judged as necessary as in reading, and/or because writing is more affected than reading by the complexity of the orthographic code, which largely differs across languages.

The scientific team in which I am working has recently elaborated a course of alphabetic literacy for Portuguese illiterate adults who are unable to read (or write) even a single word among the most frequent ones (for a phonic intervention on children, cf. Klatté, Bergström, Steinbrink, Konerding, & Lachmann 2018). After three months which allowed them to become accustomed to the sounds of words, and created situations for letting them acquire an intuition of the phoneme and go through the whole orthographic code (a semi-transparent one), six out of eight Romani women, whose social life is to sell goods, bear children and be governed by their husbands and who attend a social-religious center that helps them in many respects, could read more than 20% of words never studied or seen at our classes. The individual differences were very large, one of them reached almost 90% accuracy in reading, and the average was about 50% (Kolinsky, Leite, Carvalho, Franco, & Morais 2018). Obviously, the method was phonic and progressed from the more accessible and consistent correspondences to the more complex ones. A major characteristic of the method is that writing was trained and evaluated as much as reading (at the beginning, these women still had to be helped to draw the letters). The participants' motivation was unequal. If the governments applied a program like this, in less than one year adult alphabetic illiteracy could disappear, and all first-graders would be autonomous readers and writers.

2.5 Two Crucial Conditions: Nurturing the Parents, Teaching the Teachers

Some authors have proposed the term “emergent literacy” to refer to the acquisition of knowledge about (or awareness of) some aspects of alphabetic writing, phonology, the correspondence between letter sequences separated by blank spaces and words, the linear organization of texts, and directionality of reading. The children become familiar with most of this before they can read words. We may thus admit the existence of a pre-literacy period during which both the human (mainly parents and preschool) and the physical environment contribute to increase the sensitivity and the ability of the children to benefit from later reading and writing instruction. However, I disagree with the idea that “there is no clear demarcation between reading and prereading,” that there is “developmental continuity between emergent literacy and later reading from the early preschool period to the early elementary school period” (Lonigan, Burgess, & Anthony 2000). These authors justify these claims on the basis of correlational data involving “phonological sensitivity” (measured at different times) and reading performance. However, this term refers to a group of tasks (rhyme and alliteration oddity, and blending and deleting syllables and phonemes) without distinguishing between them. The same merging characterizes the CTOPP (Wagner, Torgesen, & Rashotte 1999), which is used to provide a very global indication of “phonological awareness,” but leads to serious mistakes when it is crucial to distinguish between phonological awareness and phoneme awareness.

Phoneme awareness develops when one begins learning to read and write, whereas all the (other) forms of phonological awareness may develop earlier. Preliterate children and illiterate adults are able to distinguish “cat” and “bat,” thus they are *sensible* to a phonetic difference without being aware of /k/ and /b/ as segments. Many of them may be able to indicate above chance level the pictures, among those of cat, fish, door and flower, whose names begin with f . . . , but very few, perhaps only those who already know a fair number of letter sounds, become capable of deleting the initial phoneme of an utterance (for example, say fish without f . . .) after a short training. Accordingly, Stanovich, Cunningham, and Cramer (1984), testing kindergartners, found much better performance on the substitution of the initial consonant of a monosyllable by another consonant (86% correct responses, on average) than on its deletion, for example “if I tell you task” say it without the /t/ sound (25%). To say it simply, one thing is the preliterate’s phonetic sensitivity; another is the awareness of phoneme that usually develops with alphabetic literacy. The purpose of this argument is to leave it clear that emergent literacy (which is actually preliterate) and literacy, in the case of alphabetic writing, are marked by a specific and crucial discontinuity, namely between the mere intuition of phonetic similarities or differences, and the development of both phoneme awareness and the ability to operate on phonemes.

The family and preschool milieus are the main determinants of the child’s emergent literacy acquisitions. In particular, the parents’ and the teachers’ influence is crucial. However, both their interactions with the child and the quality of the

overall human environment (in terms of literacy, but also of mental, physiological and physical health, well-being, affectivity, and cognitive stimulation) are greatly impacted, from the child's birth and even before, by their socioeconomic and sociocultural status and life conditions. These are extremely unequal. "Leveling the playing field" through public policies is practically impossible; the field has never been so slanted. In the upper part, individual differences are determined by genetic factors because the manifestation of these is not restrained by the social factors, whereas in the lower part it is the social factors that are determinant because they leave almost no room for the genetic differences to manifest (...). Leveling the playing field can only be obtained through two simultaneous progressive changes: restraining and eventually suppressing the heritability of the appropriation patrimony (Piketty 2013, emphasizes this source of inequality, but does not go so far) and organizing and improving in and by the communities themselves the socioeconomic and educational support of the disadvantaged families, so that all children will benefit from equal conditions for the development of cognition and literacy. This change from inside the society requires that contingents of volunteers be aware of the relevant scientific knowledge, namely of the science of literacy, to "nurture the parents" and "teach the teachers."

Some children do not suffer from a social disadvantage, but from a genetic anomaly that makes it hard for them to acquire reading and writing abilities. They are usually called dyslexics (although *dyslexia* often presents mainly patent impairments in the orthographic component of literacy). In dyslexia, the process of learning to read is disturbed since the earliest acquisitions. Phoneme awareness is more difficult to develop and often does not lead to isolated and operational conscious representations. In *L'Art de Lire*, Morais (1994), I described the case of an active and intelligent young man who managed to apparently overcome his dyslexia and eventually graduated in economy, but who still showed some errors and especially slowness in pseudoword reading. Confronted with the spoonerism task, i.e. exchange of the initial phonemes of two words (in this case, names), he was unable to find the correct answers "Kacqueline Jennedy" or "Kill Blinton." In many dyslexics, decoding is slow and inaccurate. In others, decoding reaches enough efficiency to allow them to read texts with comprehension, but not as fast as it should be, so that they seem not to be able to read with enough automaticity in word identification.

Thus, the different types of impairment shown by the dyslexics correspond to the three successive conditions of alphabetic literacy: phoneme awareness, autonomous decoding, and word identification through automatic access to stored orthographic representations. Correct diagnosis of the impairment(s) allows in principle to work out, respectively, phoneme analysis and fusion, and, through practice and targeting decoding and orthographically complexities, two kinds of fluidity in oral reading of, respectively, pseudowords and words.

Another, quite basic anomaly is the fact that dyslexics do not seem to process individual letters as normal learners do. This was observed in a same vs. different decision task on two letters presented successively, which requires ignoring a surrounding shape congruent or incongruent with the shape of the letter. Using this paradigm, designed by van Leeuwen and Lachmann (2004), it was found that

only dyslexic children processed the letter and its surrounding shape, benefitting from the shape similarity of shape and envelope. All the other groups, namely children who were normal readers, and adults (literate, ex-illiterate and illiterate) ignored the shape surrounding a letter, which never happened for pseudo-letters, showing that only dyslexics did not apprehend the letter-target independently of the potentially disturbing context (Fernandes, Vale, Martins, Morais, & Kolinsky 2014). The consistent observation that the congruency effect was negatively correlated with phonological ability in the dyslexic group, and with the knowledge of letter names in the illiterate group, suggests that the perceptual processing of a letter may be protected from extraneous stimuli by the spontaneous activation of the letter's phonological label. These dyslexics know very well the letters' names but it may be conscious knowledge, not knowledge that is mobilized unintentionally. In dyslexics, something that should have happened in the association of visual symbols and their phonological counterparts did not (Blomert 2011).

How can this letter-processing anomaly be overcome? I can only answer this question theoretically. Learning to read is learning to process written language, thus, it should never, at none of the stages, be dissociated from learning to write. At the beginning, letters must be hand-drawn solicited by their names and by their phonetic values. Like phoneme grasping, which starts and develops best when manipulating appropriately pairs of spoken-written syllables, in the other way, round letters must be imbibed by phonology from the beginning and the success of this learning process must be checked by the teacher.

Dyslexia has received diverse explanations based on visual, or visuo-spatial, or visuo-attentional factors. The latter is a particularly interesting case, because all the data supporting it as a cause of poor reading can also be interpreted as a consequence. During reading acquisition, the letter perceptual span increases (but the size of the orthographic units, which represent phonological units, also increases). In Bosse, Tainturier, and Valdois (2007), from 1st to 3rd to 5th grade, the % of letter sequences correctly identified jumped from 7% to 34%. Dyslexics aged 11.5 years on the average had a mean reading age of about eight years and could obtain only 26% correct identifications, much less than the normal readers of the same chronological age (60%). However, the fact that in the dyslexics compared to normal readers there was a greater lateral masking effect only for Latin letters, not for Korean ones (Pernet, Valdois, Celsis, & Demonet 2006), implies that the dyslexics' problems concerns a late stage where the letters are put in relation with stored knowledge (which could be phonological). The idea that some phonological impairment is involved in dyslexics assumed to present a visuo-attentional deficit is supported by the fact that they were found to be inferior to normal readers when they had to read a real text but not when they had to search for the occurrence of a target letter, for example "R," in a "text" made only of consonants (Prado, Dubois, & Valdois 2007). Thus, dyslexia, even in those who have apparently mastered decoding, does not result from a deficit in a general perceptual analyzer, but in one that is specific to orthographic structures, i.e. structures whose function is to code phonological ones. Dyslexics who can decode but rely too much on decoding are much more affected by word length and may need twice the number of fixations that normal readers have (Hawelka, Gagl, & Wimmer 2010).

The dissociation interpreted as between phonological and visual attention span disorders (Peyrin et al. 2012) may actually reflect different developmental stumbling blocks affecting decoding. In this study, two highly educated adult dyslexics, graduated by the University, have been examined. LL, presented as phonological dyslexic, managed to read and spell both irregular words and pseudowords as correctly as normal readers, but she was slower. In contrast, she failed almost completely a difficult phonemic awareness task (spoonerisms). Her result pattern is thus very similar to that of the economist I examined and mentioned in Morais (1994). LL was a phonological dyslexic and it does not make sense to keep calling her dyslexic unless we invent the expression “dyslexic literate”. FG was presented as having a visual parallel processing disorder but preserved phonological skills. Indeed, his global report of consonantal strings was very poor (but not the post-presentation cued report of one of the consonants), and he performed normally with spoonerisms. However, FG, compared to LL, was slower in reading irregular words, poorer in spelling them, and also clearly poorer in pseudoword reading. His excellent awareness of phonemes does not imply that all his phonological skills are preserved. Phoneme awareness, as indicated above, is a predictor of reading performance only in the very initial grades. Augmenting the size of the phono-orthographic units involved in decoding frees the reading process from the phoneme unit. It is either at decoding through larger units or, more likely, at the memorization of word representations automatically accessible that FG had experienced serious difficulties. This would explain why FG made numerous errors in reading and spelling pseudowords and was too slow on irregular words. When these cannot be read fast, it means that they must be read by a necessarily time-consuming “corrected decoding.”

The theory I suggest is that problems can arise at one or the other, or both, of the first two acquisitions that lead to skillful reading: phoneme awareness and decoding. The difficulties with orthography experienced at advanced decoding are also difficulties with phonology. This happens not only because the latter may result from peripheral or central hearing problems (cf. Calcus, Hoonhorst, Colin, Deltenre, & Kolinsky 2018), but also because orthographic knowledge contributes to make phonological processing more segmental (Calcus et al. 2018). Orthography exists by reference to phonology. It is a specific way of representing phonology, its code. It may be visual, or it may be tactile: orthography is not tied to a sensory modality. Phonology, too, is not strictly dependent on hearing, as we know that deaf people using “cued speech” develop phonological representation and can learn to read and write quite well (Leybaert 2000). Regarding intervention, in the same way as phoneme awareness is grasped through appropriate questioning on material arranged in such a way that the learner can “isolate” the phoneme, the repeated exposure and orientation of attention to particular phonograms in different lexical contexts should help the learner to create such units. Additionally, as proposed in Share’s (1995) theory, and confirmed experimentally by him and others, organized exposure to repeated words in teaching classes (obviously, of pseudowords in experimental sessions) should help to create the long-term word memory called the “orthographic mental lexicon.”

Does the above theory imply that dyslexia is nothing more than severely-poor reading and spelling that is or seems highly intractable? Before answering this question, it is opportune to recall the impressive study by Fluss et al. (2009). These authors examined more than 1,200 second-graders with at least 16 months of instruction, from 20 schools in Paris. They came equally from high, medium and low classes, which allowed a comparison of the prevalence of poor reading in each social class. Poor readers, according to the criterion of a 12-month delay in the reading tests, were 12.7% of the total sample, and the impact of social class was huge: only 3% came from the high class, 11% from the medium class, and as much as 24% from the low class. Thus, a low class child is eight more times at risk than a high class child of becoming a poor reader. This data shows that the estimations that 10–20% of the general population is dyslexic must be gross overestimations.

The 3% of high class poor readers cannot be all dyslexics, i.e. due to genetic anomaly. Assuming that all of them benefit from the cognitive and linguistic advantages afforded by their class, there may be high class poor readers for affective, motivational problems or, indeed, innate and severe cognitive backwardness. The 3% is thus itself an overestimation. On the contrary, to assume that only a maximum of 3% among the medium and the low class children are dyslexics may be an overestimation, as a poor milieu can influence epigenetics. We must admit that we ignore almost everything about these questions. The genes identified as being involved in dyslexia do not seem to be dyslexia-specific, but relevant for other learning domains or for learning in general. Anyway, the concept of dyslexia is vague. If it is defined only by very poor reading, it lacks of specificity; and it also lacks of specificity if it is defined by a genetic origin, given that this origin is not demonstrated to be reading and/or writing-specific.

I am not proposing to abandon the concept of dyslexia. In science one must be patient; we can keep it waiting until more relevant evidence is obtained. In the meanwhile, we should agree that, even if we should continue doing everything that can be done for the so-called dyslexic children and adults, we should also do, not more but as much, for the several times greater part of the population constituted by children and adults who are poor readers and spellers and in many cases functionally illiterate. Presently, perhaps because “dyslexics” are numerous, or seem to be numerous, in countries’ high classes, there are many initiatives such as world foundations and congresses on dyslexia. I think they are welcome. However, there should be also many initiatives, world foundations and congresses on child and adult illiteracy. Since the world’s governments became neoliberal, no more international meetings comparable to those on dyslexia worry about illiteracy. Only the UN and its dependent organization UNESCO are publishing reports that repeatedly call the world’s attention to the permanence of this serious situation and to the risks of functional illiteracy increase. This is strange because we know it seems feasible and not ruinous to eradicate illiteracy. The vexing case of the revolutionary Cuba, which eradicated illiteracy in a few years, has been voted to embarrassing silence.

What I propose, given the passivity of most of the worlds states, politicians and potential private donors, is the following. Researchers, academics and teachers can form a huge contingent of literate people to come in aid of the illiterate and

functionally illiterate people and to raise literacy preparation and learning for, respectively, preschool and primary school children. They can do it, to begin, on a local basis, by joining their efforts and expertise to those of communities, associations and committees that are already operating in the field. Two pressing objectives are (1) to nurture the parents, by showing them how to help their children, at home, to develop emergent literacy (cf. among others, Morais 2016, chapter 2.4), and (2) to teach appropriately most of the teachers (cf. among others, Rayner, Foorman, Perfetti, Pesetsky, & Seidenberg 2001) who were not taught themselves how to best teach children learn reading and writing. They were not taught so, partly because their own teachers have old and a-scientific (if not antiscientific) conceptions (see Brady 2011), and partly because the governments are not interested in transforming education in a way that would put in danger the oligoliteracy (Morais 2016).

2.6 Conclusion

To conclude, I formulate, first, what should be a preliminary question to the debate on the present issue, and, second, the question that is socially the most relevant concerning the methods of learning to read and write in an alphabetic system.

The preliminary question is: “What ethical values should inspire educational authorities, but also educators and researchers?” Hoping to contribute to triggering a debate, my answer is: The main ethical value, in what concerns the acquisition and development of literacy is a strict equality of rights to be realized in equitable educational efforts, from birth to high-level studies, whatever the social origin of the children and of the families.

The question about learning to read and write methods in the alphabetic system is: “Is there one method more democracy-friendly than the others?” My answer is yes. More precisely, it is phonics, which is based on a clear comprehension that the alphabetic characters stand for phonemes, that it is necessary to learn the decoding-recoding mechanisms in reading and writing taking into account the orthographic code, and that skillful alphabetic literacy is automatic access to word orthographic representations allowing, eventually, in connection with other cognitive capacities, a productive and creative use of the literacy abilities. This method is the only one that is really democratic because it gives each person the autonomy and automaticity of reading and writing that conditions an efficient processing and communication of information and that, by this mean, permits a personal, pondered and critical participation in collective debates and decisions.

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Chapter 3

Domain Generality and Specificity of Statistical Learning and its Relation with Reading Ability



Yi-Hui Hung, Stephen J. Frost, and Kenneth R. Pugh

Abstract Reading is to map orthographic units onto an existing phonologic-semantic system of its corresponding spoken language. Neuroimaging studies have shown that the print-speech co-activation in perisylvian networks for print-speech conversion is a universal neural signature of skilled readers. In addition to large commonality, small language differences are suggested: phonological knowledge from spoken language is useful for orthographic learning in transparent writing systems whereas visuospatial processing is more demanded for opaque writing systems, like Chinese. An emerging research suggests that reading acquisition may also reflect a general statistical learning (SL) capacity to implicitly assimilate the systematic structures of a linguistic environment. It is unclear whether visual and auditory SL play similar roles in learning to read different writing systems and whether the experience of learning of any given orthographic system changes the way one detects and computes statistical patterns. To understand the bidirectional relations between SL and reading experience, future research could examine the relative contribution of visual and auditory SL to individual differences in learning different writing systems, e.g., English vs. Chinese and track the changes of SL after learning different writing systems. Such studies will also shed light on the debate in universal account of learning difficulty in dyslexia.

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3.1 Overview

Learning to read fluently depends on neurocognitive processes that link orthographic units with phonological units and further linking these yoked orthography/phonology representations with well-specified lexical semantic (S) representations (Perfetti & Hart 2002; Pugh et al. 2013). Learning to read well both depends upon and also changes spoken language representations (Dehaene et al. 2010; Preston et al. 2016). At the brain level, speech-related neural networks in perisylvian cortex are reorganized after learning to read to support print-speech conversion and our studies have shown the degree of print-speech co-activation in these regions have been associated with individual differences in phonological processing, and reading abilities, and longitudinal reading outcomes in children (S. J. Frost et al. 2009; Preston et al. 2016). An emerging literature suggests that people who fail to establish coherent neural connections between print and speech appear to have setbacks in becoming fluent readers (Blomert 2011).

At the cognitive and neurobiological levels of analysis, the development of an interconnected orthography, phonology, and semantic organization is needed for the acquisition of high quality lexical representations and fluent word decoding skills (Perfetti & Hart 2002) and failure to create these bindings, particularly orthography to phonology underpins reading failure. We argue that this depends upon implicit knowledge of the segmental structure of speech and how speech units are linked together (phonological awareness) puts constraints on establishing connections between print and speech (as well as connection to meaning) because the statistical patterns of spoken language constrain sequences of orthographic units. This may further impact grasping the alphabetic principle (that the letters of written alphabets (usually) correspond to the phonemes of the language), thus allowing individuals to develop efficient phonological decoding routines (mapping spellings-to-sound) that are necessary for fluent word recognition (Apel, Wolter, & Masterson 2006; Liberman, Shankweiler, Fischer, & Carter 1974). When the mapping between print and speech is transparent and consistent, the phonological knowledge from spoken language is useful for orthographic learning. It could be less so for opaque writing systems like Chinese and learners may give greater weighting to morphological or semantic codes in learning to decode the writing system (R. Frost 2012), but this is far from a resolved question and some data suggest far more universality than divergence across writing systems differing in orthographic depth (Nakamura et al. 2012; Rueckl et al. 2015).

Despite an extensive literature supporting a core deficit in reading disability is rooted in phonological processing or access, recent research has generated questions regarding whether these language-specific problems in learning and consolidation of this learning are grounded in more basic domain general problems in detecting sta-

tistical regularities. Indeed, some have speculated that people who are challenged by reading acquisition because they have difficulty in detecting and/or storing statistical structures of linguistic inputs (e.g., Gabay, Thiessen, & Holt 2015; Pavlidou, Kelly, & Williams 2010). However, it is still debatable whether such statistical learning (SL, the establishment of rule-like knowledge based on reoccurring patterns) is specific to language and print or more general to other cognitive domains, (e.g., learning sound or motor sequence as examples; see R. Frost, Siegelman, Narkiss, & Afek 2013; Lum, Ullman, & Conti-Ramsden 2013).

With regard to how different aspects of learning abilities moderate individual differences in learning to read, two questions arise. First, what are the relative contributions of orthography, phonology, or semantic connections and learning skills to reading mastery across diverse languages? Second, to what degree do more basic domain general SL skills underpin literacy learning and might these domain general influences differ across writing systems? To explore these topics, we first review previous findings on typical/atypical reading behaviors and their neural correlates and will consider what is currently known about how these brain pathways intersect with more basic SL systems. Second, we will address how and why SL in non-linguistic domains can impact decoding skills. Finally, we will lay out the evidence and an argument for how cross language comparisons of subsets of SL abilities may shed light on the specificity of SL and the direction of the relationship between general SL and the ability of learning to read.

3.2 Reading Skills, Neural Correlates and Individual Differences

3.2.1 Reading Development

Learning to read involves linking existing phonologic-semantic representations acquired implicitly over the course of spoken language development with visual (orthographic) symbols acquired through literacy instruction. There is overwhelming evidence that phonological awareness is characteristically deficient (or lacking) in those with reading disabilities who, as a consequence, have difficulty mapping the alphabetic characters of print onto the spoken word. Measures of phonemic awareness predict later reading achievement (Wagner, Torgesen, & Rashotte 1994); deficits in phonemic awareness consistently separate typical and impaired readers (Stanovich & Siegel 1994); and instruction in phonemic awareness promotes the acquisition of reading skills (Foorman, Francis, Fletcher, Schatschneider, & Mehta 1998). The importance of skills such as phonological awareness for learning to read is generally found across alphabetic languages with different degrees of orthographic depth (Caravolas, Lervag, Defior, Seidlova Malkova, & Hulme 2013). Taken together, the ability to parse speech sounds provides an initial constraint on reading acquisition and the ability to associate these sound patterns with prints further supports later reading performance.

3.2.1.1 Phonological, Semantic Processing and Print-Speech Conversion in the Brain

Neuroimaging studies have consistently identified a group of brain regions involved in skilled reading, including the supramarginal gyrus (SMG), the inferior frontal gyrus (IFG), the middle temporal gyrus (MTG) and superior temporal gyrus (STG) and the angular gyrus (ANG) as well a region in occipitotemporal cortex often referred to as the visual word form area (VWFA) (Sandak, Mencl, Frost, & Pugh (2004); see Martin, Schurz, Kronbichler, & Richlan 2015; Paulesu, Danelli, & Berlinger 2014; Pugh et al. 2010, for meta-analyses and review). Functional specificity is observed in these brain regions: STG/SMG and IFG are critically involved in orthography to phonology mapping whereas the MTG and the ANG are more involved in orthography to semantic processing (S. J. Frost et al. 2005; Joubert et al. 2004; Price, Moore, Humphreys, & Wise 1997, see Cattinelli, Borghese, Gallucci, & Paulesu, 2013 for meta-analysis). The VWFA, serves as a fast mapping system which entails functional connections to both dorsal-frontal orthography to phonology and ventral orthography to semantic pathways in skilled but not in unskilled readers (Pugh et al. 2013, 2000).

Knowledge of the phonological structure of spoken language allows readers to parse continuous speech into segments to be mapped onto printed representations. Print-speech convergence at the brain level is probed by examining the extent to which a given brain region's activity is driven by both visual and auditory inputs of linguistic stimuli; that is the degree to which reading and listening come to depend on the same tissue (Preston et al. 2016); as noted several studies from our lab reinforce the claim that good reading depends on the capacity of the left-lateralized perisylvian networks for speech becoming "available" to support reading or writing behaviors. For example, our research has demonstrated that individual differences in reading skills are positively correlated with the extent of the left IFG responding to both print and speech for adolescents (Shankweiler et al. 2008) and of the left IFG and the STG for children aged between six to eight (S. J. Frost et al. 2009). The work by Preston et al. (2016) points towards a causal relation between print-speech co-activation and reading performance, showing that the extent of the left IFG, VWFA and the left inferior parietal regions responding to both print and speech in children aged between six to eight predicted their reading ability 2 years later. Moreover, a recent cross-language study by Rueckl et al. (2015) suggests that print-speech overlap is a universal signature of skilled reading. In this study, Rueckl et al. showed that a left-lateralized neural network of the VWFA, the STG, the SMG and the IFG pars triangularis was commonly involved in reading and listening to words in both alphabetic (Spanish, English, Hebrew) and non-alphabetic (Chinese) languages that also vary from transparent to opaque print-speech mappings. These findings reinforce the idea that fluent reading depends on how well orthographic units connect to existed phonologic-semantic system and this results in modifying the neural network of spoken languages to become bimodal, i.e., be tune to the input of visual words.

3.2.2 *Group and Individual Differences of Reading Skills*

3.2.2.1 **Developmental Studies of Reading (Dis)Ability**

Developmental neuroimaging studies have shown a trajectory towards the mature reading circuit described above with several studies showing early recruitment of the left STG, which is implicated for speech processing, for beginner readers; and increasing engagement of the left MTG and the left IFG as well as increasing specificity for print of the VWFA, paired with decreases in activation of the right occipital area with age (Brem et al. 2010; Turkeltaub, Gareau, Flowers, Zeffiro, & Eden 2003, see Martin et al., 2015 for a meta-analysis). In contrast, children with developmental dyslexia with core deficits in phonological processing do not show this developmental pattern, instead showing under-activation of the VWFA, left temporoparietal regions and the left ANG compared to age-matched and ability-matched children (Hoeft et al. 2007; McNorgan, Randazzo-Wagner, & Booth 2013; Pugh et al. 2000, see Pugh et al., 2010 for a review). And as noted above, dyslexic children fail to develop strong print/speech convergence in the left perisylvian cortex and this severely impacts the development of fluent reading skills (Preston et al. 2016). Thus, the normal trajectory of increased reliance on left-lateralized fast mapping systems (e.g., VWFA) is not seen in dyslexic. By contrast compensatory activity and connectivity in brain regions in the right hemisphere is often observed (Pugh et al. 2000).

Cross-Language Comparison: Reading Chinese

Although the design principles of writing systems, i.e., the mapping between print and speech, are similar across languages, different writing systems pose different challenges for its readers in terms of the systematicity of the mappings between written and spoken forms and the visual complexity of the orthography. In the Chinese writing system, most characters have one phonetic radical at the right side and one semantic radical at the left side of the character. Each character corresponds to one syllable whereas no orthographic unit corresponds to sub-syllabic phonological representations. There are 700 phonetic radicals and 200 semantic radicals (Huang & Chen 1998), implying that phonological component is more represented than semantic in orthographic structure. Hung, Hung, Tzeng, and Wu (2014) showed that the repetition effect of phonetic radicals was observed earlier than semantic radicals, indicating the mapping between phonetic radicals and syllables plays an important role for early lexical access for Chinese character recognition (see Hsu, Lee, & Marantz 2011; C.-Y. Lee et al. 2007, for similar conclusions).

A basic difference between alphabetic languages and Chinese lies in the functional unit size of the mapping between written and spoken forms – letter or letter cluster to phoneme and graph to syllable, respectively. Because of a high

prevalence of homophones in Chinese, some researchers have argued that Chinese entails less reliance on orthography-phonology and more reliance on orthography-semantic mappings than alphabetic languages (Perfetti et al. 2007). Zhao et al. (2014) compared brain activity of native Chinese speakers who were trained to read low-frequency Chinese characters by either focusing on pronunciation or semantics. A direct comparison between the phonological training and the semantic training revealed little differences: the phonological training activated more the right cingulate gyrus and the semantic training activated more the left IFG. In contrast, the results from a similar training paradigm tested on native English speakers found more differences between the division: the phonological training engaged more the VWFA, SMG and IFG and the semantic training engaged more the MTG (Sandak et al. 2004). The authors argue that a greater balance between the engagement of regions supporting semantics and those supporting phonology in Chinese relative to the patterns observed in English. Despite differences in the grain size of orthography-phonology mapping and in the weightings of orthography-phonology and orthography-semantic across languages, Rueckl et al. (2015) revealed a common neural network of the VWFA, the STG, the SMG and the IFG at the left hemisphere involved in reading and listening to words across Spanish, English, Hebrew and Chinese, suggesting that print-speech co-activation is a common neural signature of fluent reading across languages. Again, the question of universality is hotly debated but there is at least some indication that similar print/speech dependences are language invariant.

In addition to its opaque spelling-to-sound mappings, the Chinese writing system is also known for its visual complexity. As mentioned, there are 700 phonetic radicals and 200 semantic radicals, which outnumbers letters (20–50) for alphabetic languages. Chang, Plaut, & Perfetti (2016) and Chang, Chen, & Perfetti (2017) showed that compared to other alphabetic and syllabic writing systems, Chinese writing system ranked on the top of the visual complexity in both objective measurements and subjective ratings. Although the visual organization (specifically, the order of letters) is constrained by phonotactics in alphabetic writing systems, the organization of strokes of a radical is independent of meanings or sound. A cross-language comparison study showed that beginning Chinese readers performed better in visual tasks than their counterparts in Israeli and Spanish (McBride-Chang et al. 2011). These findings suggest that when prior knowledge from spoken language is less predictive of orthographic structure and the orthographic structure itself is complex, learning to read Chinese characters could call upon cognitive resources, such as visual learning skills.

We note that Rueckl et al. did observe small cross-language differences, i.e., the greater engagement of the right fusiform, the left MFG and the superior parietal gyri for print-speech co in Chinese than other alphabetic languages, may reflect an additional demand of visuospatial processing for reading Chinese characters. In addition to the common reading network of VWFA and the left IFG, a meta-analysis by Tan, Laird, Li, and Fox (2005) showed that Chinese readers additionally recruit the right hemisphere homologue of the VWFA and the dorsal part of the left lateral frontal area and the left inferior parietal area. They speculate that the first two

regions are recruited to support the high level of visuospatial processing for Chinese characters and the third region is engaged due to the attentional requirements of processing the visually complex Chinese characters. Findings from a cross-language comparison also indicate that Chinese readers recruit more extensive visual association areas (V3 and V4) for words relative to scrambled words to deal with the complexity of Chinese characters compared to readers of French readers for whom the difference between words and scrambled words was limited to V1 (Szwed, Qiao, Jobert, Dehaene, & Cohen 2014). Additionally, a study by Chan et al. (2009) showed that the activation of VWFA and the bilateral intra-parietal region was varied proportionately with the complexity of the Chinese stimuli presented, i.e., strokes, radicals and characters. These studies converge to implicate widespread ventral (bilateral OT) and dorsal (the left dorsal lateral frontal region and the left inferior parietal region) systems for handling the visual complexity of Chinese characters.

In summary, neuroimaging studies on typically-developed readers observed a common neural basis of print-speech co-activation across languages (Rueckl et al. 2015) and some language-specific differences (Tan et al. 2005). In contrast with Tan et al. who have suggested that alterations in frontal and parietal activity in dyslexia are unique to Chinese (Siok, Niu, Jin, Perfetti, & Tan 2008), Hu et al. (2010) found a large commonality for typical adolescent readers of Chinese and English in the brain, which replicated previous findings, but found no difference between Chinese and English atypical readers. The general lower activation in the left VWFA, ANG, posterior STG and middle frontal region suggest common weakness in orthography-phonology conversion and working memory in the dyslexic subjects of both languages. This raises a question regarding whether the common brain alteration of dyslexic population across languages indicates a common cause or product of reading impairment, and clearly more data is needed to resolve this question.

In summary, to become fluent readers, the tight connection needs to be established between orthography and spoken language system. Those connections are well supported by the neural network of VWFA, SMG, STG, IFG and MTG. Such brain signature of fluent reading is commonly across writing systems of various levels of orthographic depth. Previous cross-language study on typical readers revealed small differences in brain activation patterns and that may indicate the different weighting in orthography-phonology and orthography-semantic mapping and additional demands on visuospatial processing for Chinese readers. However, the language effect may not be observed in the dyslexic population (but see Siok et al. 2008; Tan et al. 2005, for counterargument). It leaves an open question about whether there is a common cause of reading problem across languages. Some studies showed individual difference in learning mechanisms before reading becomes fluent and such individual differences could arise at the perceptual level. For instance, visual, auditory, and speech perception were altered in people with dyslexia before they learned to read (Boets, Wouters, van Wieringen, de Smedt, & Ghesquière 2008; Carr, White-Schwoch, Tierney, Strait, & Kraus 2014; Goswami et al. 2011; Leppänen et al. 2002; van Zuijen, Plakas, Maassen, Maurits, & van

der Leij 2013). Furthermore, Bosseler, Teinonen, Tervaniemi, and Huotilainen (2016) showed that enhanced speech perception by infant directed speech facilitated statistical learning in newborns, implying that individual differences of perceptual ability change detection and learning of abstract patterns. Moreover, we suspect that the involvement of general statistical learning ability in learning to read might differ according to the distance between spoken language and writing systems. Particularly, when prior knowledge from spoken language is less predictive on orthographic structure, learning to read could call upon cognitive resource, such as visual learning skills, in addition to phonotactic knowledge. It is also interesting to examine whether the long-term experience of reading a writing system influence general learning ability. If visuospatial skills are more for Chinese readers, researcher could examine whether these different demands from long-term reading experience modulate general learning (e.g., non-linguistic statistical learning) or only language-specific learning (e.g., second language learning).

3.3 Relation between Reading Skills and Statistical Learning

3.3.1 Statistical Learning in Non-linguistic Domain

As described above, reading acquisition and development involves linking orthographic units to phonological units and to semantic representations. Recent research has also suggested that learning to read may also reflect a general SL capacity for becoming attuned to and implicitly assimilating the systematic structures and correlations of a linguistic environment. SL involves the establishment of rule-like knowledge based on reoccurring patterns and use the knowledge to segment continuous information into smaller units so the processing load is manageable. Such statistical knowledge can be applied to new stimuli in the same category and spares brain resource to learn new stimuli. The question of whether orthography, phonology and semantic binding problems reflect a more general deficit in SL abilities has profound implications for theory in the domain of reading disabilities. In this section, we will review the research of SL in non-linguistic domains and in linguistic domains and link them together by their contribution of predicting reading ability.

A common paradigm for measuring SL entails presenting subjects with a stream of nonsense speech syllables (e.g., babupubupadadutabapatubipidabututibu), and manipulating the probability of a given item predicting the presence of another item (transitional probability). Early work by Saffran, Aslin, and Newport (1996) showed that infants listened to the syllables with low transitional probability longer compared to the ones with high transitional probability. They suggested that this result was due to better memory for the stimuli with high transitional probability, causing the stimuli with low transitional probability to be treated as novel and hence attending to them for a longer period of time. Empirical studies showed that both

infants and adults were subject to the influence of the distributional frequency of individual visual features and the transitional probability (Fiser & Aslin 2002, 2005; Kirkham, Slemmer, & Johnson 2002). SL is observed in auditory (Saffran, Johnson, Aslin, & Newport 1999), in motor responses (Hunt & Aslin 2001) and across modality, e.g., visual and auditory (Mitchel & Weiss 2011). Moreover, visual SL is sensitive to the spatial distribution of inputs and is more efficient at the slow than fast presentation of input whereas auditory SL is sensitive to the temporal distribution of inputs and is efficient at both fast and slow presentation (Conway & Christiansen 2009). SL across modalities did not correlate with each other within individuals, suggesting that SL is not a unified ability (Siegelman, Bogaerts, Christiansen, & Frost 2017).

At the brain level, Turk-Browne et al.'s fMRI study (2009) found that the IFG, the striatum, the MTG and the ventral occipito-temporal (OT) cortex were implicated in visual SL. Karuza et al. (2013) found that Broca's area was implicated when the subjects engaged in auditory SL. A recent review of past neuroimaging studies of SL indicates that modality differences of SL in the brain, with visual SL engaging in the fusiform gyrus and the cuneus and auditory SL engaging parietal sites and posterior STG whereas the IFG, as well as hippocampal and subcortical (caudate and thalamus) regions serving as core regions for both auditory and visual SL (R. Frost, Armstrong, Siegelman, & Christiansen 2015). Doyon (2008) further dissociate functional roles of the subcortical regions (hippocampus and striatum) in learning. In the early stage, the brain mobilizes a functionally relevant network (e.g., modality specific regions for visual input) to process the to-be-learned materials and engage the hippocampus for temporary storage. In this stage, the representation of the newly-acquired information is malleable and may decay easily. After many exposures or practice, the representation of new information is more stable and is consolidated in the striatum.

Although SL starts from infancy, it is not a fixed cognitive ability but is susceptible to other factors, such as memory capacity and development. Bulf, Johnson, & Valenza (2011) showed that the visual SL was most pronounced when the sequence contained four items but the performance deteriorated when the sequence contained more than four items, suggesting the influence from memory capacity on SL. SL also changes across ages: the performance of sequence learning was improving with increasing ages and achieved peak during adolescence but afterwards declined with ages (Janacsek, Fiser, & Nemeth 2012). The authors, hence, propose that young individuals use model-free learning which allows more sensitivity to detect probability changes in the environment. In contrast, older individuals use model-based learning which rely more on existed knowledge system to predicts the patterns in new stimuli and hence decrease sensitivity to detect unpredicted changes. SL emerges at an early stage of human life and changes across the lifespan. Hence, it is open to investigation on whether the age effect of SL is simply a result of the change of cognitive efficiency across the lifespan or a consequence of the prior life experience. The later speculation is especially interesting to explore because the increase of SL until adolescence could be partly due to the accumulated reading experience.

In summary, SL are sensitive to different parameters according to the modality of inputs, that is, it is easier to detect distribution over space for visual input and distribution over time for auditory input (Kubovy & Schutz 2010). Such modality effect also reflects in the brain: visual SL in occipitotemporal area while auditory SL in the superior temporal gyrus. The central computation and storage of statistical relations involves the IFG and the subcortical network of the hippocampus, the caudate and the thalamus. The finding of the development of SL across age leaves an open question about to what extent the development of SL is a result of prior experience. We speculate that the knowledge from prior language system may modulate the sensitivity of SL to different parameters and hence exert its influence on learning new languages (and on efficient orthography, phonology and semantic binding and print/speech convergence in the area of reading).

3.3.2 SL in Linguistic Domains

SL is more than a laboratory paradigm to probe human learning processes. Languages and writing systems are full of statistical patterns, and language and literacy acquisition may draw on SL. Distributional and conditional statistics can be manipulated not only in non-linguistic stimuli, but they also can be used to describe the statistical patterns in the orthographic structure (Thiessen, Kronstein, & Hufnagle 2013). For instance, N-gram frequency describes how often of two or more letters co-occur across words (N indicates the number of letters). Orthographic neighborhood density is defined as the number of words that can be generated by changing one letter from a target word in the same letter position. Both indices measure how often a given orthographic pattern occurs across words. The statistical relationship between graphemes and phonemes or grapheme and meanings can be captured by orthographic/morphological regularity and consistency (Glushko 1979; Prasada & Pinker 1993). The former is whether the pronunciation (or meaning) of any word obeys the grapheme-to-phoneme (or meaning) correspondence rules. The latter refers whether an orthographic body across different words shares the same phonological rime or meanings. In the following section, we will review the evidence showing readers' sensitivity to statistical patterns in orthography and its relations with reading performance.

3.3.2.1 Sensitivity to Statistical Mapping between Orthography and Phonology/Semantic Codes in Readers of Alphabetic Languages

Skilled readers were sensitive to the bigram frequency and the neighborhood density (Grainger & Jacobs 1996; Massaro & Cohen 1994; Westbury & Buchanan 2002). Cunningham and Stanovich (1993) found that children's sensitivity to frequency of certain letter combination (e.g., "yikk" vs. "yinn") accounted for thirty percent of unique variance of their word recognition ability. Neuroimaging results found

that the VWFA was implicated in the effect of bigram frequency (Binder, Medler, Westbury, Liebenthal, & Buchanan 2006; Dehaene, Cohen, Sigman, & Vinckier 2005; Vinckier et al. 2007). Some studies showed that skilled readers are tune to statistical patterns in their written languages but is less so for people in another end of spectrum of reading skills. Dyslexic participants did not show reading time difference between the words composed of regular mapping between letters and sounds and the words composed of irregular mapping between letters and sounds as typically-developed participants did (Coltheart 1978, 1983; Patterson 1981). Taraban and McClelland (1987) showed that the consistent words were read faster than the inconsistent words in typically-developed participants. In contrast, Gottardo, Chiappe, Siegel, and Stanovich (1999) demonstrated that the subjects with developmental dyslexic participants showed reduced consistency effect compared to typically-developed group (but see Metsala, Stanovich, & Brown 1998; Pugh et al. 2008; Strain & Herdman 1999, for different findings and discussion). In terms of orthography-semantic mapping, skilled readers extracted regular morphemes easier than irregular ones (e.g., Ellis & Schmidt 1998; Stockall & Marantz 2006). Besides, morphological awareness, the ability to extract subword units that carry meanings or mark grammatical categories, accounted for the individual variance of reading outcomes (Carlisle & Feldman 1995). All these results indicate that the statistical mapping of orthography-phonology and orthography-semantic is established in fluent reader's mind.

Dehaene and Cohen (2007) propose that the VWFA, as part of the visual system, has the property of increasing sizes of visual receptive fields along the posterior-to-anterior of the ventral system and hence are sensitive to letter combination of different grain sizes. Interestingly, such hierarchical property was also observed in the IFG (Vinckier et al. 2007). This might suggest that the hierarchical organization of the VWFA is influenced by phonological structure of spoken language because of orthography-phonology mapping. The left ANG was implicated in the effect of orthographic neighborhood (Binder et al. 2006; Fiebach, Ricker, Friederici, & Jacobs 2007), suggesting that the lexical organization by orthographic similarity not only reflects distributional frequency of orthographic patterns but also results in semantic competition among visually similar words. That is, the lexical organization by orthographic similarity is more than a visual property and might be mediated by its association with phonological-semantic representation. Neuroimaging studies found that spelling-to-sound (or meaning) relationship is implicated in the VWFA, the left IFG (Fiez, Balota, Raichle, & Petersen 1999; S. J. Frost et al. 2009; Graves, Desai, Humphries, Seidenberg, & Binder 2010; Herbster, Mintun, Nebes, & Becker 1997; Joanisse & Seidenberg 2005; Mechelli et al. 2005; Sahin, Pinker, & Halgren 2006). These studies indicate that while typically developed readers learn the distributional frequency of orthographic patterns and the statistical relationship between prints and speech and meaning. Extracting statistical patterns from orthographic inputs involves the neural network of the VWFA, the IFG and the ANG, similar to the reading network reviewed above, suggesting that fluent reading entails the neural entrainment of statistical patterns of orthography-phonology and orthography-semantic of orthographic structure.

3.3.2.2 Sensitivity to Statistical Mapping between Orthography and Phonology/Semantic in Chinese Readers

Similar to the neighborhood density in alphabetic writing systems, radical combinability for Chinese writing system measures how often a radical appearing in across different characters. Phonetic consistency and phonetic regularity are two common factors to examine the sublexical mapping between orthography and phonology in Chinese characters. The phonetic consistency describes to what extent one character with a given phonetic radical is pronounced identically to other characters which have the very same phonetic radical regardless of tonal difference. The phonetic regularity describes whether one character is pronounced the same way that its phonetic radical stand is pronounced when the radical stands alone. Numerous studies showed that the radical combinability, the phonetic regularity and the consistency modulated Chinese readers' response time in a lexical decision or naming (Chen & Weekes 2004; Feldman & Siok 1997; C.-Y. Lee et al. 2007; C.-Y. Lee, Tsai, Su, Tzeng, & Hung 2005; Taft & Zhu 1997; Zhao, Bi, & Wang 2011).

At the brain level, Lee et al.'s fMRI study (2004) demonstrated that the covert naming of phonological inconsistent characters activated the IFG, the supplementary motor area, the insula, the superior parietal lobe, the superior temporal gyrus in the left hemisphere more than that of phonological consistent ones. Zhao, Li, and Bi (2012) reported that irregular Chinese characters elicited more the bilateral IFG, the left inferior parietal lobule and the left STG than regular characters. S.-H. Lee, Booth, and Chou (2015) observed the mapping between semantic radicals and character-level semantic processing implicated the IFG, ANG and MTG in emerging readers. Taken together, Chinese readers are tuned to the statistical relationship between radicals and pronunciation and the processing of the relationship engages the left IFG as the consistency effect in readers from alphabetic writing systems.

In summary, skilled readers are sensitive to orthographic patterns and its statistical relations with spoken language and that is generally observed across English and Chinese (but less is known at present on the latter). The statistical computation of orthographic and phonological patterns is implicated in the VWFA and the left IFG across languages. These regions are also part of the reading network in which statistical extraction of word patterns are not explicitly demanded, suggesting that extraction of statistical patterns of orthography is part of process in natural reading and should be universal with regards to typical and atypical outcomes. We next consider extent evidence on relations between basic SL and reading behaviors.

3.3.3 Correlation Between General SL and Reading Skills

SL involves detection, categorization and prediction of patterns and that are similar across linguistic and general domain. Empirical studies showed that general SL

capacity correlated with individual differences in decoding skills¹. Arciuli and Simpson (2012) showed individual differences of visual SL performance correlated with reading skills in both children and adults. Gabay et al. (2015) demonstrated that the developmental dyslexic adult participants impaired in detecting the transitional probability of sound sequences in both speech and non-speech stimuli. The ability to learn the sequence of visual objects (Pavlidou et al. 2010) and motoric responses (Howard, Howard, Japikse, & Eden 2006, see Lum et al., 2013 for a meta-analysis) also correlated with one's decoding skills. Bogaerts, Szmalec, Maeyer, Page, and Duyck (2016) showed that individual differences of learning the order of auditory words predicts first graders' decoding skills 1 year later. R. Frost et al. (2013) showed that individual differences of visual SL performance correlated with the improvement of decoding in second language adult learners of Hebrew. All these studies suggest that the ability to detect the order or statistical patterns from inputs is involved in learning to read and SL and that gives rise to the correlation between two skills behaviorally.

The correlation between SL and reading ability emerges may because they share the same neural mechanism. Christiansen, Conway, and Onnis (2012) found that after the subjects learned a sequence of elements linked by specific transitional probability, the violation of the learned transitional probability led to the brain potential change in a similar way as the violation of linguistic syntax. Davis and Gaskell (2009) propose a complementary systems account of word learning and suggest that the hippocampus is implicated in fast associative learning and temporarily storage of newly-learned materials (plasticity). The distributed cortical regions (e.g., STG and medial temporal gyrus) are implicated for maintaining existed (language) knowledge where newly-learned information could be integrated with which for long-term retention (stability). Based on the complementary systems account of word learning and Doyon's neural model of learning mentioned before, one may infer that regardless of stimulus types, learning and storage of any new information is a domain-general process which engages the hippocampus. Hence, the hippocampus might be the locus for the correlation between SL and reading ability. However, in an unpublished fMRI study from our lab (Hung et al. 2018), we found the neural overlapping between motor sequential learning and word reading in the occipitotemporal cortex, the STG, the IFG, inferior parietal lobule, the insula, the putamen and the cerebellum. More importantly, activation of the right insula and the right IFG in the reading task and the sequential learning task was associated with individual differences of reading skill, suggesting that sequential coding is commonly involved across tasks. Individual differences in the general statistical learning predict one's decoding ability and language skills and such correlation is mediated by shared neural mechanism for statistical pattern extraction.

¹The paradigms of statistical learning and implicit learning are similar in probing human learning sequence or structure from inputs without explicit instruction (Perruchet & Pacton 2006). Hence, we cited the results from both paradigms as index of statistical learning.

Spoken languages and their written forms have statistically predictable structures. Beginning readers have to use general SL mechanism detect and eventually adapt to the patterns in their native languages. Therefore, both general SL and linguistic-specific SL (e.g., orthographic neighborhood and phonological consistency) predict reading achievement. What remains less clear is whether general SL is modulated by domain-specific experience. The notion is supported by Vasuki, Sharma, Ibrahim, and Arciuli (2017) showing that musicians, compared to non-musicians had better SL performance in auditory modality but similar in visual modality, suggesting that experience from music training over 10 years exerts modality-specific impact on SL.

From a cross-language perspective, the constraint of orthographic patterns (i.e., letter sequence) is largely from phonemic sequencing in spoken language for alphabetic writing systems. For Chinese writing systems, while orthography-phonology and orthography-semantic mapping occur at the radical level, stroke pattern of a given radical is visual-based and independent of phonology or semantic. For transparent writing systems, beginning readers could rely on the knowledge from spoken language system to learn the orthographic patterns in its written language. However, for opaque writing systems, like Chinese, which has complex stroke pattern in characters, beginning readers cannot only rely on the knowledge from spoken language system but may also require visual (domain general) SL skills to learn stroke patterns within characters. Consequently, the ability to detect statistical patterns at the visual modality might be more sharpened in Chinese readers compared to alphabetic readers. Comparing SL across readers of different writing systems potentially shed lights on the extent that domain general SL is shaped by language experience. In sum, there is fairly good evidence for links between outcomes and basic domain general SL for alphabetic languages. Careful consideration of the statistical organization of Chinese suggests that differential weighting toward visual learning might be salient in discriminating good and poor readers in this language. However, more research into these bidirectional links are needed in the coming years as we consider the plausibility of universalist accounts of reading (see R. Frost 2012, for speculation).

3.4 Summary and Future Direction

Learning to read involves linking visual symbols with an existing phonologic-semantic system of its corresponding spoken language. In order to acquire the mapping, one have to segment continuous speech sounds into adequate phonological units and map them to graphemes. The print-speech mapping relations may be more or less consistent across graphemic units. The mapping units could be as large as syllables or as small as phonemes. The sensitivity to orthographic and phonological patterns in the target written language is gradually built up over the learning course to become fluent readers.

Humans are capable of detecting the structure and the regularity in nature environment, speech signals and writing systems, and such capacity begins to show in infancy. General SL mechanism is used to acquire the structures in their spoken languages and writing systems. As a result, fluent readers adapt to language-specific statistical knowledge. This might be seen that linguistic-specific SL effects (e.g., orthographic neighborhood size effect and phonological consistency effect) are the product of general SL. However, it is less clear whether the experience of learning of any given orthographic system changes the way one detects and computes statistical relations in general and cross language studies of SL and reading ability are at a premium going forward.

The literature about the critical period of language acquisition indicates that infants are capable of discriminating speech sounds that are not in their mother tongues but lose such sensitivity after they immerse in their native language environment for 12 months (Kuhl 2004). That is, human is born with general learning mechanism to pick up any statistical patterns in their surroundings but later tune to certain structure or regularity that they encounter in daily life. This shift is important because neural resource is limited and one needs to dedicate it to the most significant information so that one can efficiently process the information and quickly interact with the outside world accordingly. How these early tunings impact later language learning for written form is unknown but developmental studies across languages will provide key information on how early language and later literacy (and their dependencies on SL) differ. On a related note, the neuronal recycling hypothesis (Dehaene & Cohen 2007) argues that existing brain systems for vision can adapt to new knowledge by unlearning certain associations which are critical only to old knowledge system. If general SL and learning to read share the same mechanism for detecting statistical patterns, it is expected that general SL could be modulated by reading experience, and whether the specifics of contrastive writing systems impact these relations will be an important research topic.

Thus, we propose cross-language comparison studies to more deeply investigate the bidirectional relation between SL and reading experience. First, it needs investigation about whether SL in general domains correlate with SL in linguist domains (e.g., the phonological consistency effect, the morphological regularity effect). If so, we can test the notion that for alphabetic readers, auditory SL, compared to visual SL, might more strongly correlate with the phonological consistency effect and the morphological regularity effect at the individual level because of the transparent nature of the writing systems, whereas for Chinese readers, visual SL, compared to auditory SL, might more strongly correlate with the phonological consistency effect and the morphological regularity effect because of the opaque writing systems. Such studies are important to test whether different types of SL in nonlinguistic domain taps linguistic domains at the level of pattern extraction as a function of the statistics of the language. Another question of some interest is a bidirectionality; given that SL ability predicts reading achievements, research could explore whether long-term learning experience in native languages change nonlanguage types of SL. We tentatively hypothesize that visual SL might be more improved after learning to read Chinese characters compared to alphabetic languages (though a strong phonological

and universalist account might suggest a different result). Such research may also inform us about whether the diminished SL in dyslexic population is a product of disrupted exposure to the structure of spoken and written languages rather than a cause of their language or reading impairment; thus is a universal reading disability account to be found in the links of SL to reading?).

Learning to read is multifactorial process. Examining the relation between SL and learning to read or reading outcomes opens up a window to observe the dynamics of how humans use general learning ability to acquire and become attuned to the structure of specific linguistic environments and how domain-specific experience might modulate general learning ability later. At the neural level, learning is influenced by how the brain extracting useful information (e.g., statistical patterns) from noise. Pugh et al. (2014) showed that the glutamate concentration in the occipital regions were higher in the dyslexic population than typical children (age 6–10) and the glutamate concentration further predicted the reading outcomes 1 year later, suggesting that neuronal hyperexcitability underlies the problem of noise exclusion in learning to read in the dyslexic population. Such studies inspire the future exploration about whether and how the neural chemistry associated with noise exclusion impacts SL and how they together determine reading outcomes.

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Chapter 4

Searching for the Orthographic Lexicon in the Visual Word Form Area



Heinz Wimmer and Philipp Ludersdorfer

Abstract In this chapter, we will present findings speaking for the position that VWFA is not limited to short “legal” letter strings (as assumed by the proponents of the original VWFA hypothesis). Rather, it serves as neural equivalent of the orthographic word lexicon of cognitive dual-route models of reading aloud and spelling. In support of this we found VWFA activation to exhibit an orthographic familiarity effect (Kronbichler et al., *NeuroImage* 21(3):946–953, 2004; *J Cogn Neurosci* 19(10):1584–1594, 2007; *J Cogn Neurosci* 21(2):222–229, 2009), a familiarity by length interaction effect (Schurz et al., *NeuroImage* 49(3):2649–2661, 2010) and, recently, a capitalization familiarity effect (Wimmer et al., *Psych Sciene*, 27(9): 1240–1248, 2016). Also in accordance with the orthographic lexicon view, in recent spelling-based studies we found the VWFA to be responsive to auditory words in orthographic decision tasks (Ludersdorfer et al., *Hum Brain Mapp* 36(4):1393–1406, 2015; *NeuroImage* 124(Pt A):834–842, 2016).

Keywords Visual word form area (VWFA) · Orthographic lexicon · fMRI · Visual word processing · Spelling

4.1 Introduction

It seems rather obvious that the brain of a competent reader and writer must represent a vast number of memories which somehow specify the letters of written words. By relying on such memories we are able to distinguish between homophonic words like *maid* and *made* and to correctly pronounce words like *yacht* for which letter-sound associations are not helpful. Most importantly, efficient reading depends on such memories because even long words such as *microscope* or *parliament* are often identified with a single fixation lasting little longer than a

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quarter of a second. For writing of words, reliance on memories has to be stronger than for reading, because pronunciations – even in rather regular writing systems – hardly ever specify exactly the letters.

Following Coltheart and colleagues (2001; Coltheart 2004) we use “orthographic lexicon” to refer to the memory system which allows efficient visual word perception and correct word production. The question for cognitive neuroscience is how and where in the brain the orthographic lexicon is represented. With few exceptions, however, neuroimaging research on visual word processing either did not deal with this question or did not identify plausible candidate regions. In this chapter, an attempt is made to provide an answer to where in the brain the orthographic lexicon is localized. In doing so, we will review a set of neuroimaging studies from our lab which suggest that a left hemisphere region at the ventral border of the occipital and the temporal lobe – the left ventral occipitotemporal cortex (vOT) – functions as neural equivalent of the orthographic lexicon.

4.2 Background for Our Search for the Orthographic Lexicon

Our interest in the left vOT as a candidate brain region for an orthographic lexicon function was inspired by the work of Laurent Cohen and Stanislas Dehaene, who in an impressive set of neuroimaging studies established the importance of this region for visual word processing and coined the term Visual Word Form Area (VWFA; Cohen 2002; Cohen et al. 2000; Dehaene et al. 2001, 2002). Anatomically, the VWFA is situated in the occipitotemporal sulcus on the border between occipital and temporal lobe and extends medially into the fusiform gyrus and laterally into the inferior temporal gyrus (see Fig. 4.1). The region is part of the ventral visual pathway – also referred to as “what” pathway in visual object recognition – with adjacent regions engaged in object and face recognition.

The VWFA hypothesis was challenged by Price and Devlin (2003, 2011) who presented an alternative account of vOT function in visual word processing. The proponents of the hypothesis responded to the critique and elaborated the original proposal (Cohen & Dehaene 2004; Dehaene & Cohen 2011). A comprehensive presentation of the work of the Paris-based group around Cohen and Dehaene is provided by Dehaene’s book entitled “Reading in the brain” (2009, German: 2010).

Referring to the left vOT region as Visual Word Form Area seems to imply that Cohen and Dehaene attributed an orthographic lexicon function to the vOT. However, this is not the case. A study by Dehaene and colleagues (2002) found no difference in VWFA activation between the familiar letter strings of existing words and unfamiliar pseudowords and concluded that the VWFA – contrary to its name – is not engaged in proper visual word recognition but performs a “prelexical” function, that is, it computes an abstract representation of “legal” letter strings in a similar manner for both familiar and unfamiliar letter strings. In an elaboration of this “prelexical” function, Dehaene (2005; see also Dehaene & Cohen 2011) proposed a hierarchy of increasingly larger codes (from letter forms to abstract

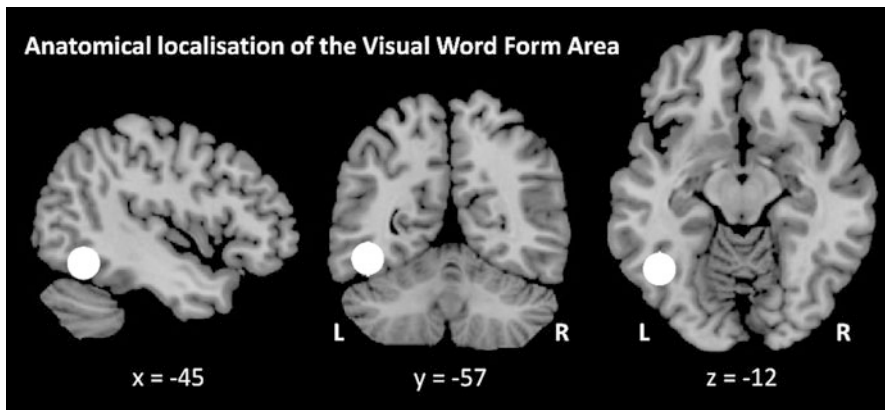


Fig. 4.1 Classic anatomical localization of the Visual Word Form Area (VWFA) in the left occipito-temporal cortex (vOT). Coordinates are taken from Cohen et al. (2000) and converted into Montreal Neurological Institute (MNI) standard space

letters to open letter pairs and short letter sequences) along the left ventral visual pathway culminating in a region slightly anterior to the original VWFA location shown in Fig. 4.1. Of specific importance in this coding scheme are neurons representing open letter pairs (referred to as open bigrams). This means that a word like *kursiv* elicits a response of neurons for *t-e*, *t-n*, *t-t*, *e-n*, *e-t*, *n-t*. A problem of such a coding device is that it results in a very large number of activated neurons for longer words like *parliament*. To avoid this implication, Dehaene and Cohen assume that the largest sequences of frequently occurring letters with a stable memory representation consist of only a very small number of letters (maximally four). This account denies that there can be stable memories for words such as *parliament* with more than four letter. This effectively denies the existence of a full-fledged orthographic word lexicon.

In initial critique of the VWFA hypothesis, Price and Devlin (2003) characterized the VWFA as myth and argued that it is a polymodal region where sensory information from various modalities interfaces with language and conceptual processing. The proponents of the VWFA hypothesis lately also spoke of the VWFA as metamodal reading region, because findings from congenitally blind readers found the VWFA region to be engaged by reading Braille words by touch (Reich, Szwed, Cohen, & Amedi 2011). In their later publications, Price and Devlin (2011) elaborated their initial position to the so called Interactive Account (IA) of left vOT function. Inspired by connectionist reading models (e.g., Seidenberg & McClelland 1989), the IA generally denies localized orthographic word representations. Instead, neuronal populations in vOT are assumed to be general-purpose analyzers of visual forms which receive top-down predictions from phonological and semantic brain regions. Orthographic representations arise only transiently in vOT from the interaction of visual inputs and top-down predictions. Obviously, in this scheme there is no place for permanently stored orthographic word memories. In summary,

the two most prominent neuroscientific accounts of visual word processing agree on the critical role of the vOT for efficient word processing but they do not attribute such processing to an orthographic lexicon.

Different from these theoretical accounts, there is empirical neuroscientific research based on the cognitive dual-route model of visual word processing (Coltheart et al. 2001) which assumes an orthographic lexicon for recognition of known written words. A modified version of this model was found to better explain a long list of experimental findings of visual word processing compared to a rival connectionist model without an orthographic lexicon (Perry, Ziegler, & Zorzi 2007). Based on a substantial number of imaging studies which presented both words and pseudowords, two meta-analyses searched for the brain locus of the orthographic lexicon (Jobard, Crivello, & Tzourio-Mazoyer 2003; Taylor, Rastle, & Davis 2013). The assumption guiding this search was that a region hosting the orthographic lexicon will exhibit higher activation to words compared to pseudowords. No evidence for such a pattern was found in left vOT regions critically engaged by visual word processing. To the contrary, these regions were found to be more activated for pseudowords than existing words. Therefore, both meta-analyses came to a negative conclusion with respect to the existence of a neural equivalent of the orthographic lexicon in vOT reading regions. However, as we argue in the following, this negative conclusion is based on the assumption that the left vOT hosts only whole-word codes and not also sublexical codes, that is, representations for letters and letter sequences associated with phonological information.

4.3 Evidence for Orthographic Lexicon Localization from Studies of Visual Word Processing

Our search for the orthographic lexicon started with Kronbichler et al. (2004). In this study we varied the frequency of words parametrically in 5 steps from high frequency to low frequency words and eventually to pseudowords of zero frequency. Presentation time was long enough to allow successful reading of even the pseudowords. The critical finding was that decreasing frequency levels were accompanied by increasing activation in regions along the left ventral visual pathway. Specifically, in a vOT region corresponding to the VWFA, there was no difference between words of high and moderate frequency, but a marked increase to low frequency words and pseudowords.

Since frequency of occurrence is a characteristic of whole words and since our study controlled for other item characteristics such as letter-pair frequency, we interpreted the comparatively low vOT activation to words of high and moderate frequency (compared to low frequency words and pseudowords) as reflecting instantiation of single orthographic whole-word memories and shut-down of the sublexical codes for letters and letter patterns which were assimilated by the orthographic word memories. Conversely, the increased activation to low frequency words and pseu-

dowords in vOT regions was taken to reflect prolonged activation of multiple sublexical codes which are required for arriving at a pronunciation. The important point is that both whole-word coding and sublexical coding of letters and letter patterns is attributed to the very same left vOT reading region. We were led to this interpretation by studies which examined the response of vOT regions (adjacent to the vOT reading region) to pictures of objects and faces (Chao, Weisberg, & Martin 2002; Rossion, Schiltz, & Crommelinck 2003; van Turennout, Bielowicz, & Martin 2003; van Turennout, Ellmore, & Martin 2000). These studies manipulated the familiarity of pictured objects or faces by repeated presentations and found that the high activation of vOT regions to initially unfamiliar stimuli decreased with increasing familiarization. The high vOT activation to an unfamiliar item was interpreted as reflecting encoding of the item with a set of visual features localized in vOT. These features are then used to build-up a memory representation in the vOT object and face regions which serve as efficient recognition unit in subsequent encounters. Similarly, we propose that the formation of orthographic whole-word codes in the vOT reading region is a process of unitizing sublexical letters and letter-sequences used on first encounters into a stable memory representation of the letter sequence of a word.

Our interpretation of the word < pseudoword vOT activation pattern as reflecting whole-word coding can be challenged based on the mentioned theoretical accounts by Dehaene and Cohen (2011) and Price and Devlin (2011). Both camps deny the existence of an orthographic lexicon so that the vOT activation difference cannot result from coding differences in the vOT reading region. Instead, it may be attributed to top-down influences from language regions. Specifically, access to whole-word phonology for familiar words may result in quick shut-down of visual feature or sublexical letter-based codes, whereas for unfamiliar words or pseudowords assembly of a pronunciation in frontal regions lasts longer so that no quick shut-down of vOT codes is possible. Therefore, differences in vOT activation observed by Kronbichler et al. (2004) and by other imaging studies comparing high and low frequency words or words and pseudowords may not result from different orthographic codes in vOT (i.e., whole-word vs. sublexical codes) but from differences in top-down influences from language regions on how long sublexical or visual codes in vOT have to be activated.

A test-case for distinguishing between these two explanations is provided by presenting correct and incorrect homophonic spellings of the same phonological words (e.g., *brain* vs. *brane*) as realized by two imaging studies from our lab (Kronbichler et al. 2007, 2009). Both types of word spellings receive the same “yes” response when presented together with pseudowords in the context of a phonological decision task (i.e., “Does xxx sound like an existing word?”). Importantly, there should be no difference in top-down influences on vOT activation because both correct and incorrect spellings activate the same phonological words in higher language regions. Accordingly, there should be no activation difference in vOT between correct and incorrect spellings when this region only codes sublexical letter patterns (Dehaene & Cohen 2011) or generic visual patterns (Price & Devlin 2011). Contrary to these predictions, Kronbichler et al. (2007, 2009) found a marked reduction of vOT activation to correct vs. incorrect word spellings, a finding replicated in other

labs (Bruno, Zumberge, Manis, Lu, & Goldman 2008; van der Mark et al. 2009). Corresponding to the reduced BOLD response to the familiar spellings, an ERP study found reduced negativity to the familiar spellings from about 200 ms onwards (Sauseng, Bergmann, & Wimmer 2004).

An even stronger test case for controlling top-down language influences on left vOT activation is provided by a recent study in which we presented two spellings of the same German words which only differed in the case format of the initial letters (Wimmer, Ludersdorfer, Richlan, & Kronbichler 2016). For nouns the familiar upper-case format of the initial letters was contrasted with an unfamiliar lower-case format (e.g., *Taxi* vs. *taxi*) and the converse manipulation was done for adverbs and adjectives (e.g., *blau* vs. *Blau*). In a lexical decision task both forms received the same positive response. Again, from the mentioned accounts of Price and Devlin (2011) and Dehaene and Cohen (2011) one would not expect a difference between the two types of spellings in the vOT reading region. However, such a difference is expected when the left vOT reading region hosts orthographic word memories which are based on how the visual word is typically perceived. Specifically, we expected reduced left vOT activation when the case-format of the initial letter of a word fits perfectly into the corresponding orthographic word representation compared to when the case-format of the initial letter violates the memory. This was indeed the case. Nouns with initial upper-case letter led to reduced left vOT activation compared to nouns with initial lower-case letter, whereas non-nouns with initial lower-case letter led to reduced vOT activation compared to non-nouns with initial upper-case letter. An important implication of these findings is that orthographic word memories do not consist of abstract letter identities but contain information on the typical case-format of the letters of the word. This is expected when orthographic word memories are based on visual experience and when the left vOT reading region is part of the ventral visual pathway.

Also of interest for the orthographic lexicon issue is an imaging study in which we manipulated the length of both words and pseudowords (Schurz et al. 2010). The short items consisted, on average, of 4.5 letters and the long items of 7.5 letters. From our hypothesis that the left vOT reading region hosts both whole-word and sublexical codes, we expect absence of a length effect for words, but presence of this effect for pseudowords. From the alternative accounts of Dehaene and Cohen (2011) and Price and Devlin (2011) which deny existence of orthographic whole-word codes – at least of codes for longer words by Dehaene and Cohen – in the vOT reading region, one would expect roughly similar length effects for both words and pseudowords. As evident from Fig. 4.2, we found support for our hypothesis. The length effect on vOT activation was limited to pseudowords and was strikingly absent of words. The similar length effect for words and pseudowords predicted by the alternative accounts was also found, but in a posterior occipital region engaged by low-level visual processes.

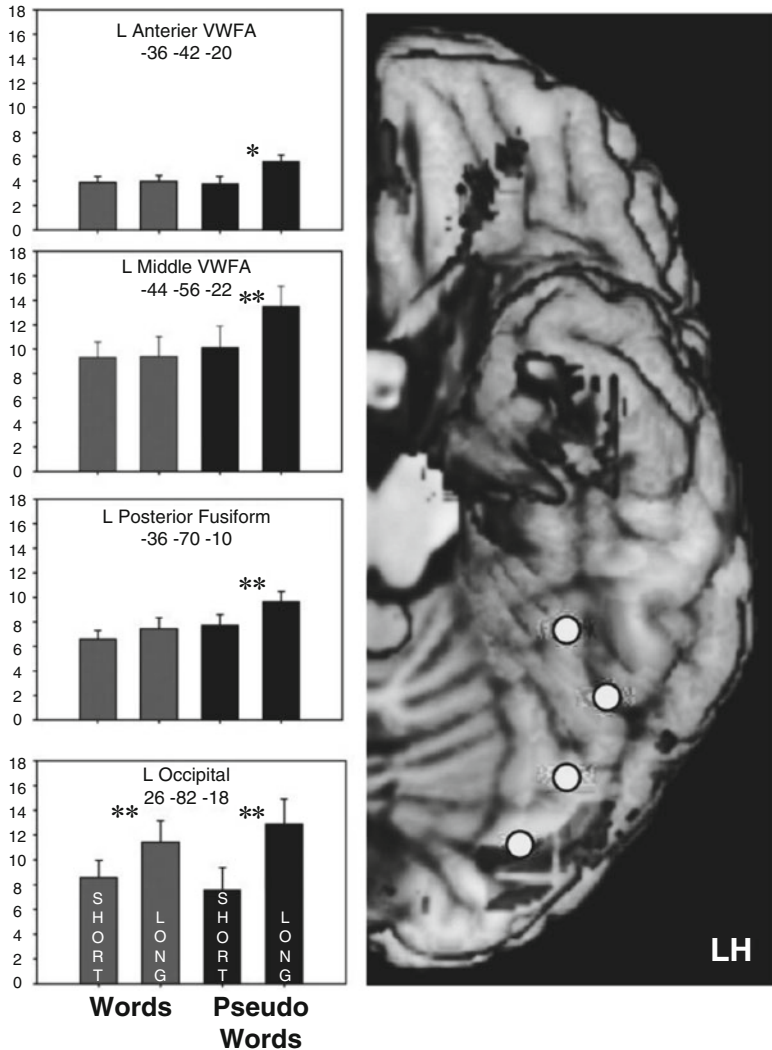


Fig. 4.2 Length effects of words and pseudowords in left ventral brain regions. Right panel: approximate locations of regions of interest. Left panel: locations of regions of interest (given in MNI space coordinates) and brain activity estimates in response to short (3–5 letters) and long (6–10 letters) words and pseudowords. (Reproduced from Schurz et al. (2010) with permission)

Our studies in support of the orthographic lexicon function of left vOT regions are nicely complemented by an imaging study of Glezer, Jiang, and Riesenhuber (2009) who used a different methodological approach. Glezer et al. examined repetition suppression effects in the left vOT reading region. In one condition, a word or a pseudoword was preceded by the same item (e.g., *boat-boat* or *soat-soat*) so that the vOT response to the second item was reduced (= repetition

suppression). Of main interest, however, was the presence or absence of such a repetition suppression effect when the target and preceding item were different but still shared the majority of letters (e.g., *coat-boat* or *poat-soat*). Importantly, while a repetition suppression effect in this condition was found for pseudowords no such effect was found for words. The vOT response was equally high when a word (*boat*) was preceded by a different word sharing most of the letters (*coat*) or by a different word sharing no letters at all (*fish*). The absence of a repetition suppression effect for even very similar words is expected when the words instantiate different orthographic word memories. This finding was interpreted by Glezer et al. as evidence for orthographic whole-word codes in the vOT reading region. To our knowledge, Glezer et al. are the only authors who similar to us have attributed an orthographic lexicon function to the left vOT reading region.

4.4 Evidence for Orthographic Lexicon Localization from Spelling-Based Studies

Compared to the large number of imaging studies concerned with visual word processing and the interest in neuroscientific accounts concerned with this issue, there is only little neuroscientific research on writing and spelling processes. Nevertheless, two recent meta-analyses collected the findings from the relatively few relevant studies (Planton, Jucla, Roux, & Demonet 2013; Purcell, Turkeltaub, Eden, & Rapp 2011). These studies presented auditory words in the context of writing or spelling tasks and the meta-analyses found converging evidence for activation in the left vOT. Furthermore, two spelling studies showed that the left OT activation in response to spelling co-localizes with activation for visual word reading (Purcell, Napoliello, & Eden 2011; Rapp & Lipka 2011). Examining the brain response to auditory words in the context of writing or spelling provides an interesting alternative approach to the study of the orthographic lexicon based on visual stimuli. Specifically, interpretational problems posed by vOT activation in response to visual stimuli such as prolonged processing of unfamiliar letter strings do not apply to vOT activation in response to auditory words. The following two studies from our lab were intended to extend the rather sparse evidence from spelling studies by methodological improvements. In Ludersdorfer, Kronbichler, & Wimmer (2015) it was ascertained that correct decisions on the presence or absence of a letter in the spelling of an auditory word were based on orthographic memory representations of the auditory words. To illustrate, in the critical condition participants heard a word like /fa:ze/ and had to decide if the letter *p* is included in the spelling of the auditory word (*Phase*). Conversely, they had to deny that the letter *k* is included in the correct spelling of an auditory word like /taksi/. In an auditory control condition, they heard the same words and had to judge whether the words were spoken by a male or female voice represented by the letter *m* or *f*, respectively. In Ludersdorfer et al. (2016) participants had to judge whether auditory

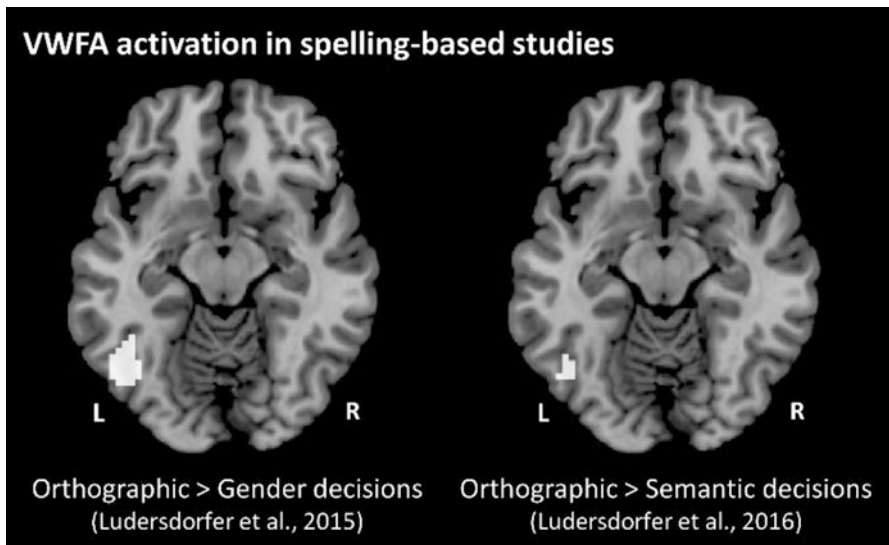


Fig. 4.3 VWFA activation in the spelling-based studies from our lab

words (all consisting of 3 phonemes) are written with 3 or 4 letters. In a contrasting semantic condition, the same auditory words were presented and participants had to decide whether the words referred to a living or non-living entity. Figure 4.3 shows that contrasting the orthographic conditions with their respective control condition in both studies resulted in increased activation in a circumscribed left vOT region corresponding to the VWFA. This response of the vOT region is expected when in response to the auditory words and the instruction, the orthographic lexicon in the vOT region were accessed.

4.5 Conclusion

As noted in the Introduction and the Background section, the orthographic lexicon issue is not at the forefront of neuroscientific research. Actually, it is a non-issue in prominent theoretical accounts of visual word processing (Dehaene & Cohen 2011; Price & Devlin 2011) which deny the existence of a localized orthographic word lexicon. It was an issue for the meta-analyses of Jobard et al. (2003) and Taylor et al. (2013) which were based on studies of word-pseudoword comparisons. However, as pointed out, the meta-analyses arrived at a negative conclusion with respect to an orthographic lexicon function of the critical vOT reading region.

The present review of neuroimaging studies arrives at the opposite conclusion. Our findings provide evidence for the position that the left vOT reading region not only hosts memories of letters and frequently occurring letter sequences, but also

for the specific letter sequences of entire words. This is the case not only for short words as in the model of Dehaene and colleagues (2005; Dehaene & Cohen 2011) but also for long ones. This proposal assumes that the neuronal codes for letters and sublexical letter sequences in the vOT region constitute the “features” which get “unified” into stable whole-word memories in response to repeated readings of certain words.

Our attribution of the orthographic word lexicon to the left vOT reading region will certainly not be the final word on this issue, but the proposal links neuroscientific research to the orthographic lexicon assumption of the well-known cognitive dual-route models in visual word processing (e.g., Coltheart et al. 2001) and in spelling processes (e.g., Hillis & Rapp 2004). In this sense, it may constitute a step towards a unified neurocognitive account of reading and spelling.

If the left vOT in typical readers is the prime reading region including the orthographic lexicon, then one would expect that dyslexic readers suffer from a dysfunction of this region. Support for this expectation comes from two functional imaging studies (Richlan et al. 2010; Wimmer et al. 2010) which extended the already mentioned work of Kronbichler et al. (2007) and Schurz et al. (2010) by including dyslexic samples. To summarize, both Richlan et al. and Wimmer et al. found that the dyslexic readers failed to exhibit those vOT activation patterns in response to visual words which we interpreted as evidence for an orthographic lexicon function in the present chapter. Specifically, the dyslexic readers of Richlan et al. failed to exhibit the length by lexicality interaction effect on left vOT activation of the typical readers. In particular, no length effect on vOT activation was found neither for words nor pseudowords despite marked behavioral length effects. The dyslexic readers of Wimmer et al. failed to exhibit the orthographic familiarity effect of the typical readers on left vOT activation although this effect was evident in their decision latencies and in left frontal regions. These findings speak for a different neural organization of visual word processing in dyslexic readers due to a dysfunction of the left vOT reading region. A dysfunction of this region in dyslexic readers finds also support in quantitative meta-analyses from our lab of functional imaging studies (Martin, Kronbichler, & Richlan 2016; Martin, Schurz, Kronbichler, & Richlan 2015). These meta-analyses found convergence between dyslexia studies with respect to underactivation of the left vOT in response to visual words. Based on these findings it stands to reason that a dysfunction of the left vOT is the main neural cause of developmental dyslexia. This differs from accounts – inspired by the phonological deficit explanation of dyslexia – which assumed a dysfunction of left posterior language regions as prime cause of dyslexia (e.g., Pugh et al. 2000).

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Chapter 5

Simple View of Reading (SVR) in Different Orthographies: Seeing the Forest with the Trees



R. Malatesha Joshi

Abstract One of the influential models of reading development may be the Simple View of Reading (SVR), according to which Reading Comprehension can be explained by two important components, decoding (D) and linguistic comprehension (LC) and is expressed as $RC = D \times LC$. Decoding refers to pronunciation of the word and listening comprehension refers to understanding of the text when read by others and listening to the text. This chapter reviews various studies in support SVR from monolinguals, second language learners and conducted in various orthographies of different orthographic depth. Findings from these studies support of SVR and the model is applicable for assessment and intervention by identifying the weak component in the model (e.g., decoding or listening comprehension) and providing systematic instruction to the identified weak component. Future research directions are also provided.

Keywords Decoding · Listening comprehension · Orthography · Reading comprehension · Second language learners

5.1 Introduction

One of the influential models that is useful in the assessment and intervention of reading problems is the Simple View of Reading (SVR) proposed by Gough and Tunmer (1986) and Hoover and Gough (1990), according to which the two most important components of reading are decoding and comprehension. The relationship among decoding, linguistic comprehension, and reading comprehension is expressed as $RC = D \times LC$, where RC is reading comprehension, D is decoding, and LC is linguistic comprehension. Thus, both decoding and linguistic comprehension are important to comprehend the written materials and thus, if D is zero, then RC

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will be zero, and if LC is zero, then also RC will be zero. Various studies have shown that SVR can account for approximately 40–80% of the variance in reading comprehension for readers ranging from 2nd through 10th grade among English speaking children. In addition to English-speaking children, we have tested SVR model with students from Spanish, Chinese, and Hebrew backgrounds as well as in bilinguals by administering decoding, LC, and RC measures at various grade levels. Similar to the findings of English-speaking children, a significant variance in RC has been explained by the two factors: D and LC. However, the percentage of variance is different at different grade levels and in different orthographies and the results are explained in terms of the nature of the orthographic depth – whether it is transparent or shallow. The results have important implications for literacy instruction. In opaque languages like English and Chinese, systematic decoding instruction should be continued for a longer period of time and comprehension instruction can be introduced at earlier time for Spanish-speaking children. Further, different decoding systems have to be applied in Hebrew literacy instruction due to its pointed (vowelized) and unpointed (unvowelized) system of writing.

Literacy, the ability to read and write, is basic for survival and hence research from various specialties have attempted to understand the components of reading and writing. One of the influential models that has practical utility in identifying and remediating reading problems is the ‘Simple View of Reading’ (SVR) postulated by Gough and his colleagues (Gough & Tunmer 1986; Hoover & Gough 1990). Even though, the concept of SVR is accepted in general, there is still some discussion about decoding whether it includes non-word reading or real word reading and whether it refers to accuracy and also speed. Additionally, the role of vocabulary is also discussed in the context of SVR, whether it should be a separate component (Braze, Tabor, Shankweiler, & Mencl 2007) or vocabulary influences indirectly through word recognition and reading comprehension (Protopapas, Simos, Sideridis, & Mouzaki 2012; Tunmer & Chapman 2012). Similarly, there is discussion about whether to include fluency as an additional component of SVR (See, Adlof, Catts, & Little 2006; Joshi & Aaron 2000). Nevertheless, it is widely accepted that the two important components of reading are decoding and comprehension and much of the variance in reading comprehension can be explained by these two components: decoding and linguistic (listening comprehension, LC). For the diagnostic purposes SVR has been applied to classify poor readers into those with decoding problems but adequate comprehension, exhibiting dyslexia-type syndrome. Aaron, Joshi, and Williams (1999) administered measures of decoding, listening comprehension, and reading comprehension to about 200 students in grades 3, 4, and 6. Applying SVR model, they found that approximately 7% of the students exhibited good decoding ability but their comprehension – both listening and reading – was not on par with their decoding ability, exhibiting hyperlexia-type syndrome. Additionally, about 8% of students were poor in decoding skills but adequate comprehension skills, who could be referred to as displaying dyslexia-type syndrome. Further, another 8% of students had both decoding and

comprehension problems and can be referred to as either low ability readers or Garden variety poor readers. Further, SVR model has also provided support for instructional applications. Contrary to using only one type of reading instruction to all poor readers, Aaron, Joshi, Gooden, and Bentum (2008) first identified the weak component of reading, whether it was decoding or comprehension and then provided systematic decoding and comprehension instruction to both the groups for 12 weeks and compared to another group of poor reader who were receiving the business-as-usual instruction in the schools. After the completion of 12 weeks, those with decoding problems showed significant gains in reading when provided with decoding instruction but did not improve when provided with systematic comprehension instruction. Similarly, comprehension instruction was more helpful for those with comprehension problems. Poor readers who did not receive differentiated instruction did not make any significant gains in reading comprehension. Hence, in order to improve reading among poor readers, first the poor component based on SVR has to be identified and then should be provided with systematic and evidence-based instruction. Thus, SVR is a simple, yet a valuable, model to identify and improve reading problems.

The effectiveness of the SVR model is further explored in this chapter by addressing issues of the contribution of decoding and comprehension at different grade levels, the role of orthography and second language learners.

Hoover and Gough (1990) presented SVR based on English-Spanish bilinguals in grades 1–4 and found that about 50–60% of the variance in reading comprehension was explained by decoding and linguistic comprehension, even though the percentage varied slightly at different grade levels. However, SVR provided an alternate way to identify reading disabilities without administering the traditional IQ measures, which, in most studies, have accounted for only about 25% of the variance.

Tilstra, McMaster, van den Broek, Kendeou, and Rapp (2009) tested SVR model among students in grades 4, 7, and 9. A couple of interesting findings from their study was that the amount of variance decreased at the upper grade levels and also the percentage of variance contributed by decoding decreased at upper grade levels but the variance contributed by comprehension increased with higher grade levels. Both the findings can be explained on theoretical grounds. The amount of variance that SVR can explain at higher grade levels becomes lower as reading comprehension at higher grade levels may require more background knowledge by wide reading and other factors such as syntax and knowledge of idioms. Additionally, decoding contributes more at early grade levels because many students are still at the ‘learning to read’ stage and at upper grade levels, when students are in the ‘reading to learn’ stage, comprehension plays an important role. Similar findings were also reported in a study by Joshi, Tao, Aaron, and Quiroz (2012) based on students from 2, 3, and 4.

5.2 SVR in Different Orthographies

The above findings were based on English-speaking participants. Does the same pattern hold true for other orthographies? Before answering this question, it is better to clarify some terms. The world's writing system can be broadly classified into three broad categories based on the smallest written unit – alphabetic, syllabic, and morpho-syllabic writing system. Letter is the smallest written unit in the alphabetic writing system; a syllable is the smallest written unit in the syllabic writing system, and a morpheme (as a character) is the smallest written unit in the morpho-syllabic writing system. Syllabic writing system is further sub-divided into syllables which cannot be further broken down into phonemic representation such as Kana of Japanese and syllables where the phonemic representation can be identified such as Korean Hangul. Examples of alphabetic writing system include English, Spanish, and French and Chinese Kanji is an example of morpho-syllabic writing system. Additionally, orthographies are also classified as transparent or shallow and opaque or deep orthographies and are referred to as 'orthographic depth'. Transparent orthographies have almost 1:1 correspondence between graphemes and phonemes such as Finnish and Spanish orthographies, while opaque orthographies may not have 1:1 correspondence between graphemes and phonemes such as French and English. However, it is better to view this classification as a continuum rather than as belonging to one or the other category. Thus, Finnish and Spanish may be at the one end of the spectrum near the transparency end and English and French may fall at the other end of the spectrum near the opaque end among alphabetic languages. By administering various reading measures in about 13 European orthographies, Seymour, Aro, and Erskine (2003), found that it may take approximately 2 years of formal instruction to master basic decoding skills in English, while it may take only about 1 year of formal instruction in transparent orthographies like Spanish and German.

Due to the interest in the orthographic influences in literacy development, SVR has been applied in different orthographies. For instance, Megherbi, Seigneuric, and Ehrlich (2006) applied SVR among French speaking children in grades 1 and 2 and found that more than 50% of the variance in reading comprehension was explained by decoding and linguistic comprehension. Similar to English speaking children, decoding contributed more at grade 1 and linguistic comprehension contributed more at grade 2. On the continuation scale of orthographic depth, French is considered less opaque than English. Even though SVR has been examined and has been found to be useful in various orthographies, the pattern is slightly different depending on the transparency of the orthography. Among more transparent orthographies such as Greek, Swedish, Finnish, and Norwegian, decoding plays a lesser role at earlier grade levels compared to English speaking children. For instance, in a study by Joshi et al. (2012), the performance of third grade Spanish speaking children resembled the performance of English speaking children in grade 4. The fact that Spanish speaking children had already mastered the basic decoding skills earlier than English-speaking children was explained in

terms of the transparency of Spanish orthography. In many of the transparent orthographies studied thus far, the findings of Seymour et al. (2003) have been found to be true as decoding contributes less even at earlier grade levels compared to English and LC starts contributing more even from early grade levels again compared to English. For instance, in the study by de Jong and van der Leij (2002) with Dutch speaking children, much of the variance in RC was explained by LC after grade 1. Similar results have also been reported among Greek-speaking children (Protopapas et al. 2012); Italian children (Tobia & Bonifacci 2015); in Portuguese (Cadime et al. 2017) and in Finnish (Torppa et al. 2016). Høien-Tengesdal and Høien (2012) validated SVR in Norwegian and Swedish orthographies, where they found about 50% of the variance is explained by D and LC. The earlier version of Turkish orthography was heavily influenced by Persian and Arabic orthographies till 1928 when Mustafa Kemal Atatürk, first president of Turkey, changed the script to Latin. Turkish orthography is transparent and SVR has been validated by Babayiğit and Stainthorp (2011) in Turkish also. Virtually, in all the orthographies presented till now, decoding and linguistic comprehension can explain about 50% of the variance, while IQ scores explain only 25% of the variance for RC, thus, SVR is a better model to apply for the identification as well as intervention of reading difficulties by identifying the weak component and then providing appropriate evidence-based systematic and explicit instruction.

5.3 SVR in Non-European Orthographies

The above review referred to alphabetic languages of varying orthographic depth, mostly from European languages. Would the SVR be applicable in other non-European languages? We explored this hypothesis by applying SVR to Hebrew and Mandarin Chinese. Hebrew is a Semitic language and has two forms of writing – vowelized and unvowelized. Children in early grade levels are introduced words with the vowels present (vowelized) and after about grade 3, vowels are removed (unvowelized) and students are expected to read without the presence of vowels. (To illustrate this concept of vowelized and unvowelized from the perspective of English orthography, children will be exposed to words like CAT, CUT, and COT, with the vowels present in early grade levels and after about grade 3, students will have only CT for CAT, CUT, and COT and students have to read the word mainly based on the context). We (Joshi, Ji, Breznitz, Amiel, and Yulia 2015) explored whether SVR is also applicable for Hebrew orthography by administering various decoding, listening comprehension and reading comprehension measures for students in grades 2–10. It was found that between 37% (at Grade 6) to 70% (at Grade 4) of the variance in RC were explained by decoding and linguistic comprehension. Further, decoding made more contribution at early grade levels and LC made more contribution to RC at upper grade levels. These findings are similar to what is reported in English and other alphabetic languages. Due to the nature of Hebrew orthography, decoding contributed for a longer period of time

compared to many of the transparent orthographies such as Spanish, Finnish, and Italian. Nevertheless, D and LC explained much of the variance in RC in Hebrew orthography. SVR was also found to be applicable in another Semitic language – Persian (Sadeghi, Everatt, & McNeill 2015). Both Persian and Hebrew are written from right to left and similar to Hebrew, in Persian also vowels are omitted in the written text from the beginning of upper elementary grade levels.

Mandarin Chinese is considered a morpho-syllabic writing system, where the basic unit is a character which is a syllable and contains a morpheme; it is quite different from many other writing systems. In one of our studies (Joshi et al. 2012), it was found that SVR can also be applied to Mandarin Chinese as character recognition (decoding) and listening comprehension accounted for much of the variance in RC even in Chinese. However, due to the complexity of the character recognition with various stroke patterns, word recognition (decoding) contributes more even at the fourth grade level. Similar results in Cantonese Chinese have also been reported by Yeung, Ho, Chan, Chung, & Wong (2013).

Akshara orthography, derived from the Brahmi script, mostly used in the Indian subcontinent, is sometimes considered as alphabetic, syllabic, alpha-syllabic, abugida. However, recently, there is a push for akshara orthography to be considered as a separate category (Share & Daniels 2015). SVR was tested in two of the akshara orthographies – Kannada and Telugu – among children from the slum areas of metropolitan cities in India. The results showed that even though about 50% of the variance in RC was explained by D and LC, decoding plays an important role even at upper grade levels (Nakamura, Joshi, & Ji *in press*; Nakamura, Koda, & Joshi 2014). The results were attributable to the complex writing of aksharas. Even though, akshara orthography is highly transparent, each akshara is visually complex and even a ‘dot’ (.) can change the sound and meaning.

Korean orthography, referred to as Hangul, is considered a transparent alpha syllabary, where the basic unit is at the syllable level but the phonetic element can be identified in the syllable. Even in Korean orthography, SVR has been found to be applicable as demonstrated by Kim, Park, and Wagner (2014).

5.4 SVR Among Second Language Learners

Even though the first major study to validate SVR was based on the data from English-speaking children with Spanish background (Hoover & Gough 1990), recently several studies have reported that SVR is also applicable to second language learners. Geva and Farnia (2012), in a longitudinal study of grades 2–5 from the same school systems in Canada, found that SVR is applicable for both monolingual English speakers (EL1) and those who were learning English as a second language (ELL) from various first language background. The amount of contribution of D and LC were about the same for the groups and decoding was more important at early grade levels. However, LC contributed more to RC earlier among EL1 compared to ELL.

Erbeli and Joshi (submitted) divided seventh graders into less skilled and advanced readers and administered various decoding and listening comprehension measures. Results from the Structural Equation Modeling (SEM) showed that about 60% of the variance in RC was explained by D and LC measures for both skilled groups, LC contributed much earlier for the advanced skill readers than the lower skilled readers. This finding demonstrates that in addition to decoding, additional skills such as good vocabulary, background knowledge and syntactical knowledge are required for comprehension.

The above two studies were related to English as a second language and English is generally considered as having a deep orthography. However, studies have produced similar results when learning a second language which has a transparent orthography. Verhoeven and van Leeuwe (2012) examined the applicability of SVR in Dutch as a second language. Dutch is considered a transparent orthography compared to English orthography. The authors found SVR to be valid for both Dutch as the first language (L1) as well as Dutch as a second language (L2). However, similar to the results found in the studies by Geva and Farnia (2012) and Erbeli and Joshi (submitted), even though decoding skills were similar in both the language groups, listening and reading comprehension had lagged behind among L2 participants than L1 participants. In a recent study by Bonifacci and Tobia (2017), it was found that SVR was applicable for Italian as a second language also. Similar to the findings of other studies from different orthographies, LC contributed more to RC from early grade levels in Italian.

5.5 Conclusions

As presented in the chapter, various findings from different orthographies in both monolinguals and second language learners, SVR has found to be valuable in explaining the variance in RC through D and LC. Even though, the researchers have used different types of assessments to measure D, LC, and RC, the results are unequivocal in demonstrating that RC consists mainly of two important components D and LC. For instance, some researchers have used non-word reading tasks and some have used real word reading to measure decoding and similarly some researchers have used different procedures such as cloze techniques, asking questions from the passages to measure LC and RC. Other factors such as working memory, fluency, and vocabulary may explain some of the variance, but the findings are not as clear cut as the two important components of D and LC. Even though some researchers have argued that an additive formula, $RC = D + LC$ can be as useful as the multiplicative model $RC = D \times LC$, which was originally proposed by Gough and his colleagues, the additive formula can be rejected on rational grounds. According to the multiplicative model, if $D = \text{zero}$, then, RC will be zero and if LC is zero, then also RC will be zero demonstrating the importance of both D and LC

components. However, in the additive model, if D is zero, then, theoretically, RC can have some value if the individual has some LC. However, this may not be possible in reality to comprehend passages when read without some decoding ability.

SVR has diagnostic value without using the IQ measures to assess reading difficulties and additional advantage of SVR is that once the weak component is identified, then appropriate systematic instruction can be provided to improve the weak component. Diagnosis based on IQ and achievement discrepancy formula may not have the same advantage. Additionally, SVR model has been found to be applicable based on the studies with monolinguals and second language learners, in orthographies that are transparent or opaque, and in longitudinal studies also (Torppa et al. 2016). Further, SVR also has received support from genetic studies that have shown that decoding and comprehension are influenced by different genetic components (Keenan, Betjemann, Wadsworth, DeFries, & Olson 2006). Future studies may explore the applicability of SVR in bilinguals as well at various grade levels and in different orthographies. Additionally, most of the studies have explained about 50% of the variance in reading comprehension, while the IQ scores, which is used in the assessment of reading problems explains only 25% of the variance. Perhaps future studies can explore contributions of other factors such as working memory, speed, and vocabulary to the SVR model.

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Part II
**Developmental Dyslexia: From Basic
Functions to Higher Order Cognition**

Chapter 6

The Role of Rodent Models in Dyslexia Research: Understanding the Brain, Sex Differences, Lateralization, and Behavior



Albert M. Galaburda

Abstract Developmental dyslexia, a reading disorder that piggybacks on some of the highest human cognitive functions, can be studied in animal models. This is because dyslexia can be decomposed into endophenotypes, some of which are present in one form or another in non-human animals, and because dyslexia has a developmental trajectory beginning at a time when the human being is not predominantly a cognitive being. In this chapter I provide four examples of animal research where insights about the pathophysiology of dyslexia can be obtained. The first one concerns cortical abnormalities initially reported in autopsied dyslexic brains and modeled in rodent brains. The second one models brain lateralization, considered to be abnormal in dyslexia, the role of cilia in somatic lateralization, and possible roles of dyslexia susceptibility genes in cilia structure and function. The third example considers sex differences in dyslexia and reports sex-differences in developmental plasticity in subcortical structures and in behavior in rodents after induction of cortical malformations. Finally, the fourth example deals with the dichotomy of cortical versus subcortical involvement in dyslexia by showing developmental interactions of cortical and subcortical structures in early cortical and genetic manipulations in rats and mice.

Keywords Phonological grammar · Auditory processing · Heterotopia · Dysplasia · Cerebral cortex · Thalamus · Brainstem · Cochlear nucleus · Male-female differences · Asymmetry · Cilia · Dyslexia risk genes

6.1 Introduction

Developmental dyslexia, or as it was known then, congenital word blindness (Morgan 1896), was noted for the first time in the late nineteenth century, and became a topic of discussion during the first decades of the 20th. Although initial

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understanding was based on adult brain models of reading, in turn derived from the analysis of brain injury (Dejerine 1892; Hinshelwood 1911; Morgan 1896), this gradually gave way to behavioral and increasingly developmental explanations (Bradley & Bryant 1978; Liberman, Mann, Shankweiler, & Werfelman 1982; Liberman, Shankweiler, Orlando, Harris, & Berti 1971). In the 1970s and 1980s, there was a renewed effort to find brain-based explanations for acquired and developmental cognitive disorders (Galaburda & Kemper 1978, 1979; Galaburda, Sherman, Rosen, Aboitiz, & Geschwind 1985; Geschwind 1965a, 1965b; Geschwind & Galaburda 1985a, 1985b, 1985c; Hier, LeMay, Rosenberger, & Perlo 1978), and the first findings in the brains of dyslexics were published in the neurological literature then (Drake 1968; Galaburda & Kemper 1978, 1979; Galaburda et al. 1985; Hier et al. 1978). Since that time, there have been many advances in our understanding of the mechanisms¹ and some etiologies² underlying this common learning disability. Discoveries have taken place at most levels of biological understanding, from risk genes, through potential cellular and molecular mechanisms in which these genes function, as well as anatomy, functional brain circuits and networks involved in language and reading, to cognitive descriptions, to behavioral characterizations and treatment approaches (Bakker 2006; Elnakib et al. 2014; Galaburda, LoTurco, Ramus, Fitch, & Rosen 2006; Hämäläinen, Salminen, & Leppänen 2013; Johansson 2006; Kere 2014; Kershner 2016; Lachmann, Steinbrink, Schumacher, & van Leeuwen 2009; Ramus 2004; Snowling & Melby-Lervag 2016; Valdois, Bosse, & Tainturier 2004; Vandermosten, Hoeft, & Norton 2016). These gains have benefitted greatly from advances in cognitive science, cognitive psychology, neuroscience, cognitive neuroscience, genetics, and molecular biology, as well as in educational psychology and the field of special education.

Although educational, behavioral, cognitive, and imaging research have shed light on mechanisms, fundamental etiology instead benefits from descriptions at genetic, subcellular, cellular, and circuit levels; rodent models can also shed light on the behaviors on interest, especially if behavioral endophenotypes can be found that are implemented in the simpler brain. Such studies of rodent behavior place constraints on the types of psychological theories that can be formulated regarding reading acquisition and dyslexia in humans. *In this paper, I will provide four examples of ways by which rodent research has contributed to knowledge about neural and behavioral features underlying dyslexia.*

¹By “mechanisms” I mean the nature of the dysfunction of the system in question. This dysfunction can be analyzed at multiple levels, from the molecular to the networks levels.

²By “etiology” I mean the event and associated factors that started the deterioration of the system and rendered it dysfunctional. These could have been a genetic mutation, a virus, a stroke or some other type of brain injury, a toxic-metabolic derangement, or a neoplastic transformation.

6.2 Nature of Cortical Abnormalities in Developmental Dyslexia

Our laboratory reported the first anatomical findings in dyslexic brains, which have been subsequently confirmed, at least in part, in structural imaging studies (Chang et al. 2005, 2007; Elnakib et al. 2014; Galaburda & Kemper 1979; Galaburda et al. 1985; Pugh 2006; Vandermosten et al. 2016). The neuropathologic findings consisted of alterations of cerebro-cortical architecture that resulted from abnormal neuronal migration at around mid-gestation. Most of the anomalies were layer 1 heterotopias, comprising nests of around 100 neurons and glia in the normally almost neuron free first layer of the cortex, often with subjacent disordered cortical lamination (Figs. 6.1 and 6.2) (Galaburda & Kemper 1979; Humphreys, Rosen, Press, Sherman, & Galaburda 1991). Occasionally we saw frank microgyria, which often occurs together with layer 1 heterotopias in cases of human developmental

Fig. 6.1 Layer 1 heterotopia in a human dyslexic cortex. Neurons are stained with cresylechviolett. The neuron cloud lying between the arrows contains cells that have migrated abnormally beyond their standard locations in the subjacent cortical layers. These malformations can occur spontaneously in the dyslexic cerebral cortex and can be induced in rodent brains for further analysis (Fig. 6.2)

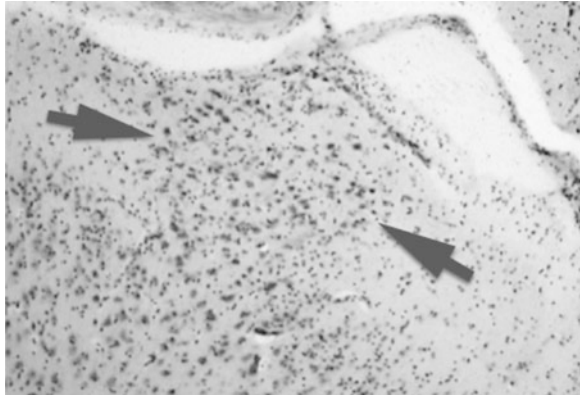
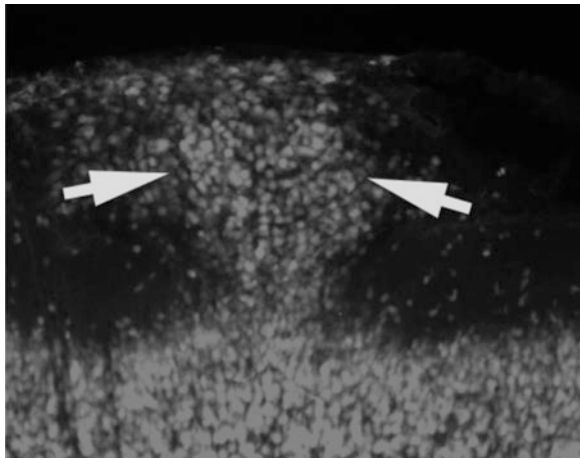


Fig. 6.2 Layer 1 heterotopia experimentally induced in the rat cerebral cortex. Neurons are stained with red fluorescent protein. The neuron cloud lying between the arrows represents cells that have migrated abnormally beyond their standard locations in the subjacent cortical layers. These malformations occur spontaneously in the dyslexic cerebral cortex (Fig. 6.1)



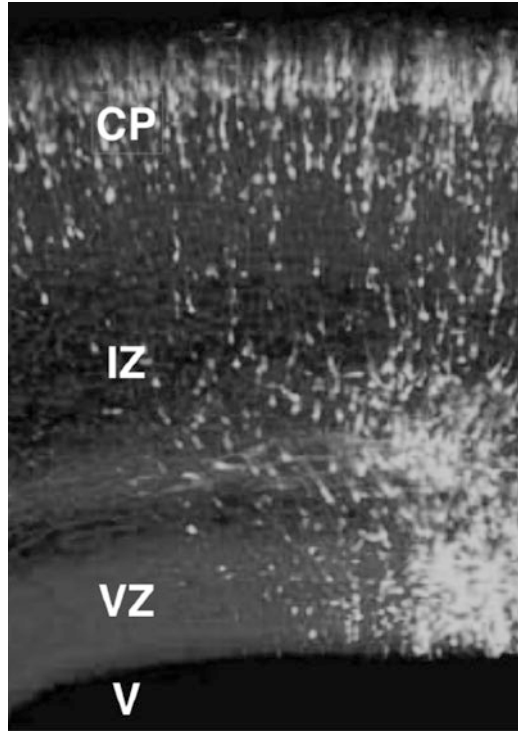
neuropathology, and which in the experimental rat model can be attributed to the same underlying mechanism (Rosen, Burstein, & Galaburda 2000). The malformations were more frequent in the left perisylvian cortex, which suggested a causal link to the language difficulties, as the perisylvian regions contain the classical language areas of the brain. Yet, however tempting this conclusion is, causal links require modeling, and in the case of malformations, modeling in animals. We chose first to model in the rodent brain, because there was precedent in the literature showing that comparable malformations to those seen in the dyslexic brains could be experimentally induced in the rat brain (Dvorak, Feit, & Jurankova 1978).

Our first rat model consisted in placing a freezing probe on the skull of the newborn rat pup for a few seconds, which resulted in a four-layer microgyrus in the cortex underlying the freezing probe (Rosen et al. 2000). Many studies using this method showed that these small malformations change the cellular profile of subjacent cortical layers and of connectionally related cortex, both in the ipsi- and contra-lateral cortex, and thalamus, and that cortico-cortical connections are rendered anomalous by the microgyria induction (Rosen & Galaburda 2000; Rosen, Galaburda, & Sherman 1989; Rosen, Jacobs, & Prince 1998; Rosen, Mesples, Hendriks, & Galaburda 2006; Rosen, Sherman, & Galaburda 1994; Rosen, Windzio, & Galaburda 2001). It would have been impossible in the autopsy dyslexic brains to get this type of detailed information regarding secondary cellular and connective changes in the brain related to the malformation, but especially about the causal relationship between the malformation and the cellular and connective changes. As it turns out, we could furthermore not document a causal link between the induced cortical malformation in the rat and the auditory processing deficits these animals exhibit. Instead, we found that the auditory deficits more likely reflected secondary subcortical changes following the induction of the cortical malformation (Herman, Galaburda, Fitch, Carter, & Rosen 1997). Here, again, it was necessary to rely on rodent research for this conclusion.

It has long been suspected that developmental dyslexia is to a large extent heritable, but it took the introduction of powerful genetic epidemiological methods to discover candidate genes. During the course of the rat freezing injury research, several such studies uncovered genes that increase the probability of dyslexia – dyslexia susceptibility genes – and some of them were confirmed in multiple studies, for instance *DCDC2*, *KIAA0319*, and *DYX1C1* (for a review, see Kere 2014). These discoveries provided an opportunity to switch from the rat perinatal freezing lesion model for producing neuronal migration anomalies to a more naturalistic, genetically-based model.³ We began with the short hairpin RNA interference (shRNAi) model perfected by our collaborator, Dr. Joseph LoTurco, at the University of Connecticut (Bai et al. 2003). In this model a plasmid containing a small sequence of RNA is made to match the native RNA of a particular gene one wishes to block, in this case a rodent homolog of a dyslexia susceptibility gene,

³There has never been evidence to show that the dyslexic cortex is lesioned or otherwise injured during its protracted course of neuronal migration *in utero*. Instead, the cause of these malformations in this population is likely to have a genetic origin.

Fig. 6.3 Section of rat cerebral cortex after electroporation of a short-hairpin RNA directed against the dyslexia susceptibility gene *DYX1C1*. Transfected cells also express green fluorescent protein (white in the photograph), which disclose their positions. Neurons initially blocked in migration in the ventricular zone (VZ), are now starting to migrate upward through the intermediate zone (IZ) into the cortical plate (CP). V – ventricle



which in effect paralyzes gene function for several days. This manipulation knocks down gene expression immediately prior to protein translation,⁴ and allows for the study of the gene's role during cortical development. It is possible to demonstrate zero protein production, and the functional knock-down is so late in the process that there is no time for the neuron to find a substitute protein.

Paralyzing plasmids were injected into the ventricle of the fetal brain *in utero*, and then electroporated⁵ into the ventricular germinal zone, where neuronal progenitors replicate and young neurons then initiate their migration to the developing cerebral cortex. Neurons taking up the plasmids (transfected neurons) develop a dysfunctional dyslexia risk gene. As the transfected young neurons migrate, their trajectories can be followed because the paralyzing plasmid is coupled with a transcript that codes for a fluorescent protein that reports its location (Fig. 6.3). In this way, we were able to demonstrate that knocking down dyslexia gene expression

⁴This reversible gene lesion leads to no protein production on-line, and there is no developmental time available for a possible compensation by other, unaffected genes, as might be the case in early embryonic gene deletions in knock-out mice.

⁵Electroporation is a technique during which an electrical field is applied to cells, in this case the brain with its neurons, in order to increase the permeability of the neuronal membrane, thus allowing chemicals, drugs, DNA or RNA to be introduced into the cell.

in cortical neurons lead to neuronal migration arrest. The bulk of the transfected neurons, and many others that had not in and of themselves been transfected, remained in the ventricular zone for several days, after which many were able to reach the cortex, albeit to the wrong layers (Adler et al. 2013). The original reports in dyslexic human brains stressed the finding of neuronal migration anomalies; these rat findings established for the first time a link between neuronal migration anomalies and dysfunction of dyslexia susceptibility genes in the rat; once more, the human dyslexic neuropathological observations alone could not have established this causal link. It should be stressed here again, that tweaking the function of a dyslexia susceptibility gene homologue in the rat brain may be slightly more naturalistic than placing a freezing probe on the skull of a newborn rat (see above), but it is still highly unlikely that this is the mechanism that is functioning in the brain of a dyslexic human during development. Therefore, it is important to emphasize that the neuronal migration findings from shRNAi may not be altogether generalizable, although they probably point to an effect of these dyslexia risk genes on cortical development. It is not even known how generalizable the finding of neuronal migration anomalies in the cortex of human dyslexics is, from the limited number of such brains that have been studied at autopsy. In fact, with the exception of the cases published by Chang and colleagues (Chang et al. 2005, 2007), where the cortical malformations are more severe than the ones we have been discussing here, MR imaging of living human dyslexic brains have not thus far been capable of disclosing cortical malformations that are as small as layer 1 heterotopias. So, we do not know how prevalent subtle cortical malformations are among dyslexics in general. Additional animal research suggests that abnormal cortical anatomy per se may not be the only way to render a dyslexic cortex dysfunctional.

Another commonly used methodology for studying the effects of genes on brain structure and function is that of targeted gene deletion, a powerful procedure that was rewarded with the Nobel Prize for its inventors (Capecchi 2005). These experiments are most commonly performed in early mouse embryos. Animals in which one or more genes have been deleted are known as “knockout mice.”⁶ The whole gene need not be removed, since most commonly removing one or a few coding regions of the gene is all that is needed to silence the gene completely.

Our collaborators deleted one exon in the mouse *DCDC2* gene, which resulted in complete lack of protein production.⁷ The brains of the adult genetically modified mice were examined in detail, including layer specific markers, and no abnormalities in neuronal migration were detected (Che, Truong, Fitch, & LoTurco 2016). In contrast to knocking down gene expression in the shRNAi rat models, which

⁶It is also possible to insert non-native genes into genetically modified mice.

⁷Although mouse deletion models are more naturalistic and imitate the human situation better than shRNAi, caution should be used against over-interpreting these deletion results, because the genetic abnormalities in dyslexia risk genes in dyslexic individuals have not involved the exonic portion of the genes but regulatory portions instead. These knock-out models, therefore, may be a more severe form of the gene defect in dyslexic humans, in whom protein production may only be partially altered.

did indeed produce neuronal migration abnormalities from *DCDC2* knockdown, deleting the *DCDC2* gene earlier in development in the mouse did not lead to such abnormalities. One could say that the animals can better compensate for the genetic lesion when it occurs earlier in development; however, another explanation could be that in one case the experiments were carried out in the rat, whereas in the other the animal model was the mouse. Even in our early freezing lesion experiments (see above), layer I heterotopias were more difficult to induce in the mouse, and we never saw a microgyrus after such a lesion (unpublished observations). One problem that occurs in shRNAi experiments, but not in knockout mice, is off-target effects. Small sequences of RNA can be shared among many genes, so it is possible that unknown genes, not the target gene, are also knocked down by the electroporation – genes that could affect the phenotype in question. One tries to control for this occurrence by rescuing the normal phenotype with overexpression constructs for the tested gene.⁸ However, even with this maneuver, the phenotype may not be completely rescued and one is left wondering whether one has discovered the function of the gene in question or that of some unknown gene or genes.⁹ Therefore, it can be argued that gene deletions in knockout mice may still be more reliable than rat shRNAi experiments because the former do not show off-target effects and the gene modification is early. Be that as it may, these statements illustrate the difficulty one sometimes encounters when using rodent models for understanding human brain development.

The anatomy of the cortex of the *DCDC2*-deleted mouse was normal, but the knockout provided an unexpected finding about the cortex, which was not normal (Che, Girgenti, & LoTurco 2014; Che et al. 2016). In the normally appearing *DCDC2*-deleted mouse cortex neurons were physiologically abnormal, showing evidence of hyperexcitability. The subcellular explanations for this increase noisiness could have included abnormalities in ion channels or excessive synaptic events. A transcriptome analysis of the deleted neurons indicated that the GRIN2B NMDA excitatory glutamatergic receptor RNA was upregulated, suggesting the likely synaptic mechanism for the increased neuronal excitability. Furthermore, exposure of the neurons to the NMDA receptor blocker APV (a.k.a. AP5) reduced the noise to normal levels, confirming the hypothesis implicating excessive synaptic events and refuting the hypothesis that the deletion affected ion channels (Che

⁸After knocking down protein translation, which produces the abnormal phenotype, the tissue is flooded with the lost protein by overexpressing it, which would cause the abnormal phenotype to normalize. If the abnormal phenotype is the result of blocking other, unknown, proteins from expressing, then replenishing with the known protein would not be expected to rescue the normal phenotype.

⁹The plasmid made for shRNAi experiments must be small in order to avoid an immunological reaction to it by the animal. However, the smaller a sequence of RNA nucleotides, the more likely it is that they will paralyze other genes in addition to the target gene (the extreme case would be a plasmid with only one nucleotide, where all genes could in principle be affected because they all share the same nucleotide). These, off-target effects can make shRNAi results uninterpretable, even if the experimenter is able to mostly rescue the phenotype by replacing the lost protein in the control experiment.

et al. 2016). Not only did these experiments demonstrate that anatomical changes were not a necessary outcome of deactivation of a dyslexia risk gene, but they also provided for a possible physiological mechanism for the temporal processing auditory abnormalities demonstrated in human dyslexics, freeze-lesioned and shRNAi electroporated rats, and knockout mice. The claim here would be that neural noise interferes with fine sound mapping required for rapid auditory and language processing (Dale et al. 2010).

6.3 Asymmetry and Lateralization

Another characteristic of the dyslexic brain is the presence of deviant anatomical asymmetries in the posterior language areas in the temporal lobe. All five autopsy brains reported by Galaburda and colleagues showed symmetric *plana temporale*, when in the general populations this occurs in only 24% of brains (Galaburda et al. 1985; Geschwind & Levitsky 1968). The probability that this would occur randomly in 5 brains in a row is about 8 in ten-thousand. It has been difficult to expand these findings to larger numbers of dyslexic brains imaged during life with magnetic resonance imaging, because it is challenging to identify on the images exactly the same landmarks used in the *post-mortem* brains. Several studies attempted to do this (Eckert & Leonard 2000; Hugdahl et al. 1998; Hynd & Semrud-Clikeman 1989; Larsen, Høien, Lundberg, & Odegaard 1990; Leonard et al. 1993), but the results were inconsistent. When the post-mortem landmarks used by Geschwind and Levitsky (1968) were finally reproduced in a large MRI study, Altarelli, Ramus and collaborators at the École Normale Supérieure in Paris reported a bias toward right asymmetry or absence of asymmetry in their dyslexic sample (Altarelli et al. 2014).

Geschwind and Galaburda (1985a, 1985b, 1985c) proposed a mechanism by which asymmetry would vary in the population to produce individuals with less leftward asymmetry, more rightward asymmetry or lack of asymmetry altogether. For this to occur they conjured up an effect of testosterone acting *in utero*, whereby left hemisphere structures related to handedness and language would be inhibited in their development. This testosterone effect presumably explained the higher prevalence of left-handedness in boys, as well as the more frequent occurrence of left-hemisphere (language) based learning disabilities. Marian Annett (Annett 1964), on the other hand, proposed that a gene (or genes) was responsible for right-handedness, the so-called Right Shift Theory, and that the absence of the gene led to random handedness, with half of those missing the gene being left-handed. However, although some genes have been shown to impact on lateralization (Sun et al. 2005; Sun & Walsh 2006), no fundamental explanation for brain asymmetry and asymmetry variation in the population and in dyslexia has been provided. However, genetically modified mice can again provide new avenues for research on the relationship between lateralization and dyslexia.

Thus, it has been shown that the dyslexia susceptibility genes *DCDC2* and *DYX1C1* impact on cilia function, which, in turn, impact on lateralization (Brandler

& Paracchini 2014; Chandrasekar, Vesterlund, Hultenby, Tapia-Paez, & Kere 2013; Girard et al. 2016; Kere 2014; Massinen et al. 2011; Schueler et al. 2015; Tarkar et al. 2013). Our collaborator, Joe LoTurco, and others (Tarkar et al. 2013) deleted the *DYX1C1* gene in mice and found two unexpected results: The animals developed hydrocephalus and demonstrated *situs inversus totalis*, with half of the mice showing dextrocardia and dextrogastria and half the standard organ asymmetry with the heart and stomach on the left side. Cilia malfunction has been linked to the production of *situs inversus* and hydrocephalus.

Differences in brain lateralization in dyslexics are not universally accepted, but they probably do exist. Problems in demonstrating anomalous asymmetry in dyslexics may stem, at least in part, from the fact that the cilia dysfunction model predicts for random distribution of asymmetry, which would have 1/2 of dyslexic being right-handed, albeit not “normal” right-handers. These random right-handers would counterbalance the random left-handers and eliminate the finding of anomalous lateralization, when in fact even those right-handers are anomalous. There is not as yet a known direct link between cilia function and brain lateralization, although *situs inversus* may, in fact, predict for changes in the patterns of brain asymmetry (Ihara et al. 2010; Kennedy et al. 1999). Therefore, these cilia results in rodents from dyslexia susceptibility gene deletions provide additional impetus for research on fundamental mechanisms underlying the relationship between dyslexia and anomalous brain lateralization.

6.4 Male-Female Differences in Developmental Dyslexia

The dyslexia literature is replete with discussions on the sometimes accepted, sometimes rejected, gender-based differences in dyslexia prevalence, although it is fair to say that most researchers today agree with a male predominance (Berninger, Nielsen, Abbott, Wijsman, & Raskind 2008; James 1992; Liederman, Kantrowitz, & Flannery 2005; Nass 1993; Rutter et al. 2004; Shaywitz, Shaywitz, Fletcher, & Escobar 1990; St Sauver, Katusic, Barbaresi, Colligan, & Jacobsen 2001; Tallal 1991). No adequate explanations for this skewed distribution can be found from strictly political-social-educational or psychological analyses, and the few neurological explanations for male predominance in developmental disorders have blamed the generally greater accident-proneness of boys, as compared to girls, including in utero and perinatal events blamed on the larger male head (Miller & Miller 2010). In 1985, Geschwind and Galaburda (1985a, 1985b, 1985c) published a three-part paper in which they developed a theory implicating the effect of testosterone in male-female differences in language-based learning disorders (but also about lateralization, see above), which stimulated research in animals. In fact, this is an area where animal research has been able to provide a deeper understanding.

A focal freezing injury to the cerebral cortex on post-natal day 1 in the rat leads to neuronal migration anomalies (see above, Humphreys et al. 1991), which vary in

morphology according to the gravity¹⁰ of the injury (Rosen et al. 2000). The results presented here are based on neonatal injury in the rat that produces focal, 4-layer microgyria, often with surrounding layer 1 heterotopias, similar to those reported in dyslexic brains (Galaburda & Kemper 1979; Galaburda et al. 1985). Qualitative and quantitative assessments of the microgyria show no differences between males and females, as may be predicted by the fact that the experimental injury in both cases is identical. However, when these animals are made to perform an auditory temporal processing task, substantial and significant sex differences emerge (Rosen, Herman, & Galaburda 1999). Two tones are presented to the animals separated by a gap that can change in duration. If the animal perceives the presence of the gap, the observed behavior – a startle reaction – is attenuated compared to the case where the animal does not detect the gap. At the shortest durations, only females are able to demonstrate that they have perceived the two tones separately; the males cannot. In fact, both unlesioned and lesioned females are better than any of the males, lesioned or not, which also demonstrates a task-related gender difference in normal animals (Fitch, Tallal, Brown, Galaburda, & Rosen 1994). But, the induced cortical malformation is the same in males and females, so what could explain the difference in the behavior?

The answer comes from examining secondary changes that follow from lesioning the cortex. In fact, an important factor in any neurological disorder is illustrated by the notion of *plasticity*, which refers to secondary changes that occur in relation to an initial event, which can be a faulty gene, a toxic exposure, a virus, a tumor, or an episode of brain damage (Butefisch 2006; Giza, Kolb, Harris, Asarnow, & Prins 2009; Rahn, Guzman-Karlsson, & David Sweatt 2013). This plasticity is a reaction to the initial event and not an attempt to fix an injury; it is a response to changes in inputs and outputs in relation to the lesioned area and the release of excitatory, inhibitory, trophic and suppressive factors; the reaction can be adaptive, and make improvements in the resultant behavior, but more often it is maladaptive and makes things worse. This is probably because plasticity did not evolve to repair injured brains, but rather as a response to normal experiences throughout life, including learning, growth, development and aging.

Changes in the visual thalamus were reported in the dyslexic brains (Livingstone, Rosen, Drislane, & Galaburda 1991), which gave rise to the magnocellular theory of dyslexia (Stein & Walsh 1997). More recent human work has implicated the thalamus in the dyslexic dysfunction (Diaz, Hintz, Kiebel, & Kriegstein 2012; Giraldo-Chica, Hegarty, & Schneider 2015). Similar to the changes in the human dyslexic auditory thalamus (Galaburda, Menard, & Rosen 1994), following freezing

¹⁰In this case gravity is defined by the length of time the freezing probe is kept on the skull. Small durations lead to layer 1 heterotopias; mid-range duration produces microgyri; and the longest durations cause frank porencephalic cysts cleaving the hemisphere from pia to ventricle. Except for the latter, the other two malformations have been noted to occur in dyslexic brains (Galaburda et al. 1985; Rosen et al. 2000)

injury to the cortex in newborn rats, the auditory nucleus of the thalamus – the medial geniculate nucleus – shows a change in the distribution of neurons of different sizes – fewer large- and more small neurons – in males; females do not show such changes (Galaburda et al. 1994; Herman et al. 1997; Rosen et al. 2006). The presence of these thalamic changes, rather than the malformations in the cortex per se, predicts for auditory deficits in this experimental rodent model: only males show the thalamic changes and only males demonstrate auditory deficits (Herman et al. 1997; Peiffer, Rosen, & Fitch 2002). However, care should be taken not to attribute a causal role in the behavioral deficits to the thalamic changes, since other changes, not assessed for in these experiments, may alone, or in combination with the thalamic changes, be the causative agent. In fact, an additional study finds other subcortical changes arising from the neonatal cortical injury in neonatal rats. Thus, examination of a brainstem auditory nucleus – the ventral cochlear nucleus – also shows changes in cell sizes, which affect males but not females (unpublished). In conclusion, these experiments show that animals are quite resistant to developing auditory deficits after induction of cortical malformations unless secondary changes occur, either in the thalamus, the brainstem, or even in areas not analyzed in the research. Since only males show these secondary anatomical and behavioral changes, additional questions arise.

Thus, the question arises as to what causes the male-female differences in the thalamus and brainstem (and behavior) in response to injury in the cortex of the newborn rat. The most obvious explanations for sex-differences, not relating to cultural or related environmental differences, implicate genes present in the Y-chromosome, dose effects of genes present in the X-chromosome,¹¹ and/or hormonal effects that differ between the sexes, namely sex steroids. In fact, in the above experiments, when pregnant females were exposed to the perinatal administration of testosterone propionate, the females of the dam now showed changes in the thalamus comparable to those seen in the males. Their behavior, however, is sufficiently variable that no consistent changes in the testosterone-treated females can be demonstrated, at least not in the adult females. This latter finding raises the issue that females have additional protective factors that keep them from behavioral deficits even in the case when the thalamus changes as a result of induction of cortical malformations. At the very least, this rodent model suggests a hormonal role for the sex differences seen in thalamic plasticity and possibly also in the auditory behavior.

¹¹Males have one X-chromosome, whereas early in development, before suppression of one of the X-chromosomes by lionization, females have two active X-chromosomes.

6.5 The Cortical-Subcortical/Cognitive-Perceptual/Phonological-Auditory Debate

Many studies have underscored the role of phonology in developmental dyslexia (Bradley & Bryant 1978; Liberman et al. 1971, 1982). However, this general idea is in need of a thoughtful review and possibly also a revision. Thus, it is not clear whether the so-called phonological deficit in dyslexia, which reflects itself in metaphonological tasks and rapid lexical access, refers to the phonological grammar, phonetics, or sound processing, or perhaps a necessary combination of these three factors or their interphases, which are known to impact on the speech processing (Berent, Vukobratovic, Nusbaum, Balaban, & Galaburda 2012, 2013; Goswami et al. 2011; Perrachione, Del Tufo, & Gabrieli 2011; Ramus 2001, 2004). Brain models in this regard would want to contrast higher-level processors in the cortex, farther from input or output channels, from lower-level and primary cortical areas, which are only one or two stations away from input or output channels, and even subcortical regions in the thalamus and brainstem, to underlie, respectively, the more cognitive *versus* the more perceptual and sensory aspects of speech processing.¹² Demonstrations of auditory and probably phonetic processing failures in dyslexia would leave open the possibility that subcortical regions are meaningfully affected by the pathophysiology of dyslexia. This said, it is important to stress that, developmentally and during on-line speech and language processing, higher- and lower-level functions and their structural underpinnings are network-linked and influence each other (Behroozmand et al. 2015; Wilke, Lidzba, & Krageloh-Mann 2009). However, results such as those presented in the previous section on the testosterone effect on plasticity would pit the relative contributions of cortical against subcortical dysfunction in the pathogenesis of dyslexic speech and sound processing, as it may pertain to dyslexia. Preliminary analysis would suggest that lower-level processing and subcortical structures are relatively more important.

An example of the possible importance of a subcortical structure such as the thalamus has been illustrated above. However, it is not clear from those experiments whether the thalamus or some other subcortical structure is playing a causative role in the auditory processing failure in the affected males and testosterone-treated females. Also, those experiments do not deal with the possibility that the cortex itself reorganizes differently *vis à vis* gender, maladaptively in the males and adaptively in the females. I am referring here to possible cortical plasticity away from the area of induced malformation, which has not been specifically explored *vis à vis*

¹²By higher-level cortical processing I mean the classic language areas in the frontal, parietal and temporal lobes, e.g., Brodmann areas 44, 45, 39, 40 and 22, as compared to primary cortices such as areas 4, 3, 2, 1, and 41. Relevant subcortical areas would include the medial geniculate nucleus of the thalamus and brainstem auditory nuclei such as the cochlear nucleus and the trapezoid body, among others.

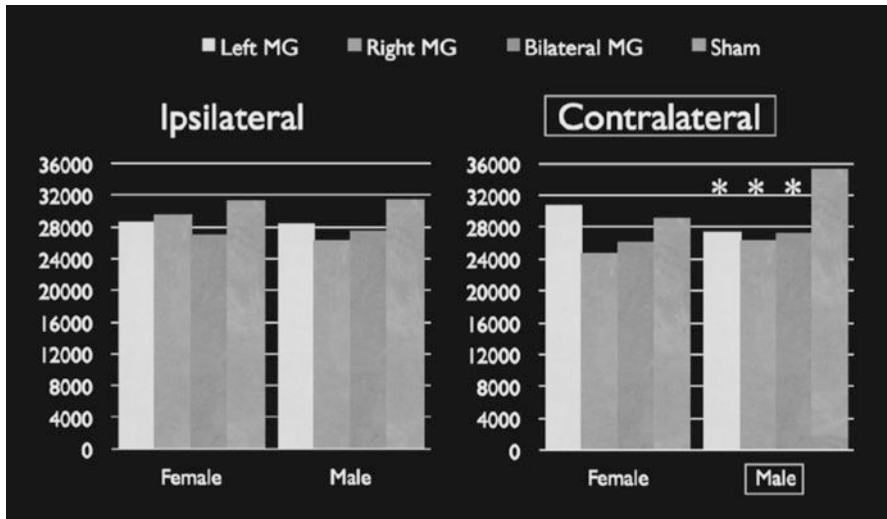


Fig. 6.4 Unilateral induction of a microgyrus in the cortex of the newborn rat induces changes in neuron numbers in the contralateral ventral cochlear nucleus, but significant changes are seen only in male rats (unpublished observations)

gender differences. Thus, although there are clear subcortical changes in the males and not in the females, it is possible that the latter do not contribute to the deficient behavior in either case, and that rather the problem lies elsewhere, either in other areas of the cortex not directly involved in the malformation, or in other subcortical structures. In fact, the same rats that showed changes in the thalamus were examined for additional changes in subcortical structures. Again, changes were found in males but not in females, this time in the ventral cochlear nucleus on the side of the brainstem opposite to that of the freezing injury (unpublished), suggesting that these changes instead of, or in combination with, the thalamic changes, could be responsible for the auditory failures in the males, which were absent in the females (Fig. 6.4). Clearly, the most parsimonious interpretation would be that the induction of a cortical malformation in the rat at birth produces a series of changes in the cortex and subcortex, the combination of which is responsible for the auditory processing deficits, and that these changes occur only in males and are influenced by testosterone.

A useful animal model is presented by mice that have been deleted for a given dyslexia risk gene. Over the past decade several dyslexia susceptibility genes have been published, some of which appear to be quite robust, e.g., *DYX1c1*, *KIAA0319*, and *DCDC2* (Galaburda et al. 2006; Kere 2014). Preliminary evidence exists showing that mice deleted for the *DCDC2* dyslexia risk gene homologue in the mouse, *dcdc2*, in addition to being expressed throughout the neuraxis (see, for instance, Shen, Overly, & Jones 2012), including olfactory lobe, neocortex, thalamus, cerebellum, and brainstem nuclei, also appears to be accompanied by

cell size changes in the cochlear nucleus¹³ (see above). In the case of this knockout mouse, in contrast to the lesion-induced cochlear nucleus cell changes in the rat, the possibility still exists that the changes in the mouse cochlear nucleus do not result from primary changes in the cortex,¹⁴ but appear spontaneously through direct gene effects occurring *in situ* in the brainstem. Alternatively, of course, the cochlear nucleus changes in the mouse represent an interaction between local effects in the brainstem and secondary to changes in the cortex, thalamus and other parts of the brainstem. Further clarification of the role of the cortex and sub-cortex – also read cognitive and sensory-perceptual factors, respectively – will require additional research. For instance, conditional knockout mice can be made that delete dyslexia susceptibility genes regionally, for instance in the cortex, thalamus, or brainstem, each of which can be assayed for auditory processing deficit. Thus, the contribution of each region to the deficit could be possibly disentangled.

6.6 Summary and Conclusions

Associations between human neuropathologic findings and developmental disorders can give only hints as to possible mechanisms and etiologies. The presence of a finding on an image or in a tissue cannot attribute causality, although a causal relationship is one possible explanation for the observed association. In order to test causal relationships it is necessary to create models in which a human finding is induced and its consequences are examined. In the case of developmental dyslexia, human neuropathologic findings have disclosed problems with cortical development caused by abnormalities in neuronal migration, changes in the auditory and visual thalami, and brain asymmetry in some language areas that deviate from standard patterns. We have used animal models, in rats and mice, to explore mechanisms and etiologies that may underlie the human brain findings. We have found that neuronal migration anomalies to the cortex may be a consequence of dysfunction of dyslexia risk genes, but this depends on the genetic model that is used. Even if not able to produce frank anatomical malformations, dyslexia risk genes may render cortical neurons dysfunctional. We have also seen that dyslexia risk genes may alter body organ lateralization, *via* dysfunction of cilia, and this suggests a possible mechanism for the findings of brain lateralization anomalies in the dyslexic brains and in dyslexic handedness. Animal models have also pointed to plausible mechanisms for the sex-differences that have been reported to occur in dyslexia. These appear to be mediated by plasticity, whereby testosterone worsens the odds of recovering from developmental cortical abnormalities acquired genetically or

¹³This remains a preliminary, non-publishable result, as the laboratory was unsuccessful in getting funding to replicate it and further pursue its consequences on behavior.

¹⁴In fact, the changes in the cortex of the knock-out *DCDC2* mouse do not include malformations and are limited to changes in the cell biology of NMDA receptors (Che et al. 2016).

through neonatal injury. Finally, experimental models indicate that even though the cortex may be abnormal in the dyslexic brain, it may be the secondary changes in the thalamus and other subcortical regions that account for the behavioral deficits. Given the localization of the dysfunction, then, it is possible that low level auditory processing may be the first step to developing speech processing and phonological problems in dyslexic individual. More animal research needs to be done to expand on the knowledge thus far gathered.

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Chapter 7

The Magnocellular Theory of Developmental Dyslexia



John Stein

Abstract The late 19th neurological concept of dyslexia had 3 crucial elements: selectively poor reading, with unaffected other cognitive skills and a genetic background. The contemporary ‘phonological theory’ has undermined the selectivity criterion because all poor readers, dyslexic or otherwise, have phonological problems. Here I argue that the phonological theory is pitched at too high a cognitive level so that it does not illuminate the mechanisms that cause reading problems in dyslexia. Recent genetic and imaging studies have confirmed their biological basis. In children with visual reading problems there is strong evidence that they suffer impaired development of the visual magnocellular (M-) system which is vital for tracking shifts of the focus of visual attention and of eye movements. This can often be ameliorated by viewing text through deep yellow or blue filters because they can facilitate the M- system. Likewise children with phonological problems seem to suffer an analogous impairment of sound sequencing, which can be ameliorated by musical training, particularly in rhythm; whilst those with impaired motor sequencing can often be helped by motor training. Thus in dyslexics the neural sub system which is required for rapid and accurate temporal processing and is distributed throughout the brain, appears to be compromised. This system’s ‘M-’ neurones express a specific surface marker that renders them susceptible to autoimmune attack, and the rapidity with which they have to respond, makes them particularly vulnerable to lack of omega 3 long chain polyunsaturated fatty acids in the diet. But its weaknesses for temporal processing may be balanced by exceptional talents for other kinds of cognitive task.

Keywords Temporal processing · Vision · Magnocellular · Colored filters · Audition · Embodied cognition · Genetics · Handedness · Omega 3s · Dyslexia talents

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7.1 Introduction

In 1896 W. Pringle Morgan described the first case of developmental, as opposed to acquired, “word blindness” in a young boy. “Percy F had not yet learned to read by the age of 14, even though he knew his letters well. In spite of laborious and persistent training, he can only with difficulty spell out words of one syllable; yet the schoolmaster who taught him for some years says that he would be the smartest lad in the school if the instruction were entirely oral.” Pringle Morgan also speculated that his inability to read was so remarkably selective and so profound, that he had no doubt that it was due to “some congenital defect”. These three crucial elements described by Morgan in Percy, namely inability to read all but the simplest words, yet reasonable intelligence in other respects, and a probable genetic cause, have set the scene for arguments about developmental dyslexia, its definition and causes, that still continue 120 years later.

7.2 The Phonological Theory

Until the 1950s most of those interested in why some children find it so difficult to learn to read, believed that it was due to a failure of visual processing; Kerr, Hinshelwood and Orton called it “word blindness”. After all, blind people cannot read ordinary text – reading obviously requires vision. But in 1957 Noam Chomsky published his seminal book, “syntactic structures”, which revolutionised linguistics and ushered in their scientific study (Chomsky 1957). This book suggested that humans alone are genetically endowed with an “encapsulated linguistic processor” which mediates a “Universal Grammar” that underlies all languages.

These ideas quickly transformed the study of language and with it, reading. Some children’s reading problems were already thought to be hereditary, and christened “dyslexic”. So they speedily became attributed to a fault in Chomsky’s linguistic processor, and any role for visual processing was abandoned. This meant that the “medical” neurological explanation for dyslexia became less attractive than a linguistic one, and its study passed into the hands of linguistic and educational psychologists. Dyslexia became a linguistic, phonological, problem, not a visual one (Lieberman & Shankweiler 1977).

However more recently it has become clear that there is no “special” linguistic processor; human language has developed out of pre-existing gestural and auditory specializations found in our primate ancestors and even in lower mammals and birds. This means that the basis of language comprehension may be found in auditory processing functions far deeper than phonology, in the sound analysis that underlies phonology. Hence today many researchers are trying to identify the faulty auditory processing that causes the phonological problems from which most dyslexic suffer (Goswami, Power, Lallier, & Facoetti 2014; McAnally & Stein 1996).

Indeed we can now view the phonological theory of dyslexia as almost a tautology – merely restating the fact that dyslexics cannot translate visual symbols into the sounds they stand for, rapidly and accurately. The phonological theory does not provide a helpful explanation for dyslexic reading problems because it is set at too high a cognitive level. A more useful explanation would trace the genetic mechanisms that set up the neural processes whose failure leads to the phonological deficits we see; this would explain why children have problems hearing the sounds in a word in their right order and seeing the letters properly.

7.3 Arguments Against the Concept of Dyslexia

This enterprise should carry with it the additional advantage of providing biomarkers of true dyslexia, so that it could be distinguished from other causes of reading failure, such as bad teaching, lack of home support or very low general ability. Unfortunately at present we do not have such identifiers; hence we cannot make this distinction reliably. This has even led some people to want to completely abandon the whole concept of dyslexia as a distinct neurological syndrome (Elliott & Grigorenko 2014).

Here I argue the opposite, that true dyslexia is a genetically based neurological syndrome which leads via aberrant brain development to a spectrum of symptoms characterized by impaired temporal sequencing. These include not only impaired ability to sequence sound streams accurately which underlies the inability to acquire phonological skills, but it also leads to inaccurate sequencing in general: of visual symbols reliably in the right order, hence poor orthography together with wider ramifications: slow learning of the order of letters in the alphabet, misordering the days of the week, months of the year, slow learning to tell left from right, poor sequencing of the actions required for tying shoelaces, poor time keeping etc. (Miles 1993). Elucidating the neural basis of these temporal sequencing problems will enable us to design principled and effective treatments for each child's individual difficulties. It would have the additional advantage of enabling us to establish diagnostic biomarkers to distinguish the neurological syndrome, dyslexia, from other causes of reading failure. In so doing I hope also to re-establish the importance of visual processing in reading.

The basic hypothesis outlined here is that genetic vulnerability early in the growth of the brain in utero in dyslexics impairs development of a system of magnocellular neurones throughout the brain (Stein & Walsh 1997). These cells appear to be a specialized lineage since they all express a common surface signature recognized by selective antibodies such as CAT 301 (Hockfield & Sur 1990). They are adapted for the rapid temporal processing that is required for accurate sequencing in all domains: auditory, visual, proprioceptive and motor.

7.4 Dyslexia Genetics

Paradoxically the one feature for which Pringle Morgan had the least evidence is nowadays the least controversial; this is his conjecture that developmental dyslexia is hereditary. The evidence is now overwhelming. Twin studies have established a heritability of at least 60% (Kirkpatrick, Legrand, Iacono, & McGue 2011; Olson et al. 2013). But note that these estimates leave 40% of reading variance in the hands of environmental factors. Hence immune attack, poor nutrition, family poverty and stress, deficient education, may all contribute to convert a vulnerable genotype into overt dyslexia. Conversely reading problems may not occur if such environmental factors can be avoided. Thus studying populations with high educational standards may actually increase our chances of identifying the real genetic contributions to dyslexia because adverse environmental factors may then be absent (Asbury & Plomin 2013).

Recent advances in molecular genetics have now become so powerful that more than a dozen gene variants (alleles) have now been identified that confer vulnerability to dyslexia. As techniques develop even more genes will undoubtedly emerge, but it is notable that the number discovered so far is very much smaller than those suggested for much more highly hereditary conditions such as schizophrenia, for which nearly 200 gene variants have been fingered (Schizophrenia Working Group of the Psychiatric Genomics Consortium 2014). Perhaps this difference is partly because reading skills can be measured so much more accurately in the phenotype, than can the symptoms of schizophrenia.

7.5 Genes and Brain Development

Normally, during the early development of the brain in utero, the neurones destined to form the surface of the cerebral hemispheres, are born around the ventricles in the center of each hemisphere and then migrate outwards to form its six surface layers. But when Galaburda and colleagues examined histologically the brains of severe dyslexics from the Orton brain bank, now at Harvard University, they found that they were characterized by significant malformations, in particular periventricular heterotopias and surface ectopias, caused either by the neurones failing to migrate at all or migrating to the wrong places (Galaburda, Sherman, Rosen, Aboitiz, & Geschwind 1985).

What has been most exciting in relation to these observations is the discovery that five of the genes which now have the strongest evidence of association with dyslexia (DYX1C1, DCDC2, KIAA0319, KIAA0319L and ROBO1) all affect the development of the brain very early in utero, by helping to control this migration. For example in the rat, if the expression of DCDC2 is prevented in embryonic brain by local electroporation of an inhibitory RNA specific to this gene, many neurones do not migrate at all, but remain stuck around the ventricles where they

were born, forming “periventricular heterotopias” (Rosen et al. 2007). Knockdown of KIAA0319 or KIAA0319L in this way causes less dramatic changes, but local electroporation of the RNAi still impairs successful migration; and this emerges in the rats when they mature being less accurate at discriminating rapid sequences of sounds (Centanni et al. 2014). The same 5 genes are also involved in the formation of the connections between these cortical neurones once they reach their final destination (axon and dendrite outgrowth, guidance and the formation of the synapses between them). Probably they control the expression of neuronal membrane surface proteins, such as those recognized by CAT 301, that direct both their migration and their interconnections.

Another set of genes which has been shown to be associated with dyslexia include FOXP2 and its partner, CNTNAP2. These are particularly important in setting up the frontal cortex language areas and they were already known to be also associated with developmental dysphasia, which often causes dyslexic problems later (Newbury et al. 2011). A further set includes C20ORF3 and MCR5 both of which play important roles in fatty acid metabolism and neuronal membrane structure (Fisher et al. 2002). This relationship with essential fatty acids will be developed later in this paper.

7.6 Comorbidities

One tricky problem with the definition and diagnosis of dyslexia is that there are very few “pure” dyslexics. Clinically there are very large overlaps between the symptoms of dyslexia with most other neurodevelopmental conditions. Indeed it is often largely a matter of chance according to which professional a child sees first, as to whether s/he is diagnosed as having dyslexia, dyspraxia, dysphasia (also known as specific language impairment – SLI), dyscalculia, ADHD, Tourette’s syndrome, obsessive/compulsive or autism spectrum disorders, because there is so much overlap in their symptoms. Accordingly, all these conditions tend to be found in the same families and it seems as if it might be better to talk of a spectrum of neurodevelopmental conditions; their overlap is so great that the points at which we categorize one way or another are highly arbitrary. For example up to 50% of children diagnosed with dyslexia would also meet the criteria for ADHD, SLI or dyspraxia (Rice, Smith, & Gayan 2009). These overlaps can probably be explained by shared genetic components. Increasing evidence shows that the same genetic factors may underlie different traits as in the case of the CNTNAP2 which is implicated in SLI as well as many other neurodevelopmental disorders (Graham & Fisher 2013). Equally, genes originally identified for either dyslexia (e.g. KIAA0319) or SLI (e.g., *CMIP*) have been found to contribute to both reading and language measures (Scerri et al. 2011).

None of the alleles so far discovered, or likely to be discovered, contributes individually a substantial component to the total genetic background to dyslexia – dyslexia is usually caused by the interaction of many genes with individually

small effects. Therefore they could not be used for diagnosis as some people have suggested. In fact the main reason for studying the molecular genetics of dyslexia is not for diagnosis, but to attempt to understand more about the mechanisms whereby the gene variants contributing to dyslexia, may do so. This should give us insights about how to help these children's problems.

7.7 Brain Differences in Dyslexia

The ectopias seen by Galaburda et al. in the Harvard Orton brain bank are too small to be seen reliably even with the most powerful current imaging techniques. But magnetic resonance imaging has shown that there are clear anatomical and functional differences between the brains of dyslexics and those of good readers. There is widespread agreement that in the cerebral cortex the left occipitotemporal, occipitoparietal and superior temporal areas have less neuropil in dyslexics compared with good readers whilst the left arcuate fasciculus projecting to the left inferior frontal gyrus (Broca's area) is also less developed. Conversely the right inferior frontal gyrus may be thicker in dyslexics (Richlan, Kronbichler, & Wimmer 2013). However the largest differences are seen in the cerebellum (Rae et al. 2002; Stoodley 2014). Here the right neocerebellum, which projects to the left cortical language areas is smaller in dyslexics and its size predicts how well they perform in both phonological and irregular word tests. It has been argued that most of the anatomical differences in both cortex and cerebellum could in fact be the result of low reading experience rather than its cause. However even though reading experience must play a part, there is now abundant evidence that some of these differences can be detected soon after birth, long before any reading exposure (Hoeft et al. 2011).

7.8 Visual Sequencing and Dyslexia

What we really want to know is how these genetically based brain differences cause the symptoms we see in dyslexic children, so that we can design scientifically based treatments to alleviate them. Here I commence with their problems with visual sequencing because visual processing is obviously the starting point for reading and because many children complain of visual symptoms when attempting to read.

How do we build up representations of the sequence of letters in a word in our visual, orthographic, lexicon? The first step in reading is to learn to recognise individual letters. Actually, dyslexics are initially just as good as good readers at recognizing single letters (Lachmann & van Leeuwen 2007). As this basic process is the same for visual recognition of any object, this shows that dyslexics do not have significant problems with the very earliest stages of visual processing. However, letters do not come singly, but in groups, and their order matters. When children are first learning to read they inspect each letter individually moving their eyes from

letter to letter. They need to know where their eyes were pointing when they fixate on each letter in order to determine its order in the word. Therefore, to remember the letter sequence, they have to learn to associate the visual form of each letter being fixated with signals about where the eyes were pointing at the time.

Many dyslexics fail to achieve this process accurately. Each time we move our eyes, images stream across the retina. Although this is happening physically, we are not usually consciously aware of this motion, because the neural command to move the eyes which is generated in the frontal eye fields in the dorsomedial part of the prefrontal cortex is also sent to the conscious vision centers in the brain, and this is used to blank out the apparent movement. At the same time, the letter seen at the new position of the eyes is automatically associated with that position, using the eye movement signal just before it, together with the image motion signal. Even though we do not see the image motion consciously it enables us to ascertain that the letter now being inspected comes just after the previous one. Thus we are able to keep an account not only of its identity but also of its position in a word. When described and dissected in this clumsy way, the process sounds very complicated and effortful. But most of us learn to sequence not only letters, but also numbers, objects and lists, fast, effortlessly and accurately.

7.9 Magnocellular Neurones

The special image motion and eye movement signals required for these associations are provided by the set of large nerve cells mentioned earlier; they are called visual magnocellular neurones (from the Latin “magnus”, meaning large). The distinctive properties of magnocellular neurones have been most studied in the visual system. The ganglion cells in the retina receive information from the light receptors at the back of the eye and project it via the optic nerves to the lateral geniculate nucleus (LGN) of the thalamus, thence to the visual cortex which is situated on the back of the occipital cortex at the very back of the brain. 10% of these ganglion cells are much larger than the others; these are the magnocellular (M-) ones. They capture information over a very large retinal area – as much as a square millimetre; this is up to 50 x larger than that of the much more numerous parvocellular (P-) ganglion cells. These are much smaller but 10 times more numerous. Being smaller they respond more slowly but they can define the fine detail and colour of objects. Therefore for reading, it is actually the P-system that provides the main input to the brain’s “visual word form area” (VWFA) where letters are identified, and a word’s orthography stored.

The magnocellular neurones cannot define such fine detail and they do not discriminate colors; but because they are larger, they respond and conduct signals much more rapidly than the parvo cells (Schmolesky et al. 1998). This means that they are much more sensitive to temporal changes in the outside world such as flicker and movement. Thus they can rapidly signal changes in the environment and capture attention.

Both the M- and P-ganglion cells project to the lateral geniculate nucleus en route to the primary visual (striate-V1) cortex. But the M- cells project to the ventral magnocellular layers of the LGN whereas the P-cells project to the more dorsal parvocellular layers. This separation is preserved in the onwards projection from the LGN via the optic radiations to layer 4 of the striate cortex. Here magnocells project to layer 4C alpha and parvo cells project to layer 4C beta. But thereafter M and P inputs converge greatly.

7.10 Dorsal and Ventral Visual Pathways

Nevertheless the magnocellular system provides the main input to the “dorsal stream” (see Fig. 7.1) which is one of the two major forward projections from the primary visual cortex in the occipital lobe to the rest of the brain: the dorsal and ventral pathways (Goodale & Milner 1992). The dorsal stream is responsible for the visual guidance of attention and of eye and limb movements. Accordingly the dorsal stream passes to the visual motion sensitive area (MT/V5) which is situated in the middle temporal gyrus at the occipitotemporal junction, and thence to the posterior parietal cortex. But we are unaware of most of what the dorsal stream does, because, serving the visual control of movement, most of its actions are automatic and do not enter consciousness.

The slower ventral pathway passes forward ventrally underneath the occipital and temporal cortex; its main function is to detect the form and color of objects in order to identify them; hence it is the system that is able to identify individual letters and it dominates our conscious visual perception.

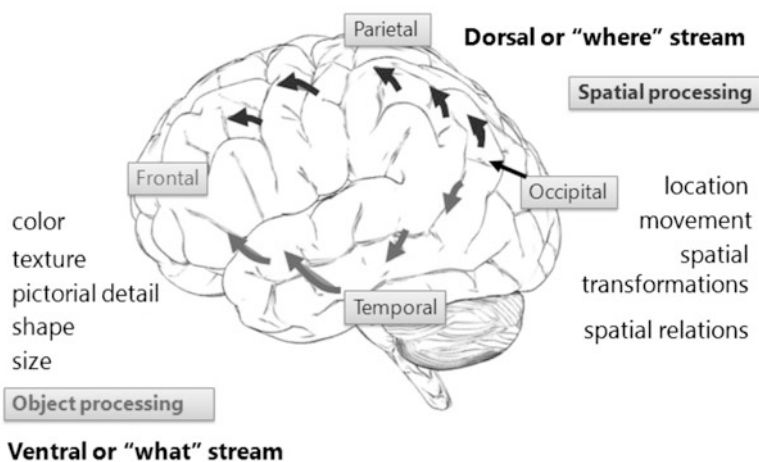


Fig. 7.1 The dorsal and ventral pathways

In the left hemisphere the dorsal stream angular and supramarginal parietal areas are particularly important for reading. They are responsible for associating the visual form of a word with its sound and meaning. An area known as the VWFA situated in the ventral, P-dominated, form analyzing, pathway, receives from and projects back to the supramarginal gyrus. It responds to the visual form of the word (Dehaene et al. 2010). The job of the angular and supramarginal gyri is now thought to be very rapidly to focus visual attention on the letters and their order in words to be read and to associate this visual word form with its pronunciation. The ventral route and VWFA system can recognize individual letters, but they cannot code their precise location, i.e. their order in a word, which is of course vitally important for reading. So the rapid dorsal route sends back to V1 and to the VWFA a signal about where to attend in order to identify letters and thereby to specify their order in a word (Cheng, Eysel, & Vidyasagar 2004). Then if this word is already in the reader's visual lexicon, it can be linked to the sound of the word which is stored in Wernicke's area situated at the back of the superior temporal gyrus. Its meaning can then be grasped by connection via the arcuate fasciculus with Broca's speech area in the inferior frontal gyrus, even if the word is not actually spoken. Thus when learning to read, the angular and supramarginal gyri focus V1 and VWFA attention on individual letters and their order; then these areas link the visual word form with the word sounds stored in Wernicke's area.

7.11 Magnocellular Impairment in Dyslexia

Thus the visual magnocellular input to the dorsal attentional stream appears to play a crucial role in reading; so the accumulating evidence that it is poorly developed in many dyslexics has especial significance. Although the theory that a visual M- cell deficit underlies visual problems in reading is still highly controversial, over 90% of the studies made over the last 10 years that have sought evidence for M- impairment in dyslexics have found it in at least some.

7.12 Subcortical M- system

Strictly speaking, visual M- cells can only be distinguished from P- cells with complete certainty in the subcortical visual system because only in the LGN are they anatomically separated from the P- system. Thereafter magno and parvo systems converge and interact strongly. Hence the only way to be sure that deficits in dyslexics are confined to the M- system is to use stimuli that are selectively processed by the subcortical M- neurones in the retina and LGN. Skottun (2015) has made this point repeatedly. But even if we confine ourselves to the M- system in the retina and LGN the evidence for M- impairment in dyslexics is strong.

7.13 Contrast Sensitivity

Perception of low contrasts in the environment is set by the properties of M-ganglion cells in the retina. The simplest way of assessing their variability in individuals is to measure subjects' sensitivity to the contrast of coarse gratings (spatial frequency <1 cycle per degree) flickered at high temporal frequencies >10 Hz, since M- cells respond much more sensitively to this combination. Since Lovegrove, Heddle, and Slaghuis (1980)'s first report, there have been many studies that have confirmed that the contrast sensitivity (CS) of many dyslexics is lower than that of controls', particularly at the low spatial and high temporal frequencies mediated by the M- system (Bednarek & Grabowska 2002; Cornelissen, Richardson, Mason, Fowler, & Stein 1995).

Interestingly, at the M- cell threshold such gratings appear perceptually twice as fine; this is known as the spatial frequency doubling illusion. It is thought to depend on the non-linear properties of the M- cells (Maddess, James, Goldberg, Wine, & Dobinson 2000). Although the details of this dependence have been contested (White, Sun, Swanson, & Lee 2002), nevertheless, whether or not the grating appears twice as fine, it is accepted that as the contrast of such a grating is decreased, the point at which observers first see the pattern is determined by their retinal M-cells. Dyslexics have been consistently shown to require more contrast to see the gratings, confirming their M- cell weakness (Pammer & Wheatley 2001).

M- cells are responsible for timing visual events, so their sensitivity in individuals can also be assessed by various timing tests. For example Lovegrove et al. (1980) tested people's ability to detect a discontinuity (a temporal "gap") in the display of a low contrast, low spatial frequency grating; they found that dyslexics needed a much longer gap to perceive it than the controls. When the frequency at which a light is flickered is increased, above around 30× per second the M- system can no longer respond fast enough and the light appears to cease flickering. This is the "critical flicker fusion frequency". Chase and Jenner (1993), Mason, Cornelissen, Fowler, and Stein (1993), Felmingham and Jakobson (1995) all showed that this frequency tends to be lower in many dyslexics.

7.14 Lateral Geniculate Nucleus

Further strong support for the idea of M- cell impairment in dyslexics came from further post mortem histological studies of dyslexic brains from the Orton dyslexia brain bank at Harvard University (Livingstone, Rosen, Drislane, & Galaburda 1991). They found that the M- layers in the LGN in the dyslexic brains were selectively impaired. Not only were the cells 25% smaller in the dyslexic as compared to control brains, but also the M- cells were not confined to their proper M- layers; many had migmigrated into the adjacent konio and parvo layers of the LGN. As we have seen this theme of migmigration relates to at least 5 of the genes that have been associated with dyslexia.

Recently the spatial resolution of MR imaging has been improved considerably with the introduction of stronger 7 Tesla magnets, so that the structure of the LGN can now be visualized in more detail (Denison, Vu, Yacoub, Feinberg, & Silver 2014). Galaburda's observations were made in only 5 dyslexic brains post mortem, but 7 Tesla MRI has confirmed in 13 young dyslexics that their left LGN, was significantly smaller in volume than controls'. This difference was confined to the magnocellular layers in the 13 dyslexics, so that their LGNs also differed in shape (Giraldo-Chica et al. 2015).

7.15 The Cortical Dorsal “Where” Pathway

The dorsal stream (see Fig. 7.1) is dominated by visual M- input as we have seen. Abnormalities have been found in dyslexics at every level in this stream from the prestriate visual motion area (MT/V5), via the posterior parietal cortex to the ultimate goal of both M- and P-systems, the prefrontal cortex (Rao, Rainer, & Miller 1997).

90% of the visual input to the motion sensitive neurons in the middle temporal visual motion area (V5/MT) is provided by the M- system and only 10% comes from other sources. One way of assessing the sensitivity of these MT neurons in individuals is to measure their responses to visual motion in “random dot kinematograms” (RDKs). Clouds of dots moving in the same direction, “coherently”, are progressively diluted with noise dots moving in random directions until the subject can no longer detect any coherent motion in the display. This threshold therefore defines that individual's motion (visual dorsal stream) sensitivity. Several researchers have shown that this is reduced in many dyslexic individuals (Cornelisssen et al. 1995; Downie, Jakobson, Frisk, & Ushycky 2003; Hill & Raymond 2002; Samar & Parasnis 2007; Talcott, Hansen, Assoku, & Stein 2000). Other work has similarly shown reduced velocity discrimination (Demb, Boynton, Best, & Heeger 1998; Eden et al. 1996) and elevated speed thresholds for motion-defined form (Felmingham & Jakobson 1995) in dyslexics.

People with low motion sensitivity can still be adequate readers however (Skoyles & Skottun 2004), so that weak M- function by no means predestines a child to reading failure. Other vulnerabilities must contribute to dyslexia as well. Nevertheless, individual differences in motion sensitivity explain over 25% of the variance in reading ability (Witton et al. 1998). In other words, individuals' dorsal stream performance, which is dominated by M- cell input, plays an important part in determining how well visual reading skills develop, and this is true of everybody, not just those diagnosed with dyslexia.

The posterior parietal cortex (PPC) receives its main visual input from V5/MT; this input plays a crucial role in guiding visual attention, eye and limb movements (Cheng et al. 2004). Dyslexics have been found to be worse than good readers at cueing visual attention (Kinsey, Rose, Hansen, Richardson, & Stein 2004), visual

search (Iles, Walsh, & Richardson 2000), visual short term “retain and compare” memory (Ben-Yehudah, Sackett, Malchi-Ginzberg, & Ahissar 2001) and attentional grouping in the Ternus test (Cestnick & Coltheart 1999). These findings confirm that dorsal stream function is often impaired in dyslexia. Many of the studies mentioned above incorporated control tests for parvo function, such as visual acuity or color discrimination; and dyslexic populations usually proved to be as good or better at these. This confirms that the tests assess dorsal stream function specifically, rather than the subjects’ attending to the stimulus or the difficulty of the task.

7.16 Cross Modal Attention

Dyslexics show not only slow deployment of visual attention but they also show difficulty shifting their attention between sensory modalities, for instance between vision and hearing. But it seems that such “sluggish attention shifting” (Hari & Renvall 2001) is worst when dyslexics shift their attention from the visual to the auditory modality, rather than vice versa (Harrar et al. 2014). Thus dyslexics are not only slower, but particularly slow when they have to attend to a visual stimulus and shift to an auditory one, as when reading.

Taken together all this evidence suggests that dyslexics’ poor dorsal stream performance can be mainly attributed to M- system weakness even in the presence of robust parvocellular function.

7.17 Eye Movement Control by the Dorsal Stream

Normally the dorsal stream not only directs visual attention to a target but also subsequently redirects the eyes towards it. Hence numerous studies have found not only that the direction of visual attention is disturbed in dyslexics (Facoetti et al. 2010; Vidyasagar 2004), but also that their eye control during reading is poor (Eden, Stein, Wood, & Wood 1994; Kirkby, Webster, Blythe, & Liversedge 2008). However it is strongly argued that these abnormalities are not a cause of reading problems but rather that they are the result of not understanding the text; hence the person has to make longer fixations and more reinspections of previous letters to try to decode words (Rayner 1985). But poor eye control in dyslexics has also been demonstrated in several non-reading situations, using tests of fixation stability (Fischer & Hartnegg 2000) and of smooth pursuit and saccadic control (Crawford & Higham 2001), implying that poor eye control comes first and may be a significant cause of reading problems.

7.18 Event Related Potentials

Recording averaged electroencephalographic (EEG) potentials in response to a moving, low contrast visual target provides a more objective measure of cortical dorsal stream processing than psychophysical techniques. Of recent visual event-related potential (ERP) studies in dyslexics the great majority have either confirmed the original observation by Livingstone et al. (1991) that dyslexics have weaker responses to moving, low contrast, targets than good readers (e.g., Kuba, Szanyi, Gayer, Kremlacek, & Kubova 2001), or they have found that dyslexics show slower, smaller and spatially abnormal visual attentional ERP responses. These observations are in line with a multitude of psychophysical results showing deficient allocation of visual attention (Hari & Renvall 2001; Lallier et al. 2011).

7.19 Criticism of Magnocellular Theory

Nevertheless even though the vast majority of new research that has looked for visual magnocellular deficits in dyslexics has confirmed that at least some dyslexics demonstrate them, the idea has not gained full acceptance. Skottun, while not ruling out that dyslexics may have some kind of visual deficit, has written over 30 critiques of the idea that these involve the M- system (Skottun 2016). But the only one of these which reports new research findings (Gross-Glenn et al. 1995) actually partially supports the magnocellular theory! Skottun's main point is that standard tests of visual magnocellular function do not entirely distinguish M- from P-inputs to the dorsal stream. Nevertheless, since the M- system provides 90% of the input this probably not a substantial problem.

In summary so far, there is now overwhelming evidence that many dyslexics have impaired development of the visual magnocellular system, so that their visual temporal processing is impaired. Because this hinders their ability to associate each letter they inspect with its position in a word, their sequencing of the letters in a word is slow and inaccurate. The degree of this deficiency predicts the severity of their visual reading difficulties. What this means in practice is that we can in principle distinguish visual dyslexia from other causes of reading problems simply by looking for symptoms of visual magnocellular impairment.

7.20 Visual Difficulties in the Classroom

Teachers should therefore always ask children with reading difficulties whether they have any visual symptoms when attempting to read. Do letters appear to blur, or split into two, or move around when you try to read? Does reading make your eyes or your head ache? Further investigations for research, such as measuring

eye fixation, convergence and accommodation, recording brain waves evoked by moving or flickering stimuli or functional MRI investigations, may then confirm their visual temporal processing problems, but these are not essential. Asking the right questions about any visual problems is.

7.21 Yellow Filters

The idea that viewing text through colored filters may help some children to overcome their visual problems and learn to read is highly controversial and sullied by commercial interest and wild claims. Part of the problem is that not all reading problems are due to visual deficits, so that the contention that all children can be helped by the right color is easy to refute. However the magnocellular theory leads to a rationale for supposing that particular colored filters may help some children.

Visual magnocells receive most of their input from the red (long wavelength) and green (medium wavelength) color receptors (cones) in the retina. Yellow light combines both those wave lengths. So magnocells are best stimulated by yellow light. Looking at text through yellow filters reduces the amount of blue light entering the eye, and causes the pupils to dilate. So the amount of yellow light falling on the retina increases and retinal magnocells are selectively activated. If the main visual problems that the child has are that letters look blurry and out of focus and they tend to split into two, which symptoms are often associated with a reduced vergence range, viewing text through yellow filters often boosts their magno input sufficiently to help them overcome these problems; hence their reading improves rapidly. Ray, Fowler, and Stein (2005) found that giving yellow filters to children with these symptoms improved their accommodation and convergence, and their reading progress tripled thereafter.

7.22 Blue Filters

However, yellow filters only appear to help about half of dyslexic children with visual reading symptoms. Another subset who complain mainly of eye and head aches and of letters moving around and over each other are more likely to be helped by viewing text though deep blue filters. These cut out most of the long wavelengths that directly stimulate magno cells. Nevertheless their reading may improve even more than in those who benefit from yellow (Clisby et al. 2000). They probably work in a very different way.

7.23 The Hypothalamic Clock

The blue filters optimally excite a newly discovered set of ganglion cells in the retina which do not contribute directly to conscious vision (Hankins, Peirson, & Foster 2008). Instead, they feed into the hypothalamic suprachiasmatic nucleus; this is where the body's internal "clock" which times out our daily rhythms is situated. The blue input is required to entrain the clock to seasonal changes in day length, so that we wake up earlier in summer and later in winter. On arousal by blue light the first neural system that is woken up is the dorsal attentional and visuomotor stream, in order to prepare us for rapid responding. Hence blue filters probably help these children by enhancing their arousal and this helps them to focus their attention more accurately, in particular by helping them to keep proper track of their eye movements, so that letters cease to appear to move around.

Not surprisingly therefore, we found that the blue filters not only improve arousal, concentration and reading, but they also reduce difficulties with getting to sleep at night because they help to improve synchronization of the body clock. What we didn't expect as well however, was that they also had a dramatic effect on these children's headaches. Even more surprising was that a mother with recurrent migraines, on noting that her son's headaches had greatly improved, began wearing blue filters herself; and her migraines disappeared. We have since found that if blue filters improve sleep patterns, they will often also improve headaches.

To summarize so far, the visual magnocellular system is crucial for reading. It is responsible for signaling the moments in time when visual events occur. Each time the eyes move, visual magnocells signal the command to move and also the resultant movements of the letters across the retina, so that these can be discounted, and the letters kept stationary. Thus, if the magnocell responses are weak, as in many dyslexics, letters may appear to blur or move around, making reading very difficult. But now we know that using either yellow or blue filters we can often help these children make the letters keep still.

With such a strong case, it may seem odd that the magnocellular theory is not yet generally accepted. However because phonology lies at the heart of reading, phonological impairments are fairly easy to show in most dyslexics, whereas the visual magnocellular impairment may be mild and not detectable at all in some. This may either be because it is absent and auditory problems predominate or too mild to be demonstrated using current techniques.

Nevertheless visual M- weakness probably contributes also to phonological problems. Morais and colleagues found that not until children, and also illiterate adults, learn that words can be represented as a sequence of separate letters, do they learn that they can also be broken down and heard as separate sounds (Morais, Cary, Alegria, & Bertelson 1979). Hence successful acquisition of phonological skill at the phonemic level probably depends to a large extent on first learning how a word can be visually represented orthographically. Indeed Chinese who have learned only to read Chinese characters, not alphabetic scripts, are not able to parse words down to the phonemic level until they learn an alphabetic script

(Read, Yun-Fei, Hong-Yin, & Bao-Qing 1986). In alphabetic scripts therefore, if orthographic analysis is compromised by weak visual magnocellular function, then acquiring good phonemic skills will naturally be secondarily affected also. This may help to explain the apparently paradoxical result of some studies; namely that the visual impairment in dyslexics correlate not only with measures of their visual/orthographic ability, but also with their phonological performance (e.g., Cestnick & Coltheart 1999).

7.24 Auditory Problems

However it is clear that some dyslexics have no visual reading difficulties at all. Particularly in dyslexics who have a background of speech and language delay, auditory analysis rather than visual sequencing inefficiency, may be their main problem (Bradley & Bryant 1978; Hornickel & Kraus 2013; Manis et al. 1997).

In order to break down a word into a sequence of phonemes, the auditory system needs to perform a series of operations analogous to those for visual sequencing. First the continuous speech sound needs to be broken down into words, then syllables and then phonemes. This is accomplished mainly by tracking changes in the amplitude and frequency of the sounds, also known as amplitude and frequency modulation (AM & FM). Words and syllables are marked by “stresses”, mainly amplitude peaks. In English often the first syllable is the primary stress. Speaking rate is about two words per second, syllable rate is about five per sec and the spoken phoneme rate is about ten per sec. Within each phoneme the amplitude and frequency changes that enable us to distinguish them occur at up to 50 times per sec.

There is a system of large neurones in the auditory system, analogous to the visual magnocells, which is specialized for tracking these sound modulations in real time. They can follow AM and FM frequencies from less than 1 to 100 Hz. Tallal was the first to suggest that developmental dysphasics may be poor at the rapid auditory temporal processing that is required for such discrimination of speech sounds (Tallal & Piercy 1973) and she subsequently showed that many dyslexics show a similar impairment. In the last few years there have been many more studies testing such basic auditory processing capabilities in dyslexics. Almost all have shown abnormalities in at least some dyslexics that could help to explain their phonological weaknesses (Hämäläinen, Salminen, & Leppänen 2013; McAnally & Stein 1996). Indeed auditory sensitivity to frequency and amplitude modulations has been shown to account for nearly 50% of individual differences in phonological skill (Witton, Stein, Stoodley, Rosner, & Talcott 2002).

Unlike in the visual system, the auditory system does not have an anatomically separate magnocellular pathway. Nevertheless it does contain large cells that seem to be specialized for temporal processing. These large neurones could be termed magnocells because, like visual magnocells, they may be recognised by M- specific antibodies, such as CAT 301 (Lurie, Pasic, Hockfield, & Rubel 1997). The detection of the frequency and amplitude changes in real time that underlies phonological

processing, seems to be mediated by these auditory magnocells. The development of auditory M- like cells may also be impaired in dyslexics. For example in dyslexic brains, like those in the dyslexic LGN, the magnocellular division of the auditory medial geniculate nucleus was found to contain fewer large cells on the left (Galaburda, Menard, & Rosen 1994). Impaired development of large M- like cells in the auditory system might therefore also underlie dyslexics' problems with acquiring good phonological skills. Interestingly deficient auditory temporal processing mediated by the auditory M- system is often accompanied by poor visual magnocellular function, suggesting a common aetiological causation, maybe involving immune recognition of the CAT 301 M- specific antigen (Talcott et al. 2000).

However not all dyslexics who show phonological problems can be shown to have either auditory or visual weaknesses; thus these are neither necessary nor sufficient to cause dyslexia. Some have argued from this that they cannot be considered causal at all (Ramus, Pidgeon, & Frith 2003). But this is like saying that because smoking is neither necessary nor sufficient to cause lung cancer it can never cause it – patently false. The probability is that impaired auditory and visual temporal processing are important, but not the only, causes of impaired phonological processing.

7.25 The Cerebellum

Impaired motor control is another potential cause of some dyslexic and dyspraxic problems. The cerebellum is the brain's autopilot responsible for automatizing motor skills. Since accurate timing of sensory feedback and motor outflow is an essential requirement for this function, the cerebellum receives a rich input from visual, auditory, proprioceptive and motor magnocellular systems (Stein 1986). The cerebellum plays an essential role in maintaining balance. Hence many people have studied balance stability in dyslexics. Almost all have confirmed that they show deficits (Fawcett, Nicolson, & Dean 1996; Gouleme, Gerard, & Bucci 2015; Stoodley, Fawcett, Nicolson, & Stein 2005). More direct evidence has come from measuring cerebellar morphology by magnetic resonance imaging (MRI) (Rae et al. 2002; Stoodley 2016). In fact differences between dyslexics and controls are seen in the cerebellum more consistently than any other part of the brain (Eckert et al. 2005).

Another way of strongly activating the cerebellum is by mean of visual tracking tasks (Miall, Weir, Wolpert, & Stein 1993). Nicolson et al. (1999) showed that cerebellar activation during learning to track a novel trajectory is greatly reduced in dyslexics. Thus in summary, there is now a great deal of evidence that cerebellar function is impaired in dyslexics, and this provides yet further indirect evidence for magnocellular involvement in dyslexic problems (Stoodley & Stein 2011).

7.26 Embodied Cognition

When reading, brain activity occurs not only in the classic “language areas” such as Wernicke’s and Broca’s areas, but also in areas which control movement. Evolutionarily our cognitive skills, particularly language, seem to have developed from neural representations of gestural movements (Corballis 2003). Hence cognitive reasoning probably engages subliminal activation of the motor neural systems that would participate in such gestures if they were actually produced. Thus cognition is “embodied”. Hence it is reasonable to postulate that training children’s awareness, “mindfulness”, of how they control their own movements might improve their cognitive skills.

We have therefore helped to develop a new classroom physical training program, called Move4words, based on embodied cognition, and administered it to pupils aged 7–13 years in 10 mainstream UK schools. We showed significant improvements in reading, writing and maths performance in those given the program compared with pupils receiving normal teaching. Struggling pupils performing in the lowest 20 percentile did particularly well (Hedges’ $g = 0.86$). Their performance gains were maintained for at least 1 year after the end of the intervention (McClelland, Pitt, & Stein 2014).

7.27 A General Magnocellular System for Temporal Processing?

Magnocells with rapid temporal processing, “transient” sensitivity and expressing M- cell surface antigens such as that recognized by CAT 301, are found throughout the whole nervous system (McGuire, Hockfield, & Goldman-Rakic 1989; Mueller, Davis, Sovich, Carlson, & Robinson 2016). As we have seen, when visual and auditory transient (M- cell) sensitivity are measured in the same individuals they tend to correlate with each other, suggesting that both might be under the same genetic neurodevelopmental control (Talcott et al. 2000). Therefore one can speculate that perhaps all the visual, auditory, memory and motor temporal processing impairments that are seen in dyslexics may be due to underlying weak development of this generalized, CNS wide, pansensory, transient processing, magnocellular system (Hari & Renvall 2001; Stein 2001). This impairment might affect different individuals more in one system than another, idiosyncratically, so that one dyslexic might suffer mainly visual problems, another mainly auditory, and a third mainly motor symptoms; he might be termed “dyspraxic”.

One can take this idea a stage further. Ramus showed in a small group of well compensated undergraduate dyslexics that only a few of them had demonstrable auditory, visual or motor problems, whereas despite their compensation most could still be shown to have residual phonological difficulties (Ramus et al. 2003). So he attributed the latter to a higher level developmental abnormality, perhaps in the

angular gyrus (Ramus 2004). Since the angular gyrus is an important node in the M- cell dominated dorsal visuomotor stream; clearly this higher level impairment might also be caused by impaired magnocellular connectivity.

7.28 The Immune System

Carla Shatz and her colleagues have shown that the development of magnocells, at least in the visual system and in the hippocampus, is regulated by the Major Histocompatibility Complex (MHC) cell recognition system which is the main controller of the immune system (Corriveau, Huh, & Shatz 1998). Most of the genes controlling the MHC system reside on the short arm of chromosome 6. It is unlikely to be a coincidence that among genetic studies of dyslexic families the most widely replicated linkage to reading problems is to just such sites on the short arm of chromosome 6 (Franks et al. 2004). Close to the MHC system genes are located the KIAA0319, DCDC2 and NRSN1 genes which are not only associated with dyslexia but also, as we have seen, involved in cell/cell signaling, helping to control the developmental migration of neurones and their interconnections (Paracchini et al. 2006).

Thus the immune system probably plays an important role in the development of “magnocellular” timing systems all over the brain. It has been known for some time that dyslexia and related neurodevelopmental conditions are associated with an increased incidence of immunological abnormalities, in the children or their families (Bashir & Al-Ayadhi 2015; Galaburda 1993; Jariabkova, Hugdahl, & Glos 1995; Lahita 1988; Warren et al. 1990).

The developmental disorder, arthrogryposis multiplex congenita is caused by maternal antibodies attacking the fetal isoform of the acetylcholine receptor (Riemersma et al. 1996). Jacobson, Polizzi, Morriss-Kay, and Vincent (1999) showed that this condition could be reproduced in mice by maternal-to-fetal transfer of the human maternal IgG antibodies. We therefore postulated that maternal antibodies might likewise contribute to the impaired brain development seen in dyslexia and other neurodevelopmental disorders. To test this hypothesis, we obtained sera from mothers of children with dyslexia and injected them into pregnant mice to see if the sera would affect the development of their pups. As we have seen, both functional and structural changes have been described in the cerebellum in dyslexia (Fawcett et al. 1996; Nicolson et al. 1999; Rae et al. 1998), so we particularly focused on alterations in cerebellar function. The pups exposed in utero to the serum of the women who had two or more dyslexic or autistic children, showed deficits in motor tests which correlated with lower cerebellar neuronal choline and creatine levels. Thus it seems that maternal antibodies can contribute to dyslexic deficits, and possibly to other neurodevelopmental disorders.

7.29 Handedness

From the very first descriptions of either acquired or developmental dyslexia, the idea that it might be related to abnormal lateralization of the cerebral hemispheres became widely accepted, mainly because it was already known that speech and language skills were centered in the left hemisphere, which was thought to be the explanation why the majority of people wrote with their right hands. Hence it was natural to conclude that among individuals with dyslexia left handedness might be more frequent than expected. The truth has turned out to be rather more complex than this, but the basic association between anomalous laterality and dyslexia still stands. Imaging has shown that handedness and hemispheric lateralization for speech are not directly related however. In most left handers speech is still controlled mainly by the left hemisphere, and most dyslexics are still right handed (Herve, Zago, Petit, Mazoyer, & Tzourio-Mazoyer 2013). Nevertheless dyslexics tend to be less skilled with either hand, therefore they are less strongly either left or right handed than typically developing children, but overall they are no more likely to be left handed for writing.

Nevertheless magnetic resonance imaging experiments have proved unequivocally that atypical brain hemispheric specialization is indeed associated with dyslexia. The left planum temporale is a structure at the back of the temporal lobe which is known to be important for the comprehension of speech. In typically developing children it is larger on the left than in the right hemisphere; but in dyslexics the right side is larger than normal, so that their hemispheric asymmetry is reduced (Leonard & Eckert 2008).

In typically developing children a network of left sided structures is activated when reading. These include the left anterior fusiform gyrus underneath the occipital lobe, the angular and supramarginal parts of the left posterior parietal cortex and the left inferior frontal gyrus, but in dyslexic children these areas activate much less during reading, whereas homologous areas in the right hemisphere activate more, again leading to reduced asymmetry (Zhao, Thiebaut de Schotten, Altarelli, Dubois, & Ramus 2016). Of course these experiments cannot by themselves tell us whether the reduced asymmetry is a cause or consequence of their impaired reading, but recent studies in young infants before reading onset show similar reduced asymmetries (Lyytinen, Erskine, Hämäläinen, Torppa, & Ronimus 2015). This suggests strongly that the anatomical differences precede reading failure and therefore probably contribute to their causation.

It used to be thought that hemispheric asymmetry is uniquely human. But it is now known that left/right asymmetries are found throughout the animal kingdom. For example marine mammals show stronger left hemisphere controlled right turning biases than even man's right handedness; and left sided dominance for vocalizations is found in frogs, birds, rats and mice. Two thirds of our closest relatives, chimpanzees are right handed. Conversely a right hemisphere/ left visual field dominance for detection and expression of emotions, visuospatial skills and attention is present in all primates so far investigated, suggesting an evolutionary

provenance extending back at least 50 million years. Thus the origin of hemispheric specialization greatly predates speech. It probably relates to early specialization of the left hemisphere for correctly sequencing actions, hence gestures which evolved into speech, whilst the right hemisphere became specialized for the holistic allocation of attention (Corballis 2009).

However the molecular basis of these asymmetries is almost unknown. We have carried out a genome-wide association study meta-analysis for a quantitative measure of relative hand skill in individuals with reading disability (Brandler et al. 2013). In dyslexics the strongest association was found with the PCSK6 gene. But this locus is not associated with relative hand skill in the general population. As PCSK6 is known to regulate NODAL in the development of left/right (LR) asymmetry in mice, we developed a novel approach to GWAS pathway analysis, using gene- set enrichment to test for an over-representation of highly associated variants within the orthologs of genes whose disruption in mice yields LR asymmetry phenotypes. Four out of 15 LR asymmetry phenotypes showed an over- representation. We replicated three of these phenotypes; situs inversus, heterotaxia, and double outlet right ventricle, in the general population. Thus it seems that handedness is a polygenic complex trait controlled in part by the molecular mechanisms that establish LR body asymmetry early in development, but particularly in dyslexics.

7.30 Omega 3 Long Chain Polyunsaturated Fatty Acids

One gene, not implicated in neuronal migration early in development, but strongly linked to dyslexia, is the MCR5 gene situated on Chromosome 18 (Scerri et al. 2011). This gene is of great interest because it is known to be involved in stress, production of sebum and pheromones, obesity, immunity and inflammation. What links all these functions is probably its role in the metabolism of fatty acids. It encodes one of the huge family of seven-pass transmembrane G protein-coupled receptors that stimulate cAMP signal transduction. The encoded protein is a receptor for melanocyte-stimulating and adrenocorticotrophic hormones and it is one of the few of these MSH receptors that is expressed in the brain; it is particularly strongly expressed in the retina and in the hypothalamus which explains its role in fatty acid and metabolic control and obesity. Although this has not been directly demonstrated, its functions in fatty acid metabolism, the retina, and immunity together with its association with dyslexia suggests that it may be particularly important in magnocellular function.

A crucial component of all excitable cell membranes, particularly magnocells because of their very large surface area, is a unique long chain omega 3 polyunsaturated fatty acid (LCPUFA), docosahexaenoic acid (DHA). This has exactly the right physical and electrostatic properties in the membrane lipid bilayer to maintain the separation of charges underlying the neuronal resting membrane potential. More importantly, due to its flexibility, it facilitates rapid reorganization of the membrane

when ionic channels open and close to allow the ionic currents that cause action and synaptic potentials to occur. Thus if it is replaced in artificial membranes by saturated fats of the same physical length, channel opening and closing times can be prolonged by up to ten times. The main source of DHA comes from eating oily fish because humans cannot synthesize LCPUFAs from scratch. But the modern Western diet contains very little fish and 75% of teenagers in the UK eat no fish at all. Our brains contain 5 g of DHA but we lose about 5 mg per day. So this has to be replaced if magnocellular membranes are going to function optimally. Yet the majority of the population has dangerously low levels of DHA together with another important omega 3 LCPUFA, eicosapentaenoic acid (EPA). In recent studies of children from disadvantaged backgrounds, discussed later, we found that their red blood corpuscle levels of DHA & EPA were less than 1/2 that regarded as optimal.

These considerations encouraged us to investigate whether giving children with reading problems omega 3 DHA & EPA supplements might help them to improve their magnocellular function and with it their reading. In double blind randomized control trials my colleagues have shown that this is indeed the case (Montgomery, Burton, Sewell, Spreckelsen, & Richardson 2013; Parletta, Niyonsenga, & Duff 2016; Richardson, Burton, Sewell, Spreckelsen, & Montgomery 2012; Richardson & Montgomery 2005). After 3 months of supplementation and without any extra reading help, children with initially low reading ability and very poor diets given extra DHA improved their reading by 9 months on average.

7.31 Antisocial Behavior

Another change we observed after improving the diet of these poor readers, noticed also by their teachers, was that they were much less prone to fight each other in the playground. This was probably not just because they were learning to read more easily, but because they were more sociable and less easily angered. In 1942, Hugh Sinclair, a pioneer in human nutrition, helped to persuade the wartime British government to supplement the diet of all pregnant mothers and their infants with cod-liver oil; along with Vitamins A & D, this contains omega 3 LCPUFAs. He had speculated that among other ills, poor diets could lead to antisocial behavior (Sinclair 1956). Since that time, ample evidence has accrued that he was right; and improving diet can reduce such antisocial behavior (Corrigan et al. 1994; Gesch, Hammond, Hampson, Eves, & Crowder 2002; Hamazaki et al. 1996; Hibbeln et al. 1998; Rutter, Giller, & Hagell 1998).

I have postulated that the link between deficient omega 3 LCPUFA levels and antisocial behavior is that the temporal processing mediated by magnocellular neurones is crucially important not only for reading but also for social communication-tracking facial expressions, anger, pleasure etc. Hence I was not at all surprised to find that our poorly reading children's behavior improved with the omega 3s they were receiving. We have therefore carried out a more formal trial in which we showed that supplements of omega 3s, minerals and fatty acids not only raised

the omega 3 levels in the blood of poorly fed teenagers, from initially dangerously low values, but it also did improve their sociability (Tammam, Steinsaltz, Bester, Semb-Andenaes, & Stein 2015). We have also carried out a large trial giving 750 prisoners in 3 jails in the UK capsules containing either fish oils, vitamins and minerals or placebo for 4 months; this also confirmed that this improvement in their diet improved their behavior significantly.

7.32 Dyslexia Talents

Dyslexia is extremely common; estimates of its prevalence range from 5 to as high as 17%. The precise figure depends on exactly how it is diagnosed. Such high numbers in a strongly hereditary condition suggests the operation of a “balanced polymorphism”. This is when an apparently deleterious version of a gene is retained in the gene pool at relatively high rates because as well as its disadvantage, it confers a selective advantage. In the case of reading this selective advantage might have been overwhelming, until recently when inability to learn to read became a general disadvantage. What might that advantage have been?

More artists than you’d expect by chance (Everatt, Steffert, & Smythe 1999; Wolff & Lundberg 2002), and architects, engineers and entrepreneurs (Logan 2009) are dyslexic. But this doesn’t prove that dyslexics are inherently more artistic or entrepreneurial. Maybe art, architecture and risky business attract dyslexics simply because there’s less reading involved in these pursuits. However this argument is not very convincing. Art and architectural courses include art history which requires a great deal of reading. Engineering and business also need a fair amount of reading.

Unfortunately scientific research on the talents of people with dyslexia is rare, even though its possible advantages are widely discussed. Because it is widely known that the left hemisphere is specialized for speech and language, it is often argued that dyslexics have “stronger right hemispheres” and that this may compensate for their poor reading skills. It may endow them with exceptional artistic and creative talents, and this may be what keeps the “dyslexia genes” in the human gene pool. However such conjectures are very difficult to substantiate scientifically. Artistic talent is difficult to measure; creativity, whether in the Arts, engineering or business, is very subjective. For every study that claims talents in dyslexics there is another that disputes them. This may result from an overly simplistic and crude conceptualization of right hemispheric function.

To settle whether dyslexics have inherently superior right hemisphere skills for art etc., first we would need to agree what general skill a right hemisphere advantage would provide them with and then agree a test or selection of tests which will reliably measure that skill. We would then need to administer them to a large group of dyslexics compared to matched controls. Despite the importance of answering this question, it has proved impossible to raise funds to do such a study. In the meantime however there have been a few small studies that point to dyslexics indeed showing superiority in some tasks likely to involve the right hemisphere. These are

tasks that do not depend on sequential local temporal processing (bit by bit), but do call on static global holistic skills. Thus dyslexics tend to show superior ability in tasks calling on the visual parvocellular system, suggesting that magnocellular weakness in dyslexics may be compensated by parvocellular strength, during the competition for connectivity that occurs during the early development of the brain. These P- strengths include higher sensitivity to stationary high spatial frequency gratings (Bednarek & Grabowska 2002), better blue/yellow color discrimination (Dain, Floyd, & Elliot 2008), quicker identification of “impossible figures” (von Károlyi, Winner, Gray, & Sherman 2003). The latter requires ability to distribute current attention over a wider area, a skill characteristic of dyslexics (Geiger & Lettvin 1987) which may be mediated by greater long range connectivity of the p-system, particularly in the right hemisphere. This aspect of dyslexics’ attention, often treated as a disadvantage, may explain their superiority in virtual reality visuospatial localization (Attree, Turner, & Cowell 2009).

In summary, although there is much dispute it seems likely that dyslexics do demonstrate superior skills in certain kinds of visuospatial tasks, in particular those that depend on wide ranging, holistic appreciation of a scene rather than moment to moment sequential processing.

7.33 Conclusion

Since Livingstone et al. (1991) first showed a selective visual magnocellular deficit in dyslexic people, the great majority of the neuroanatomical, electrophysiological and psychophysical evidence has supported the hypothesis that a significant proportion exhibit a visual magnocellular weakness. Analogous neural systems have been demonstrated in the auditory, somatosensory, motor systems, indeed throughout the whole brain, which are characterized by their expression of particular surface signature molecules. Many of these magnocellular temporal processing systems have also been shown to be impaired in some dyslexics. This suggests that a fundamental disorder of temporal processing mediated by magnocellular systems throughout the brain may underlie all the manifold and variable symptoms of dyslexia. The particular areas of the brain most affected as a result of genetic inheritance, random play of immune attack, poor nutrition make each individual completely unique, but may also endow exceptional talents.

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Chapter 8

Auditory Processing in Developmental Dyslexia: Some Considerations and Challenges



Caroline Witton and Joel B. Talcott

Abstract It is generally agreed that some people with dyslexia exhibit apparent impairments in auditory tasks, but there is no consensus about the underlying nature or aetiology of such impairments. Convergent evidence from a wide range of tasks suggests that any physiological explanation for auditory impairments in dyslexia must be centred at the level of thalamo-cortical and/or cortical mechanisms rather than low-level mechanisms such as basic neural timing. The literature on auditory processing in dyslexia shows high variability in the magnitude of the effects across studies, reflecting phenotypic heterogeneity in the dyslexic population as well as in task design. Measurement effects, especially when adaptive procedures are shortened or when participants make high numbers of “lapses”, may also mean that thresholds are inaccurate which can further add to difficulties in interpreting auditory data. These factors combined mean that auditory thresholds probably reflect a complicated mixture of pure sensory abilities and the additional neurocognitive mechanisms that are required for the overt perception and recognition of stimulus dimensions being tested in a given task, as well as task compliance. Future studies aiming to unpick auditory impairments in dyslexia should place strong emphasis on study design, including choice of psychometric variables and auditory measures.

Keywords Auditory · Temporal processing · Dyslexia · Development · Frequency · Language · Phonological awareness · Reading

8.1 Introduction

The literature exploring auditory processing in developmental dyslexia spans nearly four decades, and although it is now generally agreed that some people with dyslexia exhibit apparent impairments in auditory tasks, there is no consensus about the

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underlying nature or aetiology of such impairments. Despite a number of theories implicating particular physiological mechanisms in these impairments, the evidence in their support is often weak, because of high levels of inter-individual variability and a lack of consistency across tasks which ought to tap into the same basic mechanisms (see Farmer & Klein 1993; Habib 2000; Ramus, Pidgeon, & Frith 2003; Talcott & Witton 2002; Wright, Bowen, & Zecker 2000). In this chapter we consider what conclusions can reliably be drawn from the literature on auditory processing in dyslexia, and explore some of the challenges which have limited progress in this area of research.

8.2 Basic or High-Level Auditory Impairment?

The quantitative review by Hämäläinen, Salminen, and Leppänen (2013) provides a useful summary of the range of auditory tasks which have yielded consistent between-group effects in developmental dyslexia, including: pitch discrimination, detection of slow frequency (pitch) modulations, discrimination of amplitude rise-time, and discrimination of sound duration. Other studies have reported moderate to strong statistical relationships (i.e., correlations) between different aspects of auditory processing and phonological skills in dyslexia (e.g., Boets, Wouters, van Wieringen, Smedt, & Ghesquière 2008; Goswami et al. 2002; Witton et al. 1998) and in typically developing participants (Ahissar, Protopapas, Reid, & Merzenich 2000; Talcott, Hansen, Assoku, & Stein 2000), although a lack of group differences or associations with reading component skills has also been reported (e.g., Hill, Bailey, Griffiths, & Snowling 1999; Rosen 2003; White et al. 2006). From the perspective of face validity, these observations are broadly consistent with the historical view in the literature that dyslexia is associated with a basic impairment of auditory temporal processing (e.g., Farmer & Klein 1995; Tallal 1980), or a more generalized cross-modal impairment of the detection and discrimination of stimulus dynamism (Stein & Walsh 1997; Talcott & Witton 2002; Tallal 1993). Several commonly-used tasks depend, at least in part, on fine temporal processing: pitch discrimination at low frequencies requires accurate phase-locking of neural activity in the auditory periphery to the fine structure of acoustic waveforms, as does encoding of the rapid amplitude changes in rise-time and duration tasks (Moore 2013).

Widely-reported group differences in these different measures offer support to the auditory temporal processing impairment hypothesis. However, to robustly test this, it is necessary to also explore the literature about stimuli which have *failed* to reveal consistent group differences. The most exacting measures of auditory phase-locking are those which test binaural hearing. By resolving the fine structure of a stimulus at each ear, the auditory system is able to detect tiny inter-aural delays, as short as 50 μ s under optimal conditions. Measures such as the binaural masking-level difference, sensitivity to illusory binaural pitches (e.g., Huggins pitch), or detection of inter-aural phase modulations, can only be completed by utilizing

binaural processing based on exquisite phase-locking accuracy. If the auditory processing impairments in dyslexia were associated with a general impairment of temporal resolution, the most robust group differences would be predicted to occur in such tasks of binaural hearing. The literature on these tasks is equivocal, however, with some reports of significant between-group effects (Dougherty, Cynader, Bjornson, Edgell, & Giaschi 1998; Edwards et al. 2004; McAnally & Stein 1996; Patterson, Uppenkamp, Johnsrude, & Griffiths 2002; Putter-Katz, Feldman, & Hildesheimer 2011) and others negative results (Amitay, Ben-Yehudah, Banai, & Ahissar 2002; Chait et al. 2007; Johnson et al. 2013; Santurette et al. 2010). Another auditory task which relies on accurate encoding of waveform fine-structure is gap detection, and again, findings for this stimulus type have been inconsistent (see Hämäläinen et al. 2013). Taken together, the evidence suggests that the most consistent group differences in perception of pitch, amplitude rise-time, and sound duration, cannot be simply explained by a basic peripheral impairment in auditory temporal resolution but instead emerge at higher levels of processing. Indeed, a great deal of auditory processing takes place at levels beyond those which rely on basic temporal codes. Fine temporal resolution diminishes as the neural representation of sound progresses towards cortex, and from auditory cortex there is an increased manifestation of neural codes based on firing rates, which represent “processed temporal information” of the form required for integration with other sensory and cognitive systems (Wang, Lu, Bendor, & Bartlett 2008). The network of cortical areas ultimately engaged through auditory processing is extensive, incorporating the entire superior temporal gyrus, large portions of parietal and prefrontal cortices, and the limbic system (Poremba et al. 2003).

Instead of focussing on temporal processing per se, some authors have examined the auditory system’s ability to encode stimulus dynamics, proposing that differences for example in detection of slow frequency changes (Talcott & Witton 2002; Witton et al. 1998) or amplitude rise-times (Goswami et al. 2011) may be related to segmental processes in speech perception underlying the extraction of phonological information. Such effects need not depend on a peripheral timing mechanism, as neuronal selectivity for slow rates of frequency modulation does not emerge until auditory cortex (Altmann & Gaese 2014). While cortical levels of processing respond only to lower rates of amplitude modulations, they also show an increased tolerance to changes in other stimulus properties such as the level or type of sound (Joris, Schreiner, & Rees 2004).

Whatever the key characteristic of the auditory processing difficulties observed in dyslexia, it seems increasingly clear that any physiological explanation must be centered at the level of thalamo-cortical and/or cortical mechanisms rather than low-level mechanisms.

The genetic basis of dyslexia is proposed to be both polygenic and heterogenic, and linked to candidate genes involved in neuronal migration and axon guidance during brain development (Carrion-Castillo, Franke, & Fisher 2013). Studies exploring auditory processing in rodent genetic knockdown models of dyslexia are beginning to emerge, and indicate that differences in auditory processing may co-occur with the cortical disruption caused by genetic manipulation in utero.

For example, differences in performance on an auditory oddball task, but not for more simple tone detection or sequence discrimination tasks were observed in mice treated in utero with RNA interference of *DYX1c1* (Threlkeld et al. 2007), and for detection of frequency sweeps in *KIA0319*-knockdown mice (Szalkowski et al. 2012). This evidence supports the view that auditory processing disorders in dyslexia may result from widespread, subtle, anomalies in cortical development which lead to abnormal thalamo-cortical circuits and cascade to affect the sensory and cognitive processes which underpin the development of the skills needed for proficient reading (Galaburda, LoTurco, Ramus, Fitch, & Rosen 2006).

It is also important to consider whether higher-level processes at the interface between sensory perception and more general aspects of cognition could account for the group differences which have been reported on auditory tasks. For example, one account has suggested that the auditory processing impairments reported in dyslexia result from difficulties in stimulus identification, rather than in perception (Ramus & Ahissar 2012). Our own work (Hulslander et al. 2004; Witton et al. 2002) has highlighted the importance of accounting for the effects of cognitive variables such as working memory when exploring relationships between auditory processing and reading. In other disorders, such as congenital amusia (“tone-deafness”), impairments may only emerge as task difficulty is increased (Foxton, Dean, Gee, Peretz, & Griffiths 2004). Processing at the cortical level, even in primary auditory cortex, can be modulated extensively by “top down” factors such as attention (King & Nelken 2009) and this interface between sensory and cognitive factors, at the cortical level, may be critically important in developmental disorders such as dyslexia.

8.3 Heterogeneous Effects in a Heterogeneous Population?

While systematic reviews and meta-analyses (Benassi, Simonelli, Giovagnoli, & Bolzani 2010; Farmer & Klein 1993; Hämäläinen et al. 2013) have repeatedly confirmed the presence of moderate effect-sizes for group differences on sensory processing tasks, including auditory ones, all these findings are characterised by high variability in the magnitude of the effects across studies. Our recent meta-analysis of frequency discrimination effects in dyslexia confirm statistically that the effect-sizes are heterogeneous across studies (Witton, Swoboda, Shapiro, & Talcott, unpublished). In the literature more widely, and including our own work, significant effect-sizes at the group level are often accompanied by substantial within-group variability, which is nearly always larger in the sample of individuals with dyslexia – identifiable by larger group standard deviations for thresholds. Indeed, at least some individuals in the dyslexia groups could often be described as not having impairments.

One possible source of this variability in auditory measures in dyslexic populations is that the population itself is heterogeneous – something we know to be true, with the diagnosis of dyslexia representing a phenotype that has substantial intra-

class variability in the underlying cognitive and neuropsychological dimensions that accompany the reading impairments upon which it is defined (Pennington 2006). If auditory processing impairments are directly linked to some underlying characteristic of dyslexia (either causally or through third variables), then phenotypic variability could result in the kind of mixed results that are seen in the literature. This heterogeneity is a problem for the quasi-experimental research designs that are conventionally employed in investigations of sensory processing in dyslexia. Given the relative scarcity of pure phenotypes of dyslexia, it is likely that the significant variability between studies on sensory processing tasks results at least in part from methodological differences in sample selection or ascertainment (Hogben 1996; McArthur & Bishop 2004a,b). For example, the presence of uncontrolled cognitive or developmental factors (Dawes & Bishop 2008; McArthur et al. 2012; Roach, Edwards, & Hogben 2004) potentially contribute both to high inter-individual variability across studies, and associated differences in effect-sizes across groups. A related factor is the presence of symptom sets such as in attention capacity that are associated with other developmental disorders that have a high incidence of overlap with dyslexia.

It should be possible to account for heterogeneity through careful study design and thorough use of psychometrics. But for frequency discrimination, we have found it difficult to draw firm conclusions through our meta-analysis of the literature about why this heterogeneity has arisen (Witton, Swoboda, Shapiro, & Talcott, unpublished). This is for two main reasons: probable ceiling effects in key measures (including a lack of standardized measures of reading), and wide differences in psychophysical task design. Looking across studies, there was no significant relationship between frequency discrimination and non-word reading, the most widely-used measure of phonological skills. This was unexpected, but post-hoc examination of the group scores within studies revealed strong average non-word reading performance in control groups, close to statistical ceiling in many cases. Because of the restricted variance that this causes, it becomes statistically inappropriate to look for relationships with non-word reading either within studies or in a meta-analysis. While seven studies had used a more sensitive measure, phoneme deletion (which did yield a significant meta-regression with frequency discrimination), this was only a small subset of the overall body of work. Overall, it was extremely difficult to draw conclusions from meta-regressions which might explain the heterogeneity of effect-sizes in frequency discrimination, because of a lack of appropriate psychometric covariates. Relatively few studies had used standardized measures of reading or phonological skills which would help comparison across populations with minimal ceiling effects. Very few studies had included other psychometric measures that tap important constructs such as working memory or attention, or even other reading sub-skills. This is a clear limitation of the literature on frequency discrimination (and the wider auditory processing literature) in dyslexia, and without improvements in the choice or design of psychometric tests it is unlikely that researchers will be able to easily unpick any cognitive explanations for the heterogeneity in effects.

8.4 The Challenging Nature of Auditory Tasks

The majority of evidence about auditory processing in dyslexia comes from psychophysical tasks. Here, participants are typically asked to listen to “trials”, typically consisting of sequences of two or more stimuli separated by a silent inter-stimulus interval. One of the stimuli is designated the “target” and the listener is asked to identify this by responding verbally or with a button-press. For example, if the task is auditory frequency discrimination, the trials might contain tones that differ only in frequency, with the participant required to select the higher-frequency tone – the target. The size of the frequency difference would be manipulated by the experimenter. Or, in a gap-detection experiment, the listener might hear two bursts of noise, and the listener would be required to pick the noise containing a silent gap, with the duration of the gap manipulated by the experimenter. In all cases, the target is as likely to be in the first as in the second interval. The participant would need to listen and respond to large numbers of trials (determined by the experimenter and discussed further below), over a period of several minutes, to obtain enough data for the detection or discrimination “threshold”, a measure of sensitivity, to be computed using the principles of signal detection theory (Green & Swets 1966). Usually this consists of an adaptive procedure which will adjust the stimulus strength until the participant’s performance matches a predetermined level.

The serial nature of stimulus presentation in an auditory psychophysical task means that it relies not only on the participants’ sensory sensitivity, but also on their working memory for comparison of sounds heard in succession, and the necessary attention span to produce reliable responses. Over large numbers of trials these tasks are boring to complete and so they rely heavily on the compliance of the participant, which can be a particular challenge when working with young children.

The adaptive procedures that are most often used to determine threshold were typically designed for use with trained listeners in a laboratory setting, based on hundreds of trials. But researchers working with one-off volunteers, especially children, may decide to shorten the procedures so that they use fewer trials. This is particularly likely if they also need to collect large amounts of psychometric data during a measurement session, where saving time may be a priority. However, simulations of adaptive procedures using fewer trials shows that they can be rather inaccurate, with a tendency to over-estimate thresholds and increased “measurement noise” (Witton et al. 2017). The measurement noise (i.e., a reduction in how closely the measured threshold relates to the actual underlying threshold) can make it more difficult to detect group differences, and may account for some apparent heterogeneity in individual scores.

Further problems arise if the participants do not respond consistently. Several authors have noted that children often respond erratically in these kinds of tasks: 41% of children with dyslexia or SLI who completed up to 140 runs of an auditory frequency-discrimination task responded inconsistently with no improvement across runs (McArthur et al. 2012); and nearly 50% of children may be unable to produce response-patterns with adult-like consistency even after training (Halliday,

Taylor, Edmondson-Jones, & Moore 2008). It has been suggested that inconsistent responding produces widely varying scores on psychophysical tasks (Roach et al. 2004). In some of our previous work (Hulslander et al. 2004; Talcott et al. 2002), we introduced easy “catch-trials” into our procedures, in an attempt to index the participants’ level of vigilance during the task in a way that could be used as a covariate. We found that children were responding incorrectly on anywhere between 5% and 19% of these trials on average, and that this differed according to reading group. This observation is not surprising, given that we know that reading problems are associated with poorer working memory, and symptoms of attention-deficit/hyperactivity disorder (ADHD).

We have simulated the effects these “lapses” have on the measured thresholds using adaptive procedures, and found that they are significant (Witton et al. 2017). Lapses, modeled as occasional responses which are random rather than depending on the underlying psychometric function of the simulated observers, also increase the measurement noise in psychophysical thresholds. This means that measured thresholds bear a weaker relationship with the participant’s actual threshold, with some considerably higher. This can be enough to generate artificial group differences (Witton et al. 2017): in a simulation using 20 reversals of a Levitt 2-down, 1-up staircase, where the only difference between groups was lapse-rate (i.e., veridical thresholds were identical), comparing to the group making 0% lapses, a group making 5% lapses would show a spurious, statistically significant group difference if they contained 45 individuals. A significant group difference would emerge with only 15 individuals if the second group were making lapses on 10% of trials (2-sample t-test, 80% power, $p < 0.05$). This finding has clear implications for researchers studying auditory processing in dyslexia. It is reasonable to expect that a group of dyslexic individuals might make more lapses than controls, so researchers should consider ways of taking this into account in statistical analyses. It is impossible to know the true lapse-rate in any task, because we can never measure the reasons why a participant responded in the way they did. But we can attempt to index performance by the use of measures such as catch-trials and incorporate this information into our analyses. It is also important to do as much as possible to reduce lapses, by making tasks as interesting as possible (see for e.g., Abramov et al. 1984); and to remember that simply shortening tasks may not be the best solution, as discussed above.

Irrespective of pure measurement effects such as those discussed above, individual differences in cognitive skills may interact with sensory sensitivity to affect thresholds. Importantly, group effects may reflect dissociations in the way groups of participants execute a psychophysical task, for example, differences in memory capacity related to the maintenance of memory traces over sequential presentation of stimuli (Ahissar 2007; Ben-Yehudah, Sackett, Malchi-Ginzberg, & Ahissar 2001), or differences in perceptual learning. Dyslexia is associated with reduced working memory and digit span has been identified as a significant predictor of performance on frequency discrimination (Banai & Ahissar 2004, 2006) as well as other auditory psychophysical tasks (Hulslander et al. 2004; Witton et al. 2002). Psychophysical task design is therefore another potentially important variable determining the

results of auditory processing measures in dyslexia. In our meta-analysis of frequency discrimination (Witton, Swoboda, Shapiro, & Talcott, [unpublished](#)), we identified five different trial-designs used in different studies. These ranged from two-tone designs where participants were asked to identify whether tones were the same or different, or which was the higher in pitch, to tasks with sequences of tones that either changed in pitch or not. We presume that these each present a different cognitive load, and indeed we found a significant effect of task design on effect-size for frequency discrimination thresholds, although there were too few in some categories to perform more detailed analyses. Other studies have explored in detail the effects of certain aspects of task design and found that dyslexics may differ substantially in the way that they use information from the task-design, explored specifically in Ahissar's work regarding the Anchoring hypothesis (e.g., Ahissar 2007). Thus, auditory thresholds probably reflect a combination of pure sensory abilities and the additional neurocognitive mechanisms that are required for the overt perception and recognition of stimulus dimensions being tested in a given task.

8.5 Alternative Approaches

An alternative approach to measuring sensory sensitivity, which bypasses the need for obtaining behavioral response from participants, is to use neurophysiological measures. This approach has been fruitful in exploring auditory processing in dyslexia and has the benefit that measurements can be taken from children and babies before they exhibit signs of dyslexia. For example, electrophysiological studies have shown that atypical responses to differences in pitch are already present in children at familial risk of dyslexia before they learn to read (e.g., Leppänen et al. 2010; Maurer, Bucher, Brem, & Brandeis 2003). Hämäläinen et al. (2013) provides a systematic review of the smaller body of evidence from electroencephalography (EEG) and magnetoencephalography (MEG) work and, importantly in the context of measurement effects in behavioral work, find that it follows the same pattern as findings as the psychophysical literature.

There are nevertheless some challenges associated with these kinds of study as well. Like psychophysics, neurophysiological responses are not necessarily restricted to sensory processing. For example, the widely-used mismatch-negativity (MMN) response is modifiable by contributions from sources in the frontal lobes, and is sensitive to the cognitive symptoms of disorders such as schizophrenia, so although considered pre-attentive in origin it is not entirely free from cognitive influences. Using different approaches it is possible to construct "cortical psychometric functions" from auditory evoked responses measured with MEG, an approach which shows promise for bias-free estimates of threshold (Witton et al. 2012) although it has yet to be developed for other stimuli. There are also practical problems in successfully using neuroimaging techniques with children (Witton et al. 2014).

8.6 Conclusions

Despite a large number of studies which have shown group differences auditory processing for dyslexia, there remains a lack of consensus about the underlying reasons for this. Statistical effects are inconsistent, for at least two main reasons: First, dyslexia is itself a heterogeneous disorder, especially with respect to the underlying cognitive correlates of reading disability (e.g., Ramus et al. 2003; Talcott et al. 2013), such that group-based studies are ill-posed to identify critical relationships with auditory processing. Second, the psychophysical measures used to determine sensory sensitivity are complicated by individual differences in performance consistency, and do not lend themselves well to shortening for use with children or other naïve participants, resulting in “noisy” data. The balance of evidence from work across a range of auditory stimuli suggests that problems emerge at the cortical, rather than peripheral, level of processing and result from effects occurring at the interface between sensory and neurocognitive processes. Future studies aiming to unpick auditory impairments in dyslexia should place strong emphasis on study design, including choice of psychometric variables and auditory measures.

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Chapter 9

From Auditory Rhythm Processing to Grapheme-to-Phoneme Conversion: How Neural Oscillations Can Shed Light on Developmental Dyslexia



Marie Lallier, Mikel Lizarazu, Nicola Molinaro, Mathieu Bourguignon, Paula Ríos-López, and Manuel Carreiras

Abstract Developmental dyslexia is frequently associated with phonological difficulties such as poor phonological awareness, access, or short term memory skills, that further impede the acquisition of letter-to-sound mappings. Some hypotheses suggest that phonological disorders in dyslexia are themselves caused by a more basic auditory processing deficit. Here, we review evidence showing that a high sensitivity to auditory rhythmic cues may be critical for phonological and reading development. Moreover, the brain signature of prosodic and rhythmic processing difficulties in dyslexia may reside in atypical right hemisphere synchronization to slow frequency auditory modulations, that would then generate left hemisphere-based dyslexic reading symptoms. Overall, the data presented in this chapter suggests that interventions aimed at facilitating the extraction of rhythmic and

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temporally regular patterns in auditory sequences could improve reading in dyslexia through the enhancement of phonological skills.

Keywords Dyslexia · Speech rhythm · Neural entrainment · Oscillations · Phonology · Prosody · Music

9.1 Introduction

Developmental dyslexia (dyslexia, hereafter) affects 3–10% of the population and is diagnosed when severe reading difficulties (at least 2 years of delay compared to the chronological age level) appear whereas no apparent explaining factor can be found, such as low IQ, sensory or psychiatric disorders, or abnormal schooling. Researchers agree on the multifactorial nature of the cognitive cause(s) that lead to reading acquisition problems, since the heterogeneity of the deficits reported so far in the dyslexic population cannot be explained by one single theoretical account (Bosse, Tainturier, & Valdois 2007; Peyrin et al. 2012). Still, one type of difficulties seems to frequently and consistently occur in most dyslexic individuals (at least in alphabetic orthographies): they display impaired phonological (i.e., linguistic, auditory) processing (Landerl et al. 2013). More particularly, dyslexic individuals exhibit difficulties when they have to identify, manipulate, memorize or access the sounds of language, the phonemes. These phonemic processing difficulties are thought to directly affect the build-up of the links between the graphemes of a language and their corresponding sounds, which are necessary for decoding new words accurately (Vellutino, Fletcher, Snowling, & Scanlon 2004). The presence of phonological and decoding impairments during the first stages of reading development would severely hinder the formation of the orthographic lexicon upon which fluent reading relies (Share 1999, 2004).

The high prevalence of phonological deficits in the dyslexic population (Ramus et al. 2003) has pushed researchers to look for the possible underlying cause of these problems. In particular, data suggests that the phonological disorder in dyslexia is rooted in an abnormal development of the processes that support language acquisition and speech perception from birth (Leppänen et al. 2012; Lyytinen et al. 2001). Therefore, infants who will develop reading difficulties may fail to use the relevant cues in the speech signal in order to make sense of their phonological world and acquired the phonemic repertoire that will later be used for grapheme-to-phoneme conversion.

In the present chapter, we will first review evidence showing that rhythmic cues present in speech and auditory signals are relevant for phonological and reading development. Then, we will provide data supporting why atypical synchronization of brain signals to these rhythmic cues may lead to developmental dyslexia. Finally, we will sketch some research ideas that could help designing theoretically-based intervention programs to improve brain synchronization to speech rhythm in dyslexic individuals.

9.2 The Importance of Prosody and Rhythm for Phonological and Reading Development

The encoding of linguistic rhythmic cues by infants, such as syllabic stress, has attracted a lot of attention in the field of language acquisition and phonological development (e.g., Nazzi & Ramus 2003), and seems to have an important role to play in dyslexia. There is evidence suggesting that the encoding of stressed units in speech is a fundamental mechanism that contributes to language and word learning (e.g., Curtin 2010). Stressed speech units help infants segment and encode speech by automatically orienting the auditory attentional focus towards important information in a continuous stream of speech segments. Therefore, when lacking lexical knowledge, infants may take advantage of the distributional properties of stressed units in the continuous stream of phonemes to discover word boundaries (e.g., Jusczyk et al. 1999; Thiessen & Saffran 2003).

Stressed syllables last longer, are louder, and are higher in pitch than unstressed syllables. Interestingly, individuals with phonological disorders like dyslexia or specific language impairment exhibit poorer sensitivity to the acoustic correlates of stress (Corriveau, Goswami, & Thomson 2010; Goswami, Mead, Huss, Barnes, Leong 2013; Huss, Verney, Fosker, Mead, & Goswami 2011; McArthur & Bishop 2004; Richards & Goswami 2015; Richardson, Thomson, Scott, & Goswami 2004; Thomson, Fryer, Maltby, & Goswami 2006), which suggests that they may have difficulties to discriminate stressed from unstressed units embedded within speech streams. This has been taken as direct evidence for a stress processing deficit as a possible cause for the phonological and reading difficulties in dyslexia (Goswami, Mead et al. 2013; Jiménez-Fernández, Gutiérrez-Palma, & Defior 2015). In favor of this hypothesis, individuals with dyslexia across languages consistently show poor sensitivity to sound amplitude rise time (e.g., Goswami et al. 2002; Hämäläinen et al. 2009; Richardson et al. 2004; Surányi et al. 2009), which is another important acoustic cue to syllabic and stress information (Goswami & Leong 2013).

Amplitude rise time corresponds to the rate of amplitude change (from low to high amplitude) and codes for the onset of syllables in speech. Amplitude modulations such as those defined by the rise time will contribute to the perception of units as stressed and in turn generate the impression of the rhythmic structure of speech. These changes in amplitude (energy, intensity) will generate the speech envelope. As Fig. 9.1 shows, the speech signal is composed of various amplitude peaks (following amplitude rise times) and valleys which represent the oscillations of phonological units of different sizes according to different time-scales. Changes in amplitude oscillating every one second approximately (1 Hz, delta band) code for stress and prosodic variations which correspond to words and phrase temporal windows (Bourguignon et al. 2013, black lines on Fig. 9.1a). Amplitude modulations occurring every 125–250 ms (4–8 Hz, theta band) correspond to the oscillatory rate of syllables in speech (dotted gray lines on Fig. 9.1a). Therefore, poor sensitivity to stress and rise time is thought to

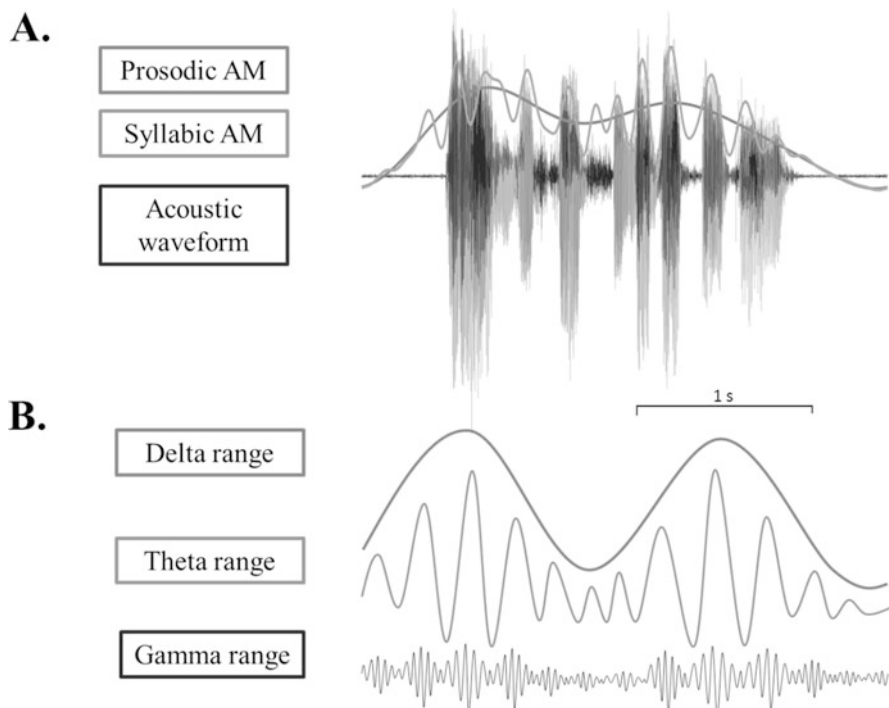


Fig. 9.1 (a) Time (x-axis)-amplitude (y-axis) representation of an acoustic signal (light gray). The envelope of the speech signal low pass filtered in the delta band (<1 Hz; prosodic information) is represented with a black plain line. The envelope band pass filtered in the theta band (4–8 Hz; syllabic information) is represented with a dotted gray line. (b) Possible representation of the hierarchical coupling (with fast oscillations nested in slow oscillations) happening between neural oscillations synchronized to the temporal properties of the speech signal. (Adapted from Lallier et al. 2016)

weaken the perception of sequential amplitude modulations alternating at “slow” frequencies (<8 Hz) in people with dyslexia (Goswami et al. 2002). In particular, Goswami & Leong (2013) propose that encoding the temporal (phase) relation between rise times at these slow time scales may be the core deficit of dyslexia such that lexical stress should provide useful information about word syllabic boundaries (e.g., position of the stressed syllable within words). Such precise temporal encoding of rise time sequences (i.e., speech envelope) will provide an oscillatory phonological framework to parse and acquire new vocabulary (Lallier, Molinaro, Lizarazu, Bourguignon, & Carreiras 2016; Leong, Kalashnikova, Burnham, & Goswami 2014).

Interestingly, not only speech but also nonverbal rhythmic stimulus sequences processing (such as tone and beat sequences) may be affected in individuals with poor reading skills and dyslexia (Carr, White-Schwoch, Tierney, Strait, & Kraus 2014; Muneaux, Ziegler, Truc, Thomson, & Goswami 2004). In particular, the

speed at which auditory attention shifts between sequential auditory stimuli might be one of the parameters that could index how well sequential rhythmic cues are encoded (Lallier & Valdois 2012, see Goswami et al., 2013, for a similar suggestion). Accordingly, we showed that auditory sequential processing whose rate falls within the temporal window corresponding to the theta band (125–250 ms) is sluggish in dyslexic individuals (Lallier et al. 2011; Lallier, Thierry, & Tainturier 2013; Lallier et al. 2009). More specifically, we used an adaptive procedure and measured the shortest tone onset asynchrony (TOA threshold) at which participants could shift their attentional focus in order to perceive two sequential tones as distinct entities. Across our studies, we found that the shortest TOAs (fastest auditory attentional shifting speed) was around 125 ms for the controls (8 Hz) whereas it was significantly longer for the dyslexic participants (around 170 ms, ~6 Hz). This set of data is in line with the hypothesis that auditory attention might act as an oscillator whose sequential shifts are tuned according to the pseudo-regular distribution of stressed and unstressed syllabic amplitude modulations (Quené & Port 2005). Sluggish attentional shifting within slow temporal windows (theta in particular) in dyslexic individuals may contribute to the prosodic and syllabic disorders described above. It is noteworthy that we highlighted a direct relation between sluggish attentional shifting speed in the theta band and phonological disorder at the *phonemic* level in dyslexia (Lallier et al. 2013). How could the failure of the auditory attentional system to shift *between* relevant auditory stimuli (e.g., tones, amplitude rise time, syllabic stress) impair the perception of fine-grain acoustic modulations *within* phonemes?

A big challenge of hypotheses focusing on “slow” temporal rhythmic deficits (~125–1500 ms) is to account for the consistent “fast” phonemic features processing difficulties (e.g., ~30 ms such as voice onset time) reported in dyslexia across languages (Caravolas et al. 2012; Caravolas, Volin, & Hulme 2005). In fact, this led to some debate regarding whether the core phonological deficit in dyslexia hinders the processing of phonological information at the level of the syllables (onset and rime – and consequently, syllabic rise time and prosodic processing) or the phonemes (Goswami et al. 2002; Hulme, Caravolas, Malkova, & Brigstocke 2005; Hulme et al. 2002). Perhaps one question of the utmost importance raised by this same debate relates to the brain bases of dyslexia: how would problems in rhythmic processing subtended by the right hemisphere (e.g., Abrams, Nicol, Zecker, & Kraus 2008; Geiser, Zaehle, Jancke, & Meyer 2008; Riecker, Wildgruber, Dogil, Grodd, & Ackermann 2002) explain the long-standing theory linking atypical left hemisphere with dyslexia and reading difficulties (e.g., Boets et al. 2013; Brem et al. 2010; Pugh et al. 2000; Vandermosten et al. 2012)? Multi-time resolutions neural models of speech perception (Giraud & Poeppel 2012; Poeppel 2014) give hints to address this question.

9.3 Atypical Neural Oscillations as the Brain Signature of Rhythm Processing Deficits and Phonological Disorders in Developmental Dyslexia

According to multi-time resolution models (Giraud & Poeppel 2012), the brain tracks speech temporal modulations at different rates simultaneously, by synchronizing neural oscillations (rhythmic fluctuations in neuronal excitability) to these specific modulation rates (see Fig. 9.1). More specifically, “slow” prosodic and syllabic modulation rates in speech trigger right hemisphere (RH) lateralized neural oscillatory activity in the auditory primary regions in the delta (<4 Hz) and the theta (4–8 Hz) bands respectively, that phase-lock (i.e., temporally align) to these speech modulations. Such phenomenon is called neural entrainment. In addition, these models propose that fast speech temporal modulations linked to phonemes are encoded through high frequency (>20 Hz) neural oscillations, that would be biased towards the left hemisphere (LH) and show a bilateral hemispheric activation.

A first logic prediction from the rise time hypothesis of dyslexia would be to find atypical neural entrainment to slow temporal modulations in right hemispheric regions, but preserved high frequency bilateral neural activity in response to fast auditory changes. However, we now know that speech perception relies on a nested hierarchical coupling mechanism between oscillatory neural activity at different frequencies: the phase of slow (delta-theta) modulations in the brain couples with the amplitude in fast neural oscillations (gamma) (e.g., Gross et al. 2013, see Fig. 9.1b). This hierarchical cross-frequency coupling provides a plausible explanation why atypical auditory phase locking to slow speech amplitude modulations would generate problems at the level of phonemic encoding, and consequently, why inaccurate rise time (or stress) processing would affect the sensitivity to phonemic acoustic features (Goswami, Fosker Huss, Mead, & Szücs 2011).

Most of the studies which have looked at neural oscillations in dyslexia did not assess neural responses in the same dyslexic participants across the whole range of relevant frequencies for speech perception (i.e., delta, theta and gamma, Giraud & Poeppel 2012; Poeppel 2014), nor did they measure cross-frequency coupling. Hämäläinen, Rupp, Soltész, Szücs, and Goswami (2012) reported impaired phase locking to amplitude modulated white noises in the delta band in the RH (2 Hz) in dyslexic adults but did not assess frequencies above 20 Hz that could reflect a possible phonemic oscillatory processing deficit. Lehongre, Ramus, Villiermet, Schwartz, and Giraud (2011) assessed neural entrainment from 10 to 80 Hz in dyslexic French adults, hence ignoring the delta and theta frequency bands. They reported a reduced left hemispheric bias in the neural oscillation in response to 30 Hz, and enhanced response for frequencies beyond 40 Hz, suggesting phonemic processing difficulties. Lastly, Poelmans et al. (2012) reported that dyslexic adults showed a reduced left hemispheric bias in neural coherence to amplitude modulated noise at 20 Hz, but not difference between skilled and dyslexic readers at 4 Hz.

Still, this study did not quantify the neural response of dyslexic participants in the delta band. Overall, little consistent support was found for the rise time/amplitude envelope hypothesis of dyslexia across these studies.

In order to shed light on these inconsistencies, we measured neural entrainment in the delta (2 Hz), theta (4 and 7 Hz), and gamma (low gamma, 30 Hz, high gamma, 60 Hz) bands in the same Spanish speaking participants with and without dyslexia using magnetoencephalography (MEG) (Lizarazu et al. 2015). Moreover, we assessed both children and adults to explore how age-related reading expertise modulates prosodic-, syllabic-, and phonemic-rate oscillatory dysfunctions in dyslexia and their associated brain lateralization patterns. Indeed, phonological sensitivity changes with the amount of experience with print (see Castles & Coltheart 2004, for a review), such that pre-readers or readers with little reading experience will be especially sensitive to large phonological units like syllables (onset and rime), whereas readers with a sufficient amount of exposure to print will start showing sensitivity to phonemes (Anthony & Francis 2005; Ziegler & Goswami 2005).

Our results showed that dyslexic participants exhibited reduced RH synchronization to syllabic-rate auditory amplitude modulation (4 Hz) as well as a reduced bilateral response for stimuli presented at 30 Hz, reflected by a stronger right lateralized synchronization to phonemic-rate stimuli in the dyslexic groups. Laterality indexes at 4 Hz correlated with reading (see Abrams et al. 2008, for similar results), indicating that a stronger RH response to syllabic-rate stimuli was associated with faster word and pseudoword reading times. Interestingly, no difference was highlighted between the deficits of the dyslexic children and the dyslexic adults, suggesting that atypical auditory oscillatory activity in dyslexia is not affected by print exposure. More importantly, dyslexic adults showed these two types of abnormal neural oscillatory specialization (i.e., at 4 and 30 Hz) compared to much younger skilled reader children. Again, this suggests that it is unlikely that these auditory processing deficits are mainly the consequence of the lack of print exposure in dyslexic individuals. Still, we found an increased response to auditory amplitude modulations presented at 30 and 60 Hz in both hemispheres in the whole group of adults compared to the whole group of children. Moreover, a stronger left-hemisphere lateralization for these high frequencies was found in adults compared to children within the whole groups of participants, which was furthermore related to the phonological short term memory scores of the participants. Although, these two last results speak in favor of the impact of print exposure on phonemic sensitivity (Castles & Coltheart 2004), our study also supports the idea that reading expertise does *not* affect the strength of phonemic-rate oscillatory disorders in dyslexia.

A last novel feature of our MEG study was the exploration of a possible brain structural underpinning of atypical auditory oscillatory activity in dyslexia. We measured the cortical thickness of participants in the primary auditory regions in their two hemispheres. First, adults had overall thinner auditory cortex in both hemispheres than children overall, suggesting a cortical pruning due to increased experience with auditory (or speech) stimuli. Interestingly, a left hemispheric asymmetry in cortical thickness (thinner cortex in the LH than the RH) was

related to a stronger left hemispheric lateralization of neural synchronization to stimuli presented at the phonemic rate in skilled readers. In contrast, the same anatomical index was related to a stronger right hemispheric dominance for neural synchronization to syllabic-rate auditory stimuli in dyslexic readers. These different relations between LH structural and functional specialization between dyslexic and skilled readers might reflect different phonological grain size sensitivity: the auditory cortex in the language-dominant hemisphere (LH) would be recruited more than that in the RH for processing syllabic units in dyslexic readers, whereas that asymmetry occurs for processing the phoneme units in skilled readers. Overall, the data of this study suggest that the acoustic sampling deficit in dyslexia might be linked to an atypical hemispheric specialization of the auditory cortex to both low (theta) and high (low gamma) frequency amplitude modulations.

The aforementioned studies fail to provide evidence for a possible causal link between atypical entrainment of the RH to low frequency speech modulations and LH dysfunction in dyslexia at phonemic-rate frequencies. Moreover, a potential limitation of most of these studies is the use of non-verbal auditory stimuli (Hämäläinen et al. 2012; Lehongre et al. 2011; Lizarazu et al. 2015) when we know that phonological difficulties stem from the atypical neural processing of speech streams.

In the following study (Molinaro, Lizarazu, Lallier, Bourguignon, & Carreiras 2016) we recorded oscillatory neural activity during ecologically valid continuous speech perception in 20 skilled readers and 20 age-matched dyslexic readers. Both groups included 10 Spanish monolingual adults (22–39 years old) and 10 Spanish monolingual children (9–13 years old). We found that the brain significantly synchronized to speech modulations occurring at 0.5–1 Hz (delta range) and at 5.8–6.3 Hz (theta range) in skilled readers. In the delta band, the analyzes revealed that the brain significant synchronization to speech occurred within a bilateral brain network with a rightward asymmetry including the right and the left auditory cortex, the right superior and middle temporal regions, the left superior temporal gyrus and the left inferior frontal regions. In the theta band, source reconstruction for the same group revealed brain-to-speech synchronization in right primary auditory areas, peaking in superior temporal regions. Group comparisons showed that dyslexic readers exhibited reduced brain-to-speech synchronization within the delta band (0.5–1 Hz) in both the right auditory cortex and the left inferior frontal gyrus. No group difference was found in the theta band (5.8–6.3 Hz).

Interestingly, this result contrasts with Lizarazu et al. (2015) where no difference between the dyslexic and the control groups was found in the delta band (note that most of the participants of the two studies were the same). However, when comparing the brain synchronization to both nonverbal and speech auditory oscillations in the delta band (although the specific delta frequencies tested were different: amplitude modulated noise at 2 Hz in Lizarazu et al. (2015) and 0.5–1 Hz amplitude modulations within Spanish sentences in Molinaro et al. (2016), respectively), a clear difference in the brain sources of the synchronization emerged: an engagement of LH activity appeared in addition to neural response in the RH in the speech condition only. This might suggest that brain synchronization to

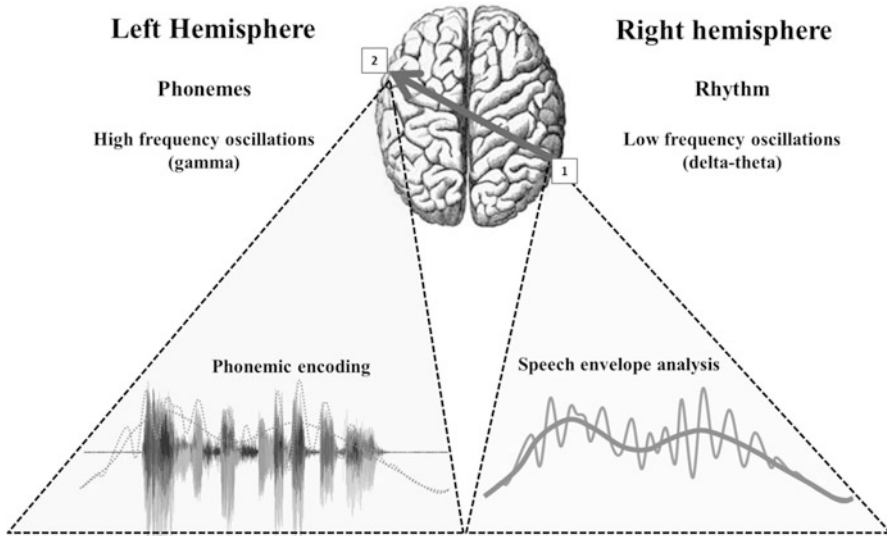


Fig. 9.2 Illustration of a possible mechanism for inter-hemispheric directional connectivity during speech analysis, contributing to reading development and dyslexia. The right auditory cortex ([1]) would be in charge of the analysis of low frequency speech modulations (delta, black line; theta, dark gray lines) coding for speech rhythm. Through parsing the acoustic wave (light gray) in relevant units (dotted lines on the left-hand side), the low frequency brain synchronization to speech arising from the right hemisphere would support phonemic encoding reflected in high frequency oscillations (gamma) in a left-sided network including the left inferior frontal gyrus ([2]). Speech amplitude modulations in the delta band might be driving the left hemisphere analysis of the speech signal, and be especially important for reading development (see Molinaro et al. 2016)

amplitude modulations in speech in the delta band may have a role to play in the LH processing of phonological stimuli.¹

Importantly, Molinaro et al. (2016) took advantage of the high temporal resolution of MEG signals to determine the direction of propagation of neural activity synchronized to speech modulations in the delta band (i.e., causal connectivity between the different regions involved in the “delta band network” described above). We found that fluctuations in the delta band in the right auditory cortex drove those in the left inferior frontal gyrus (see Fig. 9.2). In other words, RH synchronization to speech in the delta band triggered similar neural synchronization to the speech signal in the LH frontal regions, in areas known to support phonological processing and dyslexia (e.g., Peyrin et al. 2012; Temple et al. 2003). Importantly, connectivity from the right auditory cortex to the left inferior frontal gyrus was weaker in the dyslexic group than in the skilled reader group (both in children and adults). Moreover,

¹Neural entrainment at 2 Hz might not be critical for processing speech, as evidence in the similarity between neural entrainment at 2 Hz during speech processing and resting activity at 2 Hz (see Fig. 1 in Molinaro et al. 2016)

this connectivity measure correlated with the phonological skills of participants (phonemic awareness) and synchronization to the delta properties of speech in the left inferior frontal gyrus correlated with reading skills.

Overall, Molinaro et al.'s data suggest that slow delta modulations (0.5–1 Hz) driven by the prosodic contour of speech (prosodic phrase, intonation) is critical for phonological and reading development. Again, here, the hypothesis of a hierarchical cross-frequency coupling provides an explanation why atypical synchronization to delta speech oscillations would generate problems at higher syllabic-rate frequencies in dyslexia, as reported in a number of aforementioned studies.

This suggests that atypical synchronization of the brain to the low frequency modulations of speech in the RH may be the cause of atypical phonological processing leading to reading difficulties expressed mainly through a LH dysfunction. The analysis of low speech frequencies would provide the temporal framework to parse the speech signal into relevant units in order to extract phonemic information (see Fig. 9.2, and Lallier et al. 2016). Furthermore, these findings support the hypothesis linking the encoding of prosodic properties of speech (lexical stress and rise time) to phonemic processing and reading development (Goswami, Fosker, Huss, Mead, & Szücs 2011; Goswami, Power, Lallier, & Facoetti 2014).

9.4 How to Improve Auditory Neural Entrainment in Dyslexia?

Overall, the data available supports the claim that dyslexia is associated with difficulties at entraining to speech rhythm (i.e., the prosodic and syllabic amplitude envelope, <8 Hz) that would prevent them from building a temporal framework guiding speech perception. Then, how could synchronization to speech rhythm be enhanced in dyslexia? Speech rhythm is aperiodic, and although it contains some sort of regularities between stressed and unstressed syllables, these are not metrically regular. Therefore, it is reasonable to think that dyslexic individuals may benefit from more temporal regularities in auditory signals in order to boost the encoding of the phase relation between aperiodic low frequency speech modulations (see Goswami & Leong 2013) and, in turn, improve neural entrainment to these modulations.

In one of our studies we tested whether priming speech sentences with their amplitude envelope low-pass filtered at 8 Hz (i.e., with no phonemic information making the signal sound like an amplitude modulated white noise) would enhance the perception of this sentence (Rfös-López, Molnar, Lizarazu, & Lallier 2017). The rationale behind this manipulation was to present the perceptual and attentional auditory systems with the temporal structure of speech before listening to this actual speech. Because of the repetition of speech rhythm, we assumed that it would be easier to extract the low frequency temporal framework upon which higher linguistic processes involved in speech perception will rely (as suggested

in Molinaro et al. 2016, see Fig. 9.2). Accordingly, we found that children were more accurate at reporting a non familiar pseudoword embedded in sentences presented in quiet or multi-talker babble noise, when the sentence was preceded by its amplitude envelope (<8 Hz) compared to when it was preceded by an unmodulated white noise. Furthermore, the speech envelope repetition/prime benefit (pseudoword recall accuracy of the primed versus non primed sentence) correlated with the reading skills of the children, reflecting that the poorest readers exhibited the highest benefit from the speech envelope prime. Our study is in line with research showing that speech repetition may trigger some changes in our brain that make us perceive speech differently, possibly focusing on more detailed acoustic information. For example, it has been shown in the “speech-to-song illusion” (Deutsch, Lapidis, & Henthorn 2008; Tierney, Dick, Deutsch, & Sereno 2013) whereby the exact same phonemic sequences can be heard as speech or music (i.e., words being sung instead of said). This perceptual shift from speech to music only depends on whether the sound sequence is repeated. This illusion suggests that repetition of speech helps cognitive and neural resources to focus on finer grain acoustic information in the repeated speech segments (such as relative pitch differences between phonemes). It is reasonable to assume that the repetition of the amplitude envelope of speech may have facilitated phonemic processing in Ríos-López et al. (2017)’s study. As speech rhythm is pseudo-regular, repeating the rhythmic structure of a sentence might have made temporal cues in speech more regular and accessible, and boosted the perceptual and attentional synchronization to low frequencies.

Interestingly, repetition is a fundamental component of music. Hence, musical rhythmic patterns are very regular, which allows the perceptual and attentional auditory systems to predict quickly and accurately where the next beat is going to occur. Therefore, it is not surprising that dyslexia research is now opening new avenues addressing the potential beneficial effect of music on phonological and reading development. Interestingly, the processing of musical rhythm in the brain may be rooted in the RH hemisphere as it is for speech. In fact, normal development of musical skills seems to require an efficient connectivity between the *right* auditory cortex and the *right* inferior frontal gyrus (Albouy et al. 2013; Peretz 2013; Peretz, Vuvan, Lagrois, & Armony 2015). This strikingly echoes the results of Molinaro et al. (2016), reporting that brain synchronization to speech rhythm (delta band) elicited in the *right* auditory cortex and propagating to the *left* inferior frontal gyrus was strongly associated with phonological and reading skills.

The significant link between speech rhythm, music rhythm and reading is also reflected in data showing that sensitivity to rise time is associated with sensitivity to musical rhythmic parameters, which furthermore predicts phonological awareness and reading development (Huss et al. 2011). Moreover, synchronizing to a regular beat before learning to read predicts phonological and reading skills (Carr et al. 2014).

9.5 Conclusion

Overall, the studies presented in this book chapter contribute to building a possible theoretical framework that would causally associate a RH-related rhythm processing deficit to the LH-rooted reading disorder that is dyslexia. Figure 9.3 illustrates how RH low synchronization to slow frequency auditory modulations can cause LH-based grapheme-to-phoneme conversion deficits via impaired rhythmic and prosodic processing difficulties. Importantly, this framework makes it possible to investigate new ways to remediate reading difficulties, through music. Playing and listening to music is a universally fun and child-friendly activity often associated with positive emotional and motivational outcomes. If we can confirm that music training programs positively impact the development of reading and reading related skills (Chobert, Francois, Velay, & Besson 2014; Thomson, Leong, & Goswami 2013), music should become a significant part of educational and health practice, and improve durably the life of millions dyslexic children across the world.

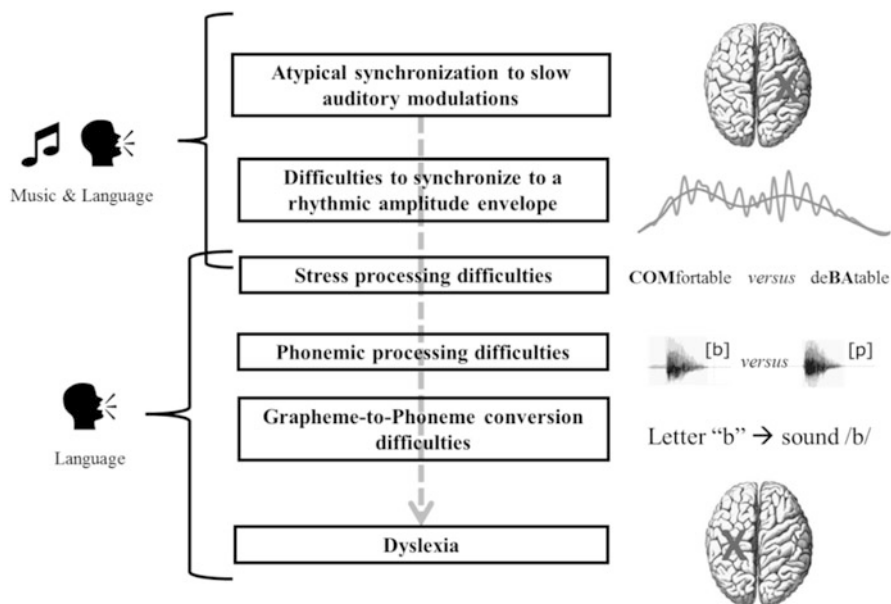


Fig. 9.3 Possible causal framework linking right hemisphere abnormalities to left hemisphere dysfunction in dyslexia. The two first processing stages in the causal chain would overlap between language and music processing. This suggests that music training could lower the strength of the original deficits leading to atypical phonological and reading acquisition in dyslexia

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Chapter 10

Dyslexia: A Failure in Attaining Expert-Level Reading Due to Poor Formation of Auditory Predictions



Sagi Jaffe-Dax, Luba Daikhin, and Merav Ahissar

Abstract In this chapter, we present evidence that alters the way Dyslexia is typically viewed and assessed. Based on accumulating findings obtained from behavioral assessments, computational modeling, imaging and ERP studies, we propose that Dyslexia results from a failure in acquiring a specific (reading and linguistic) skill that relies heavily on familiarity with stimuli distributions characterized by temporal regularities in a specific time window. Dyslexia is naturally associated with language related impairments, since learning temporal regularities is crucial for acquiring linguistic skills, but not confined to them. Studying Dyslexics' basic auditory processing from this perspective reveals specific and robust deficits in benefiting from simple temporal consistencies, which are associated with a reduced ability to accumulate stimuli statistics across time windows of $>2-3$ s. Importantly, similar impairments are demonstrated in the visual modality, supporting the cross-modal nature of the core deficit. Collectively, our findings show that Dyslexics fail to achieve expert level performance in variety of tasks, including reading, due to deficient accumulation of summary statistics, which impedes the formation of reliable predictions, which in turn facilitate switching performance to rely on efficient processing strategies.

Keywords Anchoring deficit hypothesis of dyslexia · Working memory · Perception · fMRI · EEG · Statistical learning

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10.1 A General Theory of Skill Acquisition and Cognitive Disabilities

We propose a general, principled theory of skill acquisition, which integrates a theory of learning (Reverse Hierarchy Theory, RHT – Ahissar & Hochstein, 1997; 2004; Rokem & Ahissar, 2009), with the 2-system theory of modes of cognition (Kahneman 2011). RHT proposes a separation between bottom-up (local to global) processing, and top-down (global to local) perception, and perceptual learning. It asserts that as a default our attention system and, in accordance, our perception are based on high-level, object- and scene-oriented representations, which have an ecological reality. This is useful for every day purposes, but when learning requires fine discriminations (e.g., small bars, similar letters), a backward search for allocating informative neural populations, that input the higher level, is activated. When such populations are allocated, they can gradually replace higher-level populations as part of the overall scheme of performance, with better resolution. This process leads to “pushing down” initially high-level roles, with practice, but only when this practice is successful in detecting lower-level populations, i.e. detect and integrate regularities. The dual system approach to cognition proposes two modes of cognition: fast, automatic and effortless, but prone to “perceptual biases” (system 1) versus slow, serial and effortful (system 2). We propose that these are two extremes along a hierarchy of modes of performance that characterize the gradual skill-acquisition process from the novice (system 2) to the expert (system 1). System 2 is embodied by the working-memory fronto-parietal system, which is consistently activated in novel and challenging situations (Duncan & Owen 2000), and has been termed the “multiple-demand system” (Duncan 2010). This system was associated with general intelligence (e.g. the amount of space taken by these areas is correlated with reasoning skills; Woolgar et al. 2010).

The novice has to “set the stage” to perform a task, relying mainly on the domain-general high-level fronto-parietal system, which maps new tasks to neural implementations. Successful practice proceeds along a hierarchy of processing in a reverse direction, where gradually lower-level areas encode sub-parts of the trained tasks. Crucially, the gradual reliance on lower levels depends on allocating neuronal populations that reliably encode task-related regularities, which is the neural correlate of regularity detection. Thus, the expert does not activate the same algorithms faster, but replaces them with low-level encoded schemes.

Practice does not always make better. One has to detect the repeated regularities. We propose that specific developmental disabilities result from a failure to automatically detect and use sensory or sensory-motor regularities under conditions that allow such detection for the general population. What characterizes developmental disabilities is adequate general reasoning skills, allowing understanding of novel tasks, but reduced ability to attain fast and effortless performance based on retrieved schemes, in spite of intensive practice. We propose that this conceptual account applies to a variety of developmental disabilities, though here we shall focus on the case of reading disability. This focus allows us to study the computational and

neural mechanisms that yield these complex phenomena in an unprecedented depth, leading to conceptual shift in our understanding of disabilities as part of a principled theory of learning and inference.

Dyslexia is a pervasive difficulty in attaining expert-level reading, in spite of adequate reasoning skills and adequate practice and guidance (education) opportunities (World Health Organization 2008). Within our proposed framework we view dyslexia as follows: Dyslexics' 'system 2' is adequate. However, they have difficulties in delegating reading sub-tasks to gradually lower-level areas, due to inefficiency in detecting sound regularities. These difficulties impact the processing of both simple and complex sounds, and impede the acquisition of expert level reading due to inefficiency in representing linguistic regularities. In this chapter, we describe our series of observations and modeling supporting this hypothesis.

10.2 The Anchoring Hypothesis of Dyslexia

In the early 2000s, two open questions were heatedly debated in the field of dyslexia – (1) Are dyslexics' deficits specific to speech sounds? (2) Is the deficit representational or does it affect only access to otherwise adequate representations? In an early study we performed, we found that a large fraction of adult poor readers also perform poorly in a broad set of simple auditory discrimination, and that the degree of their deficit is correlated with their reading scores (Ahissar, Protopapas, Reid, & Merzenich 2000; Amitay, Ahissar, & Nelken 2002), suggesting that dyslexics' deficits are not specific to speech sounds.

Trying to better decipher the bottleneck underlying dyslexics' poor performance in simple discrimination tasks, we administered two protocols of 2-tone frequency discrimination asking which tone is higher (Ahissar, Lubin, Putter-Katz, & Banai 2006). In one protocol there was a fixed reference frequency in each trial whereas in the other, there was no such consistency. Listeners' discrimination thresholds were substantially lower when there was a reference, even though they were unaware of its presence. Our dyslexic participants did not show this benefit (Fig. 10.1). In a subsequent study, we administered the same two protocols to dyslexics and ADHD participants (Oganian & Ahissar 2012). We replicated the effect of impaired benefit from stimulus repetition among dyslexics, whereas individuals with ADHD who were good readers did not show this effect. All their thresholds were similar to controls'.

To test for a similar deficit underlying speech perception, we measured speech perception in noise under two related conditions: with a large set of words, and with a small set, hence many word repetitions. Dyslexics' deficits were only found in the small set, with repetitions. Importantly, their pattern of errors showed similar sensory sensitivity as controls (e.g., “/tarul/” instead of “/barul/”), but in contrast to controls, it was not restricted to the trained set of words, suggesting poor usage of word repetitions. We conclude that dyslexics' deficit in implicit learning of distributional statistics also results in the formation of impoverished categorical rep-

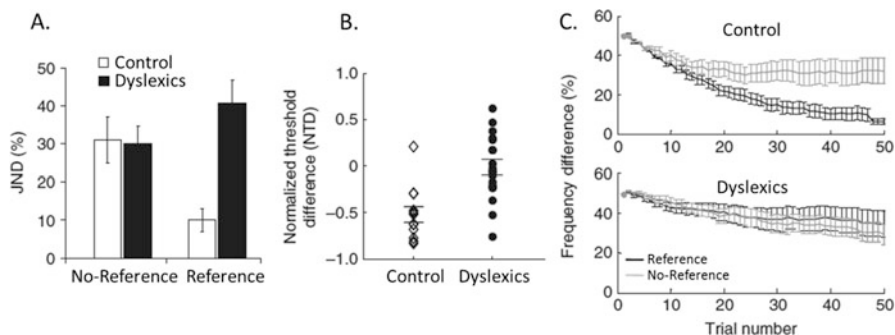


Fig. 10.1 Frequency discrimination thresholds with and without stimulus repetition across trials among dyslexics and control individuals. The effects of discrimination condition (No-reference versus Reference) differed significantly between the groups. **(a)** Average thresholds show that dyslexics had significantly higher JNDs in the Reference condition. **(b)** Single-subject data of the normalized difference in threshold between the Reference and the No-reference conditions. Filled circles: dyslexics; open diamonds: control. **(c)** Adaptive assessment protocol for control individuals (top) and dyslexics (bottom) in the two procedures shows a gradual effect of using a consistent reference for control individuals but not for dyslexics. Error bars denote SEM. (Adapted from Ahissar et al. 2006, with permission)

representations. One manifestation of this deficit is poorer performance in demanding speech discrimination tasks that rely on rich categorical representations (Banai & Ahissar 2017).

10.3 Evidence for Attaining Expert-Level Characteristics When Regularities Can Be Easily Detected

Even though context is known to crucially affect perception and learning, its specific impact had not been studied. We therefore conducted two studies to better understand the process of regularity detection, and its impact on the implicit change of underlying strategy.

The first study (Nahum, Daikhin, Lubin, Cohen, & Ahissar 2010) concerned two main questions: (1) What kind of regularities can be utilized quickly, within a single training session? (2) Is the network underlying task performance modified when regularities are detected and used to improve performance? We designed four protocols of frequency discrimination with simple regularities – Reference 1st, in which the first tone in each trial had a fixed, reference frequency (1000 Hz) and the second was higher or lower; Reference 2nd, where the second tone was fixed; Implicit Reference, where a fixed reference tone was presented five times at the beginning of the session and after that, a single tone, which could be higher or lower, was presented on every trial (evenly distributed around this reference); and Reference interleaved, where odd trials were of type Reference 1st and even

trials were of type Reference 2nd. We also applied a No-reference protocol, in which no regularities were included. Each protocol was measured in a different group of control participants, to avoid effects of more than one session training. Figure 10.2a shows the dynamics of behavior and the thresholds obtained under each of the protocols. For the Reference-interleaved protocol, the thresholds of Reference 1st trials (odd) and Reference 2nd trials (even) were obtained and presented separately. The benefit of the repeated reference tone at a fixed temporal position was the greatest. Indeed, the protocol that yielded lowest thresholds was Reference 1st, showing a steeper slope, indicating faster learning of the repeated structure. Reference 2nd protocol yielded low thresholds when presented with no competitive options, but was more vulnerable to interference, as indicated by its poor performance under the interleaved protocol. These results indicate that utilization of the repeated reference is fast (evident already by the 10th trial) and highly beneficial. Yet, there are more and less preferred conditions, which produce different degrees of benefit (i.e., higher sensitivity to regularity at the onset of a trial, i.e., event).

The similarity between the thresholds obtained under the Implicit Reference protocol and other protocols with a reference presented explicitly, questions whether the on-line comparison of the externally presented stimuli is, in fact, the strategy that is used for solving the task, since under this protocol only one tone (non-reference) is presented in each trial. To test this question we measured the performance under the Reference 1st and Reference 2nd protocols while recording Event Related Potentials (ERPs). Specifically we asked whether the ERP component, which denotes perceptual characterization, is produced at a different timing in these two types of trials. We reasoned that if listeners detect the regularity of the structure, they can compare the non-reference (target) tone to the internal reference. But for that they should detect which is the informative, non-reference tone, i.e. the structure of the protocol. If this is indeed the case, we can detect it by tracking a change in the temporal position of the P3 component. This ERP component is produced when a task-related categorization is made. We thus asked whether a clear P3 would be formed after the first tone in the trial in Reference 2nd protocol. As shown in Fig. 10.2b, this is indeed what we found! Under Reference 1st protocol, as expected, the P3 component was elicited after the second tone, since categorization could not have been made before this tone was presented. However, under the Reference 2nd protocol, where the first tone was the informative tone and the second was the repeated reference, P3 was formed ~ 300 ms after the first tone, even before the 2nd tone was presented, although participants' introspective was of on-line comparison, and their button press followed the 2nd tone. These results further show that when successfully detected, cross-trial regularity leads to a strategic shift in the operations underlying performance.

What is the role of cross-trial regularities in long-term learning? How do the specifically trained information structures (Reference 1st: repeated \rightarrow new; Reference 2nd: new \rightarrow repeated) affect long term learning? Does learning generalize with practice or, is it specific to the trained protocol? To test this we trained two groups of participants (Cohen, Daikhin, & Ahissar 2013). They were trained with each

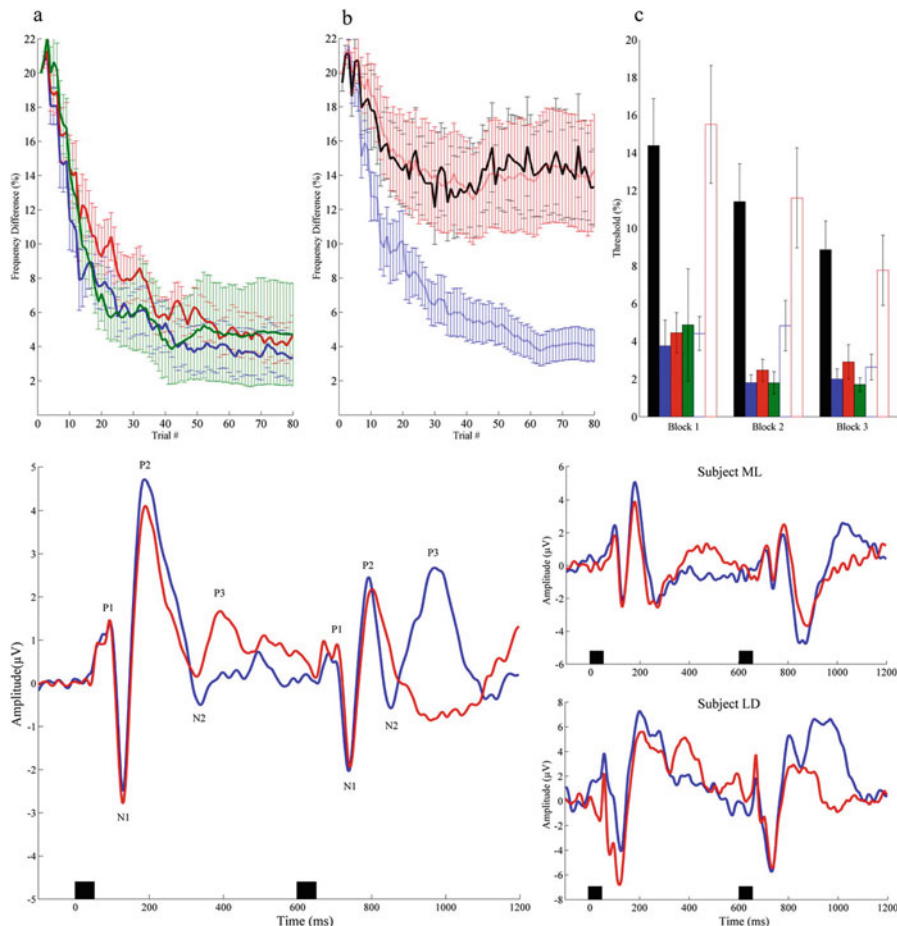


Fig. 10.2 (a) Behavioral performance under four different adaptive protocols: Left – frequency difference between tones as a function of trial number in the first assessment block under three protocols: reference 1st (blue), reference 2nd (red), and implicit reference (green). Middle – frequency difference along the first assessment for two additional protocols: No-reference (black, no repeated reference) and reference interleaved (reference 1st on odd trials (blue) and reference 2nd on even trials (red)) measured with the same adaptive paradigm. Right – thresholds (frequency difference) in three consecutive blocks performed with each of these five protocols. Although all thresholds showed improvement, their ranks were retained across assessments. Cross-subject averages and SEMs are shown. (b) ERP measured while participants performed the two-tone frequency discrimination task under reference 1st protocol (blue) and under reference 2nd protocol (red). The temporal location of the tones in a trial is marked by the black rectangles at the bottom of the plot. The relevant components are marked on the averaged waveforms. A clear P3 component can be seen after the second tone in the reference 1st protocol and after the first (non-reference) tone in the reference 2nd protocol. (Adapted from Nahum et al. 2010, with permission)

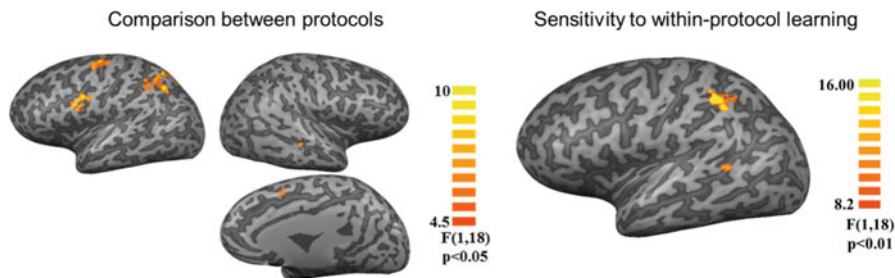


Fig. 10.3 Voxel-wise repeated-measures ANOVA (of beta values). Left – main effect of protocol; Right – main effect of within-protocol block (learning). (Adapted from Daikhin & Ahissar 2015, with permission)

of the two reference containing protocols – Reference 1st and Reference 2nd. We found that cross-session learning was largely specific to the structure of information in the trained protocol. Namely, learning did not transfer between protocols that shared stimuli, task, timing, but did not share the temporal structure of within trial regularity.

Nahum et al. (2010) study found that cross-trial regularities resulted in modification of the strategy used for solving the task. We hypothesized that detecting regularities would be associated with an increased role of auditory areas compared to the multiple-demand circuitry (i.e., task successfully delegated backward). To test this hypothesis, we conducted an fMRI study (Daikhin & Ahissar 2015), and compared the pattern of brain activation under Reference 1st (with regularity) and No-reference (no regularity) protocols, presented in an interleaved manner, so that participants were unaware of the protocol switch (every 36 trials). Figure 10.3 (left) shows the contrast between the patterns of activation under the two protocols. The cortical areas involved are associated with explicit working memory system and are part of the multiple-demand network (Duncan 2010). In each of these areas, the activation was higher under the No-reference protocol, in line with the hypothesis that utilizing the regularities leads to reduced load on the working memory system. Figure 10.3 (right) shows brain regions which were sensitive to within-protocol improvement across trials. Behaviorally, quick cross-trial improvement was found only for Reference 1st protocol. BOLD response changes were also found only under this protocol. Learning contrast was significant in two regions in the left hemisphere – an intra-parietal region, associated with controlling retention of information (Baldo & Dronkers 2006; Koelsch et al. 2009; Magen, Emmanouil, McMains, Kastner, & Treisman 2009; Sreenivasan, Curtis, & D’Esposito 2014), and a posterior superior temporal region, associated with regularity detection in simple and complex auditory stimuli (Binder et al. 2000; Davis & Johnsrude 2003; Friederici, Makuuchi, & Bahlmann 2009; Obleser & Kotz 2010). We interpret this modification as reflecting partial delegation of task performance to more posterior networks, which store effective sound regularities.

10.4 Integration of Previous Trials' Statistics in the More General Case: Contraction Bias

In the No-reference protocol listeners do not have specific item repetitions that can be used as anchors. However, even here listeners implicitly compute priors – representations of history-based knowledge, which substantially affect their performance. A related and amply documented phenomenon is “contraction bias”, which is described as follows: when the magnitude of the two stimuli is small or low with respect to the mean of the previous stimuli in the experiment, participants tend to respond that the 2nd stimulus is smaller or lower, whereas when the magnitude of both stimuli is large or high they tend to respond that the 2nd stimulus is larger or higher (Hollingworth 1910; Preuschhof, Schubert, Villringer, & Heekeren 2010; Woodrow 1933). We have shown that this “contraction bias”, can be understood within the Bayesian framework. Namely, participants form an integrated representation of the recently presented stimulus with the mean (prior) of previous trials. This integration is particularly helpful when responses are noisy and priors provide reliable predictions. Thus, one would expect that for noisier responses the weight of the priors in the integrated representation would be larger (Ashourian & Loewenstein 2011). The level of noise in the representation of the 1st stimulus is larger than the level of noise in the representation of the 2nd stimulus because of the additional noise associated with the encoding, and maintenance of the 1st stimulus in memory during the inter-stimulus interval of sequential presentation tasks (Bull & Cuddy 1972; Wickelgren 1969). Therefore, the integrated representation of the 1st tone in the trial is expected to be more biased (contracted) towards the calculated mean. Consequently, participants' responses are biased towards overestimating the 1st stimulus when it is small and underestimating it when it is large with respect to the prior.

The expected effect of the contraction bias on performance depends on the relative position of the first and second stimulus with respect to the distribution of stimuli in the experiment. Thus, there will be stimulus pairs that will gain from this bias (*Bias+*) and those that will lose (*Bias-*). *Bias+* trials are trials in which contraction of the first tone towards the mean frequency increases the difference between the representations of the two tones in the trial (Fig. 10.4, yellow zones). *Bias-* trials are those where such contraction decreases the perceived difference between the two stimuli, and hence hampers performance (gray zones in Fig. 10.4). In these trials, contracting the first tone towards the mean frequency decreases its perceived difference from the second tone and performance is thus expected to be impaired by this contraction. *Bias0* trials (white zones in Fig. 10.4) are trials in which the first and the second tones flank the mean frequency.

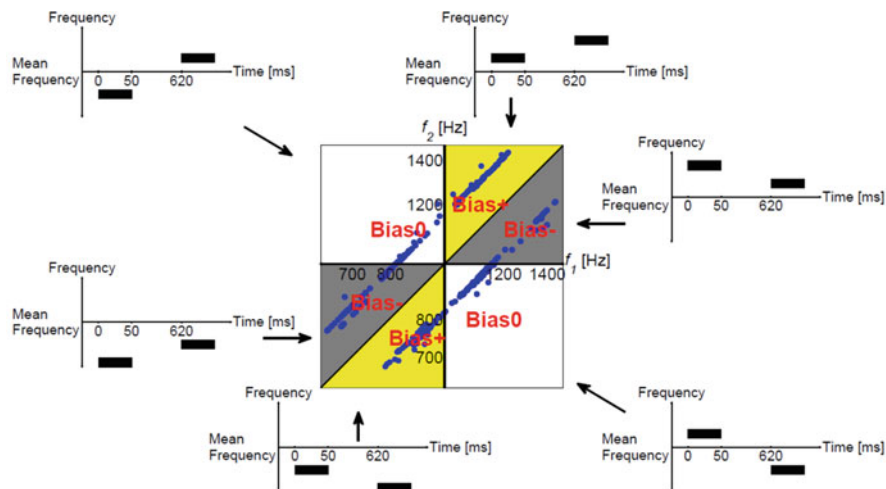


Fig. 10.4 Distribution of trials in the 2-tone discrimination task, and its impact on the perceived inter-pair frequency difference. Middle panel: Trial distributions presented by the frequency of the first and second tones [f_1 , f_2] in each trial. Each dot denotes the f_1 and f_2 of a trial. The diagonal denotes $f_1 = f_2$. Equal distance lines from the diagonal denote trials with different frequencies but fixed within trial frequency difference, as plotted here. Surrounding schematic plots illustrate contraction bias. In *Bias+* trials the first tone is closer to the mean. Hence its contraction to the mean increases the perceived frequency difference between the two tones. In *Bias-* trials the first tone is farther from the mean, and contraction of the first tone decreases the perceived difference. In *Bias0* trials the two tones flank the mean

10.5 The Magnitude of Contraction Bias Is Smaller in Dyslexics Than in Controls

Raviv, Ahissar, and Loewenstein (2012) measured the magnitude of the contraction bias (the difference in success rate between *Bias+* and *Bias-* trials, as illustrated in Fig. 10.4) in the general population under the No-reference protocol and found a substantial effect. Jaffe-Dax, Raviv, Jacoby, Loewenstein, and Ahissar (2015) used a roughly fixed frequency difference (blue dots in Fig. 10.4), chosen as the difference that yields $\sim 80\%$ correct performance in good readers (as measured by Nahum et al. 2010). Though the difficulty of each trial was the same in terms of intra-trial frequency difference, success rate varied substantially across trials, in a manner that could be largely explained by the contraction bias (Fig. 10.5a).

We examined whether dyslexics' context effects were reduced by measuring the magnitude of their contraction bias compared with controls (Jaffe-Dax et al. 2015). Overall, dyslexics performed worse than controls. However, they showed a smaller context effect; i.e., a smaller difference in performance between *Bias+* and *Bias-* trials (Fig. 10.5b). Namely, in spite of their overall noisier representations, they under-weighted previous trials' statistics. Importantly, in some behavioral situations, where priors impair performance, dyslexics' performance gains from this

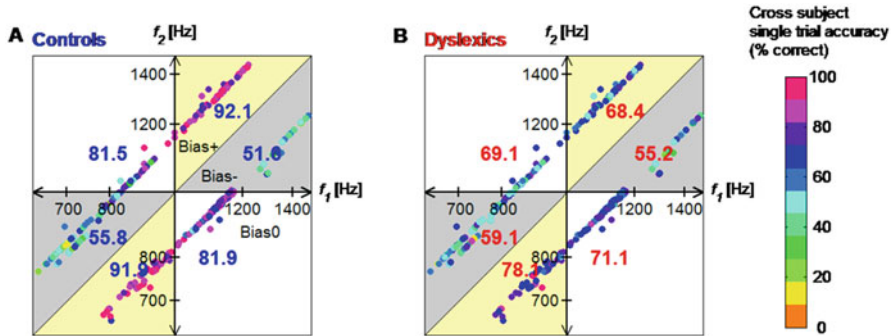


Fig. 10.5 Contraction bias (difference in performance between *Bias+* and *Bias-* zones) is larger in controls (left, average % correct in each zone in blue) than in dyslexics (right, average % correct in each zone in red). The color of each dot denotes the cross-subject average performance for that pair of stimuli. All participants were tested with the same stimuli set. Note the large color difference between dots in the *Bias+* and *Bias-* zones in the left plot (controls) versus the small difference in the right plot (dyslexics). Notations are the same as in Fig. 10.4. (Adapted from Jaffe-Dax et al. 2015, with permission)

implicit under-weighting. Thus, in the *Bias-* regions controls performed at chance level whereas dyslexics' performance was significantly above chance. Importantly, when time intervals between trials were manipulated, allowing the assessment of the dynamics of both behavioral and neural (adaptation) consequences, we found that implicit memory decays faster in dyslexics (Jaffe-Dax, Frenkel, & Ahissar 2017). Thus, dyslexics' retention is impaired with time intervals larger than ~ 5 s.

10.6 Dyslexics' Implicit Memory Trace Is Less Sensitive to Stimulus' Statistics

We hypothesized that if dyslexics' reduced weighting of previous trials stems from an impaired formation of an integrated representation, reduced sensitivity to stimulus statistics may be apparent even before the second tone is presented. To test this hypothesis, we measured event-related potentials (ERPs). We focused on the dynamics and magnitude of the P2 component, which is an automatic response evoked by the auditory cortex (Mayhew, Dirckx, Niazy, Iannetti, & Wise 2010; Sheehan, McArthur, & Bishop 2005). Previous studies, utilizing both oddball (measuring mismatch negativity (MMN); Baldeweg 2007; Haenschel, Vernon, Dwivedi, Gruzelier, & Baldeweg 2005; Tong, Melara, & Rao 2009) and discrimination paradigms (Ross & Tremblay 2009; Tremblay, Inoue, McClannahan, & Ross 2010; Tremblay, Kraus, McGee, Ponton, & Otis 2001) have shown that the magnitude of this component increases with stimulus repetitions, suggesting that this component is sensitive to the statistics of the experiment. We hypothesized that P2's sensitivity to stimulus repetitions is a special case of its sensitivity to the (frequency) distance

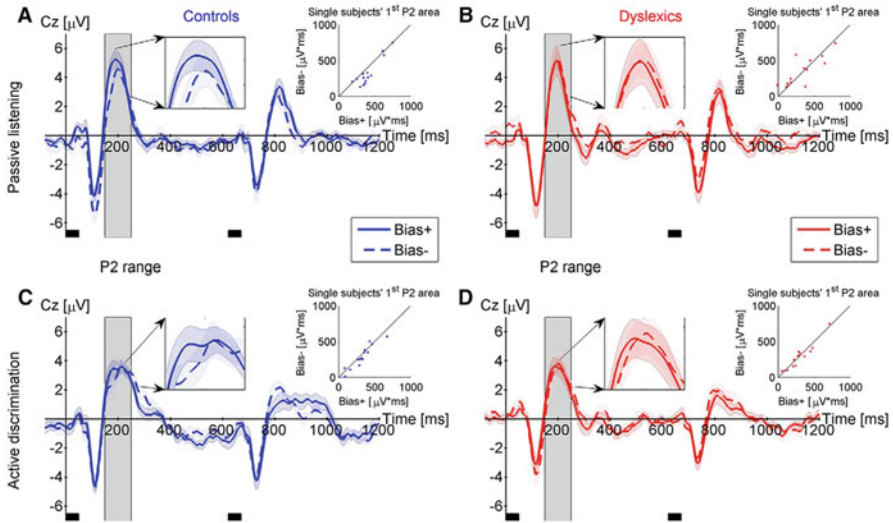


Fig. 10.6 Grand average ERP measures for the *Bias+* and *Bias-* trial types (electrode Cz). (a, c) controls (blue lines). (b, d) dyslexics (red lines). *Bias+* trials are denoted by solid lines and *Bias-* trials by dashed lines. In controls, the area of P2 after the first tone (from 150 to 250 ms, denoted by the gray rectangles) was significantly different between *Bias+* and *Bias-* trial types, in both passive listening (a) and during active discrimination (c) Dyslexics' evoked responses did not differ between the two trial types (b, d). Filled areas around the mean response denote cross-subject SEM. Small black rectangles under the plots denote the temporal location of the two tones in the trial. Insets: middle of each plot – P2 region enlarged; top right of each plot – single subject data of *Bias-* versus *Bias+* trials. In the Passive condition the difference between the trial types was significantly larger among controls than among dyslexics. (Adapted from Jaffe-Dax et al. 2015, with permission)

between the current stimulus and the mean of previous trials. Therefore, we predicted that the magnitude of control's P2 would be larger in *Bias+* trials than in *Bias-* trials, since the average distance of the first tone from the mean frequency is smaller in *Bias+* trials than in *Bias-* trials (as shown in Fig. 10.4). Consequently, the first tone in *Bias+* trials is closer to the mean (prior) than in *Bias-* trials.

We recorded ERPs with the same series of stimuli when participants either performed the task or watched a silent movie. For each participant in each of the specified trial types, we calculated the area under the curve between 150 and 250 ms after the first tone's onset as his/her individual P2 area. As predicted, we found that controls' automatically evoked response (Fig. 10.6a) was larger after the first tone in *Bias+* compared with *Bias-* trials. However, dyslexics' P2 was not sensitive to trial type, namely, *Bias+* and *Bias-* trials induced similar P2 components (Fig. 10.6b). Similar results were found under active (Fig. 10.6c,d) and passive (Fig. 10.6a, b) conditions. Taken together, these results support the hypothesis that dyslexics' computational deficit is associated with a failure to automatically integrate their on-line representations with the prior distribution.

10.7 Dyslexics' Underweight Prior also in the Visual Modality

Sensitivity to the statistics of the stimuli should not, theoretically, be restricted to a specific modality. We thus asked whether similarly reduced sensitivity would be found in the visual modality under statistically similar conditions. Previous work has shown that dyslexics' difficulty in visual discrimination tasks is restricted to sequential protocols, but their performance is intact on simultaneously presented stimuli (Ben-Yehudah & Ahissar 2004; Ben-Yehudah, Sackett, Malchi-Ginzberg, & Ahissar 2001). However, these visual studies only used typical protocols, in which one of the stimuli in each pair is constant across trials (reference) and the other (target) is randomly drawn from a limited range. Hence, serial comparisons could be aided by priors (references, anchors) based on previous trials, and consequently dyslexics' difficulties could be attributed to either poor explicit within-trial retention (explicit working memory), or to inefficient integration of priors (or both). Here (Jaffe-Dax, Lieder, Biron, & Ahissar 2016) we used the sequential spatial frequency discrimination task with a richer protocol where both gratings were randomly drawn from a wide range of spatial frequency (as in the auditory No-Reference protocol; Fig. 10.7). Thus, in addition to the groups' average performance, this protocol served to assess the magnitude of the contraction bias, which reflects the efficiency of using priors based on previous trials.

The two groups did not differ significantly in their Just Noticeable Differences (JNDs). To compare contraction bias, we compared individual performance in *Bias+* to performance on trials in *Bias-* range. Both populations showed contraction bias, i.e., better performance on trials when the first grating was closer to the mean frequency than the second grating, as shown in Fig. 10.8a. However, among dyslexics, the difference in performance between *Bias+* and *Bias-* trials was significantly smaller than among controls (Fig. 10.8a). This difference was consistent across participants: 82.5% of the control participants compared to only 57.5% of the dyslexic participants performed better on the *Bias+* trials than on the *Bias-* trials (Fig. 10.8b).

10.8 Summary and Limitations

The deficits that we found characterize more than half of the dyslexics that we assessed, though not all of our participants. These deficits are expected to impede their ability to form robust representations of their native language statistics. Yet, the actual developmental trajectory of predictive abilities and linguistic development remains a topic for future research. Additionally, we have not addressed here the relations of the anchoring, or predictive coding hypothesis to other accounts of dyslexia. One hypothesis consistent with our results, is that dyslexics' deficits stem from some type of abnormality in the dorsal stream, whose impairment had

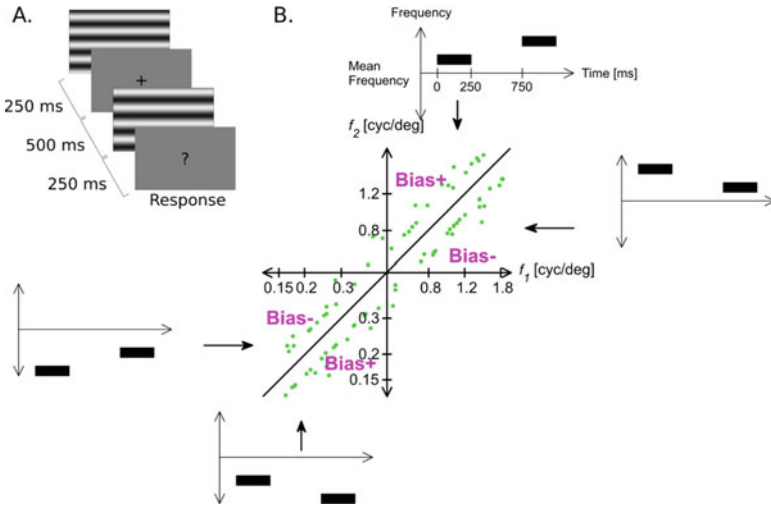


Fig. 10.7 Schematic illustrations of the sequential spatial frequency discrimination task and contraction towards the mean. (a) The temporal structure of a single trial. The first grating was presented for 250 ms, followed by an ISI of 500 ms. The second grating was presented for 250 ms. The observer was requested to indicate which of the two gratings “was denser” (had the higher spatial frequency). (b) The middle plot illustrates the distribution of single trials in the frequency plane (the frequencies of the first and second grating in each trial, respectively) for a typical subject. Each green dot denotes a pair of stimuli composing a single trial. This plane illustrates the ranges of the different trial types. Just as in the auditory frequency discrimination, in *Bias+* trials the frequency of the first grating stimulus was closer to the mean frequency; thus, contraction of its representation towards the mean increased the perceived difference between the two gratings and consequently improved performance. In *Bias-* trials the first grating was farther from the mean; thus, contraction of its representation towards the mean frequency decreased the perceived difference between the gratings and hampered performance. (Adapted from Jaffe-Dax et al. 2016, with permission)

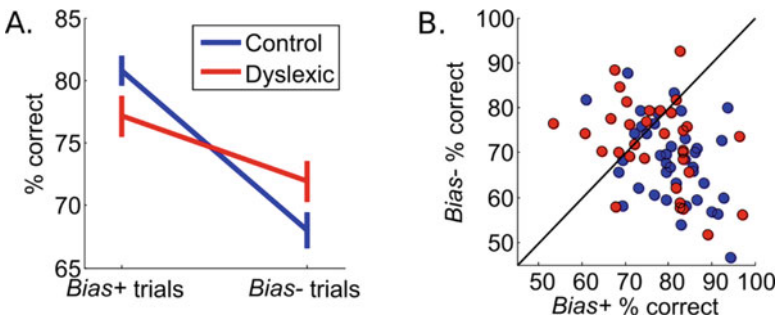


Fig. 10.8 Dyslexics’ contraction bias in the visual domain is smaller than controls’. (a) Contraction bias averaged across participants. Ordinate shows the percentage of correct responses for the two sub-divisions of trials (abscissa): *Bias+* trials (left), and *Bias-* trials (right). Controls are denoted in blue, and dyslexics in red. Dyslexics’ difference is smaller than controls’, in spite of an overall similar % correct. Error bars denote SEM. (b) Individuals’ performance (% accuracy) in *Bias-* versus *Bias+* trials. The diagonal indicates equal performance (no bias). Control participants (blue symbols) are distributed mainly below the diagonal, whereas dyslexic participants (red symbols) are more evenly distributed around the diagonal. (Adapted from Jaffe-Dax et al. 2016, with permission)

been previously suggested for dyslexia (Boros et al. 2016; Gori, Seitz, Ronconi, Franceschini, & Facoetti 2015; Paulesu, Danelli, & Berlinger 2014). According to this interpretation, the dorsal stream is involved in serial processing in perception as well as in motor plans, and based on that, perhaps in serial planning and in working memory. In fact, recent imaging studies that have tried to dissociate the role of the dorsal and ventral streams in the context of speech perception suggest that the dorsal stream involves the fronto-parietal articulatory network, which is also related to working memory (Hickok & Poeppel 2015). The left fronto-parietal network is activated both in auditory 2-tone frequency discrimination tasks (Daikhin & Ahissar 2015), and in serial spatial frequency discrimination (Reinvang, Magnussen, & Greenlee 2002, though here in both the right and left hemispheres). One of the main bundles connecting posterior and frontal parts of the dorsal stream is the arcuate fasciculus (Dick & Tremblay 2012), whose abnormality in dyslexia has been suggested by previous studies (e.g., Boets et al. 2013; Klingberg et al. 2000). These might be the candidates for the neurological origin of the deficient statistical learning in dyslexia.

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Chapter 11

The “Rowdy Classroom Problem” in Children with Dyslexia: A Review



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Abstract Over the last decades, the role of auditory processing difficulties in dyslexia has been largely debated. Recently, speech perception in noise (SIN) difficulties and their potential link with reading impairment have been investigated. However, noise has typically been considered as a unitary concept, despite the very different sort of interference it induces. Indeed, background noise typically interferes with the signal target at both peripheral and central levels of the auditory pathway. Our purpose is to review the literature to better specify SIN perception difficulties in children with dyslexia, with respect to the type of interference induced by the noise. We will first provide a description of the two main types of auditory masking corresponding to peripheral and central levels of interference. Then, we will review the existing studies that investigated SIN perception in children with dyslexia, with a detailed focus on the nature of interference induced. We hope to provide a guide to speech-language therapists, audiologists, and research scientists. In

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particular, we will specify the nature of the SIN perception difficulties experienced by children with dyslexia and will highlight the need for more precise screening and investigation tools regarding auditory processing difficulties in dyslexia.

Keywords Dyslexia · Masking · Speech in noise · Cocktail party problem · Speech intelligibility · Auditory processing

11.1 The Cocktail-Party Problem

“How do we recognize what one person is saying when others are speaking at the same time?” This question, initially formalized years ago as the cocktail-party problem (Cherry 1953, pp. 975–976), applies to most situations of human communication. Indeed, from cocktail parties to busy business meetings, understanding a given speaker is often complicated by the presence of interfering sounds, be they simultaneous speakers or environmental noises. Generations of scientists investigated speech in noise (SIN) perception in various populations, ranging from normally hearing to hearing-impaired listeners. Noise was most often considered as a unitary concept, whose presence degrades the representation of the speech target. However, psychoacoustic studies have shown that different background noises affect speech perception differently. In this chapter, we will first specify the different types of interference induced by different noise backgrounds. With this distinction in mind, we will review data of SIN perception difficulties in a specific population of normally hearing listeners who have been shown to experience unexpected difficulties in noise, namely individuals with developmental dyslexia.

11.1.1 *Peripheral Noise Interference*

Understanding a colleague’s idea, for instance, during a crowded meeting is sometimes challenging, as it requires combining efficient sensory perception and cognitive processing of the relevant speech signal while ignoring irrelevant, simultaneous speakers. The presence of background noise induces interference at two distinct levels of the auditory pathway: peripheral and central. At the peripheral level, complex auditory scenes are parsed by an auditory filterbank into their different frequency components. Schematically, the sharper the auditory filter, the better the frequency selectivity.

Because speech is a broadband signal, part of the difficulty encountered in complex auditory scenes stems from an overlap in energy coming from simultaneous auditory objects. Indeed, as long as they share common spectral components, a speech target and a simultaneous masker will interact within a number of auditory filters, hence hampering the perception of the speech target. Masking of a speech target due to spectral overlap with a simultaneous masker at the peripheral level has been called energetic masking (EM, Pollack 1975). Note that in initial studies,

the background noise had a spectrum equal to the long-term average spectrum of a speech signal (henceforth, speech-shaped noise, SSN).

Recent studies aiming at specifying the nature of the interference induced by SSN on speech intelligibility revealed that it was anything but a simple picture. Indeed, Stone, Füllgrabe, and Moore (2012) showed that notionally steady maskers (e.g., the stationary SSN used in most studies), once processed by the auditory filterbank, contain random amplitude fluctuations. These amplitude fluctuations are thought to interfere with amplitude modulations of the speech signal, hence inducing modulation masking (MM), which accounts for most of the difficulty induced by notionally steady SSN. In order to isolate pure EM, the authors presented listeners with combinations of sinusoids that were sufficiently sparse to fall within different auditory filters, hence avoiding superimposed modulations at the output of the filterbank. This “ideal” stationary masker (i.e., without random amplitude fluctuations at the output of the auditory filterbank) actually induced very limited amounts of masking.

The presence of background noise thus impedes speech intelligibility at the peripheral level through two distinct masking phenomena: energetic and by modulation, both occurring at the filter output. However, typical cocktail-party situations usually gather an important number of simultaneous speakers. Therefore, the presence of speech, rather than “simple” noise in the background induces additional difficulty in perceiving the signal of interest.

11.1.2 Central Noise Interference

In the 1980s–1990s, the first models of the auditory system were aimed at predicting auditory perception on the basis of anatomo-physiological properties (Dau, Kollmeier, & Kohlrausch 1997; Glasberg & Moore 1990). Comparing humans’ and models’ performance in cocktail-party situations led to a surprising observation: in many natural auditory environments, the listeners’ performance was lower than what would be predicted based on traditional models of the peripheral auditory system (Neff & Green 1987). The first report of this phenomenon is attributed to Pollack (1975) who termed it informational masking (IM), in opposition to the well-known energetic masking. Four decades later, the concept of IM is still ill defined (see Durlach et al. 2003, for a discussion on the definitional issues related to IM).

Canonical experiments investigating IM have focused on situations where a fixed-frequency regularly repeating target tone was embedded in a multitone background sequence whose components fell outside of a silent “protected gap” surrounding the target, a manipulation that minimizes cochlear EM (Neff, Dethlefs, & Jesteadt 1993; Neff & Green 1987). The first parametric study evaluating detection of a target using this design revealed rather staggering results: listeners experienced threshold elevations from 20 to up to 60 dB when presented in noise, compared to quiet (Kidd, Mason, Deliwala, Woods, & Colburn 1994). Threshold elevations thus confirm the presence of masking that cannot be attributed to a spectral overlap between target and maskers. Interestingly, factors typically known

to influence auditory scene analysis were shown to improve listeners' performance in IM situations, such as target repetition, relative coherence of the concurrent stream spectral content, as well as spatial and frequency separation between the target and masker (Akram, Englitz, Elhilali, Simon, & Shamma 2014). In addition to these bottom-up (perceptual) factors, top-down (cognitive) factors were shown to influence performance as well. Whereas uncertainty regarding the target to identify, fatigue, or attentional failure in focusing on the relevant target while ignoring the noise likely contribute to increase IM, musical expertise (Oxenham, Fligor, Mason, & Kidd 2003) or auditory training (Neff et al. 1993) tend to reduce IM.

Because it cannot be attributed to spectral overlap between target and maskers and is sensitive to high level, cognitive factors (experience, attention, fatigue, etc.), IM is thought to originate at a central level. Gutschalk, Micheyl, & Oxenham (2008) confirmed this distinction between peripheral and central level of background noise interference using magnetoencephalography (MEG). Their results showed that detected targets elicited a long-latency response at the level of the associative auditory areas, which was not the case for undetected targets that only elicited short-latency signals at the level of the primary auditory cortex. This result suggests that, when embedded in a background noise that maximizes IM, auditory target awareness arises between early and late stages of processing within the auditory cortex.

Nevertheless, cocktail parties are usually full of chatty human beings, rather than highly controlled robots that would only communicate with pure tone sequences. The presence of speakers of mixed gender certainly makes cocktail parties worth attending, but they also render it almost impossible to isolate the contribution of IM to listeners' perception difficulties to hear a given interlocutor. Indeed, simultaneous speech streams are broadband signals that are likely to interfere with each other at the peripheral level. Therefore, researchers aiming at evaluating IM of speech typically resorted to a very different line of reasoning than when they worked with tones.

Pioneering the investigation of IM effect on the perception of simultaneous talkers, Brungart (2001) assumed that the total masking could be split into two components, IM and EM. Only the total masking could be directly measured. Listeners' perception of a set of keywords constituting a meaningful sentence was thus evaluated when presented together with a competing speech masker. In order to evaluate the deleterious effect of IM to the listeners' performance, the author estimated the specific contribution of EM by means of a SSN with the same long-term average spectrum as the speech masker, then subtracting it from the total masking. The results of this seminal experiment have drawn general principles governing theories of speech-on-speech perception. First and foremost, IM was suggested to dominate performance in the speech-on-speech condition, as clearly evidenced by a lower intelligibility when the target sentence was presented with another simultaneous sentence than with either SSN or modulated SSN. In addition, a large proportion of the listeners' errors were intrusive words from the competing speech masker, rather than random words. Taken together, these observations suggest that in the presence of a simultaneous talker, most of the listeners' difficulty does not stem from spectral overlap between the streams, but from the linguistic content of the speech masker.

These findings were later replicated using laboratory babble (or “cafeteria”) noise (Brungart, Simpson, Ericson, & Scott 2001). Similarly to the results observed with IM of tones, both perceptual and cognitive factors were found to influence listeners’ perception of speech-on-speech. Perceptual, bottom-up cues reducing similarity (e.g., different-sex vs. same-sex speakers, Brungart 2001; Brungart et al. 2001) or increasing spatial separation between the target and babble noise (Best, Thompson, Mason, & Kidd 2013; Kidd, Arbogast, Mason, & Gallun 2005) improve listeners’ perception of the spoken target. Central, top-down factors such as prior knowledge of the target voice (Freyman, Balakrishnan, & Helfer 2004; Yang et al. 2007), syntactic coherence of the target sentence (Kidd, Best, & Mason 2008), and more generally, selective attention to the target (Zhang, Lu, Wu, & Li 2014) also contribute to improve its perception.

Whereas resorting to a “subtraction strategy” initially provided valuable insights regarding the major contribution of IM to ecological cocktail-party situations, this strategy was recently proven to have an important limitation. Indeed, most of the difficulty induced by stationary noise (such as the SSN used as an index of EM in most studies cited above) actually stems from MM, “whereby the amplitude fluctuations in the masker make it harder to detect and process amplitude fluctuations in the target” (Stone et al. 2012, p. 318). Schematically, SSN thus induces both EM and MM, whereas babble noise induces an important amount of IM in addition to EM and MM. However, because babble and SSN have very different spectral characteristics, speech being mostly periodic (for a review, see Rosen 1992) and noise being intrinsically aperiodic, it is unlikely that the amount of MM induced by a babble noise would be equal to that induced by SSN. Therefore, subtracting the amount of masking due to notionally SSN provides a rather inaccurate estimation of IM in speech-on-speech situations.

Several manipulations have been proposed to isolate the contribution of IM to complex auditory scenes. A classic solution to minimize peripheral masking is to present target and masker speech streams dichotically: as they are presented to opposite ears, target and maskers cannot interact at the cochlear level. Yet, dichotic listening provides listeners with important lateralization cues that allow them to experience spatial masking release (Freyman, Balakrishnan, & Helfer 2001; Freyman, Helfer, McCall, & Clifton 1999). Therefore, another technique was developed in order to minimize peripheral, but maximize central masking. Using speech resynthesis, spectral overlap can be removed by processing target and maskers in order to present them simultaneously but in different frequency bands, subsequently reducing EM/MM. Arbogast, Mason, and Kidd (2002) decomposed speech signals into 15 frequency bands, allocating eight frequency bands to the target speech, and the remaining seven to the masker, which was either composed of a broadband noise or of another speech signal. They observed that signal intelligibility was reduced when the processed masker was composed of broadband noise, and further decreased when it was composed of another speech signal. However, filtering speech signals drastically reduces the ecological validity of the paradigm, as it disrupts speech features (e.g., harmonicity) that are known to improve speech segregation

(Darwin 1997). Further studies are warranted to develop paradigms allowing isolating IM in ecological acoustic scenes (i.e., avoiding signal degradation).

To sum up, noisy backgrounds encountered in most typical cocktail party situations simultaneously induce peripheral and central interference with the relevant speech target. Many healthy listeners experience difficulties perceiving speech in noisy backgrounds despite normal auditory thresholds (Ruggles, Bharadwaj, & Shinn-Cunningham 2011). Crucially, the respective contribution of peripheral and central interference might vary from one listener to another, poor SIN perception hence reflecting limitations at very different levels of the auditory pathway. Therefore, there is a dire need to scrutinize the respective influence of both peripheral and central interference when investigating SIN perception in clinical populations.

The present chapter focuses on a specific population of normally hearing listeners who experience difficulties in noisy backgrounds, namely children with developmental dyslexia. Yet, so far, most studies of SIN perception in dyslexic children have considered noise as a unitary concept, acting like a “corrosive” degrading the representation of the target speech, irrespective of the nature of the interference it induces. This over-simplification likely stems from the lack of paradigms allowing clear distinction between EM, MM and IM of speech. Therefore, the following section will provide an extensive literature review on SIN perception in children with dyslexia, with a specific focus on the respective influence of all three types of masking on SIN perception.

11.2 Dyslexia and the Rowdy Classroom Problem

If they rarely attend cocktail parties, most of the children’s social life nevertheless takes place amongst noisy backgrounds: lively playgrounds, busy refectories, etc. With average noise levels largely above the World Health Organization guidelines regarding basic acoustical requirements for community noise (General Accounting Office, 1995; cited by Jamieson, Kranjc, Yu, & Hodgetts 2004), most elementary schools are the scene of what could be called a “rowdy classroom problem”. Recent studies showed that the amount of background noise in classrooms impacts typically developing children’s academic performance (e.g., Bradley & Sato 2008; Shield & Dockrell 2003, 2008). The rowdy classroom problem seems particularly challenging for a specific clinical population: children with developmental dyslexia.

11.2.1 Underlying Causes of Developmental Dyslexia

Learning to read requires accessing meaning from printed symbols, a process that, for alphabetic systems, relies on the ability to map distinct visual symbols onto phonemes (for a review see Morais 2018). If most children achieve fluent and effortless reading in their early school years, a significant proportion of the school

age population suffers from developmental dyslexia, namely persistent reading difficulties despite normal intelligence, adequate educational opportunities and in the absence of any neurological or sensory deficiencies (World Health Organization 2008). In their guidelines for teaching to children with learning difficulties, the Belgian minister for Education claimed a ratio of one dyslexic child per classroom of about 20 pupils in elementary school, which is consistent with the 5% prevalence of dyslexia usually reported around the world (Lindgren, Renzi, & Richman 1985, but see Fluss et al., 2009 for a discussion of the socio-economic status influence on this figure).

Because its hallmarks are extremely slow and error-prone reading, poor non-word decoding and weak spelling, dyslexia was initially described as a form of visual word blindness. Surprisingly, first experimental works on the causes of dyslexia soon unveiled another picture: even though they were perfectly able to identify visual letters, poor readers were unable to map them into their corresponding phonemes (Liberman 1973; Shankweiler & Liberman 1972). This process is known as phonological coding (Share 1995), and is defined as the ability to use speech codes to represent information in the form of words and parts of words. Over the last decades, the vast majority of studies confirmed this observation, and the phonological processing deficit is now widely acknowledged as the most prominent hypothesis accounting for dyslexics difficulties learning to read (for a review of the various causes of reading difficulties, see Vellutino, Fletcher, Snowling, & Scanlon 2004). Evidence of poor phonological awareness, poor verbal short-term memory and slow lexical retrieval, three abilities that contribute to phonological processing, pile up to account for the reading difficulties experienced by dyslexic children (Wagner & Torgesen 1987), and are thought to persist well into adulthood (e.g., Law, Vandermosten, Ghesquière, & Wouters 2014). Reduced neural integration between letters and sounds (as indexed by neural activation in temporal auditory cortices when letters and sounds mismatch) further support the hypothesis of a phonological deficit in dyslexic individuals (Blau et al. 2010; Blau, van Atteveldt, Ekkebus, Goebel, & Blomert 2009). Nevertheless, theories regarding the causes underlying reading difficulties are still hotly debated, and can be broadly classified as to whether the phonological deficit is directly or indirectly assumed to lead to reading difficulties.

Several authors claimed that the phonological processing deficit was the side effect of a broader deficit. Abnormal visual processing in dyslexic individuals (Stein 2001, 2018, but see Amitay, Ben-Yehudah, Banai, & Ahissar, 2002, for a discussion of this hypothesis) and atypical learning abilities (Ahissar 2007; Ahissar, Lubin, Putter-Katz, & Banai 2006; Jaffe-Dax, Daikhin, & Ahissar 2018, but see Ziegler, 2008, for a discussion of this theory) have been proposed as broader deficits underlying the reading difficulties associated with dyslexia. While these two hypotheses are still debated, the present review focuses on a third hypothesis, according to which dyslexics' phonological difficulties are linked to a broader auditory processing impairment.

11.2.2 *Nonspeech Auditory Perception*

The hypothesis of a broad auditory impairment that would account for phonological processing deficits, hence leading to reading difficulties, was initially proposed by Tallal (1980). Even though this claim was largely debated for both methodological and theoretical reasons (for discussions, see Landerl & Willburger 2010; Rosen 2003), it has stimulated an unprecedented amount of research on auditory processing in dyslexic individuals.

Various auditory processing abilities have been suggested to be impaired in dyslexic individuals, ranging from frequency discrimination (Ahissar, Protopapas, Reid, & Merzenich 2000; Baldeweg, Richardson, Watkins, Foale, & Gruzelier 1999; Hari & Kiesilä 1996; McAnally & Stein 1996) to perception of amplitude modulation (Hämäläinen, Rupp, Soltész, Szücs, & Goswami 2012; McAnally & Stein 1997; Menell, McAnally, & Stein 1999), stream segregation (Helenius, Uutela, & Hari 1999; Lallier et al. 2011; Sutter, Petkov, Baynes, & O'Connor 2000) and spatial processing (Smith & Griffiths 1987). Interestingly, dyslexic children revealed poorer detection of a complex tone target presented in sequences inducing pure IM compared to both chronological age (CA) and reading level (RL) matched controls (Calcus, Colin, Deltenre, & Kolinsky 2015a). The analysis of variations in response time throughout the experiment did not reveal significant fatigue or attentional effects on dyslexic children's performance.

An important question arises from these consistent observations of dyslexics' poorer performance on elementary auditory tasks: how does it relate to reading difficulties? Rosen and Manganari (2001) hypothesized that impaired performance in backward masking might lead to poorer perception of a /ba-/da/ contrast, as the crucial second formant transition is followed by a vowel (that has more power than the initial consonant), whereas preserved performance in forward masking would not affect the perception of an /ab-/ad/ contrast. Yet, they failed to report specific impairment for the /ba-/da/ contrast: dyslexics' speech perception performance was overall poorer than their controls'. Surprisingly, Sebastian and Yasin (2008) showed impaired neural discrimination of pure tones, but not of speech syllables in dyslexic adults. However, the very different nature of the stimuli used as speech and non-speech material somewhat limits the interpretation of their results. On the contrary, Serniclaes, Sprenger-Charolles, Carre, and Demonet (2001) compared dyslexic children's discrimination of sinewave analogues of speech (Remez, Rubin, Pisoni, & Carrell 1981) that are perceived either as simple non-speech whistles or as speech sounds, depending on the instructions. Taking advantage of this ambiguous material, the authors showed that dyslexic children's auditory deficit was specific to speech.

It is also noteworthy that, at the individual level, only a subgroup of dyslexic individuals (about 30%; for a review see Ramus, Pidgeon, & Frith 2003) show non-speech auditory processing impairments. Importantly, speech intelligibility requires listeners to not only perceive simultaneous acoustic cues, but also integrate them over multiple temporal scales (e.g., Hickok & Poeppel 2007). Taken together, these observations have led researchers to consider the possibility of a specific difficulty related to linguistic material in dyslexic individuals.

11.2.3 *Speech Perception*

Progress in signal analysis allowed investigation of speech perception by evaluating listeners’ categorical perception (CP). Speech perception is categorical as long as discrimination between two tokens depends on their labelling, rather than their acoustical differences (Liberman, Harris, Hoffman, & Griffith 1957). Inefficient categorization of speech sounds would likely affect the processing of speech sounds, consequently impeding acquisition of the phoneme to grapheme conversion code. Various studies have thus evaluated CP in individuals with dyslexia, as poor CP might be causally related to reading difficulties. If these studies provided one consistent finding, it is that dyslexics’ perception of speech is anything but a clear picture.

Many studies have reported poor CP in dyslexic individuals (Brandt & Rosen 1980; Godfrey, Syrdal-Lasky, Millay, & Knox 1981; Mody, Studdert-Kennedy, & Brady 1997). Yet, this deficit was either limited to few phonological contrasts (Cornelissen, Hansen, Bradley, & Stein 1996), to synthetic but not natural speech (Blomert & Mitterer 2004), or again, to only a subgroup of dyslexics (Adlard & Hazan 1998; Manis et al. 1997). Altogether, the CP deficit associated with dyslexia was thus proposed to be “fragile” (Blomert & Mitterer 2004).

In addition, not all researchers agree on the idea that phonological representations are merely underspecified in dyslexia. In fact, recent studies support the hypothesis that phonological representations might in fact be overspecified in dyslexic individuals. Indeed, Serniclaes et al. revealed that dyslexic children were actually better at discriminating intra-category variants of the same phoneme (e.g., two acoustically different /ba/) than typical readers. This surprising finding suggests that dyslexics might experience allophonic speech perception (Serniclaes et al. 2001; Serniclaes, van Heghe, Mousty, Carre, & Sprenger-Charolles 2004; Varnet, Meunier, Trolle, & Hoen 2016). However, other studies failed to provide clear support for either underspecified (Hazan, Messaoud-Galusi, Rosen, Nouwens, & Shakespeare 2009; Robertson, Joanisse, Desroches, & Ng 2009) or overspecified (Messaoud-Galusi, Hazan, & Rosen 2011; van Beinum, Schwippert, Been, van Leeuwen, & Kuijpers 2005) phoneme representations in dyslexic individuals.

Recently, another explanation has emerged regarding the possible cause for the fragile and inconsistent speech perception difficulties observed in dyslexic individuals. Indeed, all the studies reported above focused on optimal, quiet listening situations. Yet, everyday communication usually happens in deleterious noisy backgrounds that reduce the redundancy of acoustic cues available in the target speech (e.g., Zeng et al. 2005). Therefore, recent researches have explored SIN perception as a new potential source for the cascading difficulties encountered by dyslexic individuals.

11.2.4 *Speech Perception in Noisy Backgrounds*

Pioneering investigation of SIN perception and its potential influence on reading, Brady, Shankweiler, and Mann (1983) revealed that 8 year-old poor readers identified monosyllabic words presented in quiet similarly to CA controls, but performed significantly lower when words were presented in SSN. Over the last decades, speech perception deficits in noise, but not in quiet have been largely replicated in various studies (e.g., Messaoud-Galusi et al. 2011; Rüsseler, Gerth, Heldmann, & Münte 2015; Ziegler, Pech-Georgel, George, & Lorenzi 2009), hence confirming the hypothesis of a subtle, but consistent impairment in speech perception, which reveals itself in adverse listening conditions. It has been noted that studies investigating dyslexics' speech perception often led to ceiling scores in the quiet condition (e.g., Brady et al. 1983; Rüsseler et al. 2015; Ziegler et al. 2009). To circumvent this limitation, recent studies provided measures of speech perception thresholds in more demanding tasks (i.e., discrimination and identification of a voicing contrast). Varying the method used to measure listeners' thresholds, the results confirmed preserved performance in quiet that significantly worsened in noise when dyslexics were compared to typical readers (Hazan et al. 2009; Messaoud-Galusi et al. 2011).

In the vast majority of studies, group comparisons consistently showed poorer performance in dyslexics' than CA controls (e.g., Bradlow, Kraus, & Hayes 2003; Chandrasekaran, Hornickel, Skoe, Nicol, & Kraus 2009; Dole, Hoen, & Meunier 2012; Poelmans et al. 2011). To our knowledge, only three studies failed to report poorer SIN perception in dyslexic individuals. The first one was conducted on 6 year-old children at risk for dyslexia with no later confirmation of the diagnostic outcome (Elbro, Borstrøm, & Petersen 1998). The second was an extensive study showing that, unlike children with specific language impairment, dyslexic children were not impaired in a SIN perception task when compared to both CA and RL controls (Robertson et al. 2009). The third study was performed on adults who were selected from a university population. Some of them performed within the normal range on reading and spelling, which the authors acknowledged might reflect successful compensation mechanisms (Law et al. 2014). Thus, on the whole, the available data confirm the claim of a subtle speech perception deficit associated with dyslexia, which would only reveal its true prevalence in adverse listening conditions.

In an extensive study of the nature of the SIN perception deficit, (Ziegler et al. 2009) evaluated dyslexic childrens consonant identification. The consonant was selected from the following set: /p,t,k,b,d,g,f,s,ʃ,m,n,r,l,v,z,j/, and presented within /vCv/ logatomes (v being always /a/), together with both fluctuating and stationary background noises. Indeed, the presence of “dips” in a fluctuating background noise is known to favour masking release in adult listeners, a phenomenon that relies on rapid spectro-temporal analysis of the information available when the “local” signal-to-noise ratio (SNR) exceeds a certain threshold (Gnansia, Jourdes, & Lorenzi 2008). The authors thus presented dyslexic children with both fluctuating and stationary background noises. The results confirmed previous evidence of SIN

perception impairment when dyslexics were presented with both stationary and fluctuating noises. Crucially, dyslexics’ performed significantly worse than both CA and RL control children, which allowed the authors to conclude that the impairment in SIN perception reflects a core difficulty inherent to dyslexia rather than a maturational delay or a feedback of reading acquisition on speech perception (Goswami 2003). Moreover, SIN perception predicted significant unique variance in reading, even after controlling for sensory and cognitive factors. Yet, the dyslexics’ SIN deficits were not due to poor spectro-temporal, low-level auditory resolution, as the magnitude of their masking release was similar to the controls’. Similar SIN perception deficits were observed when presenting dyslexics with internal noise (i.e., speech was degraded in order to preserve only its slow envelope modulations). Calcus, Deltenre, Colin, and Kolinsky (2017) confirmed impaired SIN perception in dyslexics compared to CA, but not RL controls along with preserved masking release abilities, even at SNR of -12 dB. Other studies replicated and extended observation of a SIN perception deficit in 5 year-old pre-schoolers who later developed dyslexia, with SIN perception uniquely contributing to reading level observed at the age of eight (Boets et al. 2011). Significant correlations between reading and SIN perception were also reported in 11 year-old children with dyslexia (Poelmans et al. 2011). Taken together, these results suggest that SIN is a core deficit associated with dyslexia, due to a lack of robustness of speech representation in the presence of both internal and external noise.

However, so far, most studies examining SIN perception in dyslexics have considered noise as a unitary concept, overlooking the importance of the nature of the interference induced by the noise background. Yet, preserved ability to analyze the spectro-temporal content of the auditory scene, as indexed by preserved masking release in dyslexic children, rules out a purely sensory explanation to their SIN perception difficulties. Therefore, there is a dire need to specify the nature of the interference induced by a specific background noise, as each noise type may reflect a different processing mechanism.

Table 11.1 provides a brief description of the most prominent experiments that investigated SIN perception in dyslexics. Most of them used SSN as a masker, which, as we commented on, induces interference at the peripheral level of the auditory system through a combination of MM and EM. Only one study aimed at specifying the respective influence of pure MM and EM on dyslexic children’s difficulties perceiving SIN. The results confirmed poorer SIN perception in dyslexic than CA, but not RL controls in both noise conditions (Calcus, Lorenzi, Collet, Colin, & Kolinsky 2016). With respect to IM, some studies resorted to babble noise, composed of 4- (Dole et al. 2012), 12- (Elbro et al. 1998) or 20- interfering speakers (Hazan et al. 2009; Hazan, Romeo, & Pettinato 2013; Messaoud-Galusi et al. 2011). Even though the presence of an interfering speaker induces mainly IM of a speech target (Brungart 2001; Brungart et al. 2001), the amount of masking induced by an N-talker babble noise greatly varies with N (Simpson & Cooke 2005). If a 4-talkers babble maximizes IM, the presence of 20 simultaneous speakers drastically reduces the informational nature of the masker, which mostly induces EM/MM.

Table 11.1 Experiments that have investigated SIN perception in dyslexic children (in chronological order)

| Study | Participants | Target and task | Masker | Dyslexics' performance |
|------------------------------|--|---|--------------------------------------|---|
| Brady et al. (1983) | Poor reader children (n = 15) vs. CA controls | Monosyllabic words identification | Quiet and Modulated SSN (0 dB SNR) | Preserved in quiet, but affected by the presence of noise when compared to CA |
| Snowling et al. (1986) | Dyslexic children (n = 19) vs. CA and RL controls | High and low frequency words; pseudo-words identification | Quiet and SSN (0 and -3 dB SNR). | Deficit in all listening conditions (including quiet): lower than CA when repeating low frequency words; lower than both CA and RL for pseudo-words |
| Cornelissen et al. (1996) | Dyslexic adults (n = 10) vs. CA controls | CV identification | White noise (+19, 0, -2, -3 dB SNR) | Poorer for some CV contrasts (/ta/ with /a/ and /pa/ with /fa/) |
| Elbro et al. (1998) | Children at risk for dyslexia (n = 23) vs. CA controls | Phoneme discrimination (minimal pairs) | 12-talkers babble noise (+15 dB SNR) | No deficit |
| Bradlow et al. (2003) | Children with learning disabilities (n = 63) vs. CA controls | Keyword identification in simple sentences | White noise (-4 and -8 dB SNR) | Poorer than CA, especially when SNR decreases |
| Chandrasekaran et al. (2009) | Poor reader children (n = 15) vs. CA controls | Keyword identification in simple sentences | SSN (adaptive threshold measure) | Poorer than CA |

| | | | | |
|-------------------------|--|--|--|--|
| Ziegler et al. (2009) | Dyslexic children (n = 19) vs. CA and RL controls | Consonant identification /vCv/ | Quiet, stationary and modulated SSN (0 dB SNR) | Preserved in quiet. Poorer than both CA and RL in stationary noise. Preserved masking release |
| Hazan et al. (2009) | Dyslexic adults (n = 17) vs. CA controls | Phoneme discrimination and identification (CP) /pea/-/bee/ contrast | Quiet and 20-talkers babble (adaptive threshold measure) | Preserved in quiet. Overall poorer than CA with fixed-step threshold measure of discrimination abilities |
| Robertson et al. (2009) | Dyslexic children (n = 14) vs. CA and RL controls | Phoneme discrimination and identification (CP) /ball/-/doll/ continuum | Quiet and white noise (+12 dB SNR) | No deficit |
| Boets et al. (2011) | 5 year-old children at risk who were subsequently found to be dyslexics (n = 16) vs. CA controls | Monosyllable identification | SSN (-1, -4, -7 dB SNR) | SIN performance in kindergarten uniquely contributed to reading in Grade 1 |
| Poelmans et al. (2011) | 11 year-old dyslexic children (n = 13) vs. CA controls | /vCv/ logatomes and sentences repetition | SSN (-4, -7, -10 dB SNR) | SIN deficit persists until 11 year-old, and correlates with reading |

(continued)

Table 11.1 (continued)

| Study | Participants | Target and task | Masker | Dyslexics' performance |
|---|--|--------------------------------------|---|--|
| Inoue, Higashihara, Okazaki, & Maekawa (2011) | Dyslexic children (n = 10) vs. CA controls | Phoneme identification /ba/ vs. /da/ | Quiet and multi-talker babble (0 dB SNR) | Preserved in quiet, but poorer than CA in noise (especially when the target appears with a high probability) |
| Messaoud-Galusi et al. (2011) | Dyslexic children (n = 62) vs. CA controls | CP (cf. Hazan et al. 2009) | Quiet and 20-talkers babble (adaptive threshold measure) | Similar to Hazan et al. (2009): preserved in noise, but poorer than CA with fixed-step threshold measure of discrimination abilities |
| Hazan et al. (2013) | Dyslexic children (n = 34) vs. CA controls | /CV/ identification | 20-talkers babble (0 dB SNR) | Poorer than CA when intonation of the target varies |
| Dole et al. (2012) | Dyslexic adults (n = 16) vs. CA controls | Disyllabic word identification | SSN modulated SSN 4-talkers babble | → preserved → impaired when spatialized → impaired when monotic |
| Law et al. (2014) | Dyslexic adults (n = 36) vs. CA controls | Keyword sentences & /CVC/ words | → SSN (-5, -10, -13 dB SNR) → SSN (adaptive threshold measure) | No deficit |

| | | | | |
|------------------------|---|---|---|--|
| Rüsseler et al. (2015) | Dyslexic adults (n = 12) vs. CA controls | Disyllabic words (congruent or incongruent with visual information) | Quiet and white noise (+30 dB SNR) | Preserved in quiet, but poorer than CA when congruent AV information in noise |
| Calcutt et al. (2015b) | Dyslexic children (n = 10) vs. CA and RL controls | /CV/ identification | Quiet SSN; modulated SSN; 4-talkers babble (dichotic listening; SNR = -30 dB) | Preserved in quiet, but poorer than CA (but not RL) in all noise conditions |
| Varnet et al. (2016) | Dyslexic adults (n = 20) vs. CA controls (n = 18) | Phoneme categorisation /da/-/ga/ contrast | White noise (adaptive thresholds) | Poorer than CA |
| Calcutt et al. (2016) | Dyslexic children (n = 16) vs. CA and RL controls | /vCvCv/ identification | Quiet; SSN; modulated SSN (SNR: 0 to -12 dB) | Preserved in quiet, but poorer than CA (but not RL) in all noise conditions. Preserved masking release |
| Calcutt et al. (2017) | Dyslexic children (n = 14) vs. CA and RL controls | /vCv/ and /vCvCv/ identification | Quiet combination of sinusoids; SSN; modulated SSN; 1-, 4-, and 8-talkers babble (SNR = 0 dB) | Preserved in quiet, but poorer than CA (but not RL) in all noise conditions |

Very few studies specifically investigated pure IM on dyslexic individuals' speech perception difficulties. However, the available data suggest that central interference contributes to their difficulties. In a disyllabic word identification task, Dole et al. (2012) presented target and maskers in various spatial settings, including dichotic presentation. Presenting target and masker dichotically prevents spectral overlap at the peripheral level, hence minimizing EM/MM. Adults with dyslexia had a lower performance than typical readers in this condition, indicating masking at a more central level of the auditory pathway. Investigating /CV/ identification in various background noises presented dichotically, Calcus, Colin, Deltenre, and Kolinsky (2015b) showed an overall lower performance in dyslexic than CA, but not RL, control children. These studies converge to point toward a central contribution to dyslexics difficulties perceiving SIN.

11.3 Discussion in the Rowdy Classroom

A consistent finding emerges from the various studies reviewed in this chapter: dyslexic children are affected by the presence of background noise, or at least more so than CA controls. If evidence regarding the specific contribution of peripheral and central interference of noise on dyslexics' SIN perception remains scarce, the existing data suggest that they both contribute to the difficulties encountered by dyslexic children. The following section will be dedicated to highlight similarities and divergences in results regarding SIN perception in dyslexic children.

First, the vast majority of studies reported lower SIN perception performance in dyslexic children compared to typical readers when a SSN background induced mostly peripheral interference. Interestingly, preserved masking release has been consistently reported in dyslexic children, as their identification performance improves to the same extent as in typical readers when presented with fluctuating background maskers (Calcus et al. 2016; Ziegler et al. 2009). Dip listening is known to require high spectro-temporal resolution at the level of the cochlea (Festen 1990). As stated by Ziegler et al. (2009, pp. 733), "normal masking release [in dyslexics] therefore suggests that low-level auditory or peripheral processes are intact". Accordingly, Zettler, Sevcik, Morris, and Clarkson (2008) showed that on average, dyslexic children performed similarly to typical readers in a task that required them to integrate amplitude fluctuations across multiple frequency bands to segregate signals from noise. According to the authors, this suggests that dyslexic children adequately use temporal and spectral information in noise to identify a signal. Altogether, these observations are at odds with a purely sensory explanation of dyslexic children's SIN perception deficit. Hence, they rule out theories of poor temporal auditory processing (Tallal 1980). In sum, dyslexic children are impaired in noisy situations inducing peripheral interference with an auditory target, but this difficulty is not attributable to poorer peripheral auditory processing. This apparent paradox has led us to further examine the role of central mechanisms contributing to the SIN perception difficulties in dyslexics.

Dyslexics’ consonant identification performance has been reported to be lower than CA controls’ with babble noise, be it presented monotonically (Dole et al. 2012), dichotically (Calcus et al. 2015b) or diotically (with target and maskers presented simultaneously to both ears; Calcus et al. 2016; Inoue, Higashibara, Okazaki, & Maekawa 2011). Because the presence of babble in the background is thought to induce mainly IM (Brungart 2001), which takes place at a central level of the auditory pathway (Durlach et al. 2003; Gutschalk et al. 2008), this observation points to a central contribution to the SIN perception difficulties experienced by dyslexic children.

As an interim conclusion, we can note that dyslexic children perform poorer than CA controls in auditory environments that respectively induce purely peripheral and purely central interference. Yet it is worth noting that their performance does not significantly worsen as compared to CA controls’ in conditions inducing peripheral and central interference simultaneously (e.g., Calcus et al. 2017). Taken together, the data thus suggest that both peripheral and central interference respectively contribute to the dyslexics’ SIN perception difficulties, but do not seem to interact. Crucially, as stated before, peripheral auditory processing seems to be preserved in dyslexic children. Therefore, difficulties in both peripheral and central masking likely stem from non-sensory (i.e., cognitive) processes. Whereas this was expected in the case of central masking, it is somewhat more surprising regarding peripheral masking, which is typically thought to reflect the limits of the cochlear frequency selectivity. This apparent paradox is likely explained by the fact that peripheral auditory processing operates under central control. Indeed, an extensive efferent auditory pathway provides anatomical substrate to top-down modulation of auditory perception, especially in noise (for a review, see Winer 2006). Whether dyslexics’ difficulties in noise stem from a purely central deficit or from a disruption of the efferent auditory pathway remains an open question. Exploration of the top-down modulation of speech encoding in noisy backgrounds might break new ground in this respect.

Remarkably, dyslexic children seem to consistently benefit from various types of acoustic cues to improve their perception. The perceptual cues that were considered in the literature include spatial lateralization, repetition of the target, fluctuations in the background noise, or variation in the number of interfering talkers (Calcus et al. 2015b, 2016; Dole et al. 2012). Other factors such as the lexical frequency of the target words (Snowling, Goulandris, Bowlby, & Howell 1986) or the consistency in the speakers’ intonation (Hazan et al. 2013) also help them improve SIN perception. Note however that, if their performance improves thanks to perceptual cues (as compared to performance without such cues), it does not normalize (as compared to CA controls’ performance).

Last, studies generally report only weak support for a link between auditory processing and reading abilities. Indeed, most studies failed to reveal significant correlations between reading abilities and all of the auditory tasks that were evaluated in dyslexic children (e.g., Robertson et al. 2009). Scrutinizing individual profiles unveiled a similar picture, with only a subgroup of dyslexic children consistently impaired in a majority of the noise conditions tested within each study (31% of the

dyslexic children in Calcus et al. (2016); 28% in Calcus et al. (2017)). Among this subgroup, not all individuals were also impaired in phonological processing (21% of the dyslexic children tested in Calcus et al. (2016)) or categorical perception (12.5% of the dyslexic children tested in Calcus et al. (2016)). This figure is consistent with previous data suggesting that the vast majority of children perform within the norms on auditory tasks, with only about 30% of them being impaired in auditory processing (Adlard & Hazan 1998; Amitay et al. 2002; Ramus et al. 2003; Rosen, Windzio, & Galaburda 2001). The fact that only a minority of dyslexic children are consistently impaired in auditory tasks might explain the small to medium size effects reported in most studies (e.g., Calcus et al. 2015a, 2016). Further research is required to determine whether dyslexic children exhibiting consistently poor auditory performance share other commonalities and hence would form a specific subgroup.

Almost as informative as the convergence between studies are discrepancies found across experiments. Two major inconsistencies are observed. The first one concerns the comparison of dyslexics' to typical readers' performance. Indeed, whereas two studies reported that dyslexics' performance was lower than both RL and CA controls' (Snowling et al. 1986; Ziegler et al. 2009), recent SIN perception data are mixed on that matter. If dyslexic children perform consistently lower than CA controls, this is not the case when compared to RL controls. Dyslexic children were reported to perform similarly (Calcus et al. 2015a, 2016) or even better in some conditions (Calcus et al. 2017) than RL controls.

Two (not necessarily incompatible) explanations might account for the absence of a significant difference when comparing dyslexics to RL controls. The first one is that dyslexic children might experience a mere developmental delay in speech perception abilities, which would bring them to the same level as younger children (i.e., RL controls). Indeed, SIN intelligibility improves with age in typically developing children, especially in situations inducing mainly IM (Lutfi, Kistler, Oh, Wightman, & Callahan 2003; Wightman, Kistler, & O'Bryan 2010). However, this developmental delay remains to be explained. In addition, this explanation might hold for speech perception but does not account for more general impairments, namely for the fact that dyslexic children were found to be impaired when compared to both CA and RL controls in a nonspeech detection task inducing pure IM (Calcus et al. 2015b).

Another explanation might be that reading acquisition itself impacts the quality of and/or access to phonological representations, hence favoring SIN perception. Goswami (2015, p. 44) recently discussed more generally the idea that the "reduction in reading experience that is inherent in being dyslexic can itself cause differences in sensory processing between participants with dyslexia and controls". In agreement with this view, when listening to speech, adults who remained illiterate for strictly socio-economic reasons show reduced activation (compared to literates) of the planum temporale (Dehaene et al. 2010), a region known to host phonological representations (e.g., Chang et al. 2010; Jacquemot, Pallier, Le Bihan, Dehaene, & Dupoux 2003). Similar reduced activation has been observed in dyslexics (Blau et al. 2010, 2009; Monzalvo, Fluss, Billard, Dehaene, & Dehaene-Lambertz 2012).

Reading acquisition may in fact help in finely tuning phonemic boundaries and hence in increasing the precision of phoneme identification in literates compared to illiterates (Serniclaes, Ventura, Morais, & Kolinsky 2005), which would be most helpful in suboptimal listening conditions (for a review, see Kolinsky 2015). Experiments using word identification in dichotic listening suggest that in suboptimal conditions, literate people also use an attentional mechanism focusing on the phonemic structure of speech (Morais, Castro, Scliar-Cabral, Kolinsky, & Content 1987), which seems to be strategic as it is modulated by instructions to pay attention to phonemes (Morais, Castro, & Kolinsky 1991). Such a strategy might help in reconstructing poorly perceived SIN sequences and is obviously less available to dyslexics and illiterates, who are very poor at phoneme awareness (e.g., Morais, Cary, Alegria, & Bertelson 1979; Morais, Cluytens, & Alegria 1984). In addition, in typical readers, auditory words activate brain regions associated with orthographic processing, an effect that is not observed in illiterate adults (Dehaene et al. 2010) and that is strongly reduced in children with reading difficulties (Desroches et al. 2010; Monzalvo et al. 2012). Behaviorally, in literates, orthographic representations have been shown to influence spoken word recognition (e.g., Ziegler & Ferrand 1998), an effect that is particularly strong in noisy backgrounds (Pattamadilok, Morais, & Kolinsky 2011) and that is also reduced in dyslexic adults (Pattamadilok, Nelis, & Kolinsky 2014). Altogether, these studies concur to support the idea that at least some sensory deficits observed in dyslexic children might result from the effects of reduced reading experience on their brain.

In any case, further studies are warranted to disentangle the respective contribution of maturation and reading acquisition on SIN perception. As commented on by Goswami (2015), only a few research designs can to some extent control for the effects of reading experience, and if similar outcomes are found using combinations of these designs, causality is likely to be present. These designs involve not only reading level-matched studies, but also research with illiterate adults, studies on pre-readers who go on to be diagnosed with dyslexia, and, most critically, longitudinal studies that follow the same children over the whole learning process as well as well-controlled training studies.

The second finding that is inconsistent across studies concerns the potential cause(s) underlying dyslexics’ auditory processing deficit in noise. Preserved masking release rules out the hypothesis of a strictly sensory deficit in dyslexics’ auditory processing. Accordingly, several studies claimed that peripheral auditory processing is preserved in dyslexic children (e.g., Zettler et al. 2008; Ziegler et al. 2009), whose difficulties would thus stem from and/or be exacerbated by non-sensory factors (Hazan et al. 2013; Messaoud-Galusi et al. 2011). As discussed above, high inconsistency in dyslexics’ impairment across various noise conditions of the same study is in line with this interpretation. Notably, up to 40% of children diagnosed with reading difficulties also exhibit attention-deficit/hyperactivity disorder (see Eden & Vaidya 2008, for a discussion on comorbidity between dyslexia and attention-deficit/hyperactivity disorder). Altogether, this suggests that dyslexics’ difficulties perceiving SIN might in fact stem from an attention deficit. However, recent studies reporting high intra-individual inconsistencies within the

dyslexic population focused on children who were free of a formal diagnosis of attention-deficit/hyperactivity disorder. Yet, dyslexic children might experience subtle attentional lapses that might only reveal in the more complex auditory tasks. Therefore, there is a need for methodological tools allowing investigation of auditory attention in complex acoustic environments.

Overall, the nature of the relationship between auditory processing and reading abilities in dyslexic children remains an open question. One possibility is that because most everyday listening situations are noisy, a difficulty in SIN perception would hamper the acquisition of precise phoneme representations, ultimately hampering the acquisition of phoneme-to-grapheme conversion. Another possibility is that poor SIN perception reflects imprecise phoneme representations (or difficulties in accessing this information, e.g., Boets et al. 2013), whose detrimental consequences remain unnoticed in favourable listening conditions, i.e., in quiet, revealing themselves only in more adverse, noisy, conditions. Support for a causal relationship comes from correlations observed between auditory processing and reading abilities (Poelmans et al. 2011; Tallal 1980), from observations that dyslexic children are impaired in SIN perception tasks even when compared to RL controls (Ziegler et al. 2009), and from the fact that basic auditory processing difficulties have been reported in newborns at risk of dyslexia (Leppänen et al. 2010) which, together with SIN perception deficits (e.g., Boets et al. 2011), are thought to predict later reading abilities.

Yet, not all individuals with dyslexia show an auditory deficit and, conversely, not all individuals with such a deficit have dyslexia (for a review, see Hämäläinen, Salminen, & Leppänen 2013), and although many at-risk preliterate children show impaired basic auditory processing when compared to controls, it is impossible to discriminate on that basis between those who, later on, will become typical or atypical readers (e.g., Plakas, van Zuijen, van Leeuwen, Thomson, & van der Leij 2013). Other results provide only weak support for a direct link between poor speech processing and poor reading skills. Indeed, correlations between these abilities were not consistently reported (Calcus et al. 2015b, 2016; Robertson et al. 2009). Accordingly, only a subgroup of dyslexic children seems consistently affected by the presence of background noise, but not all of them are also impaired in literacy-related tasks. Lastly, comparing dyslexic to RL control children led to contradictory results (Calcus et al. 2017; Ziegler et al. 2009). Taken together, these findings do not support the hypothesis of a causal relation between SIN perception and reading abilities (Hazan et al. 2009; Messaoud-Galusi et al. 2011; Robertson et al. 2009). As stated by Rosen (2003, pp. 524), “This [...] is at the heart of what appears to be the uselessness of the auditory measure as a gauge of the language/literacy deficit”. If we cannot rule out the possibility of a relationship between auditory processing and reading abilities, most studies support the idea that both difficulties tend to co-occur in dyslexia, but are not causally related. The available evidence is rather in line with a risk factor model (e.g., Bishop 2006; Pennington 2006; van Bergen, van der Leij, & Jong 2014), which proposes that no single deficit is either necessary or sufficient to lead to dyslexia, but that a number of factors may interact to lead (or not) to reading difficulties. Studies investigating the relationship between

SIN perception and reading abilities in other populations (e.g., children with mild to moderate sensorineural hearing loss, illiterate adults) might pave the way to a better understanding of the complex link between auditory processing and reading difficulties.

11.4 Conclusion

Developmental dyslexia is a multidimensional disorder that affects a significant proportion of the school age population. The most prominent hypothesis regarding dyslexia postulates a phonological impairment as the core deficit leading to reading difficulties. This chapter intended at evaluating SIN perception difficulties in children with dyslexia, with a special focus on the respective contribution of peripheral and central processes to these difficulties. Taken together, the findings reviewed here suggest that dyslexic children are impaired in noisy environments inducing both types of interference, as evidenced by studies inducing mainly peripheral (EM/MM) and central (IM) masking. Interestingly, recent results concur to support the hypothesis of preserved sensory (i.e., peripheral) processes. However, they consistently point to a deficit in nonsensory factors that might contribute to the SIN perception difficulties, although the role of auditory attention factors to SIN perception in dyslexic children remains unclear. Further studies are needed to investigate whether SIN difficulties stem from a purely cognitive deficit or from an impairment of the efferent auditory pathway in dyslexic children.

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Chapter 12

From Subtypes to Taxons: Identifying Distinctive Profiles of Reading Development in Children



Adrian P. Burgess, Caroline Witton, Laura Shapiro, and Joel B. Talcott

Abstract The longstanding debate between dimensional and categorical approaches to reading difficulties has recently been rekindled by new empirical evidence and developments in theory. At the heart of the categorical perspective is the tenet that dyslexia is a taxon, a grouping of cases that can account for both intra-group similarities and inter-group differences. As developmental dyslexia is characterized by a diverse constellation of symptoms with multiple underlying risk and protective factors, the key question in dyslexia research has shifted from “What is dyslexia?” to “How many taxons or subtypes of dyslexia are there?” The primary objective of this chapter is to consider methods that can be used to objectively define these groupings, starting with the current practice of defining subtypes of readers using normative scores with pragmatically dened cut-offs, the “Quadrant Analysis” approach, and progressing towards more theoretically sound and statistically rigorous procedures. We review and test several candidate approaches that can be readily adapted to realistic conditions that are problematic for Quadrant Analysis. Specifically we propose a method that can be used to identify subgroups in the bivariate case when the two indicator variables are correlated. We conclude by evaluating the strengths and weaknesses of this and other methods and include implications for their future application toward identifying and validating putative dyslexia taxons.

Keywords Dyslexia · Subtypes · Orthography · Phonology · Taxon · Development · Reading · Statistical modelling

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12.1 Introduction: Categories or Dimensions?

That some children are poorer readers than others is beyond dispute and that some are poorer than might be expected given their overall cognitive ability is also certain. However, it remains unclear whether poor readers are simply statistical outliers (as is inevitable in any norm-based measurement system) or whether they form a subgroup that differs from typical readers in certain key characteristics (see e.g., S. E. Shaywitz, Escobar, Shaywitz, Fletcher, & Makuch 1992). This debate, between dimensional and categorical approaches to children with reading difficulties, is longstanding but has recently been revived by the decision to replace the term “dyslexia” in DSM-5 with “specific learning disability (reading)” (*Diagnostic and statistical manual of mental disorders: DSM-5*, American Psychiatric Association 2013) and by resurgent claims that dyslexia is an unscientific construct that does not really exist (Elliott & Grigorenko 2014). This is no arcane debate because the true nature of specific reading difficulties has powerful implications for the diagnosis, intervention and service provision for those affected and their families.

The heart of the categorical perspective on reading difficulties is that dyslexia forms a taxon. That is, dyslexia constitutes a grouping of cases that share underlying commonalities that not only account for the similarities between group members but also explain how and why group members differ from non-group members. Sex is an example of a taxon; males and females share many common features but differ in certain fundamental characteristics that justify considering sex as a categorical construct. More formally, a taxon is a fundamental, objective, non-arbitrary and reasonably enduring latent structure (Ruscio & Ruscio 2004). To justify dyslexia as a taxon therefore, children with reading difficulties should either show some characteristics that are qualitatively distinctive, or the distribution of their latent abilities should be discontinuous from those of typical readers.

In fact, given that developmental dyslexia is characterized by a diverse constellation of symptoms with multiple underlying risk and protective factors (Pennington 2006) the question often raised is not “*Does dyslexia constitute a taxon*” but “*How many taxons or subtypes of dyslexia are there?*” (Pennington 2006; Peterson, Pennington, Olson, & Wadsworth 2014) and this is currently an active area of research. To date, this pursuit has been most successful in identifying individuals with presumed dissociations between cognitive skills closely linked to reading achievement, for example phonological and orthographic skills consistent with that predicted by the dual route model (Castles & Coltheart 1993), or separable dimensions of phonological decoding and articulatory naming speed (i.e., rapid automatized naming (RAN)) as consistent with the double deficit hypothesis (Bowers & Wolf 1993). Both models predict the occurrence of discrete subtypes of individuals, with relatively isolated deficits in a single component process and comparatively normal functioning in the other and each approach has reported subtypes of dyslexia that are largely consistent with its own theoretical perspective.

The definition of subtypes of dyslexia primarily derives from normative performance and membership is assigned to those who score below a specified level

of performance on one or more theoretically relevant cognitive tasks. Defined in this way, any continuous bivariate distribution will necessarily divide the whole sample into quadrants: those who score above the threshold on both dimensions, those who score below the threshold on both dimensions and those who score above the threshold on one dimension and below the threshold on the other. The choice of threshold is at least semi-arbitrary and may be determined pragmatically by non-theoretical considerations. For example, the threshold might be set to ensure that there are sufficient cases in each group to allow statistical analysis or to equate to a round number of stanines, z-scores or percentiles. Of course, the arbitrary nature of such thresholds precludes these subtypes from being considered as taxons.

It is not that there is anything fundamentally wrong with using cut-off scores with continuous variables (categorization of continuous data is commonplace and often useful) but we should be clear that is what we are doing, if that is what we are doing. If reading ability is dimensional, then we should be clear that the subtypes identified are not fundamental and are comparable to groups like the “tall” or the “rich” which can be defined in many different ways. If, on the other hand, reading profiles are taxonic, we should be explicit about how the subtypes differ and set cut-off scores at a level that optimally separate the groups.

The primary objective of this chapter is to consider ways in which we can progress from defining subtypes of readers using normative scores with pragmatically defined cut-offs and move towards a more rigorous approach to identifying reading taxons, if they exist. In the first part, we address the use of normative scores with cut-offs to identify subtypes of readers, a method we refer to as “Quadrant Analysis”. Specifically, we show how to estimate the proportion of children in each subtype and argue that deviations between the observed and expected numbers of children in each quadrant might provide useful information about where cut-off scores should be set. In the second part, we develop this idea and propose a method that makes the choice of thresholds less arbitrary and illustrate its use with data from a previously published study (Talcott et al. 2002). Finally, we briefly consider alternative approaches to answering the “*dimension or category?*” question.

12.2 Using Normative Scores to Define Subtypes: Quadrant Analysis

The identification of one or more subtypes of children who show qualitatively different profiles of reading ability from typical developing children has grown into an area of considerable research interest, if little consensus. There is, currently, no agreed definition or characterization of these subtypes and this absence has encouraged the emergence of multiple arbitrary and ad-hoc definitions of subgroups of children based on performance on one or more measures of reading ability (Boder 1973; Castles & Coltheart 1993) or other cognitive dimensions (Bosse, Tainturier, & Valdois 2007; Bowers & Wolf 1993) For example, there is evidence to suggest

that the ability to read phonetically regular non-words such as “*tegwog*” is at least partially dissociated from the ability to read phonetically irregular real words such as “*yacht*” in learner readers. To develop this example, one can define subgroups from these two measures (non-words vs. exception words) by defining some criteria for good and bad performance on each of these tests (Sprenger-Charolles, Siegel, Jiménez, & Ziegler 2011). This defines four groups: (i) those who are good at reading both non-words and exception words, (ii) those who are bad at reading both types, (iii) those who are good at reading non-words but poor at exception words and (iv) those who are good at reading exception words but poor at reading non-words. The term “Quadrant Analysis” derives from the fact that this approach inevitably creates four groups, but the groups are not normally equal in size and the cut-offs may be used to define two groups (one quadrant vs. the other three). It should be obvious that procedures of this type will inevitably identify a subset of children regardless of whether dyslexia exists or not and so is of no use in identifying taxons.

The choice of cognitive skills to measure (e.g., non-word reading vs exception word reading) is at least driven by theoretical considerations. In contrast, the choice of threshold of determining “good” and “bad” performance is almost completely open. The threshold is typically defined such that any score less than a certain number of standard deviations below the mean is considered to indicate poor performance. In the absence of any good evidence as to how such a cut-off should be defined, it is usual to select a non-arbitrary seeming number of standard deviations below the mean (1/2, 1, 1 1/2), a whole number of stanines (1, 2, 3 or 4) or a round number percentile (10%, 20%, etc.). Such cut-off scores are chosen to appear principled and may allay the suspicion that the definition is opportunistic (imagine your response to reading that a definition of 0.6745 standard deviations below the mean was used¹) but these cut-offs are essentially arbitrary and are at least as much determined by convenience and an affection for round numbers as they are by empirical evidence.

More sophisticated, multivariate methods, such as identifying poor readers based on a discrepancy between their actual reading ability and their predicted reading ability (for example, that based on a regression of reading achievement on cognitive ability) may have some advantages but every method that uses normative scores and performance cut-offs to define subtypes of dyslexia, shares the same limitations. First, they will always identify subtypes of poor readers whether or not any distinctive taxon, such as dyslexia, exists. Second, they are fundamentally arbitrary and are not based on the distinctive characteristics of the taxon (assuming the taxon exists at all).

The absence of any objectively defined criterion or cut-off scores means that individual researchers are at liberty to define reading subtypes in whatever way suits them and it is a liberty freely exercised. Worse still, they can change their criteria from study to study at their own convenience. Unsurprisingly, the consequence of this approach is that reading subtypes are inconsistently defined in the scientific

¹Actually 0.6745 is the 25th percentile so perhaps not as arbitrary as it first appears.

literature and the justification for the cut-off scores used is not explained. It seems likely that many of the inconsistencies that have been reported, and the controversies they inspire, result at least in part from such failures of definition. None of this should be a cause for surprise because Quadrant Analysis is not designed to identify subtypes, let alone taxons. Indeed, the use of Quadrant Analysis to define reading subtypes can only be defended because the current state of knowledge makes it difficult to know what else to do and, although there are alternative approaches (e.g., Cluster Analysis, Finite Mixture Modeling, and Taxometric Methods – see Sect. 12.5), these too have their problems. Nevertheless, as Quadrant Analysis is widely used, it is worth exploring how the method might be improved.

In one wished to argue that these Quadrant Analysis “subtypes” are taxonic, it would be necessary to provide some principled justification for the choice of cut-off. For example, the subtypes should show some characteristic that would not be predicted by assuming the observed scores came from a continuous bivariate distribution. One such characteristic might be that the proportion of individuals in each quadrant differs from what would be expected if the data derived from a continuous bivariate distribution and this is the case we consider here. This is of interest because if learner readers consist of one or more subtypes, then the scores will not be normally distributed because each of the subtypes will have their own mean, standard deviation and covariance. In extreme cases, this will result in a bimodal distribution of scores although this is relatively uncommon in human performance data. More generally, a unimodal distribution will be seen but the number of individuals in each quadrant (defined by cut-offs determined by the mean and standard deviation of the whole sample) will differ from what would be seen if the data were normally distributed. This means that the proportion of individuals observed in each quadrant, compared to the expected number may offer some information on whether a subtype exists or not and where the cut-offs should be positioned. To this end, in the following section we show how to estimate the observed and expected proportions of cases in each quadrant.

12.3 How Many Individuals in Each Quadrant?

Let’s consider the example of reading non-words and exception words, and consider the case where we wish to know the proportion of individuals that score below a given cut-off score on these two measures and compare that to the proportion of cases that would be expected if the data came from bivariate normal distribution with no subtypes. If the scores on the two tests are independent (i.e., not correlated) then the problem is quite straightforward. To start, let’s use the mean as the cut-off score (i.e., 0 standard deviations below the mean). If the data are normally distributed, we would expect to find half the sample scoring below the mean on each of our tests and half above. That is, the probability of scoring below the mean on either test is 0.5.

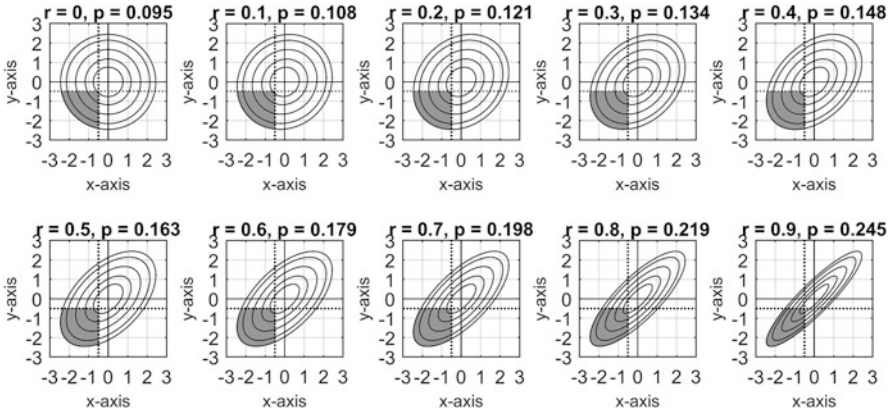


Fig. 12.1 Showing the PDF of the bivariate normal distribution with correlations from 0.0 to 0.9 and standard deviation = 1. The rings indicate the regions containing (from inner to outer) 25%, 50%, 90% and 95% of the population. The shaded area indicates the proportion of the population that is more than 1/2 standard deviation below the mean on both measures which increases from 9.5% when $r = 0$ to 24.5% when $r = 0.9$

If the test scores are uncorrelated then, by the multiplication rule,² the proportion of individuals scoring below the mean on both tests will be $0.5 \times 0.5 = 0.25$. For other cut-offs, we need to know the probability density function (PDF) of the normal distribution which is given by:

$$P(x) = \frac{1}{\sigma\sqrt{2\pi}} \exp\left[-(x - \mu)^2 / (2\sigma^2)\right] \tag{12.1}$$

Where μ is the population mean and σ^2 is the population variance. In order to find the proportion of the population expected to score below any given cut-off, x_{cut} , we need to calculate the cumulative probability from $-\infty$ to x_{cut} , which, can be estimated through numerical integration of Eq. 12.1 and is readily available in Tables. For example, if we use a cut-off score of 1/2 standard deviation below the mean, we know from the cumulative PDF of the normal distribution that approximately 31% of individuals fall below this level. It follows that about 9.5% of individuals would be expected to score 1/2 standard deviation or more below the mean on two tests ($0.31 \times 0.31 = 0.095$).

If the scores on the tests are correlated (as they usually will be), the situation becomes more complicated and the proportion of participants expected to score below any given cut-off of two tests will increase with the correlation (Fig. 12.1). For example, the proportion of individuals scoring 1/2 standard deviation or more below the mean on both tests ranges from 0.095 when the correlation is 0 to 0.31,

²The multiplication rule, $p(A \cap B) = p(A) \cdot p(B/A)$ which is $p(B/A) = p(B)$ when A and B are independent.

when the correlation is +1 (Fig. 12.1). To estimate this proportion for any given correlation, we need to know the PDF of the bivariate normal distribution which is given by:

$$P(x, y) = \frac{1}{2\pi\sigma_x\sigma_y\sqrt{1-\rho^2}} \exp \left[\frac{\left(\frac{(x-\mu_x)^2}{\sigma_x^2} - \frac{2\rho(x-\mu_x)(y-\mu_y)}{\sigma_x\sigma_y} + \frac{(y-\mu_y)^2}{\sigma_y^2} \right)}{2(1-\rho^2)} \right] \quad (12.2)$$

Where ρ is the correlation between x and y . To estimate the proportion of the population expected to score below a specified cut-off, x_{cut} , on test x and below a specified cut-off, y_{cut} , on test y , we simply have to estimate the cumulative probability of Eq. 12.2 $-\infty$ to x_{cut} and $-\infty$ to y_{cut} . For the convenience of the reader Table 12.2 was produced which shows the proportion of the population expected to score below a given cut-off score (ranging from 0 to -2 standard deviations below the mean) on two tests for correlations between 0 and 0.9 and a summary of the same data is represented graphically in Fig. 12.2. As can be seen from Fig. 12.2, the proportion of participants scoring below both cut-offs tends towards zero as the correlation, ρ , approaches -1 and tends towards the univariate marginal probability defined by the cut-off as ρ , approaches $+1$.

It would also be useful to have confidence intervals for these proportions for use with empirical data and this can be estimated assuming the binomial distribution. It follows that if p is the proportion of the population that will score below a specified cut-off, x_{cut} , on test x **and** below a specified cut-off, y_{cut} , on test y , then the expected value of sampling from the population is np where n is the sample size and the variance of the estimate will be $np(1-p)$. From these values, it is straightforward to estimate confidence intervals for and desired combination of cut-off score, correlation and sample size.

To illustrate the process, we show how to estimate the proportion of participants in each quadrant and illustrate the method using data from the Oxford Primary School Study (Talcott et al. 2002). This sample comprised 353 children (183 girls, 170 boys) between the ages of 83 and 150 months (mean 112.8, s.d. 14.9). All of the children attended mainstream primary schools within the local education authority. Children who did not have English as a first language were not included in the study, but no other selection criterion was applied.

Whichever approach is used to identify latent taxonic structure, it is necessary to select appropriate indicator variables that measure the construct in question. In the case of reading, a very large number of indicators have been used that purport to discriminate between typical and dyslexic readers but, influenced by dual-route models of reading (Coltheart, Rastle, Perry, Langdon, & Ziegler 2001), we elected to use two distinct measures of reading ability. Specifically, we chose non-word reading as the putative measure of phonological processing and exception word reading as the putative measure of orthographic processing. In languages with opaque orthographies as English, the inconsistency of the mapping between

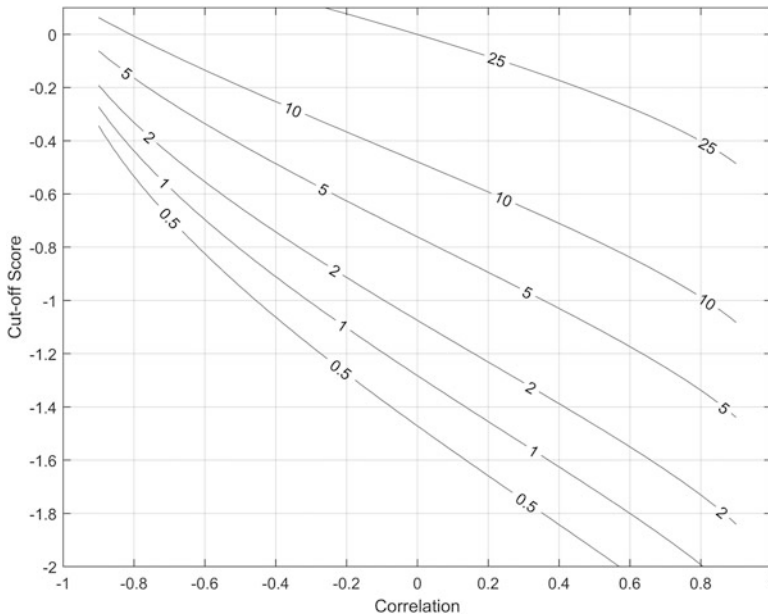


Fig. 12.2 Contour plot of the proportion of participants expected to score below a given cut-off score both of two tests by the correlation between the tests. The cut-off scores are defined as the number of standard deviations below the mean. Labels on contours are percentiles

letters and sounds poses a difficult task for the beginning reader, and ultimately requires a development of a reading system that is flexible, with lexical access facilitated by both the phonological and orthographic characteristics of words (Coltheart 1978). Although the strongest determinant of reading aptitude in typically developing children, and of reading impairments, is the competency to which phonological decoding skills are acquired and employed (Coltheart 1978). Lexical access in simple reading tasks (Rack, Snowling, & Olson 1992; Wagner & Torgesen 1987), impairments of other reading sub-skills, such as in orthographic coding, also explain variance in literacy skill in some individuals with developmental dyslexia (Badian 2005; Castles & Coltheart 1993). There is also strong evidence that phonological and orthographic impairments contribute independently to the heritable and presumed genetic component of risk for specific reading difficulties (Castles, Datta, Gayan, & Olson 1999). Evidence that subtypes of dyslexia based on dual route models may have different developmental trajectories, with implications for assessment and intervention (Manis, Seidenberg, Doi, McBride-Chang, & Petersen 1996; Talcott, Witton, & Stein 2013).

Children were assessed on a wide range of measures (Table 12.1) but the indicator variables we chose for the following examples was the Castles and Coltheart Reading Test (1993) which provides reading scores for exception words and non-words in the range 0–30.

Table 12.1 Cognitive Performance on selected tests for the Oxford Primary School Study (Talcott et al. 2002)

| Cognitive test | Mean (s.d.), skew |
|---------------------------------------|--------------------|
| BAS ^a verbal IQ | 58.8 (9.0), -0.28 |
| BAS matrices | 56.1 (8.8), -0.20 |
| BAS reading | 54.8 (10.9), -0.16 |
| BAS spelling | 52.4 (8.9), 0.02 |
| C&C ^b regular word reading | 24.8 (6.8), -1.70 |
| C&C non-word reading | 19.4 (8.0), -0.65 |
| C&C exception word reading | 16.4 (6.1), -0.71 |

^aBAS = British Ability Scales (Elliot, Murray, & Pearson 1983)

^bC&C = Castles and Coltheart Reading Test

The mean score for non-words was 19.4 (s.d. = 8.0) and for Exception words was 16.4 (s.d. = 6.1) and the correlation between the two was 0.76. A scatterplot for the observed scores from this sample is shown in Fig. 12.3a along with the estimated marginal PDF, estimated using kernel smoothing, for non-words (Fig. 12.3b) and exception-words (Fig. 12.3c). Each of the marginal PDFs is shown with a normal PDF with the same mean and standard deviation as the observed PDF. PDFs were obtained using Kernel Density Estimation (also known as the Parzen-Rosenblatt window method) which is a non-parametric method for estimating the PDF of a random variable that provides more robust and reliable estimates of the true PDF than traditional histogram methods.

Seventy children out of 353 obtained a score of less than 1/2 standard deviations below the mean on both tests which is 19.8% of the whole sample. The question we address here is whether this proportion is higher than would be expected if the data followed a bivariate normal distribution? To begin, it is worth noting that neither marginal distribution appears to be normally distributed and this suspicion is supported by the Shapiro-Wilk test which shows that both distributions deviate significantly from normal (Non-words: $W = 0.915$, $p < 0.01$; Exception Words: $W = 0.954$, $p < 0.01$) and that, in consequence, the data cannot be bivariate normal. For both tests, the data were truncated by the range of possible scores and there is evidence for a ceiling effect on nonword word reading. In addition, the scores can only take integer values and are not truly continuous as would be expected in a normal distribution. Nevertheless, the PDFs of the marginal distributions are not so abnormal that the idea of using conventional parametric statistical analysis with them (e.g., t-tests, analysis of variance (ANOVA), etc.) would cause much concern (Fig. 12.4).

We can compare the proportion of participants in the observed sample who score below the cut-off of $-1/2$ standard deviations on both tests with the proportions expected from a bivariate normal distribution using the data in Table 12.2. The closest entries to a cut-off of $-1/2$ and a correlation of 0.76 are 0.198 and 0.219 for $r = 0.7$ and 0.8 respectively. By linear interpolation this gives 0.211 for $r = 0.76$ which is close to the observed proportion of 0.198 but it remains to be determined whether a difference of this size is likely to be real or simply due to chance

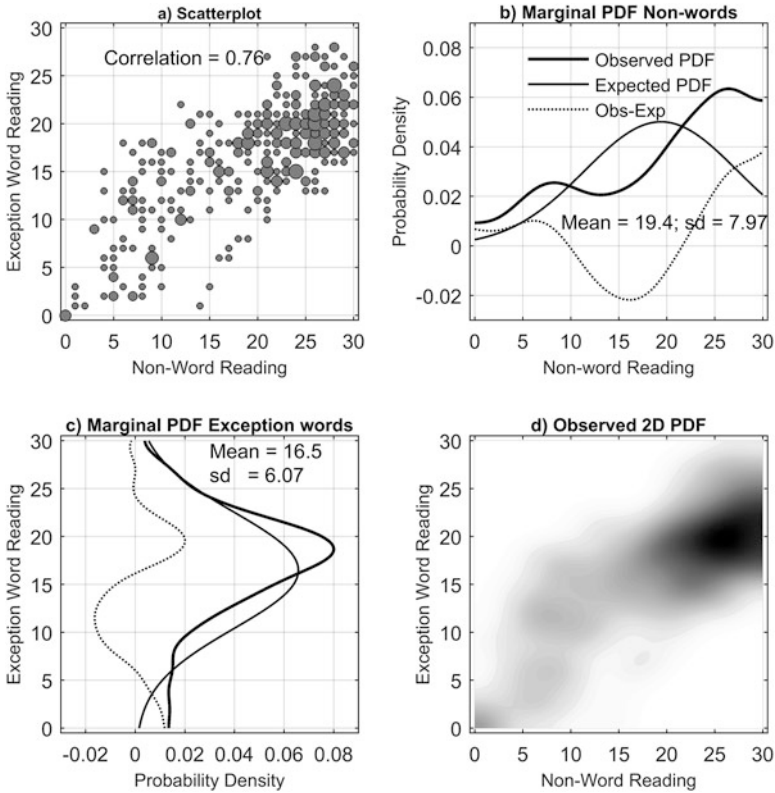


Fig. 12.3 Oxford Primary School Study cohort showing (a) the scatterplots of non-word reading by exception word reading for, (b) the marginal distribution of non-word reading (thick line) with the best fitting normal distribution (thin line), (c) the marginal distribution of exception-word reading (thick line) with the best fitting normal distribution (thin line) and (d) the 2-dimensional PDF of non-word reading by exception word reading (darker colours indicate higher density)

variation. Using binomial theory, and assuming we were sampling from a bivariate normal distribution, the mean expected number of people we should expect to see scoring below cut-off on both tests is np i.e. $353 \times 0.211 = 74.5$ and the variance is $np(1 - p)$ which is $353 \times 0.211 \times (1 - 0.211) = 58.8$ giving a standard deviation of $\sqrt{58.8} = 7.7$. To get the 95% confidence intervals we calculated the mean score ± 2 standard deviations which gives $74.5 \pm 2 \times 7.7$ giving 59–90 (rounding down and up to the nearest integer respectively). As the observed number of cases ($n = 70$) was within this interval, we can conclude that the number of cases observed is within the bounds that would be expected if we were sampling from a bivariate normal distribution.

The illustration here is used to examine individuals who scored below the cut-off on both tests but the same ideas can be applied to each of the other quadrants if desired. We know from the cumulative probability function of the

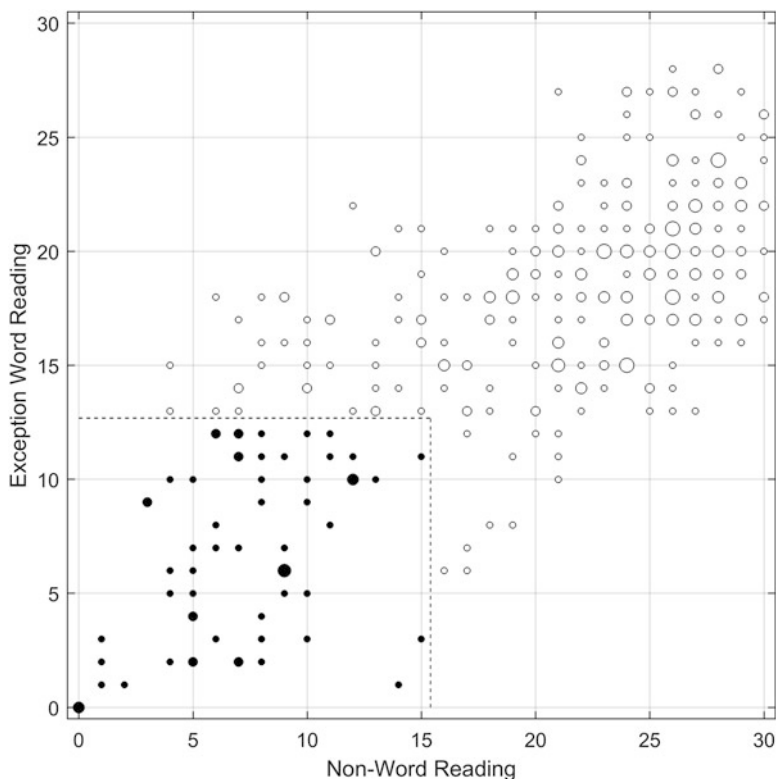


Fig. 12.4 Oxford Primary School Study cohort showing the classification of cases by the Quadrant method using cut-off scores based on the point of maximum difference between the observed and expected PDFs. Dotted lines indicate the borders of the lower-left hand quadrant

normal distribution that the probability of a score less than $-1/2$ standard deviations below the mean is 0.309. From the example above, we know that the proportion of participants who would be expected to score less than $-1/2$ on both tests was 0.211 so it follows that those who scored below $-1/2$ on Test 1 and above $-1/2$ on Test 2 is $0.309 - 0.211 = 0.098$. By symmetry, the proportion who scored above $-1/2$ on Test 1 and below $-1/2$ on Test 2 is the same, 0.098. Knowing the proportion of cases in three of the four quadrants, the fourth quadrant is not hard to find.

The finding that the proportion of participants in each quadrant is within the bounds of what would be expected with a bivariate normal distribution with the same correlation is consistent with data from other studies. Using the same procedure with published data on dyslexia subtypes (Castles & Coltheart 1993; Genard, Mousty, Content, Alegria, Leybaert, & Morais 1998; Jimenez, Rodriguez, & Ramirez 2009; Manis et al. 1996; Sprenger-Charolles, Colé, Lacert, & Serniclaes 2000; Ziegler 2008) suggests that the frequencies observed in each group in empirical studies often fail to differ significantly from what would be expected if the data had been

Table 12.2 Showing the proportion of cases expected to score below the z-score cut-off on both tests for given levels of correlation between the tests assuming a bivariate normal distribution with correlations between 0 and 0.9. Probabilities derived from Eq. 12.2 were estimated by numerical integration in MatLab (R2015a) using function `integral2.m` over the range -4 to x_{cut} and -4 to y_{cut} with $dx = dy = 0.01$

| z-score cut-off | Correlation between tests | | | | | | | | | |
|-----------------|---------------------------|-------|-------|-------|-------|-------|-------|-------|-------|-------|
| | 0.0 | 0.1 | 0.2 | 0.3 | 0.4 | 0.5 | 0.6 | 0.7 | 0.8 | 0.9 |
| 0.00 | 0.250 | 0.266 | 0.282 | 0.298 | 0.315 | 0.333 | 0.352 | 0.373 | 0.398 | 0.428 |
| -0.10 | 0.212 | 0.228 | 0.243 | 0.260 | 0.277 | 0.294 | 0.313 | 0.334 | 0.358 | 0.389 |
| -0.20 | 0.177 | 0.192 | 0.208 | 0.224 | 0.240 | 0.258 | 0.276 | 0.297 | 0.320 | 0.350 |
| -0.30 | 0.146 | 0.161 | 0.175 | 0.191 | 0.207 | 0.223 | 0.241 | 0.261 | 0.284 | 0.313 |
| -0.40 | 0.119 | 0.132 | 0.146 | 0.161 | 0.176 | 0.192 | 0.209 | 0.228 | 0.250 | 0.278 |
| -0.50 | 0.095 | 0.108 | 0.121 | 0.134 | 0.148 | 0.163 | 0.179 | 0.198 | 0.219 | 0.245 |
| -0.60 | 0.075 | 0.087 | 0.098 | 0.111 | 0.124 | 0.138 | 0.153 | 0.170 | 0.189 | 0.214 |
| -0.70 | 0.059 | 0.069 | 0.079 | 0.090 | 0.102 | 0.115 | 0.129 | 0.144 | 0.162 | 0.186 |
| -0.80 | 0.045 | 0.054 | 0.063 | 0.073 | 0.083 | 0.095 | 0.107 | 0.121 | 0.138 | 0.160 |
| -0.90 | 0.034 | 0.041 | 0.049 | 0.058 | 0.067 | 0.077 | 0.089 | 0.101 | 0.117 | 0.136 |
| -1.00 | 0.025 | 0.031 | 0.038 | 0.045 | 0.054 | 0.062 | 0.072 | 0.084 | 0.098 | 0.115 |
| -1.10 | 0.018 | 0.023 | 0.029 | 0.035 | 0.042 | 0.050 | 0.059 | 0.069 | 0.081 | 0.097 |
| -1.20 | 0.013 | 0.017 | 0.022 | 0.027 | 0.033 | 0.039 | 0.047 | 0.056 | 0.066 | 0.081 |
| -1.30 | 0.009 | 0.013 | 0.016 | 0.020 | 0.025 | 0.031 | 0.037 | 0.045 | 0.054 | 0.066 |
| -1.40 | 0.007 | 0.009 | 0.012 | 0.015 | 0.019 | 0.024 | 0.029 | 0.036 | 0.044 | 0.054 |
| -1.50 | 0.004 | 0.006 | 0.009 | 0.011 | 0.015 | 0.018 | 0.023 | 0.028 | 0.035 | 0.044 |
| -1.60 | 0.003 | 0.004 | 0.006 | 0.008 | 0.011 | 0.014 | 0.017 | 0.022 | 0.028 | 0.035 |
| -1.70 | 0.002 | 0.003 | 0.004 | 0.006 | 0.008 | 0.010 | 0.013 | 0.017 | 0.022 | 0.028 |
| -1.80 | 0.001 | 0.002 | 0.003 | 0.004 | 0.006 | 0.008 | 0.010 | 0.013 | 0.017 | 0.022 |
| -1.90 | 0.001 | 0.001 | 0.002 | 0.003 | 0.004 | 0.006 | 0.007 | 0.010 | 0.013 | 0.017 |
| -2.00 | 0.001 | 0.001 | 0.001 | 0.002 | 0.003 | 0.004 | 0.005 | 0.007 | 0.010 | 0.013 |

drawn from a bivariate normal distribution and, by definition, a bivariate normal distribution suggests a single population with no subtypes or taxons. This provides a challenge to the use of this simple dissociation logic to define subgroups of dyslexia in this context and suggests that stronger evidence is required in order to reify the existence of distinct diagnostic entities from such data.

12.4 An Improved Method for Choosing Cut-Off Scores

In the case of the Oxford Primary School Study (Talcott et al. 2002), the proportion of cases in each quadrant is very close to what might be expected had the data been drawn from a bivariate normal distribution but that does not completely exclude the possibility that subtypes exist. Indeed, the distribution of performance on the non-word reading test appears to show a bimodal distribution (Fig. 12.3b) and the two-dimensional-PDF (Fig. 12.3d) shows two distinct regions of high density. This suggests that if the cut-off scores had been better placed, they might have provided a subdivision of the sample into the clusters that visual inspection seems to suggest exist. It is obvious that the optimal location for the cut-off scores is where the difference between the observed and expected numbers of cases is greatest and this can be found by extending the logic of the above example in which the difference between the observed and expected number of cases are calculated with cut-offs of $-1/2$ standard deviations to all possible cut-offs. To do this, it is only necessary to estimate the observed and expected PDFs.

By assuming a bivariate normal distribution with the same means, standard deviations and correlation as the observed data, it is straightforward to estimate the expected PDF and this is shown in Fig. 12.3c. Estimation of the observed PDF can be done in a number of different ways but we used a 2D kernel estimator (Botev, Grotowski, & Kroese 2010) using their MatLab function “kde2d.m”, available from Botev (2015). The estimated observed PDF is shown in Fig. 12.3e. Figure 12.3f shows the difference between the observed and expected PDFs. The difference between observed and expected values represents the greatest difference and shows a minimum at non-words = 15.6 and exception Words = 12.9, which corresponds to z-scores of -0.473 and -0.583 respectively. Assuming bivariate normality with $r=0.76$, the probability of scoring below both cut-off scores, estimated, using numerical integration, was 0.201. This gives the expected number of cases as 71 ± 15 (i.e., 56–86). The first group was defined as those individuals who scored below the cut-off score on both measures and this amounted to 63 individuals making up 17.8% of the sample which did not differ significantly from what was expected. The second group consisted of all those individuals not in the first group and, by definition, scored higher on both measures.

One indicator of the validity of this classification comes from the characteristics of the two samples. Whilst it is inevitable that one group will score higher on both tests than the other, it need not be the case that they show the same relative performance on the two measures. In this example, the higher scoring group

were significantly better at non-word reading than exception word reading (Non-words = 22.1, *s.d.* = 5.9; Exception Words = 18.6, *s.d.* = 4.0, $t = -11.14$, $p < 0.001$) but there was no such advantage for the lower scoring (Non-words = 6.7, *s.d.* = 3.9; Exception Words = 18.6, *s.d.* = 4.0, $t = 11.14$, $p < 0.001$). Such a discrepancy in performance suggests that the groups showed a qualitatively different pattern of performance and did not simply differ in overall level of ability. Although this evidence falls short of what would be required to demonstrate that the groups form taxons (Ruscio & Ruscio 2004), it is the sort of difference that might be expected if they did. This difference is also readily interpretable. The high scoring group were significantly stronger at reading non-words than exception words. The natural interpretation of this is that the high scoring group includes those who are well on their way to mastering phonics whereas the lower scoring group has not.

It is notable that although the lower scoring group represents those who are less successful in reading than their peers, the group does not readily map onto dyslexia as generally conceived. First, the proportion of the sample in the lower-scoring group (17.8%) is much higher than the typical prevalence estimates of dyslexia of 5–10% (Rodgers 1983; B. A. Shaywitz, Fletcher, Holahan, & Shaywitz 1992). However, as this was a cross-sectional study, we do not know, whether the reading difficulties seen endured or whether some of the children went on to read well so only some of those identified may have been dyslexic.

In addition, the poor readers form a homogeneous group and there no evidence of the subtypes of dyslexia expected by theory but this, of course, depends upon the sample studied. Quadrant analysis will split the groups into 4 regardless of whether such grouping exist or not and this split will occur at the point of maximal difference between the observed and expected PDFs. In a representative sample of readers, the maximal difference split might well be between skilled and less-skilled. In a sample of dyslexic children, Quadrant Analysis would identify subtypes of dyslexia.

The estimation of the expected PDF was based on the assumption of bivariate normality but, as the marginal distributions of the data were not normally distributed, the joint distribution cannot have been truly bivariate normal, so the assumption was wrong. For this reason, one should not take the attribution of *p*-values in this case too seriously. The assumption of bivariate normality places a major limitation on this approach to selecting optimal cut-off scores because test data is frequently not normally distributed. Only rarely is test data strictly continuous (the possible test scores used here were restricted to integer values from 0 to 30) and there are often floor or ceiling effects (there is a clear ceiling effect on the non-word reading test used here) and both of these are deviations from a strictly normal distribution. As it stands, this means the method described above for choosing cut-off scores is of limited value but it could be revived if a more realistic method for estimating the expected PDF, taking into account deviations of the marginal distributions from the normal, could be devised.

In summary, Quadrant Analysis involves a procrustean imposition of the individual researcher's will on the data and, unsurprisingly, this fails to achieve consensus or validity. For the same reasons, it has no value in identifying taxons. Quadrant Analysis could be improved if there were some explicit, non-arbitrary rationale

for setting the value of threshold used and we propose that the location of the greatest difference between the observed and expected PDFs could be used for this purpose. However, the specific method illustrated here depends upon assumptions of normality that are often unrealistic with cognitive test scores. Despite this limitation, the method does provide an explicit rationale for choosing thresholds and we believe it has heuristic value. Any variant of Quadrant Analysis, however, will share two other serious limitations. First, the method will only be able to identify subtypes that lie within a quadrant; other shaped clusters may be missed. Second, the method is difficult to generalize to more than two or three variables. In short, Quadrant Analysis does not meet the needs of the task in hand and cannot easily be fixed to do so. What is needed is a more objective and rigorous way of identifying subtypes or taxons.

12.5 Alternative Approaches

There are several alternative approaches for identifying latent taxonic structure that offer the prospect of being able to define more objective criteria for identifying distinctive profiles of reading development in children and these include Cluster Analysis (Bonafina, Newcorn, McKay, Koda, & Halperin 2000; King, Giess, & Lombardino 2007; Morris et al. 1998), Finite Mixture Modelling (FMM, McLachlan & Basford 1988; McLachlan & Peel 2000) and Taxometric Analysis (Beauchaine 2003; Meehl 1995). In some senses, each of the methods can be considered as cluster analyses, and Finite Mixture Models often are often classified as such. However, there seems to us to be an important distinction between methods that rely on the pairwise measures of similarity or distance between observations (which we call cluster analysis) and those which generate an explicit parametric model of the data (FMM). The Taxometric approach has its own distinctive history and philosophy of use and, in consequence, deserves separate consideration. One thing that each of these methods has in common, however, and which gives them a considerable advantage over Quadrant Analysis, is that they are all readily generalizable to cases where the subtypes are distinguished by more than two observed variables.

12.5.1 Cluster Analysis

Cluster analysis is a disparate family of methods that identifies groupings in multivariate data sets based on some statistical measure of distance or similarity between observations. There are three main families of cluster analysis: hierarchical clustering, k-means clustering and density based clustering and each of these comes in multiple variations. Hierarchical cluster analysis is particularly fecund and offers a wide choice of similarity (or distance) measures and methods of agglomeration.

For example, SPSS (ver21) offers 37 measures of distance and 7 methods of agglomeration giving 259 possible combinations. Although the choice of distance measure is usually constrained by the type of data being analysed, choosing the appropriate method of agglomeration is more challenging. This abundance would not matter if the methods tended to converge on a consistent result but this is frequently not the case, which makes the decision about which clustering method to use critical.

Both hierarchical and k-means cluster analysis share a common problem. That is, how many clusters should there be? There are multiple methods to help one choose the correct number of clusters including the Akaike Information, Bayes Information, the Calinski-Harabasz and the Davies-Bouldin criteria, and the Silhouette and Gap tests. Unfortunately, these methods often disagree and the question of how many clusters to extract remains unresolved. The third approach to cluster analysis is density-based clustering and unlike the other clustering methods, the number of clusters emerges from the analysis and is not directly pre-determined by the user. However, the user is required to specify other values (critical distance and the minimum cluster size) which, effectively determine the number of clusters so there is no avoiding the issue.

Perhaps surprisingly, cluster analysis has been little used in dyslexia and reading research (Bonafina et al. 2000; King et al. 2007; Morris et al. 1998). Each of these studies primarily addressed the issue of subtypes of children with reading disability and so, unlike Talcott et al. (2002), they did not use a representative sample of all children and only King et al. (2007) compared children with and without reading disability. Each of these studies used k-means clustering (or a variant of it) either alone or with other clustering methods but in other ways (the cognitive measures used and the populations sampled) the studies were very different. Consequently, the character and number of clusters identified were inconsistent. The most sophisticated of these studies King et al. (2007) used cognitive measures derived from theory, state-of-the art criteria for selecting the correct number of clusters and bootstrap sampling to ensure the reliability of the clusters. They found that children without reading disability did not form clusters but that those with reading disability clustered into four groups, consistent with the double deficit hypothesis. As this conclusion was derived from a relatively small sample of 93 children with reading disability, driven by a particular theoretical perspective, the conclusions cannot be considered definitive. Nevertheless, this study, makes a very important contribution to the literature and shows the potential value of cluster analysis in this field of research.

12.5.2 Finite Mixture Modeling

Finite Mixture Modeling (FMM) is a parametric method for identifying clusters in a data set and aims to find the k multivariate PDFs that best account for the observed data. Typically, the PDFs are Gaussian, hence the alternative name for this

approach, Gaussian Mixture Modelling, but other probability distributions can be used if desired. One specific method of note is Latent Class Analysis (LCA) which is a special case of FMM in which the observed variables within each class are uncorrelated.

In conventional FMM, the best fitting combination of PDFs are identified using an expectation maximization algorithm which is equivalent to a maximum likelihood estimate of the parameters of the model. More recently, Bayesian approaches have been introduced that have advantages in robustness and stability of the models and which produce probability distributions of the parameters of the model. In both cases, and like Hierarchical and k-means clustering, the user needs to specify the number of clusters in advance. Unlike hierarchical and k-means clustering, which allocate each observation to a single cluster (“hard” clustering), each individual observation will have a given probability of belong to each of the k clusters defined by the cluster’s PDF (“soft” clustering). Individual observations are allocated to the cluster that has the maximum probability for that case.

As far as we are aware, FMM has not been used to identify subtypes of reading disability or to separate typical from atypical readers. As FMM has some advantages over other forms of cluster analysis, this is an omission that should be corrected.

12.5.3 *Taxometric Analysis*

The final approach to identifying subtypes in dyslexia that we will consider here is taxometric analysis. Taxometrics is a term used to describe a family of methods developed by Paul Meehl and colleagues for determining whether a multivariate data set consists of a latent taxonic structure or not (Beauchaine 2007; Meehl 1995; Ruscio, Haslam, & Ruscio 2006). Taxometrics consists of several distinct methods known as “*coherent cut kinetics*”, that each seek to identify abrupt discontinuities in what appear to be continuous parameters of the measures that distinguish putative members of the taxon in question from non-taxon members. These key measures are referred to as “indicator” variables and the existence of a discontinuity between taxon and non-taxon group members on the indicator variables is taken as evidence for taxonic structure. There are five distinct methods known as MAXSLOPE, MAMBAC, L-Mode, MAXCOV AND MAXEIG that each look for a discontinuity in a different parameter (local regression slope, local mean, latent factor, covariance and eigenvalues, respectively). Taxometrics places a strong emphasis on converging evidence from across these different methods.

Taxometrics has many advantages over other approaches in that it is objective, quantifiable and uses convergent evidence to establish taxonic structure. However, large sample sizes (average size ~600) and large effect sizes (Cohen’s $d > 2$) are needed. Taxometric methods are insensitive in cases where the taxon makes up only a small proportion of the total sample (<15%) and are susceptible to sampling bias and distributional skew in the indicator variables. In addition, the methods do not work well when there are substantial within-group correlations ($r > 0.4$) between

the indicator variables. Despite these limitations, taxometric methods have made significant contributions to the classification of many adult mental health disorders and have been notably successful in delineating the relationship between personality and adult psychopathology (see Chap. 10, Ruscio et al. 2006).

Only one study has used this approach with dyslexia (O'Brien, Wolf, & Lovett 2012). Using a large sample of 671 children with severe reading disorders aged 6–8 years old assessed on a range MAMBAX, MAXEIG and L-mode (Ruscio et al. 2006). They concluded that there was evidence of two taxa of dyslexia, those with and without phonological deficits. This approach appears most promising and merits replication and extension.

12.6 Discussion

There are two important and related questions that we address here. First, do children with dyslexia show fundamental and enduring non-arbitrary and objective differences from children with typical reading profiles? Second, is dyslexia a single condition or does it consist of multiple subtypes, each differing in some fundamental and enduring, non-arbitrary way from the others. In the language of taxometrics, these questions are whether the variation in developmental reading profiles can best be considered as dimensional or taxonic. Our objective was not to answer these questions, but to consider ways in which these question might be answered.

We showed that the common practice of performance thresholds on one or more measures of cognitive ability to delineate dyslexics from the typical readers, a method we refer to as Quadrant Analysis, can never hope to answer this question. Quadrant Analysis, as the research literature attests, is essentially arbitrary and will identify subtypes of readers regardless of whether they exist in any fundamental or objective sense. As such, it simply will not serve to answer the question as to whether we are dealing with dimensions or taxons.

Despite this, the temptations of simplicity of concept and ease of use make it likely that Quadrant Analysis will continue to be used. Acknowledging this, we proposed a modification to Quadrant Analysis that makes the choice of threshold less arbitrary and which might prove useful on occasion. We do not claim, however, that this will overcome all the problems of Quadrant Analysis, let alone resolve the “*category or dimension?*” issue.

Instead, we believe that alternative approaches, including cluster analysis, FMM and taxometrics will prove more useful. Unlike Quadrant Analysis, each of these approaches can be readily adapted to more than two observed variables, and are probably more powerful when used this way. However, these methods are not panaceas and each has limitations and presents the potential user with their own unique challenges. With all such methods, the output depends upon the input. The choice of observed measures, indicator measures in the parlance of taxometrics, is particularly critical. Measures that show some ability to discriminate between the putative subtypes should be preferred and the inclusion of too many

irrelevant measures can obscure real differences. Contrariwise, too many highly correlated variables, whether they discriminate individually or not, simply increases redundancy in the model and can impair the ability of the statistical algorithms to reach a stable solution. The sampling strategy is also critical. Choosing a representative sample of learner readers might be useful for answering the “*Is dyslexia a taxon?*” question, but would be of little use in identifying subtypes of dyslexia as too few individuals of each subtype would be present in the sample. To address this question, a representative sample of problem readers would be more useful. Whichever question is addressed, large samples sizes of several hundred cases will be required to give reliable results.

To date, attempts to use these methods with reading profiles have been few and far between with a mere handful of studies available in the published literature (Bonafina et al. 2000; King et al. 2007; Morris et al. 1998; O’Brien et al. 2012). This is a shame but one that could be corrected quite easily. There are multiple databases of problem readers and representative samples of learner readers that could be used. There are also multiple theories about the nature of dyslexia to guide the choice of indicator variables. This being so, the application of cluster analysis, FMM and taxometrics to these important questions should be straightforward.

No matter what methods we use, however, we should not expect a rapid resolution to the taxonomy of dyslexia question. The much-delayed publication of DSM-V (American Psychiatric Association 2013), where the “*category or dimension?*” issue was the focus of debate around several mental disorders, does not provide grounds for optimism. In none of these cases can the issue be considered closed, including those where the evidence base from cluster analysis and taxometrics is much better established than it is with dyslexia (e.g., schizophrenia). As for dyslexia itself, the wide range of disparate views, strongly held opinions and absence of evidence one way or the other makes consensus seem a distant destination. Nevertheless, the accumulation of evidence derived from statistical tools specifically designed to address the “*category or dimension?*” question seems to us to be a good place to start.

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Chapter 13

Procedural Learning, Dyslexia and Delayed Neural Commitment



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Abstract In this chapter we address the underlying causes of dyslexia, the developmental learning disability, rather than reading disability. In our breakthrough research over 25 years ago we demonstrated that the process of skill automatization of skill was a problem for dyslexic children not only in reading-related skills but also in a range of other skills not obviously linked to reading. In this chapter we report the subsequent development of this framework, first with identification of the cerebellum as a key structure in the automatization deficits, then a “neural circuit” analysis suggesting that many developmental disorders are related to proceduralization problems, with dyslexia associated with specific difficulties in the language/cerebellar procedural circuits. Here we bring this research to the present day, extending the analysis to consider how networks are grown from birth onwards, and introduce the concept of Delayed Neural Commitment (DNC) as a powerful explanatory developmental framework, proposing that dyslexic children are slower to build the connectivity networks on which reading acquisition and fluency depend. This analysis links back to the development of executive function and language networks, and is consistent with all the major theories of dyslexia. We hope that DNC will provide a fruitful and integrative framework for further theoretical and applied research.

Keywords Automaticity · Cerebellum · Declarative · Procedural · Neural commitment · Positive dyslexia

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13.1 Introduction

In this chapter we make the key distinction between two, often confused, approaches to dyslexia – dyslexia as a learning difference, and dyslexia as a reading disability. We first consider briefly the history of dyslexia research, highlighting where these two approaches began to diverge, and then take the road less traveled, ignoring reading and attempting to determine the underlying cause of dyslexia as a learning difference. Since our work over the past 25 years has, uniquely, been targeted on the learning differences in dyslexia, this provides us with the opportunity to provide the historical “story”, in terms of automatization deficit (Nicolson & Fawcett 1990), cerebellar deficit (Nicolson, Fawcett, & Dean 2001), procedural learning deficit (Nicolson & Fawcett 2007), together with our latest synthesis in terms of Delayed Neural Commitment.

13.1.1 *Developmental Dyslexia: A Brief History*

The first systematic approach to identification and support of dyslexia was by the neurologist, Samuel Orton (1937), who believed that the underlying problem was visual. The first international approach to dyslexia resulted in the classic definition by the World Federation of Neurology in 1968 – “*a disorder in children who, despite conventional classroom experience, fail to attain the language skills of reading, writing and spelling commensurate with their intellectual abilities*”. In the 1970s developmental disabilities were considered in terms of commonalities rather than differences. In the UK the standard diagnosis was in terms of minimal brain dysfunction (Wender 1978) or “soft neurological signs” (Touwen & Sporrel 1979). This definition was used as a catch-all term for childhood learning difficulties ranging from clumsiness to speech problems.

For dyslexia, difficulties in motor coordination were long established. Young dyslexic children up to the age of 8 have atrocious handwriting and difficulty in tying shoelaces (Miles 1993); and in speed of tapping, heel-toe placement, rapid successive finger opposition and accuracy in copying (Denckla 1985; Rudel 1985). In an extensive longitudinal study, the British Births Cohort study, (Haslum 1989) examined aspects of health in a cohort of 12,905 children at each age. Two motor skills tasks emerged among the 6 variables significantly associated with dyslexia at age 10, namely failure to throw a ball up, clap several times and catch the ball, and failure to walk backwards in a straight line for six steps. Interestingly, a recent meta-analysis of the incidence of motor difficulties in SLI, ASD, DCD and typical development (Leonard & Hill 2014) identified 43 studies, many of which highlighted a significant relationship between motor skills and the development of social cognition, language and social interactions, thereby highlighting the pervasive role of motor skills in non-physical domains.

There was also a range of approaches to visual problems in dyslexia. The Orton-Gillingham remediation programme (Gillingham & Stillman 1960; Orton 1966) was designed to scaffold the links between vision, letter knowledge and sensori-motor processing by means of multi-sensory stimulation. More recently, the magnocellular deficit hypotheses hold that problems in dyslexia are attributable to abnormal sensory pathways in vision (Stein & Walsh 1997) or audition (Tallal 1993).

By contrast, Vellutino (1979) demonstrated that, although dyslexic children did have problems in combining visual and auditory processing (cross-modality learning) these occurred specifically when verbal processing was required. He claimed that the difficulties were attributable to verbal working memory limitations, which he claimed to be a linguistic system.

Nonetheless, by the mid 1980s there was actually a reasonable consensus as to dyslexia. The case was put well in the seminal book by Miles (1993) “Dyslexia, the pattern of difficulties”, in which he identified a range of signs that were not related to reading and overlapped with other learning disabilities. It was known, even in the late 1980s, that there was great comorbidity between dyslexia and ADHD (Gilger, Pennington, & DeFries 1992). The overlaps were well-represented by the first coherent description of the phonological deficit hypothesis (Stanovich 1988) – the “phonological-core variable-difference” model for dyslexia – in which he proposed that children with dyslexia suffered from a specific deficit in phonological skills, whereas as one moves down the IQ continuum towards “garden variety poor readers”, deficits in phonological processing will remain, but the specificity will diminish, with deficits showing up in more and more skills, even those not related to reading. This captured the wisdom of the earlier years in terms of the “soft neurological signs” being associated with a range of developmental disabilities, but put phonology at the heart.

However, in the mid-1980s onwards a multi-disciplinary, innovative initiative was devised that transformed the dyslexia landscape. An alliance of phonological theorists, psycholinguists and educationalists in the USA put forward the radical view that dyslexia was a language difficulty attributable to phonological weakness, leading in turn to problems in reading unless a dedicated phonologically based intervention program was administered. This multi-disciplinary consensus view led to the relabeling of dyslexia as “Reading Disability” (RD) to highlight the core problem; RD was given the status of a learning disability, and dyslexic children were therefore entitled to a range of support. The National Institute of Child Health and Human Development (NICHD) funded an ambitious multi-site research program (which continues to the present day) into the causes and remediation of RD. Unfortunately the interventions proved to be very much less effective than hoped (Camilli, Wolfe, & Lee Smith 2006; Hammill & Swanson 2006; Stuebing, Barth, Cirino, Francis, & Fletcher 2008), and the scientific research aimed at establishing the causes of the fundamental problems in the “phonological module” proved to be frustratingly inconclusive (Blomert & Willems 2010; Nicolson & Fawcett 1995; Ramus & Szenkovits 2008).

13.1.2 Specific Learning Difficulties

Our brief history highlights the rise of RD in the late 1980s, and this academic / political initiative coincided with the rapid differentiation of the learning disabilities into separate pressure groups and representative organizations for dyslexia, attention-deficit/hyperactivity disorder (ADHD), developmental coordination disorder (DCD), specific language impairment (SLI) and autism spectrum disorders. As with progress in any area, the developments led to specialization in specific areas – in the case of dyslexia an intensive investigation of the phonological basis of dyslexia and of the effectiveness of different approaches to reading intervention – at the expense of other approaches. The two neglected approaches we highlight in this chapter are the underlying causes of dyslexia, and the links between dyslexia and other learning disabilities. We start by providing a brief overview of theoretical approaches to dyslexics.

13.1.3 Theories of Dyslexia

There is a wide range of theories of dyslexia, and this is not the place to provide a full analysis, but we can outline here some of the more important theories that contribute to a plausible developmental framework – see Démonet, Taylor, and Chaix (2004) and Nicolson and Fawcett (2008) for a full analysis. Phonology-centric overviews are provided in Peterson and Pennington (2012) and Vellutino, Fletcher, Snowling, and Scanlon (2004). Here we adopt the well-known levels of explanation framework (Morton & Frith 1995): considering behavior (for example reading), cognition (such as memory, language and learning) and the brain (neural structures and processes).

13.1.3.1 Cognitive Level

Many theories have tried to explain the underlying cause of the reading difficulty, with the dominant theory the phonological deficit hypothesis (Stanovich 1988). This proposes that difficulties are due to an inability to break down words into their constituent sounds, leading to problems in segmentation and blending, key skills underlying early reading and spelling.

Despite the extensive research base on phonological deficits, there are many other plausible theories that can inform our understanding of dyslexia, all with valid contributions in terms of both theory and practice.

The double deficit hypothesis (Wolf & Bowers 1999) goes beyond phonology in identifying speed deficits in addition to the phonological deficits, with the poorest outcomes for those children showing both problems in speed and in phonology. Many theorists have proposed that this is a subset of phonological deficits, and include working memory and processing speed within the phonological framework

(Vellutino et al. 2004). An alternative causal hypothesis linked to speech rhythm deficits (Goswami et al. 2002) proposes that phonological problems are based on difficulty in identifying syllable boundaries in the onset of the amplitude envelope which underlies prosody.

There are a number of theories linked to attention. The visuo-spatial attention deficit hypothesis (Facoetti, Lorusso, Paganoni, Umilta, & Mascetti 2003) suggests that problems in switching attention underlie reading deficits, that is “covert orienting”. In order to read fluently, it is necessary to fixate on one word while preparing covertly to switch to the next words. An alternative theory (Bosse, Tainturier, & Valdois 2007), suggests problems are attributable to reduced visual attention in dyslexia.

Visual hypotheses for dyslexia are not new, and reflect an established range of problems in fixation, stability, saccadic accuracy, and eye movement differences (Eden, Stein, Wood, & Wood 1994; Stein 1989), with more recent evidence of effects of visual crowding and larger fonts (Moores, Cassim, & Talcott 2011; Schneps, Thomson, Chen, Sonnert, & Pomplun 2013). In terms of other modalities, a similar mechanism to Specific Language Impairment has been proposed for rapid auditory processing in dyslexia (Tallal 1993) with both frameworks reflecting magnocellular deficits in the brain.

Cross-modality integration of auditory and visual information was the original discovery of Vellutino (1979) and was revisited by Blomert and his colleagues (Froyen, Willems, & Blomert 2011), who showed that problems lie in integrating visual letters with their sounds. Interestingly, unlike Vellutino, who attributed this to language problems, Blomert and colleagues attributed the difficulties to automatization difficulties, directly consistent with our automatization deficit hypothesis (Nicolson & Fawcett 1990), which we discuss below.

13.1.3.2 Brain Level

These theories explain deficits in terms of the underlying structures, with the most well-established the magnocellular deficit reflecting sensory processing, although evidence can be inconsistent. Visual magnocellular deficits are measured by tasks such as the detection of low contrast moving visual gratings (Eden et al. 1996). By contrast, Tallal and colleagues have identified problems with rapid auditory temporal processing. In an attempt to consolidate these findings has led Stein (Stein 2001) to propose a pan-sensori-motor deficit in the magnocellular systems for audition, vision and action.

Our cerebellar deficit hypothesis (Nicolson & Fawcett 1995; Nicolson et al. 2001) argues that all the cognitive deficit theories, (automatization deficit, phonological deficit and speed deficit) can be explained in terms of the cerebellar brain structures and mechanisms underlying learning. We revisit below the strengths and weaknesses of the cerebellar deficit, and this leads on to our Procedural Learning Deficit framework in the following section.

13.1.3.3 The Genetic Level

There is strong evidence for a genetic component in dyslexia, and this gives the family studies their power, given that 50% of children with a dyslexic parent or sibling will also develop dyslexia (Pennington 1991). However, despite extensive progress in genetic research (Carrion-Castillo, Franke, & Fisher 2013), the underlying genetic basis of dyslexia has been tantalisingly elusive. This seems inevitable, because studies are based on the reading symptoms of dyslexia across a range of complex manifestations. Nevertheless, genetic analyzes have the potential to provide converging evidence on a range of theories, not least on differences in cell migration during the early formation of the brain in fetus.

We set the scene by re-presenting our original research on learning differences in dyslexia.

13.1.4 *Nicolson and Fawcett Research Phase 1: Dyslexia and Learning*

Our expertise is in human learning, and we were convinced that analysis of the learning processes in dyslexia was a pre-requisite for understanding the underlying cause of the “specific learning difficulties”. In our view the processes of learning to read are similar to the processes of learning to play chess, learning to type, or learning to drive – a combination of cognitive and physical skills that are slowly built up by experience. From a learning perspective, there is nothing special about reading. It is not wired into the brain like language might be, there are no special reading-acquisition-devices in our heads. Historically there have been many illiterate societies. Our general initial hypothesis was that, given the commonality between motor learning and cognitive learning, we proposed that learning differences should be evident even for motor tasks.

This learning framework led directly to what remains as the simplest and most complete framework for understanding dyslexia – the Dyslexic Automatization Deficit hypothesis (Nicolson & Fawcett 1990). The hypothesis states that dyslexic children have problems making skills automatic and therefore need to “consciously compensate” even for simple skills. So if a task is not too difficult, dyslexic children might well be able to perform within the normal range, but they would be doing it by explicitly concentrating hard on it, whereas a non-dyslexic child would just “download” the task to some unconscious automatic mechanism.

The logic behind our initial research investigation was the simple but counter-intuitive Popperian approach. In order to discriminate between theories, one should choose a domain where they make different predictions – ideally domains where some theories predict there should be no deficit. This was a departure from established wisdom in the field. For many RD researchers it seemed close to heresy to look outside of the reading domain when trying to find the cause of reading disability.

13.1.4.1 Study 1: Balance and Dyslexia

Our key study was on the gross motor skill of balance, using the “one foot in front of the other” Romberg task. The critical test for automaticity is the “dual task” setup, where the participant not only does the primary task (balance) but also has to undertake another (secondary) task that takes up controlled processing resources. If the primary task is achieved automatically, it should be possible to perform the dual task with little or no interference, whereas if it too requires controlled processing there will be a substantial decrement.

Our participants – 13 year old dyslexic and control adolescents matched for IQ and age – stood in the Romberg position for 30s and we measured the number of wobbles. For the single task condition (just standing) there was no difference between the two groups. Participants made very few wobbles – as predicted from the literature. But for the dual task (standing and counting) there was a dissociation, even though the counting task was individually calibrated to be easily accomplished (when not standing balancing) for each participant. The control participants’ balance was unimpaired, whereas the dyslexic participants wobbled a lot more.

In short, under optimal conditions the dyslexic children could balance as well as controls. But the controls were balancing automatically, whereas the dyslexic children were not. There seemed to be automatization problems even for balance. The same results were obtained for different balance tasks (Fawcett & Nicolson 1992) in that the dyslexic children had balance deficits when blindfolded and thus unable to use visual cues.

This was strong evidence for an automatization deficit account. It was completely contrary to the predictions of the Phonological Deficit Hypothesis – and indeed counter to the predictions of almost every theory we have just outlined. In our view, therefore, this study by itself falsified all the competing frameworks, although other theorists queried or dismissed this inconvenient finding as being irrelevant to reading.

In fact, there have been several replications of the findings. The consensus of these replications was that – given a sensitive age-appropriate design – many dyslexic children, at least half – do show balance difficulties compared with children of the same age (Ramus, Pidgeon, & Frith 2003) but that these are not causal with regard to the reading difficulties (Rochelle & Talcott 2006).

This initial study set the scene for our research programme.

13.1.5 Nicolson and Fawcett Phase 2: Cerebellar Deficit Theory

We had actually completed the above research by the mid 1990s, and we realized that to further investigate the learning processes we needed to look at the underlying neuroscience. The major change in neuroscientific knowledge in the early 1990s was

realization that the cerebellum was not just a motor skill coordinator but actually an “all skills coordinator and learner”, and in particular the emerging evidence that the cerebellum was centrally involved in language fluency. This formed the basis for our cerebellar deficit framework.

13.1.5.1 Theory: The Cerebellum

The cerebellum is one of the major organs in the brain, with direct neural connections to almost all brain regions and all the body, containing more than half the brain’s neurons. It has long been known to be centrally involved in fluency of skill execution, but brain imaging revealed its role in processing tasks from taste to speech to reading to automaticity. The first claim of cerebellar involvement in cognition was made in 1989: “. . . *the 2-way connections linking the cerebellum to Broca’s area [a pre-frontal region known to be centrally involved in speech production] make it possible for it to improve language dexterity, which combines motor and mental skills*” (Leiner, Leiner, & Dow 1989).

Their hypothesis – in terms of connectivity and function – has been completely vindicated by subsequent cognitive neuroscience research (Balsters, Whelan, Robertson, & Ramnani 2013; Marien et al. 2014; Strick, Dum, & Fiez 2009). An influential summary by Desmond and Fiez (1998) confirmed the multiple skills involving cerebellar activation, from classical conditioning and pursuit learning to the core “cognitive” skills of explicit memory retrieval, language/verbal working memory, verbal working memory, and sequence learning. Furthermore, the nature of the activation was distributed, with different regions involved in different tasks, but with also overlapping distributions of some skills.

In view of the phonological deficit account, and its extension to include both verbal working memory and speed of processing, the cerebellar link to the working memory, language and reading and both declarative and procedural knowledge is particularly suggestive.

13.1.5.2 Dyslexia: An Ontogenetic Causal Chain

In a range of studies we established clear, direct and indirect evidence of cerebellar deficits associated with dyslexia – functional imaging (Nicolson et al. 1999); anatomy (Finch, Nicolson, & Fawcett 2002); prism adaptation (Brookes, Nicolson, & Fawcett 2007); eye blink conditioning (Nicolson, Daum, Schugens, Fawcett, & Schulz 2002), and cerebellar signs (Fawcett & Nicolson 1999).

This led to a significant achievement – the creation of an “ontogenetic” (developmental) model of how the reading, writing and spelling deficits arise (Nicolson et al. 2001). In Fig. 13.1 development moves from birth through 5–8 years from left to right. The life journey starts with a cerebellar impairment – possibly attributable to abnormal brain neural migration processes – from birth (or pre-birth). We also note that it is possible that the other parts of the brain – the cortico-cerebellar loops – are involved.

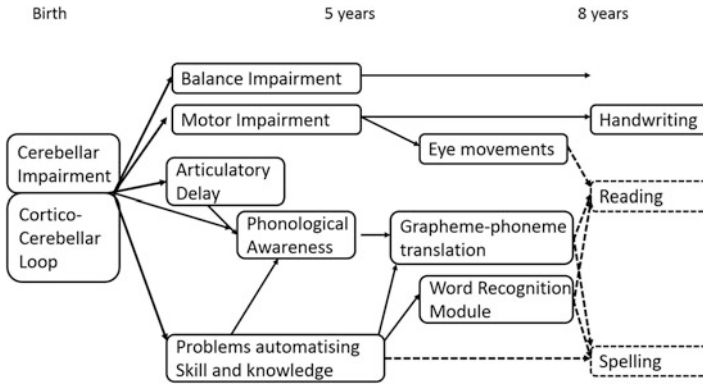


Fig. 13.1 Dyslexia: an ontogenetic causal chain

Moving from left to right, for a 3 year old child, cerebellar impairment will frequently lead to balance delays (depending upon the specific regions of the cerebellum and its neural connections that are affected) and also to motor skill delays, articulatory delay (articulation is a motor skill) and also automatization deficit. In the original article, we left open the form of the differences before the age of 5 years, preferring to focus on the ages for which there was solid data. The Delayed Neural Commitment framework (see later) provides clear predictions for pre-school differences, indicating that differences will occur where typically developing children have constructed neural networks that support more rapid processing (or more integrated processing) of information. By contrast, dyslexic children will be slower to develop these networks and will therefore show developmental delays in a range of pre-school skills from language perception to executive function. Differences would therefore be revealed through analyzes of white matter and functional connectivity.

For the early school years, balance impairment will have relatively little impact on school academic performance, although sporting skills might well be impaired. More general motor skill impairment would not be too problematic, though it might lead to impaired eye movement control (which would impact on reading), and would have a direct effect on handwriting skill. Because expressive speech and receptive speech – phonology – are directly linked (Gervain & Mehler 2010; Price 2012), articulatory delay would be hypothesized to lead to phonological differences. Inefficient automatization would lead to problems with identifying the individual letters and the spellings. Furthermore, because of the central role of the cerebellum in speech internalization (Ito 2008) and in verbal working memory (Ben-Yehudah, Guediche, & Fiez 2007; Hayter, Langdon, & Ramnani 2007), working memory would also be impaired.

As is made clear in the figure, this combination of impairments would lead to significant difficulties in learning to read, both for reading and for spelling, but actually a different set of problems. This is consistent with the findings of different genetic underpinnings to phonological and orthographic (that is, the spelling) problems (Olson 2002).

We remain particularly proud of this chart. It provides a principled explanation of the three criterial difficulties for dyslexic children (reading, writing and spelling). And it explains why there are phonological deficits and orthographic deficits. And it explains why there are various secondary symptoms not related to literacy. It provides a principled method for screening for dyslexia BEFORE a child fails to learn to read. For the first time in any discipline, it provided a link between development, the brain and school achievement, explaining our highest level cognitive skills of literacy in terms of the underlying developmental processes.

The framework has been supported by subsequent research. An early meta-analysis of imaging studies (Eckert et al. 2003) concluded “*The cerebellum is one of the most consistent locations for structural differences between dyslexic and control participants in imaging studies. This study may be the first to show that anomalies in a cerebellar-frontal circuit are associated with rapid automatic naming and the double-deficit subtype of dyslexia*”.

More recently Pernet, Poline, Demonet, and Rousselet (2009) concluded “*The right cerebellar declive and the right lentiform nucleus were the two areas that significantly differed the most between groups with 100% of the dyslexic subjects (N = 38) falling outside of the control group (N = 39) 95% confidence interval boundaries. . . . Conclusion: These results provide evidence for the existence of various subtypes of dyslexia characterized by different brain phenotypes. In addition, behavioral analyzes suggest that these brain phenotypes relate to different deficits of automatization of language-based processes such as grapheme/phoneme correspondence and/or rapid access to lexicon entries*”.

There seems little doubt that the cerebellum is somehow involved in the automatization deficits suffered by many dyslexic children. However, the framework still lacked specificity. After all, the cerebellum contains over half the brain’s neurons, it is implicated in almost all the brain’s intrinsic connectivity networks (Bernard et al. 2012; Buckner, Krienen, Castellanos, Diaz, & Yeo 2011). Half of the critics of the cerebellar deficit framework claimed that it was inconceivable that the cerebellum was centrally involved in reading difficulties, and the other half said that the framework had no greater specificity than suggesting that somehow the brain was involved!

In addressing the issue of specificity, we started with a further analysis of the learning processes in dyslexia. Here we go back to first principles, relating the discoveries relating to the different types of learning available to the human brain.

13.1.6 Three Fundamental Forms of Human Learning

A major distinction in human memory (Tulving 1972) is between procedural memory (how to undertake a skill), semantic memory (general knowledge about the world) and episodic memory (specific knowledge about events that have happened to oneself). Both episodic knowledge and semantic knowledge are available to conscious thought and are considered forms of declarative knowledge (Cohen &

Squire 1980). The distinction between declarative memory and procedural memory is considered the major distinction between knowledge types, and corresponds to different brain circuitry (Squire 1987), with the hippocampus being the hub for declarative processing and the basal ganglia and cerebellum forming the hubs for procedural processing. More recent analyzes have identified a range of subtypes of procedural processing with Thompson (2005) specifying five basic types (skills, categories, priming, associative and habituation) together with episodic and semantic memory for the declarative category.

The above analysis highlights one of the fundamental distinctions (Schneider & Shiffrin 1977) in cognitive psychology, that between Controlled Processing, which requires attentional control, uses up working memory capacity and is often serial, and Automatic Processing, which, once learned in long-term memory, operates independently of the participant's control and uses no working memory resources. This distinction is central to the major model of cognitive learning (Anderson 1983), which has been shown to apply not only to acquisition of physical skills but also mental skills. It distinguishes between three stages of skill acquisition – the declarative stage where one works out what to do, the procedural stage where one works out how to do it and the tuning stage where one gradually becomes more expert and automatic.

Putting together the neuroscience descriptions with the learning processes, declarative processing is the means by which controlled processing works – it uses conscious, attentional resources (often, but not always language-based) to work out a “high level” method of doing the task – “take the gearstick, move it to second” etc. It has the advantage of flexibility and variety, but has two major drawbacks: first it is far too slow and second, it clogs up the controlled processing, in that one can only concentrate on one thing at a time.

By contrast, procedural processing is “low level”, fast and takes little up conscious capacity. Basically it is a series of precisely timed instructions (often in parallel) to the muscles involved – “*Muscle A contract by amount B at time C for duration D...*” and is not available to conscious awareness.

A key difficulty therefore is “proceduralization” – going from the declarative stage to the procedural stage. Whereas declarative processing is easy to set up (but easily forgotten), procedural processing takes extensive practice over many days, as revealed by Shiffrin and Schneider.

This framework is now supported by the cognitive neuroscience of learning. In fact we distinguish three fundamentally different types: procedural learning, which includes three sub-types (statistical learning, reward-based learning and trial-and-error learning), which is basically for the acquisition of skills; declarative learning (which is for knowledge); and neural circuit building (which is the basis of child development, changing the architecture of the information processing system in Piagetian terms, or developing new intrinsic connectivity networks in current terminology). The three procedural forms are a legacy of the many million years old vertebrate machinery, the “primitive” types that we have inherited from the first vertebrates – fish, crocodiles, lizards. Declarative learning is much more recent, a human speciality, more dependent on thought. And neural circuit building is

the unsung and under-explored basis for human development (and reading). As with everything else in the brain, these learning systems combine (or sometimes compete). We take them in turn.

13.1.6.1 Procedural Learning

Statistical learning (also known as unsupervised learning) occurs automatically via repeated exposure. It is a consequence of the ability of networks of neurons to “self-organise”, that is automatically to adjust the “weights” (that is the connectivities) between elements to identify objects despite changes in size, angle, lighting etc. in vision, and to identify phonemes despite changes in pitch, emphasis, loudness for speech, and to automatically link visual objects with simultaneously occurring auditory information.

In reinforcement learning an action results in a reward (and hence is “reinforced” in behaviorist terms). A reward is anything that improves the animal’s state – food when hungry, water when thirsty, and so on – the carrot rather than the stick. Any organism that can optimize its ratio of reward to effort will have a competitive advantage, and therefore most animals have specialized neural machinery designed to identify just what actions or context (or both) led to the reward. Unlike statistical learning, this machinery is designed to gain the maximum possible out of even a single reward, and vertebrates have specialized neural circuits, running via the basal ganglia, for reward-based processing.

Trial-and-error learning (sometimes known as supervised learning) occurs when the animal knows what it is trying to achieve and gets an “error signal” to indicate how close the action was to achieving the planned outcome – for example, a child trying to imitate an adult’s action or speech. There are two forms, imitation when one is trying to get started and then tuning as the actions get more fluent.

All regions of the brain support statistical learning. Only the basal ganglia support reinforcement-based learning, the success-based learning. However – and this is a crucial point – only the cerebellum has the circuitry to support trial-and-error learning, where there is a target and an error signal. Consequently, if error-based learning is required, then it is necessary to involve the cerebellum as part of the circuit, along with the other parts of the brain involved. Given that speech, language, eye movements, physical actions and almost all human activity involves error-based learning, it becomes clear why the cerebellum is such a central structure in brain networks, and why it has grown massively (relative to other primates) in humans.

13.1.6.2 Declarative (Consciously Accessible) Learning

Declarative learning is one of the ways that we use our knowledge of the world to improve our memory and performance. The most obvious form of declarative learning is “learning by being told” rather than “learning by doing” or “learning by observing”.

The key structure for declarative learning is the hippocampus, and it is generally considered that, in the same way as the procedural learning circuits need to involve the basal ganglia and/or cerebellum, declarative processing needs to involve the hippocampus.

13.1.6.3 Neural Circuit Building

Automatization tends to build smaller but more efficient systems. By contrast, there is a further level of neural circuit, which Anderson (1983) refers to as the “architecture” of the information processing system – the way that the various parts of neural processing combine to produce a highly efficient system. The immensely influential Piagetian framework (Inhelder & Piaget 1958), which highlights the way that the child builds more and more sophisticated operating systems over a period of 10 or more years, is essentially chronicling the development of more sophisticated intrinsic connectivity networks through maturation and experience. Furthermore, the information processing and cognitive viewpoints that highlight the development of processing efficiency, executive function, working memory and response inhibition (Demetriou et al. 2014).

13.1.7 *Procedural Learning and Dyslexia*

It will be clear from the previous discussion that we had incontrovertible evidence that there were difficulties in skill proceduralisation, and that these were consistent with impaired cerebellar function. Furthermore, cerebellar abnormality is one of the most consistent features of brain imaging studies of dyslexia.

Nonetheless, there is a legitimate question over cause versus correlate. One intriguing possibility is the “innocent bystander” hypothesis put forward by Zeffiro and Eden (2001), namely that the cerebellum was functioning fine, it was just that it was being given impaired data from other sources (such as the magnocellular sensory system). While the imaging data do strongly implicate cerebellar abnormality, there is certainly a possibility that for a subset of dyslexic children the problems might arise elsewhere in the learning circuits involving the cerebellum.

A further topic which has emerged more recently is the acceptance that many brain regions are involved in the acquisition and the execution of cognitive and motor skills, and that therefore it is important to consider the system as a whole, not just parts of it (Doyon & Benali 2005). This neural systems approach formed the basis for our integrative recent procedural learning difficulties framework.

We have introduced the core distinction between procedural learning and declarative learning, with declarative learning directly related to language and thought, with conscious accessibility, but not to motor skills, which are procedural. However, Ullman (2004) radically enhanced this view by demonstrating that in fact the language system also splits into declarative and procedural branches. This integrated

a range of findings in the literature, in particular the fact that there are different types of language skill, some of them explicit, available to conscious introspection, and some procedural and not consciously penetrable.

According to Ullman, the Declarative Memory System involves the “mental encyclopaedia”, through the temporal lobe and hippocampus, storage and use of knowledge of facts and events. It is a part of the “ventral route” anatomically, and has direct conscious access. In contrast, the Procedural Memory System handles the mental grammar, the rules of grammar. It is a network involving the basal ganglia, and specific frontal, parietal and cerebellar structures. It underlies procedural memory, which supports the learning and execution of habit-based language skills, especially those involving sequences. A high proportion of our language is in fact attributable to non-conscious, procedural skills, such as the ability to turn thoughts into words, or to just “know” whether a statement is grammatically correct.

13.1.8 The Procedural Learning Deficit (PLD) Hypothesis

This insight led to directly to our third framework for dyslexia, which is the Procedural Learning Deficit hypothesis (Nicolson & Fawcett 2007). Following Ullman, we speculated that most developmental disorders might be attributable to problems in some form of the Procedural Memory system (Fig. 13.2).

Many developmental disorders are attributable to abnormal function of the Procedural Memory (brain-based) system – we label it the Procedural Learning system, to highlight its role in plasticity as well as memory. Following Ullman, we distinguished two branches of the Procedural Learning systems, the motor

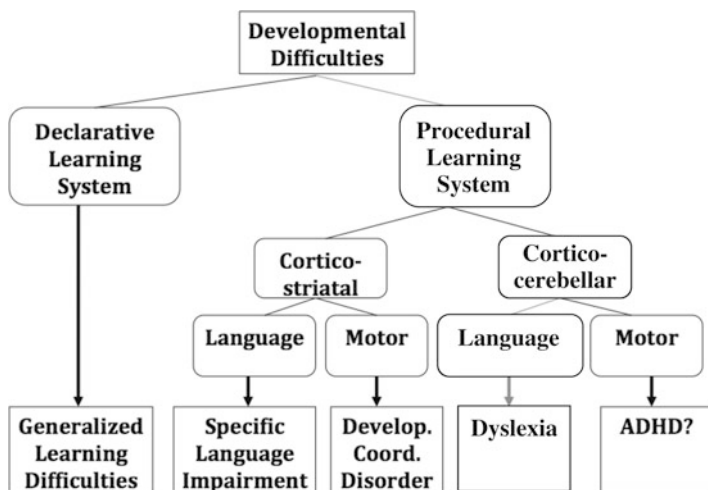


Fig. 13.2 Classification of developmental disorders in terms of declarative and procedural skills

Procedural Learning system and the language Procedural Learning system. Following Doyon and his colleagues (Doyon 2008; Doyon & Ungerleider 2002) we distinguished two further branches, the cortico-cerebellar branch, which relates to skill adaptation and tuning, and the cortico-striatal branch, which relates to skill selection and execution.

Following Ullman's approach we allocated Specific Language Impairment to abnormal function in the language aspects of the cortico-striatal system. Developmental Coordination Disorder (clumsiness) to the motor component of that system. If we turn now to the cortico-cerebellar system, again we can distinguish between language and motor. We assigned the motor cortico-cerebellar system to attention deficit disorder, whereas the language component – the Specific Procedural Learning Difficulty – was assigned to dyslexia. For dyslexia, therefore, we argued that we have Specific Procedural Learning Difficulty – specific to the language-cerebellum, but involving other Procedural Learning components to a greater or lesser degree.

This is in fact a refinement of the classic four levels analysis from behavior – cognition – brain – genetics. Our automatization deficit hypothesis was at the cognitive level, and our cerebellar deficit hypothesis was at the brain level – with the cerebellar deficit hypothesis providing a deeper explanation of the automatization deficit findings presented earlier. The procedural learning deficit hypothesis lies in between the brain and cognition levels, at what we called the neural systems level. Placing the deficit at the level of the circuit rather than the structure directly addresses the “innocent bystander” issue. And we believe that this neural systems level of explanation is a particularly fruitful one that provides the framework for explaining a range of different developmental disorders as I show in the next section.

Taking the Declarative circuit, if there is a problem in declarative learning, this will lead to generalized learning problems, and hence poor reading but with no discrepancy, because there will be poor performance across the board intellectually. So, for us, we would call those generalized learning difficulties.

This new framework not only provides a natural explanation of the well-known phonological deficits, but also incorporates our earlier automaticity and cerebellar theories. This network analysis is well supported by evidence from a range of studies, in particular serial reaction time studies and procedural learning are consistently impaired (Lum, Ullman, & Conti-Ramsden 2013). We have found problems in the consolidation of motor skills in high achieving dyslexic adults (Nicolson, Fawcett, Brookes, & Needle 2010), with even stronger impairment in letters than motor sequences (Gabay, Schiff, & Vakil 2012). A functional co-ordination deficit has also been proposed for dyslexia, based on problems in automatizing a novel synthesis of procedural learning (Lachmann & van Leeuwen 2014). In terms of declarative strengths, Ullman's group have demonstrated a distinctive superior performance on immediate and delayed declarative memory in dyslexia (Hedenius, Ullman, Alm, Jennische, & Persson 2013). This is in itself a highly unusual finding in the literature, and fully consistent with the Procedural Learning Deficit hypothesis.

Reviews of the literature on the other learning disabilities are beyond the scope of this manuscript. Ullman and Pierpont (2005) had previously provided evidence

of procedural learning problems in SLI, and subsequent research established difficulties in implicit learning (Lum, Conti-Ramsden, Morgan, & Ullman 2014).

Evidence relating to the DCD route is surprisingly sparse, given the expected role of the cerebellum in physical coordination. A study investigating prism adaptation in dyslexia established that children with DCD and dyslexia did have difficulties with prism adaptation – a cerebellar task (Brookes et al. 2007), whereas a small study of prism adaptation found differences in some but not all children with DCD (Cantin, Polatajko, Thach, & Jaglal 2007).

There is good evidence for motor skill deficits in ADHD. A recent meta-analysis (Kaiser, Schoemaker, Albaret, & Geuze 2015) concluded that *“More than half of the children with ADHD have difficulties with gross and fine motor skills. The children with ADHD inattentive subtype seem to present more impairment of fine motor skills, slow reaction time, and online motor control during complex tasks”*.

13.2 Development of Dyslexia: The Delayed Neural Commitment Framework

In presenting the first 25 years of our research program on the underlying causes of dyslexia, we have championed the importance of investigating the learning processes in dyslexia, which we characterized as delayed or impaired automatization of skill, leading to difficulties in any task that require speeded processing or multi-tasking. We then identified the cerebellum as a structure likely to be centrally involved in automatization difficulties, and then specified the neural circuits involved in procedural learning, especially for language-related activities, as the likely functional connectivity circuits to be under-performing or under-developed. This represents a significant corpus of coherent research, unsupported by research from others or any funding bodies. It provides explanations at the behavioral level, the cognitive level, the brain level, and (a level between these) the neural circuit level. It provides an outline explanation of how the problems arise from gestation to ten years old, and why there are problems in reading, writing and spelling, and why there are comorbidities with other learning disorders. The framework clearly has the benefit of great generality.

Nonetheless, the framework clearly still lacks specificity, in that it does not explain the heterogeneity in dyslexia, and it still falls short of being able to provide a complete analysis of the development of dyslexia from birth to early school. It falls short of providing an answer to the key question of whether, for a given child, their dyslexia represent a developmental delay or a developmental disorder. The implicit assumption in referring to “disorder” is that there is some underlying brain difference that is intrinsic, and therefore unlikely to be alleviated just by normal maturation and experience. In the case of reading, there has been considerable discussion as to how one might distinguish delay from disorder. A reasonable principle was posited by Bryant and Goswami (1986), who argued that it is good

practice when assessing performance of dyslexic children to have a control group of the same Chronological Age (CA) and also – to control for reading experience – a group of the same Reading Age (RA).

Consider key issues in terms of the development of children with dyslexia, at a stage before the problem has been identified, namely between birth and age 6. Typically, dyslexia is not identified until the age of 8, and by this stage only retrospective accounts of development can be obtained. Our understanding of the early years has been enriched by major European longitudinal studies of children with a family history of dyslexia (Lyytinen et al. 2004; van der Leij et al. 2013), building on earlier work that recognized the need for a full developmental analysis (Goswami 2003; Karmiloff-Smith 1998). Despite the strengths of this longitudinal approach, only those whose parents have a previous diagnosis of dyslexia have contributed, and this inevitably leads to a somewhat atypical sample and the possibility of parental intervention based on an awareness of the difficulties their child will face. Consequently, although an important source of converging evidence, these studies cannot provide a complete theoretical understanding of the area and would benefit from converging evidence from a range of studies.

13.2.1 Two Studies of Development of Dyslexia

It is important to provide here further empirical data to support our theoretical approach, drawn from earlier studies, that can now be integrated more fully into our latest research framework.

13.2.1.1 Study 1: Skill, Development and Dyslexia

In our earlier research it was very clear to us that other research groups could provide strong evidence for the approach they adopted, but they typically failed to stray far from their own areas of interest and expertise in considering the evidence. By contrast, we therefore resolved to adopt an approach that would facilitate an examination of a broad range of deficits and address the effects of maturation. In order to do this, we used a strong cross sectional design based on 6 groups of children, including dyslexic children aged 8, 11 and 15, matched with controls of the same age and intelligence level. This not only allowed for age match comparisons but also for reading age matched comparisons, examining the effects of maturation on skills. A deficit in comparison with younger children of the same reading age suggests a causal impairment, not just a symptom, and a disorder rather than a delay.

We adopted tests designed to give no opportunity for “conscious compensation”, and tapping a range of skills known to be affected in dyslexia – phonological skill, working memory, information processing speed, and motor skill (Fawcett & Nicolson 1995). The data, converted into age equivalents using data from our control group are shown in Fig. 13.2.

As predicted, there were severe problems for most phonological skills, including phonological discrimination and segmentation, apart from nonsense word repetition, a phonology/memory task. Interestingly, disorders were also indicated by articulation speed, which was significantly worse even than the RA controls, as were letter naming and picture naming. Finally, the four motor skill tasks (bead threading, pegboard, balance on one foot and balance on one-foot blindfold) also showed a disorder, with performance worse than the RA controls. See Nicolson and Fawcett (1994) for a detailed analysis.

Finally – and this is crucial for the issue of heterogeneity – we undertook analyzes in terms of individual performance. Most of the dyslexic children are at least 1 standard deviation below their controls on every task in the battery presented, indicating developmental delay (or disorder). However, more encouragingly, the dyslexic children improved steadily with age, suggesting delay rather than outright disorder. These data were cross-sectional, and so for a follow-up study we undertook a longitudinal study.

13.2.1.2 Study 2: Extended Training on a Keyboard Game

The next study examined extended learning on a keyboard-based computer game, with the player moving round the board, using the specified keys. See Fig. 13.3 for the overall results. The dyslexic group were much slower initially, and they took longer to reach maximum speed, which was also slower than controls. This was consistent with the automatization deficit and procedural learning deficit frameworks. Slower learning, impaired final performance on a procedural task.

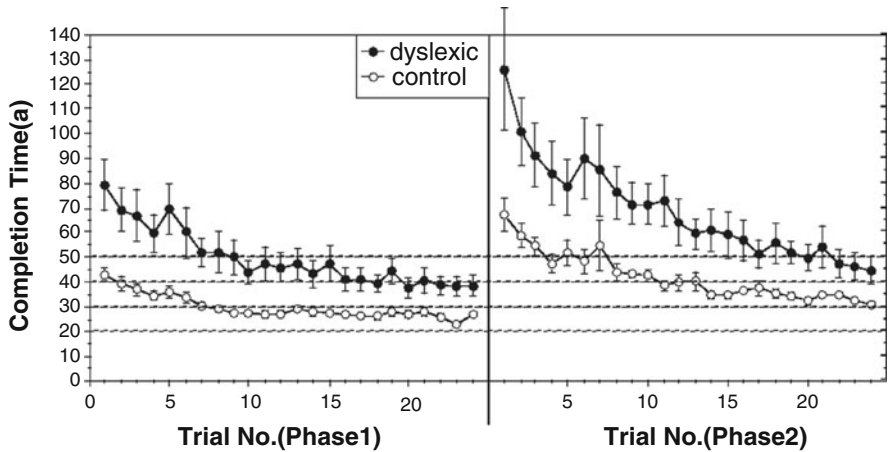


Fig. 13.3 Speed of circuit completion with practice

However, the study provided an important clarification of the framework. A key question here was the potential impact of changing the keys – essentially establishing how difficult it was to unlearn the original mappings. Interestingly, we found that the dyslexic group were more impaired by the change than the controls. Moreover, we found that when we checked their performance 6 months later, after intensive relearning the keys, they were less affected by interference than controls. The dyslexic participants were able to become automatic (showing the expected effects of unlearning and some effects of interference) but the quality of their automaticity remained impaired (as shown by problems in speed and accuracy).

In our view this study is particularly important in the development of neural networks. The result showed that their neural networks were less efficient even when developed under optimal conditions for learning – those of consistent mapping. Perhaps more important still, and an issue we consider below, was the difficulty shown by the dyslexic group in unlearning their previously learned skills.

13.2.1.3 Unlearning: The Issue of “Brown Field” Learning

In architecture there is a fundamental distinction between a “green field” site, where there is no impediment to the building, and a “brown field”, inner city, site where there are already buildings on the site, and it necessary to adapt or demolish them before starting. The same analogy may be applied for human learning. Most learning theories apply to green field learning, that is, learning a new skill for which there are no existing skills that interfere with the learning process. In the real world, most learning is brown field, and we need to adapt or inhibit interfering skills and networks. This is particularly clear in the case of the long road to learning to read fluently, where first a child learns the laborious letter-at-a-time grapheme-to-phoneme translation methods, and then over the course of the next few year has to unlearn these skills in order to achieve the “word as a whole” reading skills.

The issue of how to build up complex skills has been extensively explored in physical skills, for which the “part-whole transfer” approach in skill development (Shea & Morgan 1979) has been found optimal – one needs to ensure that the intermediate skills are practised in the context of the full skill, since otherwise the intermediate skills become encapsulated (Fodor 1983; Pylyshyn 1999), and impossible to integrate with the target skill. We are not aware of any skill building program outside the reading domain that advocates the learning and then the unlearning of an intermediate skill, and so it may be that skill unlearning issue has not been well enough explored in the literature.

Consequently we attempt to characterize the problems that are likely to be encountered by a dyslexic child in building the necessary neural networks. We make minimal theoretical commitments so as to optimize the generality of the analysis. Before turning to this analysis, we outline the developments in infant language development that inspired our work on delayed neural commitment.

13.2.1.4 Infant Speech Development and Neural Commitment

Given the wide range of difficulties that have been identified for dyslexic children in the literature, it would not be surprising if these showed up first in the acquisition of language in the early years. However, most dyslexic children can achieve this highly challenging task with minimal difficulty and only subtle differences in their language development. Nevertheless, they show extraordinary difficulty in what seems to be a somewhat comparable task, learning to read. In order to investigate this more deeply, it is important to consider what is known about the complex developmental processes of early language acquisition. Interestingly, knowledge of how we learn to speak was not known when the phonological deficit was first proposed. However, there is now a clear consensus (Kuhl 2004) and the follow-up theoretical analysis (Meltzoff, Kuhl, Movellan, & Sejnowski 2009) on how language and speech develop over the 1st year in infancy.

This understanding is divided into two dimensions, receptive (perception) and expressive (production) respectively. In the first 6 months of life, children have an equipotentiality for all languages and will develop whatever language they are first exposed to, with a sensitivity to any sounds within spoken language. Consequently, at this stage, Japanese infants can discriminate /l/ from /r/. This capacity is refined between the ages of 6–12 months, with the effect that infants become specialists in the language they hear, that is their own native tongue. At this stage, Japanese infants lose the ability to differentiate phonemes, that are redundant in their language, thus losing the capacity to discriminate /l/ from /r/. This works on the principle of identifying regularities in speech, a specialized system of statistical learning that provides a platform for effective language learning in humans.

It seems that in this, as in so many other areas, a process of statistical learning is involved, based on the infant tuning into the mother's speech patterns to acquire the elements of phonology and prosody in their own native language. Simply being exposed to a range of experiences in hearing spoken language develops the processes of neural commitment and classification. Moreover, the elements of social interaction and turn taking developed in this process, as well as the trial and error process of producing the sounds, shapes the infant's language development. This builds on the infant's generalized learning capacity, scaffolded by the relationship with their caregivers, to enhance both speech production and perception (Meltzoff et al. 2009).

Once commitment has been made to this natural language – a process Kuhl and her colleagues term “neural commitment” – there is no capacity for change. The neural circuits have made a commitment, and the infant in an English environment can no longer distinguish the sounds of other languages, which are deemed irrelevant and no longer accessible to conscious processing. This makes it almost impossible to unlearn our mother tongue, impacting on the ability to both hear and produce alien sounds, based on neural circuits for signal processing that are outside our ability to control or influence. We are now in a brown field situation where prior experience in learning has built structures that constrain and channel subsequent learning.

13.2.2 Dyslexia: The Neural Noise “Minimal Hypothesis”

Let us consider first the impact of differences in brain organization, related to the process of migration and differentiation during the formation of the infant brain in the womb. Here we can resonate with the impaired brain organization identified by Galaburda in his early work (Galaburda 1990; Galaburda, LoTurco, Ramus, Fitch, & Rosen 2006). This provides a less than optimal platform for learning to take place, based on both dysplasias and ectopias (Galaburda 1986), or a coarseness in the cortical columns (Casanova, Buxhoeveden, Cohen, Switala, & Roy 2002). Of course, this is not in itself an issue with the neural structures or connectivity, but it seems plausible to suggest that this could also lead to differences in expression of the neurotransmitters or at the level of the strength and timing of impulses within the neuron. Whatever the underlying cause, this would be likely to lead to inconsistencies in speed of information processing, reflected in greater noise (jitter) in the sequential, spatial and temporal aspects of signal processing, either in specific or more generalized areas of the dyslexic brain. Indeed, there is evidence that dyslexic children do indeed suffer from greater neural noise, in that their signal detection thresholds are more seriously affected by signal degradation than those for typically achieving children (Sperling, Lu, Manis, & Seidenberg 2005).

The development of skill depends on developing automaticity. A seminal finding in cognitive psychology (Shiffrin & Schneider 1977) is that the two fundamental requirements for the transition from controlled processing to automatic processing are, first, extensive experience over many days, and, crucially, second, “consistent mapping” between stimulus and response, such that the same response is made to the stimulus every time it occurs (as opposed to “varied mapping” where the relationship between stimulus and response is not consistent). The increased neural noise would essentially lead to more randomness, less consistent mapping and hence would interfere significantly with the process of automatization, meaning that either a greater quality of stimulus or longer experience would be necessary in order to build up skills to the same level as their normally achieving peers. Therefore, as we predicted in our earlier automaticity research, dyslexic children would have to “consciously compensate”, in order to achieve in even basic skills, thus reducing the resources available for further learning.

Moreover, once dyslexic children have painstakingly acquired a skill, they will have greater difficulty in unlearning it, in order to build new skills on top of the existing skills. Unfortunately, this is a key requirement in skills such as learning to read, where for example moving from segmenting and decoding to reading whole words is necessary in order to achieve fluency.

In addition, there will be greater variations in timing for dyslexic children, and this will impact on the development of skills that are dependent on accurate and explicit control. These include not only cognitive processing but also the precise timing underlying the acquisition of a range of motor skills, such as kicking or catching a ball.

Finally, and crucially, we had not previously realized that this variability in timing would impact on the successful construction of a whole range of functionally connected neural networks. The Hebb rule – cells that fire together wire together – is the key principle in the construction of new neural networks. Greater intrinsic noise would lead to greater variability, which would interfere significantly with network construction (and deconstruction). Unlike for the individual skills, failure to develop multi-purpose neural networks, for executive function, eye-hand coordination, cross-modality integration, proprioceptive/sensory coordination would lead to functional coordination difficulties in many forms (Lachmann & van Leeuwen 2014). These would include not only the network for fluent reading, but also for balance and executive function, indeed the full range of sophisticated network systems that older children and adults rely on for skilled processing in all modalities.

In summary, all procedural and implicit learning would be affected. Conversely, declarative skills, which may be less susceptible to neural noise, would be normal or even superior, providing a suitable platform for strengths in dyslexia to emerge.

We have explored the differences between early language perception and production and later learning. What else needs to be in place before a child is ready for school?

13.2.3 Development of Executive Function

Piaget (Inhelder & Piaget 1958) outlined a developmental framework demonstrating the extraordinary changes in cognitive processing in the first seven years, moving from the sensorimotor stage through the pre-operational stage to the start of concrete operations. Of course, there are wide individual differences in the speed of this progress and for most children development is uneven, demanding the construction and refinement of a series of neural networks. Eventually these will allow them to take control of the full range of automatic systems. Piaget's insights have largely been subsumed under the information processing framework for "executive function" (EF), an alternative perspective on controlled processing. In her recent review of executive function and its development, Diamond (2013) identified the three core "cool EF" processes, based on cognitive processing. These include inhibition [inhibitory control, including self-control (behavioral inhibition), interference control (selective attention and cognitive inhibition)], working memory, and cognitive flexibility (mental flexibility, or mental set shifting and closely linked to creativity). However, it is also important to note the existence of "hot EF" processes. These are also top down cognitive – driven processes that allow the developing child to control elements of their behavior, such as anger, aggression, impulsivity and anxiety, – that are driven by emotion (Zelazo & Carlson 2012). These all develop with experience and improve throughout childhood and adolescence (Bauer & Zelazo 2014).

In terms of the developing child and their readiness to learn, it seems that at age three, there is little differentiation between hot and cool, but these distinctions develop over the next few years. The growing literature on the development of

executive function between the ages of 3–6 is particularly important here in terms of “readiness for school” (Fitzpatrick, McKinnon, Blair, & Willoughby 2014). Moreover, Blair (2002) proposed that “hot EF” such as emotional control and socialization also play a key role at this stage.

13.2.4 The Delayed Neural Commitment Hypothesis

We have now prepared the ground thoroughly. Our earlier work revealed problems that extend well beyond the reading domain for dyslexic children, and can be attributed to the automatization and execution of procedural skills. This analysis copes well at a general level with the specific and general features of dyslexia, together with commonalities with a range of other learning disorders. Our research has also shown that these problems are consistent with the idea of generalized developmental delay, and also highlighted the difficulties of unlearning primitive skills. Kuhl’s developmental framework for speech acquisition highlights the importance of neural commitment (which is a form of procedural learning based both on statistical learning and categorisation) that leads to an encapsulated, hard-to-modify skill network. We have highlighted the importance of the development of a range of such networks, from walking to talking to thinking to controlling, which are developed in the pre-school years.

This leads to the Delayed Neural Commitment hypothesis for dyslexia. Stated formally, DNC holds that dyslexia is associated with minimal brain differences, often arising from the development of the fetus in the womb, that lead to increased noise in the neural circuits for hearing and speech (and beyond). This has several major implications.

First, delay and imprecision in language-related skills, which impact on the development of the phonological network, leading to impairments in phonology. Second, a more generalized problem with the automatization of a range of skills, notably those linked to articulation and including both implicit and explicit skills. Third, DNC also means it is difficult to shed earlier habits that need to be unlearned to allow new skills to develop and consequently, it is more difficult to build the networks needed for executive learning and reading.

These points, of course are consistent with findings in dyslexia derived from a range of theories. This is consistent with the evidence for differences in sound processing from birth in dyslexic infants (Guttorm et al. 2005; Molfese 2000), and with the data presented in Fig. 13.4. In terms of developmental transitions, primitive reflexes are vital for survival at birth, but if these are retained they prevent the development of age-appropriate skills. There is also clear evidence for the retention of primitive reflexes in dyslexia (McPhillips, Hepper, & Mulhern 2000), and this problem in unlearning is illustrated graphically in Fig. 13.4. Unfortunately the literature on executive skills and dyslexia in pre-school children is relatively sparse, although evidence of difficulties is building (Clark et al. 2014; Moura, Simoes, & Pereira 2015; Varvara, Varuzza, Sorrentino, Vicari, & Menghini 2014).

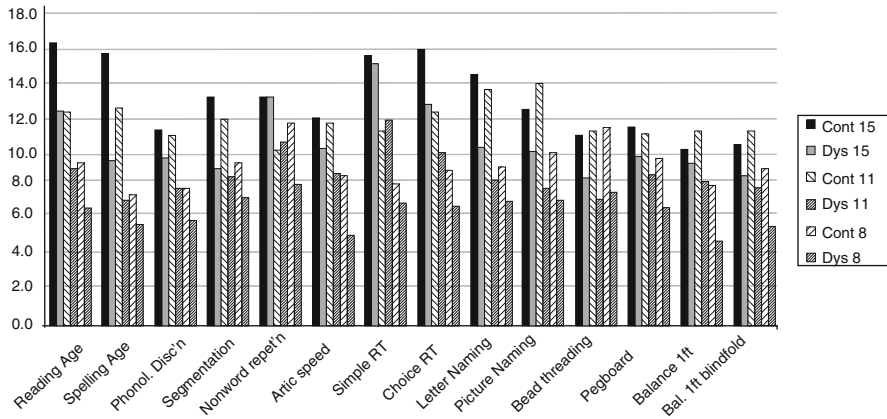


Fig. 13.4 Age equivalent scores across the range of primitive skills

More directly, there is extensive evidence that even after successful phonological training, dyslexic children have difficulty in moving on from decoding, to the more fluent whole word recognition (NICHD 2000). This suggests they have difficulty in unlearning the phonological scaffolding stage in order to progress to mature processing in reading. There is also strong evidence that dyslexic children do not demonstrate the typical “Visual Word Form Area” that is associated with fluent reading (Shaywitz et al. 2007) and any evidence for this is linked to whole words rather than sub-lexical items (van der Mark et al. 2009).

By contrast with deficit theories of dyslexia, DNC suggests that there may be advantages to dyslexia in dealing with a system based on greater noise. We speculate that there would be less “pruning” of primitive capabilities, thereby extending the critical period for combining skills. Furthermore, weak procedural skills are likely to lead to greater reliance on declarative processing and this is reflected in emerging empirical evidence of declarative advantage in dyslexia (Hedenius et al. 2013). This is entirely consistent with the literature on declarative strengths in dyslexia (Eide & Eide 2011; Geschwind 1982; West 2009), but represents the first real evidence for such abilities. So how would DNC work to confer advantages in dyslexia? The extended sensitive periods associated with DNC allows an extended learning period into late adolescence and adulthood, producing the late bloomer so often noted in the literature. Moreover, reduced commitment/compartimentalization allows greater cognitive processing and flexibility. This would be associated with more cross-talk between different brain regions and senses, allowing for the development of more integrative processing.

The DNC is of course a development from our earlier theories of automaticity, cerebellar and procedural learning deficit. However, the reformulation reflects a major change in perspective. Let us clarify here the importance of the DNC for our readers on five counts.

First, DNC provides the direct linkage to the earlier processes of speech and language development that has been searched for in vain by the phonological deficit theorists and links directly with the development of phonological skills. This allows the phonological deficit theorists to re-integrate their work with current theories on the cognitive neuroscience of language and its development.

Second, DNC allows us to see the “big picture” in child development, rather than focusing down on individual skills. It has the potential to incorporate the full information processing architecture, bringing into play not only current levels of achievement, but also underlying development and its trajectories. This incorporates Piaget, executive function, memory, inhibition, and learning in all its aspects, including the early stages of learning by being told. Furthermore, the explicit focus on the construction of neural networks provides a direct link to the cognitive neuroscience of intrinsic connectivity networks (Bostan, Dum, & Strick 2013; Fox et al. 2005; Friederici & Gierhan 2013; Kipping et al. 2013; Sporns, Chialvo, Kaiser, & Hilgetag 2004; Zuo et al. 2012).

Third, DNC moves away from value laden concepts of deficit, to reflect a system which can endow both advantage and disadvantage. Although DNC represents a limitation in many of the early stages of learning particularly during the early school years, it can also provide advantages that were not easily explained under any of the deficit frameworks, including our own. This means where it is useful to maintain earlier skills, or combine skills that do not naturally occur within the same time frame, dyslexic children may score an advantage. Moreover, it explains the mechanism by which many dyslexics achieve success, biasing them to favor their strengths in declarative skills, rather than emphasizing their weaker procedural habits. Concentration on declarative strengths allows the dyslexic adult to build their expertise for specialization in their specific areas of interest.

Fourth, DNC does not simply apply to dyslexia but can be applied across the range of developmental disabilities, and even to specific aspects of development in normally achieving children. It therefore can provide a bridge with normal development.

Most significantly, the DNC has strong implications for how we tackle early learning and support for reading. It seems that it is not simply the phonological skills that need to be mastered in order to build success, but also executive function and its development, in order to ensure classroom readiness. Interestingly, although there has been considerable recent research on improving executive function in disadvantaged pre-school-children (Diamond 2013; Fitzpatrick et al. 2014), this has not yet been extended to dyslexia. There is clearly considerable potential for an approach that ensures readiness to learn, whatever the nature of the difficulties the child may encounter. This enhanced understanding of how children learn constitutes a significant breakthrough in providing effective support.

13.2.5 Delayed Neural Commitment and Theories for Dyslexia

Let us return here to the theories of dyslexia we outlined in our introduction, and consider how the DNC deals with these. DNC is an explanatory framework able to deal naturally with all of these theories, because it includes both a learning and a temporal framework, thus handling all the major evidence in dyslexia. Moreover, DNC also has the capacity to deal naturally with the extensive evidence for comorbidity that has been established in dyslexia, with DCD, SLI, and ADHD.

In terms of Procedural Learning Deficit, DCD, although less fully researched than dyslexia, seems to relate to the motor rather than the language component of the learning networks (see Fig. 13.3). We have identified cerebellar-type problems in DCD in a prism adaptation study (Brookes et al. 2007) and a deficit similar to automaticity seems to underlie evidence for impairment that increases with task difficulty (Cantin, Ryan, & Polatajko 2014). Interestingly, there is also evidence of EF deficits in DCD (Rahimi-Golkhandan, Steenbergen, Piek, & Wilson 2014; Saban, Ornoy, & Parush 2014).

This links naturally with ADHD, where EF problems are well-established (Barkley 1997; Willcutt, Doyle, Nigg, Faraone, & Pennington 2005) particularly in control of inhibition, but also in speech internalisation. Strikingly, both gross and fine motor skill deficits are found in more than 50% of children with ADHD (Kaiser et al. 2015), with the inattentive subtype of ADHD showing problems in slowed reaction times and fine motor control, particularly in complex tasks.

There are similar links with SLI, which often leads on naturally to difficulties in reading, and clear evidence of similar procedural learning problems (Lum et al. 2014; Ullman & Pierpont 2005) as well as deficits in executive function (Henry, Messer, & Nash 2012; Im-Bolter, Johnson, & Pascual-Leone 2006).

To summarize, all of these developmental disabilities are consistent with difficulties in some or all components of the procedural learning system, including motor, language and executive function, with the cluster of difficulties and precise pattern for any given child depending on the aspects of learning most severely affected.

13.2.6 Implications for Dyslexia Theory and Practice

The DNC framework suggests that dyslexia will manifest as a delay in acquiring a wide range of skills, and this will lead to delays in building the neural network needed for successful learning, as well as the need to consciously compensate in routine tasks.

Let us consider first why reading seems to pose a particular challenge for dyslexia? As a complex and cumulative skill, it is bound to present a significant challenge for anyone with difficulties in learning, even in the early stages. Moreover, in order to become fluent in reading we need to co-ordinate a wide range of skills, across modalities. So the reader needs to directly focus his/her attention on the word

s/he is reading as well as prepare the fixation for the next word in the sentence. In order to read aloud, our eyes and our mouth need to function together, but critically our eyes need to be ahead of our voice as we process the stimuli on the page before trying to read with expression. This form of multi-tasking between two different modalities is almost impossible for even a compensated automatic system, because it is simply not possible to consciously attend to two skills simultaneously.

In terms of identifying dyslexia before a child fails to learn, there is now good evidence that using a screening test at age 5 coupled with a structured intervention designed by teachers to target multiple aspects of early learning can be successful. Furthermore, the framework provides strong support for any intervention designed to facilitate the development of cross-modality networks that will prove important for fluent reading, and therefore is consistent with the multisensory approach traditionally advocated for dyslexia (Gillingham & Stillman 1960). By contrast, it reveals the dangers of over-reliance, over-soon on a dedicated phonological awareness program, and highlights the need to ensure that phonological support is undertaken in such a way that the skills do not become encapsulated.

Above all, whatever type of difficulty a child may encounter, the key issue here is whether or not they have the necessary skills and networks in place to be able to benefit from classroom teaching. If they are struggling then we should provide support not just with phonological skills, but also with the executive skills that underpin learning. This will impact not just on learning, but also on behavior in the classroom.

The DNC also suggests that it is particularly important not to try to hot-house the development of reading before children are ready. There is long-standing evidence (Hatcher, Hulme, & Ellis 1994) that young children benefit from support in single letter reading in order to build up their fluency, in addition to the well-known phonological support, and that vocabulary is a further key in preventing reading failure. In addition, the DNC suggests that we need to target a range of non-reading skills that are at risk in dyslexia. In our view this framework highlights the need for delay in introducing reading, until a child is ready to learn, by contrast with the current approach of introducing complex tasks before a child is ready or able to engage with them. However, this is clearly an issue for policy and lies beyond the scope of the present chapter.

13.3 Conclusions

In this chapter we have taken the road less traveled, the investigation of dyslexia as a learning disability. Even without considering the reading process, it has been a long and winding road, moving from automatization to the cerebellum to procedural learning, with each corner turned accounting for a greater range of findings, but leading to further vistas and avenues for exploration. We have just turned the corner to our current framework, Delayed Neural Commitment, and the vista from our new viewpoint is particularly engaging.

We have seen that dyslexic children are likely to show problems not only in the development of individual skills, but also in the integration of those skills into new and more efficient circuits. Delayed neural commitment explains not only issues with automaticity, but also in developing the circuits underlying executive functions and internalized speech requisite for learning, and in discarding earlier circuits that are no longer efficient. The framework is compatible with all major theories of dyslexia and provides a fruitful framework for the development of educational practice to enhance language and learning before a child tries (and fails) to learn to read.

DNC provides the necessary linkage back from school to birth and infancy, it provides a link to the cognitive neuroscience of functional networks and their development, it provides an opportunity to understand and help pre-school dyslexic children, and has considerable theoretical and applied potential. We encourage intrepid researchers and educators to explore this new landscape.

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Chapter 14

Reading and Dyslexia: The Functional Coordination Framework



Thomas Lachmann

Abstract The Functional Coordination approach of reading acquisition claims that beginning readers draw on established cognitive functions that are (1) recruited, (2) modified, and (3) coordinated to create a cognitive procedure for reading text, which forms the basis of subsequent (4) automatization. In this chapter we will focus on visual functions and how they are modified and coordinated with other cognitive functions involved in a reading specific cognitive procedure. Evidence relating to the emerging prevalence of analytic processing in letter perception is discussed. It is argued that the process of learning to read does not lead to a loss (recycling) of perceptual skills, but to a novel synthesis of functions, which are coordinated for reading and then automatized as a package. Developmental dyslexia is explained as a Functional Coordination Deficit (Lachmann 2002), since the coordination stage is assumed to be most liable to manifest deficiencies. Developmental dyslexia is not seen as a consequence of a deficit in a single function or in automatization, but as result of automatizing a suboptimal functional coordination. This integrative approach is a mere framework, rather than an explanatory theory, and is open to multi-causal explanations. Rather than solving the puzzle, the framework offers a structure for integrating various theories on reading and dyslexia.

Keywords Literacy acquisition · Developmental dyslexia · Training and remediation · Automatization · Functional coordination · Alphabetic route · Letter recognition · Analytic/holistic processing · Global precedence effect · Symmetry · Mental rotation · Flanker · APPLE

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14.1 Reading: It's Amazing!

Reading is a cultural achievement. Initially we are all illiterates. Reading is an acquired skill; it must be taught, and it must be learned. Usually this is accomplished in primary school. At least as far as we may remember, most in the class learned this skill easily and rather independent from their cognitive abilities. Later, reading becomes so much part of everyday life that we may not think about the fact that, actually, this is one of the most intriguing, most amazing feats of our cognitive system. In order to read a page like this we need to manage, in an extremely fast and accurate manner, the decoding of several thousands of tiny visual configurations – codes for letters that form words and sentences – into language and thus into meaning, in order to get the message the writer intended to tell us.

Reading is a complex skill involving reams of cognitive functions in the process. These functions involve basic visual and auditory perception, as well as higher order cognition, memory and executive control, attention, various functions of language processing, oculomotor control and many others. However, none of these functions is specific for reading; evolution does not provide us with any reading specific brain structure. We have to deal with what we have to create a new reading specific cognitive procedure. We are usually not aware of how complex this procedure is, and we feel like reading is an easy task and may even be fun. This is because this complex cognitive procedure is highly automatized.

This chapter addresses the question of how this reading specific cognitive procedure is learned before it becomes automatized. The majority of studies on learning to read, and on related disabilities, focus on the development of phonological processing abilities. Here we will focus on visual functions involved in letter recognition, in particular how these become modified and coordinated with other functions during reading acquisition.

14.2 Learning to Read

14.2.1 Development of Different Reading Sub-skills

According to Frith (1985), at the beginning of reading acquisition, in the logographic phase, letters will be perceived just like similar non-letter configurations. The order of letters in a word and other phonological factors are largely ignored. Unfamiliar words and non-words cannot be read. In fact, even familiar words are not really “read” but rather “recognized,” because in this phase, the child recognizes a word as a whole configuration, based on salient graphic features, just as in visual object recognition. For “real reading” the knowledge of individual graphemes and phonemes, and their correspondences (i.e., letter knowledge) is required. If this knowledge is available for use, the alphabetic sub-skill is developed (alphabetic phase, Frith 1985). The child is now “aware” of graphemes (letters) and phonemes

(sounds) and of their relations. This sub-skill allows decoding of graphemes into the corresponding phonemes one by one. Phonemes are then merged together into pronounceable syllables and words. Note, that in some cases, grapheme clusters represent a phoneme (e.g., in German “au” or “sch”).

Regarding visual processing, fine details of each individual grapheme, its orientation, and the order in relation to other graphemes are crucial in this phase. On the other hand, graphemes may have different fonts, requiring categorical representations. This holds also for auditory processing, where the development of a categorical phoneme representation (Liberman, Harris, Hoffman, & Griffith 1957) is essential. The system of phonemic representation gains prominence in the process of learning to read (Christmann, Berti, Steinbrink, & Lachmann 2014; Serniclaes, Ventura, Morais, & Kolinsky 2005) evolving along with the graphemic representation (Port 2007).

At this point, words can be pronounced even though they are rare or unknown (including non-words) as long as the correspondences between the graphemes and phonemes follow the learned rule (regular words), which is the case for most words in transparent orthographies (e.g., German). After long training, letters become represented in a cross-modal fashion (Froyen, Bonte, van Atteveldt, & Blomert 2009; Froyen, van Atteveldt, Bonte, & Blomert 2008; Froyen, Willems, & Blomert 2011; Lachmann & van Leeuwen 2014). They are not simply visual items anymore, but connected with auditory (phonological) information.

Even for skilled literates alphabetic reading is still relatively slow. Moreover, using this reading sub-skill, irregular words, which do appear often in non-transparent orthographies (e.g., English), but do exist also in transparent orthographies, cannot be pronounced correctly. The development of the orthographic sub-skill (orthographic phase, Frith 1985) enables the instant analysis of grapheme units and words (orthographic units), including allographs. Words can now be read without a one-by-one grapheme-phoneme conversion, as wholes. However, in contrast to the logographic phase, here, the whole word reading is not purely based upon visual recognition, but on a cross-modal representation of this word in a mental lexicon (Coltheart 1978, 2007).

Compared to the alphabetic sub-skill, the orthographic sub-skill allows relatively effortless and fast reading. Nevertheless, even in expert readers the alphabetic sub-skill remains available for unfamiliar or foreign words (Coltheart 1978, 2007; Davelaar, Coltheart, Besner, & Jonasson 1978; Morton 1969, cf. Steinbrink & Lachmann, 2014, for an overview) and for both transparent and non-transparent orthographies (Lachmann & van Leeuwen 2010). The same holds for the logographic sub-skill, for instance, when skilled readers are presented with logos of brands or institutions, such as the NASA sign. Thus, principally, all reading sub-skills, the logographic, the alphabetic, and the orthographic, remain available in skilled readers and are applied during the reading process in a flexible way, depending on the requirements of reading task.

In this chapter, we will focus on the acquisition of the alphabetic sub-skill. Here, letter knowledge is essential, i.e., the establishment of a cross-modal representation of letters and, based upon that, the ability of fast decoding of individual graphemes

as visual codes into the corresponding phonemes as the smallest units of spoken language. No matter how important phonological processing may be for letter recognition (Snowling 2001), the process always starts with a visual analysis. However, predominant strategies of visual processing, which include orientation-invariance and context sensitivity, are not suitable for establishing a connection with phonology. This not only because, obviously, a “b” is not a “d” nor a “p” nor a “q” either (Orton 1925), but generally a phoneme must be represented by an orientation-specific symbol, that, independent from font, must be recognized when surrounded by context.

According to the *neuronal recycling hypothesis* (Dehaene & Cohen 2007; Dehaene et al. 2010), this problem is solved by getting pre-existing functions recycled for reading. Thus, it may be possible that original information processing skills are reduced, or even lost, since original resources are being redeployed for achieving the newly required functionality. We (Lachmann & van Leeuwen 2014) argued that, as the learning of any complex skill, literacy may train certain functions, but does not lead to a reduction, or a loss, in the sense of a “replacement” of any perceptual skill outside the context of reading (and even not within, Borst, Ahr, Roell, & Houdé 2015). Instead, learning to read leads to a novel synthesis of pre-existing modified functions, which are coordinated for reading and then automatized as a package, i.e., a reading specific cognitive procedure (Lachmann & van Leeuwen 2014). This procedural learning process (Fitts & Posner 1967; Foerde & Poldrack 2009; Nicolson & Fawcett 2018, 2011; Nicolson, Fawcett, Brookes, & Needle 2010) is described in the Functional Coordination Framework (Lachmann & Geyer 2003; Lachmann & van Leeuwen 2008a) of learning to read in terms of four stages (see Fig. 14.1; Lachmann & van Leeuwen 2014). We will introduce these in the following.

14.2.2 Reading as Functional Coordination

The Functional Coordination Framework (Lachmann & Geyer 2003; Lachmann & van Leeuwen 2008a) describes reading acquisition in terms of four stages (see Fig. 14.1; Lachmann & van Leeuwen 2014) of procedural learning (Fitts & Posner 1967). In the first stage, reading instruction leads to the recruitment of pre-existing reading relevant functions: from the auditory domain, for example, pre-existing phonological processing skills; from the visual domain, for instance, the ability to distinguish details in two-dimensional line drawings and pre-existing grapheme awareness (Wesseling, Christmann, & Lachmann 2017). In this stage, children understand that text consists of smaller units, words, and graphemes that represent phonemes (Wesseling et al. 2017).

In the next stage, the applied functions become modified in order to optimize their usage in the context of reading: in the visual domain for instance by the suppression of orientation invariance and symmetries (e.g., Borst et al. 2015); in the auditory domain for instance by the fine-tuning of the phonological system (Port 2007; Serniclaes et al. 2005).

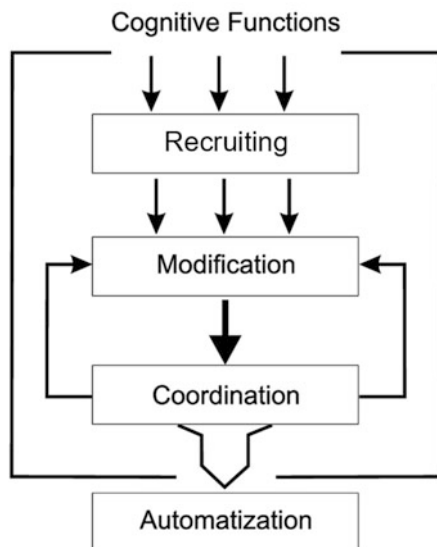


Fig. 14.1 The Functional Coordination Framework describes learning to read as a form of procedural learning in which pre-existing functions and skills from different domains, are recruited, modified and coordinated, leading to a reading specific procedure. After training this procedure gets automatized, after which experienced readers are biased against processing strategies for letter perception that do not form part of the procedure. Coordination stabilizes the modifications. A failure of coordination will result in automatization of an abnormal procedure, leading to reading and writing problems (Lachmann 2002). The whole process, including the structural and functional changes related to it, takes several years (Froyen et al. 2009)

In the third stage, the modified functions become coordinated, giving rise to grapheme-phoneme correspondences (alphabetic sub-skill) and the direct activation of word representations in the mental lexicon (orthographic sub-skill). This will result in cross-modal codes of letters and words (Blomert 2011), which, in turn, stabilizes and strengthens the modifications for the involved functions and strategy preferences (see Fig. 14.1).

Coordination leads to a procedure, which forms the basis of subsequent automatization, the final stage in the framework. Given the complexity of the procedure, automatization needs a lot of practice and takes time (Froyen et al. 2009; Lachmann & van Leeuwen 2008a). Even though children may be able to read by using both alphabetic and orthographic sub-skills, the implementation of the underlying structural and functional basis for its automatization process in the brain may take about 3–4 years (Froyen et al. 2009; Lachmann & van Leeuwen 2008a).

In relation to the phase model of reading acquisition (Frith 1985), the Functional Coordination Framework explains both the acquisition of alphabetic and of orthographic sub-skills (within one stage), not, however, of the logographic sub-skill, since for this, no modification or coordination is required, and, in fact, we consider this not as reading in the narrow sense. In the following we will describe how the

reading specific modification of visual functions will affect letter recognition and how a failure in this modification may lead to a deficit in functional coordination and consequently to developmental dyslexia.

14.3 Developmental Dyslexia

14.3.1 *Developmental Dyslexia as Functional Coordination Deficit*

Within the aforementioned framework, developmental dyslexia is understood as a multicausal deficit in functional coordination. A coordination suboptimal for reading will result in learning a procedure suboptimal for reading, which will subsequently be automatized (Functional Coordination Deficit (FCD), Lachmann 2002, 2008). This suboptimal functional coordination can result from various mild early-stage deviations, including deficient auditory processing (Ahissar, Protopapas, Reid, & Merzenich 2000; Calcus, Hoonhorst, Colin, Deltenre, & Kolinsky 2018; Christmann, Lachmann, & Steinbrink 2015; Goswami et al. 2011; Groth, Lachmann, Riecker, Muthmann, & Steinbrink 2011; Hämäläinen, Salminen, & Leppänen 2013; Jaffe-Dax, Daikhin, & Ahissar 2018; Lallier et al. 2018; Richardson, Thomson, Scott, & Goswami 2004; Talcott et al. 2002; Tallal & Jenkins 2018; Vandermosten et al. 2010; Witton & Talcott 2018), visual instabilities and processing deficits (Becker, Elliott, & Lachmann 2005; Slaghuis & Ryan 1999; Stein 2002, 2018; Stein & Talcott 1999, see Stein & Kapoula, 2012, for an overview) or a combination thereof (Au & Lovegrove 2007, see Farmer & Klein, 1995, for a review). In these cases, deficits are present already in the recruitment stage; yet they are manifested only in the coordination. This is the case, because these small deviations at the early processing levels (e.g., contrast sensitivity; Slaghuis & Ryan 1999; Stein 2002, temporal processing, Galaburda, 2002; Steinbrink, Groth, Lachmann & Riecker, 2012; Tallal, 1980) are not severe enough to lead to modality-specific deficiencies by themselves (it makes, of course, no sense, to assume that dyslexics cannot see or hear correctly or cannot move without falling; note, that none of the pioneers of dyslexia research, including Orton 1925, ever proposed that; see Lachmann & Geyer 2003), but they may cause problems in fine-tuning during the coordination stage. Early-stage deviations have a neurodevelopmental origin (Galaburda 2002, 2018). They do, however, not necessarily, lead to problems in coordination, but they may be compensated, e.g., by coping strategies or brain plasticity (Frith 1986).

Alternatively, problems may arise in the modification stage, for instance by a failure to suppress symmetry in visual perception (e.g., Borst et al. 2015; Pegado, Nakamura, Cohen, & Dehaene 2011; Perea, Moret-Tatay, & Panadero 2011) or problems in developing phonological (e.g., Fawcett 2002; Snowling 2001) or orthographic processing skills (Badian 2005; Seymour & Evans 1993; van Orden, Pennington, & Stone 1990). Yet again, even though these problems may arise at this stage, they will be manifested in the stage of coordination.

Functional coordination deficits may arise even without any deficiencies in the recruiting and the modification stage, originating from within the coordination process (e.g., Blomert 2011), or resulting from deficiencies in automatization (Fawcett 2002, Nicolson & Fawcett 2011, 2018). However, rather than automatization, the coordination level is most liable to manifest the deficiencies, because this is the level where the greatest degree of fine-tuning of complex functions and procedural organization is required. Note, that this idea is consistent with the cerebellar approach of Nicolson and Fawcett (2011, Fawcett, 2002, Nicolson & Fawcett, 2018) since the cerebellum seems to be essentially involved in such fine-tuning and coordination processes (Stoodley & Stein 2011), including language processing (Ackermann & Hertrich 2000, see Foerde & Polrack, 2009, for an overview). The coordination process can also be affected by external factors, such as teaching methods and environmental conditions (e.g., noisy classroom, Klatte, Spilski, Mayerl, Möhler, Lachmann & Bergström 2016; Klatte, Bergström, & Lachmann 2013).

An established coordination, i.e., a learned reading-specific procedure, will also feedback to the involved functions and will stabilize and advance their specific modification (see Fig. 14.1). This means, the modifications and preferred processing strategies will be strengthened and trained toward the procedure to which they contribute. Phonological processing, for instance, will advance with reading practice. In case of a suboptimal coordination, this will lead to the fortification of suboptimal modifications and processing strategies. Therefore, deficits researcher found to be associated with developmental dyslexia, for instance in phonological processing or symmetry suppression, could also reflect a consequence of suboptimal reading experience (Hüttig, Lachmann, Reis, & Petersson 2017). This sometimes makes it hard to distinguish cause from effect.

14.3.2 Conclusions for Intervention

There are a number of consequences for intervention and training with poor readers and developmental dyslexics that follow from the assumptions of the Functional Coordination Deficit approach. Firstly, a training of deficient functions should start as early as possible. Using the right diagnostic tools, some deficits can be identified even before learning to read has started and should be addressed (e.g., auditory discrimination, phonological processing). Usually, however, reading problems are identified after automatization. This means, a suboptimal functional coordination has already been implemented in a cognitive procedure. Therefore, it makes little sense to train only reading as such (at behavioral level, Frith 1986), because more reading experience (“more of the same”) would result in training an automatized coordination that is suboptimal for reading. This leads to the paradoxical effect that the deficiency of involved functions even increases. Therefore, deficits associated with developmental dyslexia may also be a consequence of suboptimal reading experience (Hüttig et al. 2017). Instead, the coordination has to be reorganized first, and then to be trained. The procedural learning has to start from the very beginning

and the whole process must run through. One thing is clear: this takes time and energy. This holds also for illiterate adults who get reading instruction.

Secondly, it makes little sense to train exclusively the cognitive function supposed to be the cause for a failure in learning to read (at cognitive level, Frith 1986), for instance exclusively auditory perception. Children may get better in this particular skill, but the transfer to advanced reading is unlikely (Schumacher 2012). Even if the function under training is, in fact, the one that has caused the problems in learning to read, the reading-specific procedure can only be optimized if the coordination with the other functions is trained. This may be the reason why, for instance, an isolated training of phonological awareness, if not conducted before or at the beginning of literacy acquisition, was found to increase phonological processing skills, but had only little effect on literacy skills (Bus & van IJzendoorn 1999), while a combination with grapheme-phoneme conversion skills was beneficial (e.g., Klatte, Spilski, Mayerl, Möhler, Lachmann & Bergström 2016). This combination helps to reorganize the functional coordination and thus supports the transfer to literacy. Then, instead of getting smaller with temporal distance from the training, the effect on reading performance becomes even gradually larger over time (Klatte, Bergström, Steinbrink, Konderding, & Lachmann 2018; Klatte et al. 2014).

Finally, since there is no doubt that dyslexia is multicausal (Lachmann, Berti, Kujala, & Schröger 2005; Ramus, Pidgeon, & Frith 2003), it will help only some individuals if a training focuses too much on one particular function or sub-skill. Therefore, it makes sense to use a computer-based adaptive program for training, containing training of multiple functions with an increasing degree of coordination, aiming on alphabetic as well as on orthographic reading sub-skills. This will allow individualized intervention (Klatte et al. 2014).

14.4 Analytic Visual Processing for the Acquisition of the Alphabetic Sub-Skill

14.4.1 Are Letters Special?

The acquisition of the alphabetic sub-skill of reading requires the fast and accurate recognition of letters as visually presented symbols. This cannot be managed by visual functions that are preferably used for visual object recognition. These must become modified for learning a reading specific procedure. The automatization of this procedure makes letters special; we then perceive letters differently than non-letters.

In a number of studies we investigated the following questions: whether there is, in fact, a letter-specific visual processing strategy; and if so, whether this processing strategy is always applied for letter recognition; and further, whether this strategy

leads to a loss of any visual ability in perceiving letters; and finally, whether a failure in functional coordination leads to developmental dyslexia.

Why should letters be special at all? Why should they be processed differently? Principally, letters are not different in their physical characteristics from meaningless small scribbles or shapes, except that we know they have a meaning. Elements from an unknown writing system are in fact nothing more than small scribbles. Obviously, before learning to read, letters and similar non-letters are processed in the same way.

However, even prior to learning to read, letters are not natural objects, because they are 2-dimensional. Natural 3-dimensional objects can be perceived from different viewpoints in different orientations, they can move in space over time, and they can occur in cluttered environments, in which they may be partially occluded. These characteristics necessitate that for natural objects, we make the best out of what is visually available. When an object is partially occluded, we may use global object characteristics, for instance symmetry, to complete them perceptually. We make the most out of an object, if we concentrate on its invariant properties, for instance, properties that remain unchanged under positional transformations and different orientations, and we are poised to take clues from the context as to what the nature of the object may be (cf. Lachmann & van Leeuwen 2014). This makes our perceptual system effective and flexible to a changing environment.

Can we use the visual strategies that have been developed for the 3-dimensional world for letter recognition? We know from experimental psychology that these strategies are also preferred for the perception of 2-dimensional objects and shapes. Mental rotation effects, for example, are similar for both 2- and 3-dimensional objects (Cooper & Shepard 1973; Shepard & Metzler 1971). Symmetry facilitates the recognition of simple 2-dimensional shapes (Lachmann 2002) and visual completion is based on criteria of mergeability of 3-dimensional volumes, both in actual 3-dimensional occluded objects and in 2-dimensional drawings of them (Tse 1999). Thus, we may generally assume that there is a robust over-all preference to perceive natural objects in a way that we term as *holistic*, and that this extends to 2-dimensional configurations. Here, we define holistic processing very roughly as a tendency to perceive visual objects as dependent of their context, to emphasize the whole over properties of the parts, of an unilateral influence of global features on local perception, of independence from transformational invariants and/or symmetries, and other strategies that make perception fast and effortless and are therefore preferred in object recognition (Lachmann & van Leeuwen 2014).

However, this processing strategy is not always optimal. In many situations and for certain tasks we need to analyze details, to ignore irrelevant context and global properties, or to take the orientation of an object into consideration. We term the strategy applied in these situations here as *analytic* processing as opposed to holistic. The analytic processing strategy is available too, but it is slow and takes more effort than holistic processing. If analytic processing is required for a task, the faster holistic strategy needs to be suppressed, which is not always easy (Hogeboom & van Leeuwen 1997; Roelfsema & Houtkamp 2011). The analytic-holistic distinction is a broad one, known under a variety of sometimes conflicting terminology laden with

theoretical baggage (Piepers & Robbins 2012; Wagemans et al. 2012). Here, we will use the terms holistic and analytic processing simply as a description of opposing processing strategies, as described above (cf. Lachmann & van Leeuwen 2014).

In sum: Both letters and similar 2-dimensional visual objects can be perceived using either a holistic or an analytic visual processing strategy. For object recognition we have a tendency to prefer holistic processing, since it is fast and effortless. For letter recognition, which is essential for acquisition of the alphabetic sub-skill of reading, analytic processing is required. Therefore, according to the Functional Coordination Framework, the visual processing must become modified in terms of a suppression of holistic processing preferences toward an analytic processing preference for letter recognition.

14.4.2 The APPLE: Analytic Processing Preference for Letters Effect

In a number of studies it was shown that letters are processed faster than similar non-letter shapes (e.g., Burgund, Schlaggar, & Petersen 2006). This facilitation could be the results of high stimulus familiarity, resulting from extended exposure to reading material (Ambler & Proctor 1976; Burgund & Abernathy 2008; James, James, Jobard, Wong, & Gauthier 2005). Evidence suggests, however, that letters are not just be processed faster, but also differently from other likewise familiar stimuli (Burgund et al. 2006; Fernandes, Vale, Martins, Morais, & Kolinsky 2014; van Leeuwen and Lachmann 2004) and elicit a different neural response (Pernet, Celsis, & Démonet 2005). As argued before, according to the Functional Coordination Approach, this specific letter processing strategy is established during the early stages of reading acquisition and is automatized over extensive reading and writing practice (Burgund et al. 2006; Fernandes et al. 2014; Lachmann & van Leeuwen 2014). Thus, it is reading acquisition what makes letters special: analytic processing, we argue, is then preferred for letters, as compared to a preference for holistic processing of similar 2-dimensional non-letter shapes.

We tested the prediction of an Analytic Processing Preference for Letters Effect (APPLE) in a number of experiments involving different tasks which, according to our definition, are suitable to distinguish between holistic and analytic processing, such as orientation invariance, symmetry processing, context sensitivity and global preference. We further tested, whether literacy will make it impossible to process letters holistically.

In Lachmann and van Leeuwen (2007) letters and dot patterns (from a set of patterns first used by Garner & Clement 1963), with different degrees of symmetry were presented in a same-different task. A categorical instruction was used: a *same* response was required independent of the possible reflection or rotation of the items, which were shown in succession (see Fig. 14.2). It had been shown (Carmo et al. 2017; Checkosky & Whitlock 1973; Hermens, Lachmann, & van Leeuwen 2015; Lachmann 2002; Schmidt & Ackermann 1990) that for this task, symmetrical dot patterns are processed faster and more accurately as compared to

| | symmetrical | asymmetrical | correct response |
|----------|-------------|--------------|------------------|
| patterns | | | same |
| | | | same |
| | | | different |
| letters | A A | F F | same |
| | D U | R B | same |
| | A T | L R | different |

Fig. 14.2 Design of the same-different judgment task used in Lachmann and van Leeuwen (2007), explained by examples of pairs of symmetrical and asymmetrical dot-patterns (Garner & Clement 1963, first used by) and letter stimuli

asymmetrical ones. As underlying mechanisms for this effect, we introduced the concept of Symmetry Generalization (Lachmann 2002, 2008, see also Mirror Image Generalization, Corballis & Beale, 1993, for review): items that are related to each other by symmetry operations were shown to be represented by the same collective code (Lachmann 2002; Lachmann & van Leeuwen 2005, 2010); an instruction to judge them as different led to a response conflict (Lachmann & van Leeuwen 2005; Proctor 1986). We (Lachmann & van Leeuwen 2014) concluded that Symmetry Generalization is a sign of holistic object processing. If there is a preference of analytic processing for letters (the APPLE) as a consequence of literacy acquisition, we should observe no symmetry advantages for letters. Indeed, for normally reading primary school (Grade 3 and 4) children in Lachmann and van Leeuwen (2007), we found symmetry effects for the dot patterns but not for letters. For the latter, we concluded, learning to read led to an automatic activation of an analytic processing strategy, i.e., symmetry generalization was suppressed. In contrast, age-matched children with developmental dyslexia showed a symmetry advantage for both patterns and letters (see Fig. 14.3). The remarkable consequence is that, on this task, children with developmental dyslexia were even faster, in particular with letters, than normally reading children. We interpreted this seemingly paradoxical result (i.e., that developmental dyslexics performed better than controls in a letter task) as indicating that normally reading children differentiate in their processing strategy between letters and non-letter shapes, i.e., they show an APPLE, whereas dyslexics do not. In the modification stage, the latter failed to apply analytic processing in terms of suppressing symmetry generalization for letters. For the particular task, i.e., letters as well as shapes of different orientation have to be rated as same, this led to a processing advantage for children with developmental dyslexia.

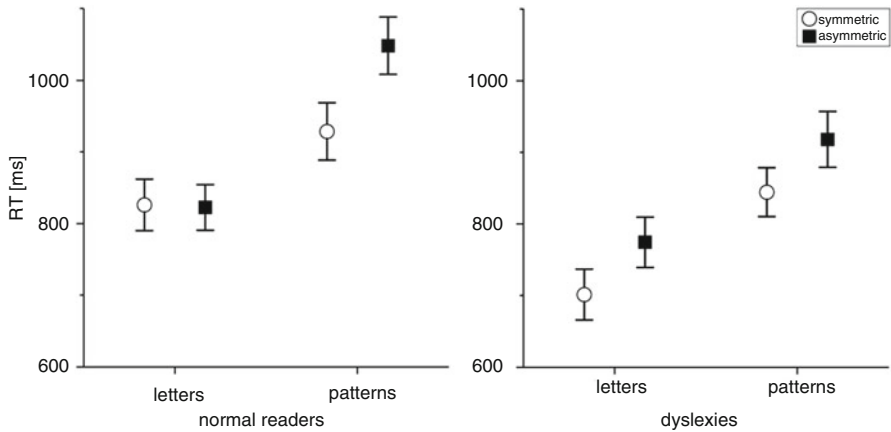


Fig. 14.3 RT (ms) for symmetrical and asymmetrical items for children with developmental dyslexia and age-/IQ-matched control children with normal reading skills. (Data from Lachmann and van Leeuwen 2007)

If, as argued before, both analytic and holistic strategies are available to normal readers, why, then, is it the case that for this particular task the normally reading children did not apply the holistic strategy to letters too, since this seems to work best for the given task? The reason is that skilled readers have automatized the analytic strategy for letters, and it seems they cannot suppress this processing preference without costs. Children who failed to modify the preference of visual processing, and thus to implement analytic processing in the coordination stage of learning to read, may have a disadvantage in acquiring the alphabetic sub-skill, but they have an advantage for the particular version of the *same-different* task used in Lachmann and van Leeuwen (2007), which involved responding to letters of different orientation as same. If, in contrast, the task explicitly requires an orientation specific letter processing (as in reading), children with a preference for holistic letter processing would have a disadvantage. We showed this in a number of studies (Lachmann, Steinbrink, Schumacher, & van Leeuwen 2009; Rusiak, Lachmann, & Jaskowski 2003; Rusiak, Lachmann, Jaskowski, & van Leeuwen 2007) in which we used versions of the well-known Mental Rotation task (Shepard & Metzler 1971). Letters were presented in different angles of rotation, half of them mirrored (see example in Fig. 14.4). Cooper and Shepard (1973) showed, that the time to decide whether the letter is presented “normally” or “mirrored” depends of the degree of rotation of these letters from their upright position. An analogue of a rotation is performed mentally, before a decision is made on whether the letter is “normal”; note that the latter translates into: whether or not the letter is presented in the way it was learned in the alphabetic phase (the orientation defined in the script system).

We compared Mental Rotation performance in young adults diagnosed with developmental dyslexia and IQ-matched controls with normal reading skills and found identical mental rotation processing in both groups (same slopes), but a

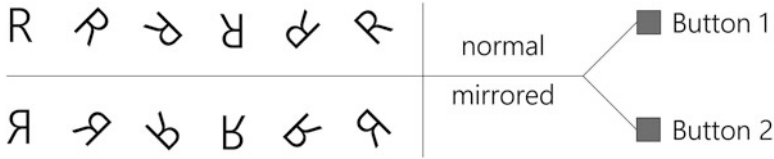


Fig. 14.4 Illustration of the Mental Rotation task (Cooper & Shepard 1973)

dramatically increased over-all reaction time in dyslexics. Visuo-spatial processes involved in mental rotation, we concluded, are not impaired in dyslexics (Rusiak et al. 2003, 2007). Instead, the overall group difference occurs late in the time course of processing, after mental rotation (Lachmann et al. 2009), when it comes to the decision whether the letter is normal or not (Lachmann 2008). Skilled readers can make this decision quickly because, based on an automatized analytic processing of the letters, symmetry generalization is suppressed. In contrast, dyslexics, who may still process letters holistically, have problems suppressing symmetry generalization. This results in extra time at the decision stage of processing (i.e., response selection, see Lachmann et al. 2009). For them, the problem is about a fast decision whether the presented letter is shown “the correct way”, i.e., as in text, as learned in the alphabetic phase. In Rusiak et al. (2007) we tested this interpretation by using letters and non-letters in same-different version of the Mental Rotation task (Shepard & Metzler 1971). We replicated the over-all group effect for the letter condition. For non-letters, in contrast, no group difference was found (see Fig. 14.5). While skilled readers showed an APPLE, here expressed by faster processing of letters, developmental dyslexics showed no significant processing difference between letters and non-letters.

In another set of experiments on letter specific processing strategies we used stimuli such as those displayed in Fig. 14.6 (Lachmann & van Leeuwen 2004, 2008a,b; van Leeuwen & Lachmann 2004, see also Fernandes et al., 2014). Using a flanker task design (Eriksen & Eriksen 1974), we investigated effects of congruence of the surrounding context on the processing of the central target. Pseudo-letters (as the one displayed in Fig. 14.6) and other non-letter targets (for instance a triangle or a circle) all showed positive effects of flanker congruence, i.e., processing was facilitated, if the surrounding was similar in shape to the central target (congruent condition). According to our terminology, this implies that the non-letter targets were processed holistically. For letters, in contrast, the surrounding shape had no effect or even interfered with processing, leading to a negative congruence effect. Negative congruence effects have also been reported in the literature for non-letter shapes (Bavelier, Deruelle, & Proksch 2000; Briand 1994; van Leeuwen & Bakker 1995), but only if the stimuli are complex and the target hard to detect among the distracting features. Bavelier et al. (2000) therefore suggested that, akin to crowding, attentional suppression of the surrounding nontarget information is needed to successfully perform such tasks. Congruence would then make it more difficult to suppress the nontarget information.

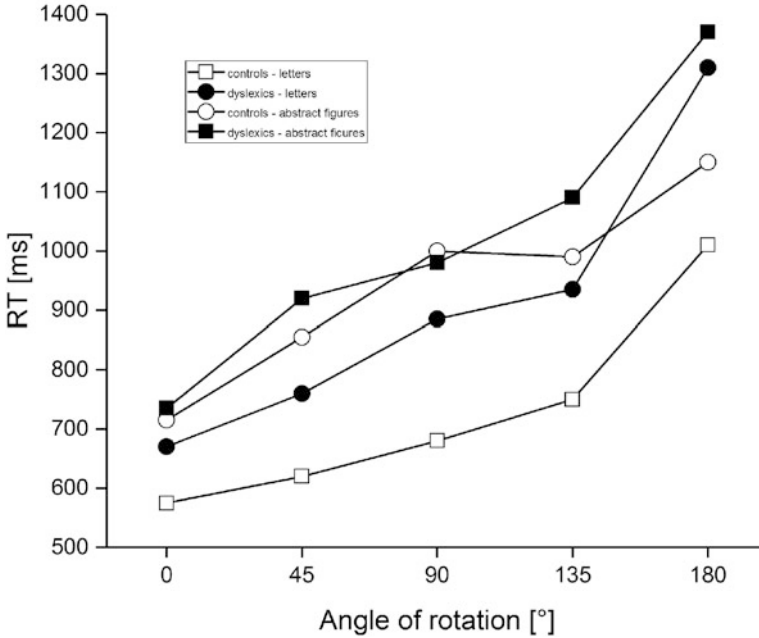
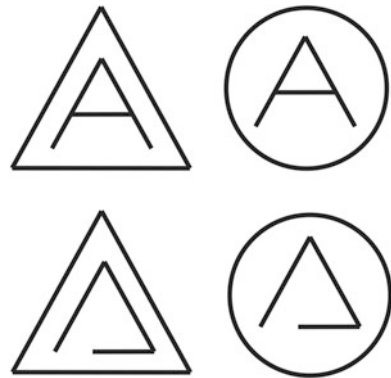


Fig. 14.5 Reaction Times (ms) as a function of angle of rotation of letters and abstract figures in children with developmental dyslexia and age-/IQ-matched controls. (Data from Rusiak et al. 2007)

Fig. 14.6 Examples of set of letters (top) and pseudo-letters (bottom) in congruent (left) and incongruent (right) surroundings as used in Lachmann and van Leeuwen (2004). See also Fernandes et al. (2014), Lachmann and van Leeuwen (2004, 2008a, 2008b), van Leeuwen and Lachmann (2004) for similar stimuli



We interpret this dissociation in processing of letters and non-letters as evidence for an APPLE: letters, in contrast to non-letters, are processed analytically. In cases where the surrounding context makes analytic processing difficult, the context is actively suppressed, resulting in smaller or even negative congruence effects: more effort is needed to suppress a congruent than an incongruent context (van Leeuwen & Lachmann 2004).

Variations of this paradigm have been informative about the strategic character of the processing dissociation between letters and non-letter shapes. First, the dissociation is task-dependent. In fact, using this paradigm, positive congruence effects in letters are possible. They appear in conditions where the task can be performed by identifying the global shape of the items (Lachmann & van Leeuwen 2004; van Leeuwen & Lachmann 2004). In one version of our experiment, van Leeuwen and Lachmann (2004) varied the task in the following way: one condition had response alternatives based on feature content (e.g., Category 1 = “A” or “circle” versus Category 2 = “C” or “triangle”); a distinction between letters and non-letters is beneficial (e.g., “A” versus “triangle”); the other condition had response alternatives based on global shape similarity (Category 1 = “A” or “triangle” versus Category 2 = “C” or “circle”). As a result, the negative congruence effect for letters was maintained in the former, but eliminated in the latter condition, where letters now showed the same congruence effects as non-letters. This shows that the holistic processing strategy is still available for letters, and is likely to be recruited if beneficial to the task. Secondly, this flanker paradigm was also used for studies including children with developmental dyslexia (Fernandes et al. 2014; Lachmann & van Leeuwen 2008b). Fernandes and colleagues replicated the aforementioned dissociation between letters and non-letters in normal readers, and found that it is absent in developmental dyslexics, depending on their phonological recoding skills. In other words, dyslexics in this study failed to apply the analytic strategy. This is in line with our results presented before, from the experiment using the symmetry effect (Lachmann & van Leeuwen 2007).

In a further study, which we (Lachmann & van Leeuwen 2014) conducted to investigate the nature of the APPLE phenomenon, we used the well-known Navon-task (Kinchla 1974; Navon 1977). As typical for this task, compound letters were used: a large F, i.e., the global level, composed of a number of identical small Fs, i.e., the local level (see the example in Fig. 14.7, left), or a large H composed of small Hs. Both form the congruent condition. In the incongruent condition, the large F was composed of small Hs or a large H composed of small Fs. Participants were instructed to respond either to the local or to the global level, while ignoring information provided at the other level, respectively.

The pattern of results typically found for this kind of task has been established in the literature (see Kimchi 2015, for a review) as the Global Precedence Effect (GPE): faster processing of the global than the local level (global advantage effect), and an asymmetric congruence effect in a way that incongruency interferes with the local-level target responses but not with global level ones. We may consider this pattern of results (GPE) as reflecting holistic processing. Thus, the GPE reported in the literature might seem to be in contrast to the proposed APPLE, i.e., that letters are preferably processed analytically. Note, however, that in previous research, the GPE was found to strongly depend on the presentation mode (Amirkhiabani 1998; Amirkhiabani & Lovegrove 1996; Han, Yund, & Woods 2003; Hübner 1997; Lamb & Robertson 1988, 1990; Poirel, Pineau, Jobard, & Mellet 2008, see Kimchi, 2015, for a review), and that the viewing conditions in which the effect was typically found do not resemble those of our flanker/symmetry studies introduced before.

Fig. 14.7 Examples of the set of compound stimuli (Kinchla 1974; Navon 1977) used in Lachmann and van Leeuwen (2014). Left side: example of a congruent letter stimulus, i.e., the local and the global level consists of the same letter F; right side: example of an incongruent non-letter stimulus (different non-letter at the local and global level)

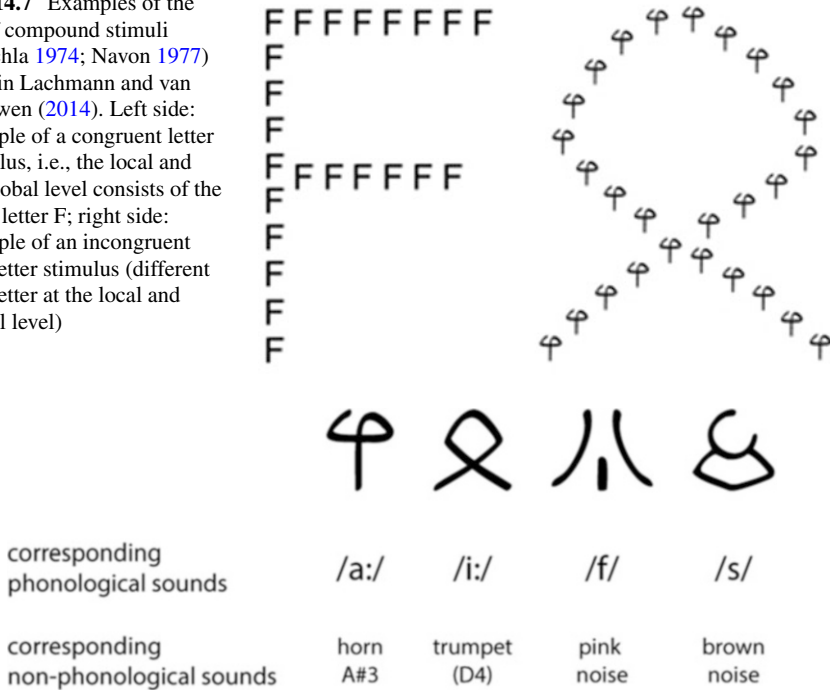


Fig. 14.8 Non-letter items in the experiment of Schmitt et al. (2017) and sounds that were associated to these in a phonological and a non-phonological subgroup

Therefore, in Lachmann and van Leeuwen (2014) we used conditions for which, according to our approach, the APPLE should be present: foveal presentation and a visual angle more closely resembling conditions of fluent reading, so the automatized reading specific visual processing strategy was more likely to be activated. With the global stimulus size close to the functional visual field in word reading and local stimuli close to the critical size for fluent reading of individual letters (Jordan & Martin 1987; Lamb & Robertson 1990; Legge & Bigelow 2011; Legge, Pelli, Rubin, & Schleske 1985), we compared the global precedence effect for letters and non-letters (see examples in Fig. 14.7, see also Fig. 14.8) in central viewing. For these conditions, we found the GPE to remain robust for non-letters. For letters, in contrast, the effect disappeared.

The dissociation in analytic and holistic processing between letters and non-letter shapes seems to depend not only on task demands (as shown above), but also on the presentation mode. The APPLE is manifest in the Navon-paradigm too, but only if viewing conditions are akin to reading. As a consequence of literacy acquisition, in reading-like conditions (cf., Legge & Bigelow 2011) and relevant task demands, an analytic visual processing strategy for letters is automatically activated, while non-letters are robustly processed holistically.

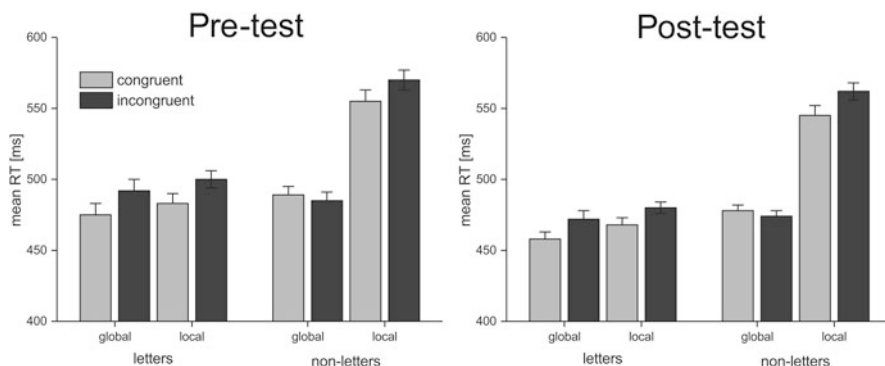


Fig. 14.9 Results from Schmitt et al. (2017). RT (ms) for letter (left side) and non-letter stimuli (right side) for congruent and incongruent trials in the local and global conditions, displayed separately for the pre-test (top) and the post-test (bottom)

Is the APPLE restricted to those configurations for which the grapheme-phoneme conversion rules were learned in the alphabetic phase, i.e., to the defined letters of the learned alphabet? What happens when the non-letters in this experiment are “converted” into “new letters”? Will these “new letters” then simply replace the cross-modal representation of the original graphemes, and will then, consequently, the APPLE appear for these too? We tested this in a training study (Schmitt et al. 2017). The same Navon-design as in Lachmann and van Leeuwen (2014) was used, but two sessions were performed, with a training in between them. In one group of participants, non-letter shapes were associated with non-phonological auditory stimuli. The other group learned phonological associations to the non-letter shapes (including fluent non-word reading with them) making these “new letters” (see Fig. 14.8). The question was, whether a relatively short training (short in comparison to the process of learning to read) could reduce or even eliminate the effects of holistic processing (GPE) for the “new letters”, i.e., only in the group with phonological training. The answer is “no”. We found no change whatsoever in the pattern of results between Session 1 and 2 in both groups; there was an APPLE for letters and there were effects holistic processing for non-letters (i.e., the GPE), no matter what kind of associations were trained (see Fig. 14.9).

A short phonological training, it seems, is not sufficient to transfer the letter-specific processing strategy to the non-letter items involved in this training. What the study by Schmitt et al. (2017) showed, however, is that the distinction in processing of letters and non-letters can be replicated between and within participants. At the same time, the results showed that an increase in familiarity with the stimuli, which is evident in the data (cf., Schmitt et al. 2017), has no effect on this distinction; familiarity alone is not sufficient to elicit analytic processing. Neither is it sufficient to have successfully learned distinctive associations to the non-letters.

14.5 Summary

We introduced the Functional Coordination Framework which describes the process of learning to read as a form of procedural learning in which pre-existing functions, mainly from the visual and auditory domain, are recruited, modified and coordinated to create a cognitive procedure optimal for reading text, which forms the basis of subsequent automatization (Lachmann 2002, 2008; Lachmann & van Leeuwen 2014). This learning process takes several years and comes along with functional and structural changes in the brain, as the learning of any complex skill.

In the present chapter, we focused on the modification of preferences of visual processing strategies, in particular, on the prevalence of analytic processing strategies for single letter processing. This modification is required in order to learn the alphabetic sub-skill of reading during the coordination stage. Analytic processing is, however, a resident skill, also present before learning to read, in dyslexics and in illiterates (Lachmann, Khera, Srinivasan, & van Leeuwen 2012), just as the other functions involved in the procedure. None of these functions is originally specific for reading what is specific is their new synthesis. It is not the case that reading acquisition implies a loss of any perceptual skills; for instance we are still able to perceive non-letter items analytically or letters holistically (Lachmann & van Leeuwen 2004; van Leeuwen & Lachmann 2004). However, while analytic processing is normally demanding and slow, automatization leads to a fast analytic letter processing, in fact faster than holistic processing.

We reviewed a number of our studies investigating the question whether literacy makes letter recognition distinctive to the processing of similar non-letter configurations in skilled readers. We showed that for letters, analytic processing is preferred, which we termed as the Analytic Processing Preference for Letters Effect (APPLE), but only as long as these are taken as part of a reading process. The habitual tendency to do so is strong enough to be manifest in our experiments, even though these used single letters outside of a reading context, as long as the task and the presentation mode are sufficiently similar to those of reading and/or if alphabetic decoding is requested or beneficial. It is the reading skill, which is special, not the letter configurations.

We further tested whether learning of pairwise associations of non-letter shapes to phonology makes these items new letters. We showed that a short training of replacing letters by non-letter is not sufficient to elicit the APPLE. It seems that the novel symbols will not easily be incorporated in the automatized skill. We still believe, however, that reading is not a matter of certain letters and sounds. These are only concretizations within a complex, higher-order procedural learning process and its automatization. After automatization, perceiving letter stimuli from the learned script may yield difficulty in suppressing their modified visual and auditory processing preferences, even though it is principally possible. Letters are then part of the automatized coordination, and they are habitually processed as such. Thus, altogether, letters are special, to an experienced reader (Duñabeitia, Dimitropoulou, Estévez, & Carreiras 2013).

Developmental dyslexia is explained within our framework as a Functional Coordination Deficit (Lachmann 2002, 2008), since at the coordination stage (see Fig. 14.1) a high degree of fine tuning of complex functions is required. Thus, developmental dyslexia is not seen as a consequence of a deficient automatization per se, but of automatization of suboptimal functional coordination.

From the point of view that a failure in learning to read is the consequence of suboptimal coordination followed by the process of automatization, it makes little sense to search for a single cause of reading problems. There might be many possible reasons for failure to become a fluent reader, like those described in various chapters of the present book or by other theories of developmental dyslexia (e.g., Bishop et al. 1999; Farmer & Klein 1995; Goswami et al. 2011; Ramus et al. 2003; Snowling 2001). All of these may lead to failures in functional coordination. Therefore, the Functional Coordination Framework is not yet another explanatory theory, but is open to multi-causal explanations and offers a structure for integrating various theories on reading and dyslexia.

Following the logic of the Functional Coordination view, isolated training of basic functions, such as basic visual-auditory integration or temporal processing alone, has only limited effects on literacy once automatization is already advanced (e.g., Schumacher 2012). After automatization, the skills must be reorganized from the beginning and then reautomatized (Klatte et al. 2014; Lachmann & van Leeuwen 2014). This also implies that learning to read is not a matter of imprinting or phase-sensitive learning related to a particular age or a life stage. As soon as the required functions, including language acquisition, are developed sufficiently, at around an age of 4 years, reading can be learned by children and by adult illiterates, all in the same way. How fast the procedure becomes automatized, depends, besides individual factors, mainly on reading practice (Duñabeitia et al. 2013).

When the learning process failed, however, as in developmental dyslexics, training in terms of increasing reading practice (i.e., “more of the same”) also has only a limited effect. It would even strengthen the suboptimal coordination and hence also the suboptimal modification of the involved cognitive functions and sub-skills, including, but not limited to, those which were the cause for the failure in coordination. Consequently, deficits in isolated functions and sub-skills that have been identified in studies to be associated with developmental dyslexia, including phonological awareness, may also reflect a consequence of suboptimal reading experience. This makes it hard to distinguish cause from effect (Hüttig et al. 2017). In the end, however, this is not even necessary, because learning must start from the beginning anyway.

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Part III
Training and Remediation

Chapter 15

The Birth of Neuroplasticity

Interventions: A Twenty Year Perspective



Paula Tallal and William Jenkins

Abstract *Fast ForWord*[®] was the first, computer/Internet delivered, neuroplasticity-based training program ever developed to enhance neural performance. It grew out of over 25 years of basic and clinical research in two distinct scientific disciplines. One utilized behavioral, electrophysiological and neuroimaging methods to study individual differences in language development and the etiology of developmental language-based learning disabilities (including Specific Language Impairment, Autism and Dyslexia). The other utilized neurophysiological and behavioral methods in animals to study neuroplasticity, that is, changes at the cellular level driven by behavioral training techniques. This chapter reviews (1) how these two lines of research were integrated to form the scientific basis of *Fast ForWord*[®] and (2) the steps taken to translate and instantiate our collaborative laboratory research into clinical and classroom interventions that could be scaled up for broad distribution around the world, while remaining efficient, effective and enduring. In 1996, Scientific Learning Corporation (SLC) was co-founded by four research scientists (Paula Tallal, Michael Merzenich, William Jenkins and Steve Miller). To date, nearly three million children in 55 countries have received *Fast ForWord*[®] interventions. On any given school day approximately 100,000 children log in to train on one of twelve *Fast ForWord*[®] *Language, Literacy or Reading* programs. More recently, *Fast ForWord*[®] language and reading programs are being used increasingly as an effective method for improving English as a second language (ESL), including success for ESL children whose first language is non-alphabetic.

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Keywords *Fast ForWord*[®] · Language impairment · Dyslexia · Central auditory processing · Autism · Neuroplasticity-based training programs · English as a second language (ESL) · Phonological processing · Phonological awareness · Reading impairment

15.1 Introduction

When we began our collaboration in 1993, the now rapidly growing fields of “cognitive neurotherapeutics” and “neuroeducation” did not exist, nor did the concept of using neuroplasticity-based training to improve “brain fitness”. The methods we developed, and subsequently were the basis of over 50 patents, were the first to use video gaming technologies with the explicit goal of improving human performance. Over the past 20 years since inception, Scientific Learning Corporation (SLC) has developed a large series of perceptual, cognitive, language, literacy and early math training exercises, “disguised” as interactive, individually adaptive, computer games, trade marked *Fast ForWord*[®]. SLC products, now delivered over the Internet, include a wide variety of individually adaptive assessment and intervention exercises that provide real time feedback and rewards as well as ongoing, digital data analysis and detailed reporting and interpretation to educators and clinicians.

15.2 Research on Language Development and Disorders

The most basic unit of any language is the phoneme, the smallest unit of sound that can change the meaning of a word. For alphabetic languages, in order to learn how to read and become a proficient reader the child must become aware that words can be segmented into smaller units of sound (phonemes) and it is these *sounds* that the letters represent. This is referred to as *phonological awareness*. Phonemes are the basic building blocks for spoken language, as well as for alphabetic written languages. Not surprisingly, phonological skills have been shown to correlate both prospectively as well as concurrently with both aural and written language ability throughout life (Tallal 2003). To improve literacy outcomes, we need to understand the *language to literacy continuum*, and how at a fundamental level phonemes of a language come to be organized or “represented” in the brain. As phonemes are, themselves, comprised of smaller, dynamically changing, acoustic spectrotemporal features, the role that complex auditory processing plays in the development of phonological systems has been a major topic of research, and one that was fundamental to the design of the series of *Fast ForWord*[®] *Language and Literacy* products.

As is the case in all other sensory modalities, the acoustic information within the complex waveform of speech can be broken down into distinct physical features (frequency, duration, amplitude), each of which is represented in fine grained detail in the auditory system (Kraus, McGee, Carrell, & Sharma 1995). According to Hebb (1949), when a complex signal occurs, all of the neurons that are activated by this complex set of features, per unit time, fire together. The likelihood that a particular pattern will come to be “represented” increases with each additional exposure of a neural firing pattern ensemble. It has been increasingly documented that phonological systems are developed through exposure to the native language(s) (Kuhl et al. 1997). Each language has its own set of phonemes that must be learned from experiencing repeating acoustic patterns within the ongoing speech waveform. As infants are exposed to a continuous speech stream from the environment, they must parse the incoming acoustic signal into consistent, replicable chunks of time that occur in statistically predictable sequences (Saffran, Aslin, & Newport 1996). It is hypothesized that when sensory inputs enter the nervous system differentially in time, the neuronal representation develops as distinct and separable. However, when information enters the nervous system either simultaneously, or within a critical window of time (tens of milliseconds (ms)) that is too rapid to separate, the information is “bound” together and thus is neurally coded as a unit (Wang, Merzenich, Sameshima, & Jenkins 1995). It is in this way that the many different physical features of a complex stimulus, such as speech, are combined (bind together) to form a unified phonological percept (Tallal 2004). For speech, statistically consistent acoustic patterns occur frequently and consistently within the ongoing acoustic waveform in chunks of various durations. Chunking within the tens of ms time window will allow for the fine grain analysis needed to represent the acoustic differences between individual phonemes such as /b/ and /d/. Chunking over longer periods of time (hundreds of ms) will result in firing patterns consistent with syllable length representations (Hickok & Poeppel 2007). It is these chunks of acoustic information, that form the building blocks for language, that can be infinitely combined to form both spoken and written words. Furthermore, based on optimal control theory (Todorov 2006), it is hypothesized that similar unsupervised statistical learning processes are at play for learning how words are combined to form sentences that are consistent with the specific grammatical rules that govern how each language is constructed.

While most children develop language without the need for explicit training, a growing number of children are entering formal education without sufficient language skills to support proficient written language and literacy development. Children who fall within the lower end of the continuum of individual differences in language development are diagnosed as having “language-based learning disabilities”. In addition to linguistic studies that focus on describing the differential pattern of language development in these populations of children (Leonard 1998), research has focused on delineating basic domain-general sensory, motor and cognitive deficits that consistently co-occur with language and reading impairments (for reviews see, Tallal 1980, 1993, 2004).

15.3 Research on Neuroplasticity-Based Training

Neurophysiologists have mapped the features of the sensory world at the single cell level. This research has shown that within each sensory modality the features that represent the physical world come to be mapped at the cellular level in a highly organized fashion. For example, in the auditory modality, there is a tonotopic representation of frequency such that cells that fire to a specific frequency are located physically adjacent to cells that fire to the next higher frequency, in a continuous manner throughout the frequency range (Clopton, Winfield, & Flammino 1974). In addition to tonotopic (frequency) representation in A1, there are neurons that code selectively for temporal features of sound (Eggermont, Aertsen, Hermes, & Johannesma 1981), as well as “inseparable” temporospectral combinations such as frequency sweeps similar to those occurring within formant transitions of speech (Orduna, Mercado, Gluck, & Merzenich 2001). That these sensory maps must be learned from environmental exposure is evidenced by neurophysiological research showing the effects of sensory deprivation or alteration (Neville 1985). Exposure to altered acoustic input during critical periods of early development, for example continuous or pulsed noise, significantly disrupts the development of tonotopic representation in primary auditory cortex, and these developmental changes continue to be evident into adulthood. Merzenich and colleagues have shown that beyond this early period these sensorineural maps can no longer be altered by mere exposure. Rather, alteration of neural maps requires active attention to highly repetitive and explicit input features in the context of an intensively trained, individually-adaptive task, with timely feedback and reinforcement of correct responses (Chang & Merzenich 2003; Jenkins, Merzenich, Ochs, Allard, & Guic-Robles 1990; Recanzone, Schreiner, & Merzenich 1993; Zhang, Bao, & Merzenich 2001). This form of training is referred to as *neuroplasticity-based training*.

15.4 The Birth of *Fast ForWord*[®]: Translating Theory into Practice

Considering the amount of speech directed to the infant, it is easy to understand how important speech is in shaping the auditory cortex during critical periods of human development. Three decades of research with children with developmental language and literacy impairments has shown that these children are at high risk for having specific deficits in processing the brief, rapidly successive, temporospectral cues within ongoing speech (Tallal 2003). Furthermore, we demonstrated as early as 1975, using computer synthesized speech syllables, that language impaired children’s perception could be substantially normalized, at least at the single syllable level, by *extending in time* rapid temporospectral, intra-syllabic cues (Tallal & Piercy 1975). These findings from research with children with language-learning

impairments (including dyslexia) were combined with neuroplasticity-based training research to develop *Fast ForWord*[®].

In 1993, the Tallal lab at Rutgers University, Newark began collaborating with the Merzenich lab at UCSF. We began with parallel research goals: (1) determine whether basic non verbal auditory temporal thresholds could be improved (decreased) in children, specifically those with language learning impairments (LLI) who were characterized by rapid auditory processing (RAP) deficits, (2) develop a speech processing algorithm that would selectively find, amplitude enhance and extend in time (*acoustically modify*) the most rapidly changing acoustic changes (3–30 Hz) that occurred in real time within ongoing speech (see Nagarajan et al. 1998, for details) and (3) determine if training children with LLI to process basic auditory temporospectral cues faster, while simultaneously training speech and language skills using this *acoustically modified speech*, would lead to improvements in speech and/or language abilities. Specifically, we hypothesized that the “scientific learning principles”, that had been shown in studies with monkeys to drive neuroplasticity in sensory maps, might be adapted for use with children with LLI to ameliorate their rate processing constraints. These “scientific learning principles” include training that is: (1) applied with a heavy schedule of intensive practice trials (repetition, repetition, repetition), (2) spaced across a series of successive days, (3) individually-adaptive – moving from easy to harder trials – while maintaining high levels of performance (about 80%) to drive continuous performance improvements, and (4) conducted under conditions of high motivational drive (continuous feedback, correction of errors and salient rewards for correct trials).

Simultaneously, we hypothesized that we would be able to improve aural language skills by training a wide variety of linguistic skills, initially using *acoustically modified* speech. However, rather than having children depend long-term on acoustically modified speech for improved speech processing, our aim was to develop neuroplasticity-based training procedures that would individually adapt based on a child’s linguistic performance from the acoustically modified “slowed down” speech to natural “fast” speech. Specifically, a hierarchy of training exercises, “disguised” as computer games (that ultimately evolved into a series of computer/Internet base training programs marketed under the trade name *Fast ForWord Language*[®]) was developed (1) to drive neural processing of rapidly successive acoustic stimuli to faster and faster rates, (2) to improve foundational cognitive skills such as attention, memory and sequencing and (3) to improve speech perception, phonological analysis and awareness, and language comprehension. We aimed to do this by providing intensive daily training exercises within various linguistic contexts (phonological, morphological, semantic and syntactic) that utilize speech stimuli that had been acoustically modified to amplify and temporally extend the brief, rapidly successive intra-syllabic cues within ongoing speech.

15.5 Designing Neuroplasticity-Based Training Games

For our first study we designed and developed a series of verbal training exercises ranging from speech discrimination to grammatical comprehension, disguised as “games”. Some of these games were implemented on computers, while trained professionals using tape-recorded stimuli presented others. In addition to explicitly training perceptual and linguistic skills, all were developed in a training format that also aimed to simultaneously increase foundational cognitive skills including auditory attention, speed of processing, sequencing and memory span. For example, as seen in Fig. 15.1a, one of the speech processing exercises was designed in the format of a “concentration game”. In this game a series of squares were laid out on the computer screen in a visual grid. When each square was clicked, a syllable was presented acoustically. The goal of this game was to find two syllables that matched. When clicked sequentially they would disappear and a point would be earned. To enhance attention, memory and motivation, bonus points were given when the screen was cleared with the fewest number of clicks. The task began at an easy level, with only 4 squares and two pairs of syllables that were acoustically

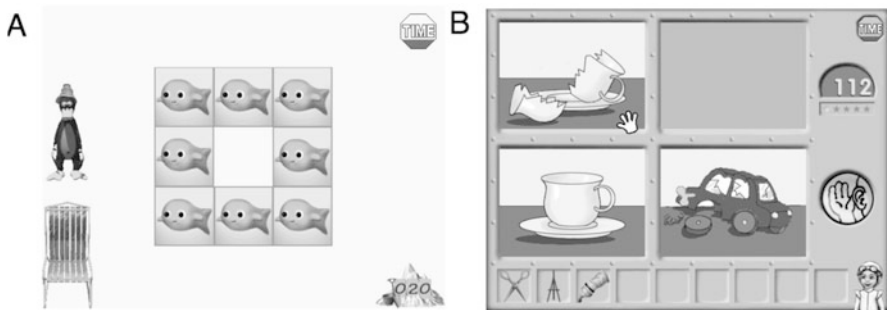


Fig. 15.1 Screen Shots from two *Fast ForWord*® Language V1 games are shown. (a) (top) shows one of the speech processing exercises (Phonic Match) that was designed in the format of a “concentration game”. The task begins at an easy level, with only 4 squares and two pairs of syllables that were acoustically easy to discriminate. As each player progresses, the number of squares in the grid increases (this screen shot shows 9 squares) while the acoustic difference between syllables decreases. (b) (bottom) shows the game Language Comprehension Builder, adapted from Curtiss and Yamada Comprehensive Evaluation of Language (Curtiss & Yamada 2013). This exercise trains each rule of English grammar. To initiate a trial a participant clicks on the hand on the ear button to indicate that they are ready to listen. A command is presented acoustically, in this example, “Point to, the cup is broken”. One correct picture as well as two or three foils were carefully designed to assure that comprehension of a specific grammatical rule, not just vocabulary, is required to answer each command correctly. Correct responses are rewarded by a “ding” sound and winning of a sticker along the bottom and a point is added to the point counter. Incorrect responses are indicated by a “clunk” sound, the command is repeated, and the correct picture is highlighted. To assure that memorization of commands does not occur, the same set of picture is also used for the command, “The cup is not broken” and picture positions are randomized. (From Scientific Learning Corporation)

easy to discriminate. As each player progressed, the number of squares in the grid increased (memory training) while the acoustic difference between syllables decreased (speech discrimination training). Thus, both speech discrimination and memory skills were simultaneously being trained and individually adapted. Another exercise was presented as a board game with colored circles and squares. Children pressed an orienting button to receive a command. Commands, initially presented with the highest degree of acoustic modification, began at a very easy level such as “touch the red square”. As a child progressed through the game the number and size of circles and squares presented, as well as the difficulty of the commands, increased based on each participant’s trial-by-trial response. The goal was to present commands of increasing length and grammatical complexity, such as, “Before touching the large blue circle, put the small red square between the large white square and the large blue square”. As participants advanced, the degree of acoustic modification decreased back to normal, fast speech. This exercise “cross-trained” sustained attention, sequencing, serial memory and grammar in the context of listening comprehension. Yet another exercise (shown in Fig. 15.1b) was designed to train English grammatical rules. The commands used in this game were licensed from the Curtiss and Yamada Comprehensive Evaluation of Language (CYCLE ©). We adapted this comprehension assessment that was based on years of laboratory research on the progression of normal and delayed language development (Curtiss & Yamada 2013) in to a training exercise designed to train the rules of English grammar. These speech discrimination and language exercises were developed to be individually adaptive; the goal being to find for each child a level of cognitive and linguistic functioning that could be responded to at a high rate of accuracy (approximately 80% correct). As the exercise progressed the goal was to move towards more rapid and less amplified, natural speech following correct linguistic responses.

In addition to the speech discrimination and language comprehension exercises, one game was designed to increase the speed of auditory processing. This computer game used adaptive training with the goal of driving more efficient (shorter) temporal integration thresholds for rapidly successive acoustic sweep tones (computer generated tones that sweep from either high frequency to low frequency, or low to high). To increase generalization across the entire frequency range of speech, three different frequency ranges were selected that covered the frequency range of human speech. As a child progressed in this game, the sweep tone stimuli were individually adapted to decrease in the duration of the tones, the ISI between tones, based on each child’s trial-by-trial performance. The overarching goal of these combined verbal and non-verbal training exercises was to drive, through adaptive training, each child into the normal processing rate of tens of milliseconds (the range important for phoneme perception) while simultaneously increasing each child’s ability to process more complex linguistic structures.

15.6 The First Laboratory Studies: Rutgers Summer Camps 1994–1995

Our initial laboratory studies were conducted with children who each met the criteria for language learning impairment (LLI). Two groups matched on age, IQ and language skills were quasi-randomly assigned to receive the same language intervention program. However, one group (Experimental group) received the intervention with acoustically modified speech and auditory temporal training with sweep tones, while the other (Active Treatment Control group) received the same language intervention, but with normal speech, and a non-temporally adaptive visual exercise instead of the auditory temporal training.

The results of these experiments were published in two back-to-back papers in *Science* in 1996. The language results are reported in Tallal et al. (1996). Traditionally, few children with language-learning impairments receive more than one or two short (30–60 min) sessions of individual or group speech therapy per week and little progress is expected to occur within only four weeks of clinical intervention. In contrast, results from this study demonstrated that the rigor, scope and consistency of the training (100 min per day, five days per week, for four weeks) resulted in significant improvements in speech discrimination, language processing and grammatical understanding for *both* groups of children. This result has important clinical implications for the intensity of speech therapy that needs to be provided for children with language-based learning disabilities. In addition to showing the benefit of more intensive intervention, this controlled laboratory study also demonstrated the added benefit of providing language intervention using acoustically modified speech and auditory temporal training. Results showed that the Experimental group, who received language intervention training *with* acoustically modified speech, demonstrated *significantly better outcomes* in speech discrimination, language processing and grammatical understanding than the Treatment Control group who received the same language intervention, but with natural (unmodified) speech.

The second paper reported the results of the non-verbal rapid auditory sequencing training that the Experimental Group received for 20 min a day, five days a week, for four weeks (Merzenich et al. 1996). This study showed for the first time that basic auditory thresholds are highly modifiable in children by behavioral training. Importantly, this study also showed that the measured improvement in a child's auditory temporal threshold for correctly segmenting and sequencing successive nonverbal auditory sweep tones was significantly correlated with post-training outcomes in real-time language processing ($r=0.81$, $p < 0.05$). That is, the amount of improvement a child made in non-verbal auditory processing speed was highly correlated with the amount of improvement that child made in language comprehension.

The results of these active, treatment controlled laboratory studies demonstrated the immediate efficacy of this novel training approach. However, it was also important to determine the longer-term effectiveness of this brief, but intensive, training. To address this question the children were assessed again at six weeks

as well as six months after training had concluded (Bedi, Miller, Merzenich, Jenkins, & Tallal 1999). Results of these follow up studies showed that (1) all children continued to make progress relative to their performance after four weeks of training, (2) the Experimental group continued to perform significantly better than the Treatment Control group and not only maintained their initial gains, but continued to improve at an accelerated pace during the six weeks following the conclusion of the program and (3) these results were maintained out to six months. These results provided strong evidence for the longer-term efficacy and endurance of this new training approach.

15.7 Scaling Up: The “Neurotherapeutic Revolution”

15.7.1 Fast ForWord® Language v1

15.7.1.1 First Multi-site Clinical Field Trial (1996–1997)

It is one thing to obtain results in well-controlled studies in a research laboratory under the direct supervision of skilled research scientists. It is quite another to demonstrate that efficacy can be achieved in “real-world” clinics and classrooms where children most commonly receive intervention. Soon after founding Scientific Learning Corporation (SLC) our first goal was to convert the games used in our laboratory studies into a fully computerized training program (*Fast ForWord*® Language v1), and then to conduct large-scale field trials in clinical and educational settings to assess its “real-world” efficacy. The purpose of this trial was to determine whether the efficacy that was demonstrated in the laboratory could be replicated in clinics and classrooms with fully computerized exercises, under the supervision of clinicians and teachers (rather than trained researchers).

The first field trial included over 500 children, aged 4 to 14 years, identified by 60 professionals, at 35 clinical or educational sites. We designed a two-day training workshop to assure that each of the professionals participating in this field trial understood the criteria for subject selection, how to set up and deliver the *Fast ForWord*® Language v1 training, and were clinically certified to select and give standardized pre- and post-training assessments. At each site, these independent speech language professionals collected all of the pre- and post-standardized test data as well as administered the *Fast ForWord*® Language v1 training. Children who were receiving speech therapy and scored at least one or more standard deviations below the mean on standardized tests in the area of central auditory processing, speech discrimination and/or language comprehension were eligible for inclusion. Case history records indicated that children who met these study criteria had one or more of the following diagnostic classifications: specific language impairment (SLI), attention deficit disorder (ADD), pervasive developmental disability (PDD), autism, central auditory processing disorder (CAPD), dyslexia or learning disability (LD).

A summary of results from this first field trial are presented in Miller et al. (1998), comparing pre-*Fast ForWord*[®] Language v1 training standardized test scores to post-training standardized scores. On average, the 35 sites reported convincing evidence that clinicians can be trained to provide *Fast ForWord*[®] Language v1 in their clinics and classrooms according to the prescribed protocol. 90% of the children experienced significant gains in one or more tested area. Most children made significant gains in multiple areas, including central auditory processing, phonemic awareness, listening, speaking, attention, language fundamentals, grammar, and ability to follow directions. On average, children advanced 1 to 2 years, based on standardized tests, following 4 to 8 weeks of *Fast ForWord*[®] Language v1 participation. Significant improvement was obtained not only in areas targeted by *Fast ForWord*[®] Language v1 training, but also generalized beyond areas directly trained. Areas that were not directly trained, but showed significant improvement after *Fast ForWord*[®] Language v1 training included: improved expressive (spoken) language abilities, as well as some early reading (phonological awareness) abilities, despite the fact that no letters were included in any of the *Fast ForWord*[®] Language v1 exercises.

There was considerable variability across children as to the degree and pattern of improvements they made across domains, as would be expected, based on the variety of symptomatology and clinical classifications of this large heterogeneous group of children with language learning problems. Figure 15.2 shows that significant efficacy was obtained for a much broader group of children than had been included in the initial laboratory studies. Further analysis showed that differences in the degree of efficacy were *not* based on the child's clinical diagnostic classification, age, gender or degree of impairment. That is, this intervention method was shown to improve language skills in both male and female children between the ages of 4 and 14 years of age with a variety of clinical diagnoses and across a broad range of language functioning. These results are significant not only in magnitude of improvement, but specifically in light of the very brief period of time (weeks rather than years) over which the intervention (training) was provided.

15.7.2 *Fast ForWord*[®] Language v1: University-Based Studies

After our first studies were published in *Science* there was a flurry of university-based studies that followed. Most of these studies focused on investigating the effectiveness of *Fast ForWord*[®] Language v1 for children with specific language impairment or dyslexia using behavioral, electrophysiological and/or neuroimaging outcome measures.

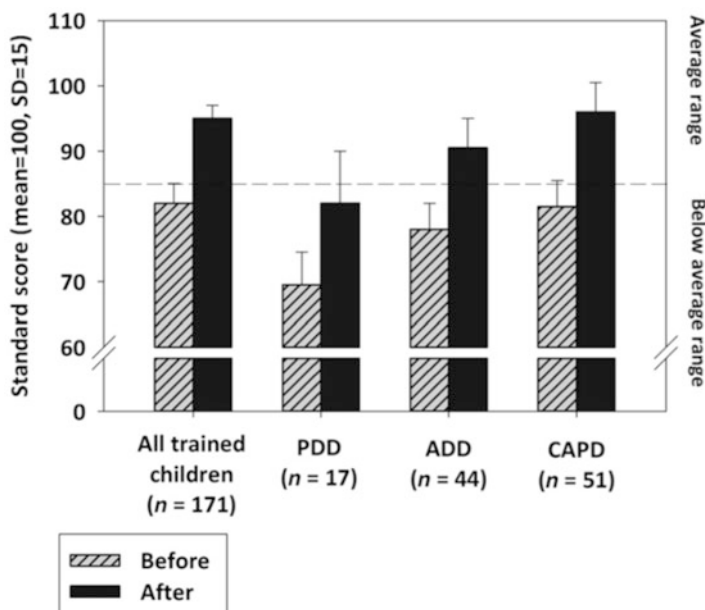


Fig. 15.2 Comprehensive language quotients (Test of Language Development and Comprehensive Evaluation of Language Function) for all children pre- and post-*Fast ForWord*[®] training. Scores are shown for all children with language impairment (LI) combined, as well as for LI children diagnosed clinically as having pervasive developmental disorder (PDD), or with co-morbid attention deficit disorder (ADD), or central auditory processing disorder (CAPD). Although the degree of language deficit differed at pre-test among these groups of children (with children diagnosed as PDD having the most severe language disorder and CAPD having the least severe), there were no significant differences in the magnitude of improvement across groups achieved with training. All groups were 1 or more SD below the mean at pre-test and showed significant improvement ($p < .0001$) from pre- to post-training. Although the PDD group improved significantly following training, they still remained more than 1 SD below the mean following training, based on these test batteries. The children with language impairments co-morbid for ADD or CAPD entered the study with pre-test scores more than 1 SD below the mean, while their average test scores approached the normal median post-training. (Adapted from Tallal et al. 1998)

15.7.2.1 Neurophysiological and Neuroimaging Studies of *Fast ForWord*[®] Language v1

The first study reporting neurophysiological changes following *Fast ForWord*[®] Language v1 training, was conducted in the laboratory of John Gabrieli at Stanford University in collaboration with a school specialized in educating children diagnosed with developmental dyslexia (Temple et al. 2003). Children with dyslexia received *Fast ForWord*[®] Language v1 training at their school under the supervision of a licensed *Fast ForWord*[®] Language v1 provider. Both pre- and post-training standardized language and reading tests as well as fMRI were performed on 20 children with dyslexia (8–12 years old) and age match typical readers. fMRI

data were collected while children performed a phonological processing task in the scanner. Behavioral results showed that training significantly improved both oral language and reading performance in the dyslexic children. Physiologically, children with dyslexia showed “normalized” metabolic activity in multiple brain areas that had been shown in the pre-training scan to be aberrant in comparison to the typically reading matched control group. After training, the children with dyslexia showed increases in left temporo-parietal cortex and left inferior frontal gyrus, bringing brain activation in these regions closer to that seen in typical readers. Increased activity was observed also in right-hemisphere frontal and temporal regions and in the anterior cingulate gyrus. Importantly, children with dyslexia showed a significant correlation between the magnitude of increased activation in left temporo-parietal cortex and their improvement in oral language abilities. These results suggest that a partial remediation of auditory and language-processing deficits, resulting in improved reading, ameliorates disrupted function in brain regions associated with phonological processing and also produces additional compensatory activation in other brain regions.

A second paper (Gaab, Gabrieli, Deutsch, Tallal, & Temple 2007) was published based on the same population included in the Temple et al. (2003) study. These authors had previously reported that, in comparison to normal readers, adults with developmental dyslexia have a disruption in the *left* prefrontal cortex neural response to non-linguistic acoustic stimuli, designed to mimic the spectro-temporal structure of consonant-vowel-consonant speech syllables, when presented with either rapid (20 ms) or slower (200 ms) transitions (Temple 2002). The goal of this study was to replicate the previous study, but using children instead of adults. Using the same non-speech analogue stimuli, whole-brain fMRI was performed on twenty-two children with developmental dyslexia and twenty-three typical-reading children. As had been found in adults, fMRI results with these non-speech stimuli, differing only in onset and offset transition durations, demonstrated that while typical-reading children showed activation for rapid compared to slow transitions in *left* prefrontal cortex, children with developmental dyslexia failed to show any differential metabolic response in these regions to rapid versus slow transitions. Remarkably, after only eight weeks of remediation with *Fast ForWord*[®] Language v1 the children with developmental dyslexia not only showed significant improvements in language and reading skills, but also exhibited “normalized” activation for rapid relative to slow transitions in left prefrontal cortex. These authors concluded that the presence of a disruption in the neural response to rapid stimuli in children with developmental dyslexia prior to remediation, coupled with significant improvement in language and reading scores and increased brain activation after remediation, gives further support to the inclusion of training aimed at increasing rapid auditory processing in interventions for children with reading disabilities.

These fMRI studies were followed by studies using electrophysiological measures. A study done in Helen Neville’s lab, (Stevens, Fanning, Coch, Sanders, & Neville 2008) focused on selective attention. Specifically, they examined whether six weeks of high-intensity (100 min/day five days per week) training with *Fast ForWord*[®] Language v1, would influence neural mechanisms of selective auditory

attention previously shown to be deficient in children with specific language impairment. Twenty children received *Fast ForWord*® Language v1 training in the Neville lab, including 8 children diagnosed with SLI and 12 children with typically developing language. An additional 13 children with typically developing language received no specialized training (NoTx control group), but were tested and retested after a comparable time period to control for maturational and test-retest effects. Before and after training (or a comparable delay period for the NoTx control group), children completed standardized language assessments and an event-related brain potential (ERP) measure of selective auditory attention. Relative to the NoTx control group, both groups of treated children showed significant increases in standardized measures of receptive language as well as larger increases in the effects of attention on neural processing following training. The enhanced effect of attention on neural processing represented a large effect size (Cohen's $d=0.8$), and was specific to changes in signal enhancement of attended stimuli. These findings indicate that the neural mechanisms of selective auditory attention, previously shown to be deficient in children with SLI, can be remediated through training and can accompany improvements on standardized measures of language. This was also the first study to show that significant enhancement of language and attention could be achieved not only in children with language impairments, but also in typically developing children. Kujala et al. (2001) found dyslexia remediation effects by presenting non-linguistic audiovisual pattern formations to children over a period of 6 weeks time. This study shows that reading gains and their underlying neural improvements can be achieved without any type of linguistic material. The results by Gaab et al. (2007) are consistent with those reported previously by Kujala et al. (2001). Taken together these studies provide strong support for the importance of including explicit non-linguistic audio-visual pattern training as an essential component in remediation methods aimed at improving reading in dyslexia.

This chapter presents work based on a training program designed in the US and Chap. 16 presents work based on a training program designed in Germany, both of which are alphabetic languages. It is interesting to question the extent to which training programs designed to target acoustic, phonological and morphological aspects of language and reading might be efficacious for helping individuals who are struggling to learn English as a second language (ESL), even those whose reading orthography in their first language is character based, such as Chinese. In alphabetic language systems, converging evidence indicates that developmental dyslexia represents a disorder of phonological processing, both behaviorally and neurobiologically. However, it is still unknown whether, impaired phonological processing remains the core deficit of impaired English reading in individuals with English as their second language, and if so, how it is represented in the neural cortex. A study by You et al. (2011) addresses this interesting question. Using functional magnetic resonance imaging, this study investigated the neural responses to letter rhyming judgment (phonological task) and letter same/different judgment (orthographic task) in Chinese school children who either were, or were not, struggling to learn to read English as a second language. Recall, this was a similar study design used by Temple et al. (2003) with English speaking typical and dyslexic readers.

For the Chinese children struggling to learn to read English, an independent region of interest analysis showed reduced activation in occipitotemporal regions during orthographic processing, and reduced activation in parietotemporal regions during phonological processing. These results are consistent with the results reported for English native speakers reported by Temple et al. (2003). These authors concluded that similar neural deficits are involved for children struggling to learn to read English, regardless of whether the orthography in their first language is alphabetic or character based. These findings have implications for reading remediation, educational curriculum design, and educational policy for Chinese children learning English as a second language.

Also using electrophysiological methods, Heim, Keil, Choudhury, Thomas Friedman, and Benasich (2013) examined the extent to which early oscillatory responses in auditory cortex change after *Fast ForWord*[®] Language v1 training. They use combined source modeling and time-frequency analysis of the human electroencephalogram (EEG). Twenty-one elementary school students diagnosed with LLI received *Fast ForWord*[®] Language v1 training for an average of 32 days in a clinical setting under the supervision of a speech language pathologist experienced in delivering this program. Pre- and post-training assessments performed in the laboratory included standardized language/literacy tests and EEG recordings in response to fast-rate tone doublets. Twelve children with typical language development were also tested twice, with no intervention given. Results showed that the LLI children made significant gains in receptive language skills post-training. Furthermore, during the first EEG assessment (pre-training), reduced amplitude and phase-locking of early (45–75 ms) oscillations in the gamma-band range (29–52 Hz), specifically in the LLI group, was observed for the second stimulus of the tone doublet. After *Fast ForWord*[®] Language v1 training, amplitude reduction for the second tone in the sequence was no longer evident for the LLI children, although these children still exhibited attenuated phase-locking. These findings suggest that specific aspects of inefficient sensory cortical processing in LLI are ameliorated after training.

Physiological improvements in auditory function were also reported for children on the Autism spectrum after *Fast ForWord*[®] Language v1 training (Russo, Hornickel, Nicol, Zecker, & Kraus 2010). Children with Autism Spectrum Disorders (ASD) share many common deficits to children diagnosed with SLI, including receptive language and auditory processing deficits. While these children have been included in field studies previously, and shown to benefit from *Fast ForWord*[®] Language v1 training, this was the first laboratory-based study to address whether the overarching language impairment that characterize children with pervasive developmental disorders (PDD), such as children with autism spectrum disorders (ASD), may potentially be alleviated through training-induced improvements in auditory processing. To assess the impact of auditory training on auditory function in children with ASD, brainstem and cortical responses to speech sounds presented in quiet and noise were collected in the laboratory of Nina Kraus at Northwestern University from five children with ASD. These children received training by their clinician who had extensive clinical experience providing *Fast ForWord*[®] Language

v1 training to children with ASD. The results showed that relative to six control children with ASD, who did not complete *Fast ForWord*[®] Language v1, training-related changes were found in brainstem response *timing* in three of the five children with ASD, while changes in pitch-tracking was found in only one ASD child. In addition, all five of the trained children with ASD showed improvement in *cortical response timing* after *Fast ForWord*[®] Language v1 intervention.

15.7.2.2 Studies Using Behavioral Outcome Measures Only

It cannot be overemphasized enough that *Fast ForWord*[®] differs from standard clinical and educational approaches in many ways. Perhaps the most important, based on extensive laboratory and field research, is the extent to which efficacy depends on rigorously following a prescribed training protocol and method of delivery. It is for this reason that Scientific Learning Corporation only provided *Fast ForWord*[®] Language v1 to speech/language professionals who had been trained and certified to use this program according to the prescribed protocol. Unfortunately, many of the early studies published in the research literature failed to use certified professionals or to follow the prescribed protocol. As a result many of these early studies suffered from very poor or non-standard implementations and, not surprisingly, failed to demonstrate efficacy. Five of these early studies constitute the only data used in an influential meta-analysis published by Strong, Torgerson, Torgerson, and Hulme (2011). Unfortunately, there were many flaws in this publication. Of the over 200 studies on *Fast ForWord*[®] Language v1, based on the extensive laboratory and field research that Strong et al. reviewed, remarkably they decided that only six studies met their arbitrarily selected inclusion criteria, and one was dropped for lack of available data. Thus, only five of over 200 studies that were included in this supposed “meta-analysis”. It should be pointed out that several studies published in excellent peer-reviewed scientific journals, that had positive results, were excluded for unconventional reasons. Surprisingly, of the five studies that did make the cut for this “meta-analysis”, three acknowledged in the publication that they had very poor compliance; for example, one stated that nearly 40% of the post-test outcome data were either missing or unreliable! Another study used parents rather than trained professionals to deliver the program to their own impaired child at home. It also is important to emphasize that regardless of when the study was actually published, all of the data included in this “meta-analysis” were collected prior to 2005 using two very early, and now discontinued, versions of *Fast ForWord*[®] Language v1.

Of the five studies included in the Strong et al. “meta-analysis” 2011, the Gillam, et al. NIH funded trial published in 2008 had by far the best implementation, albeit did not use certified *Fast ForWord*[®] professionals. The results from this study demonstrated that students who used the *Fast ForWord*[®] Language v1 products for 50h achieved statistically significant improvements in language and reading skills – improvements comparable to an active control group receiving 50h of *one-on-one* speech therapy with a licensed speech language pathologist (SLP). In

this trial, 74% of the language impaired children who received *Fast ForWord*[®] Language v1 had follow-up scores that were significantly higher than their pre-test scores six months after treatment ended. In addition, those children who received computer-based interventions significantly outperformed the active control groups (who received a comparable amount of *one-on-one* speech therapy) in early reading skills, specifically phonological awareness. Despite these highly positive outcome data, Strong et al. (2011) published the following highly misleading conclusion: “There is no evidence from the analysis carried out that *Fast ForWord* is effective as a treatment for children’s oral language or reading difficulties.” This statement is blatantly incorrect, biased and in direct conflict with the actual data reported in the NIH trial.

In the years since these early studies were conducted, SLC has significantly improved the *Fast ForWord*[®] language programs and developed two new versions: *Fast ForWord*[®] Language v2 for elementary school children and *Fast ForWord*[®] Literacy for middle and high school students. We have also developed a comprehensive series of five *Fast ForWord*[®] Reading programs (Levels 1–5). Efficacy for using these programs serially has been shown for children from pre-K to 12th grade. Recently using a combination of *Fast ForWord*[®] Literacy and *Fast ForWord*[®] Reading programs, the efficacy for older students struggling with both reading and writing skills was established in college students (Rogowsky, Papamichalis, Villa, Heim, & Tallal 2013). We have continuously used feedback and actual data from our end users to create much more efficient and effective protocols and provided better training and support to schools and clinics. We encourage scientists and educators to review the data derived from more than two hundred independent studies conducted by schools and clinics in “real-world” settings as well as more current laboratory studies. These studies show unequivocally that when used according to protocol, with high fidelity and compliance, the suite of *Fast ForWord*[®] training programs are highly effective in improving language, reading, writing and cognitive skills from kindergarten through college students.

A more recently published comprehensive review of laboratory studies, that explored the behavioral and neural basis of changes induced by auditory or phonological training in dyslexia, SLI and LLI, is consistent with the results from “real world” school studies. This review showed that auditory and phonological training strengthened previously weak auditory brain responses in children with language based learning disorders (Ylinen & Kujala 2015). Specifically, for individuals with dyslexia, results consistently showed increased or normalized activation of previously hypoactive inferior frontal and occipito-temporal areas. Ylinen & Kujala (2015) concluded that the combination of results across many studies, showing remedial gains derived from both behavioral and brain measures, not only increases our understanding of the causes of language-related deficits, but also helps target remedial interventions more accurately to the core problem.

15.8 Independent Agency Evaluations of *Fast ForWord*®

Studies on the effectiveness of educational and/or clinical interventions are inherently difficult, in part because of the many skill sets and multidisciplinary collaborations required to conduct these studies in “real- world” clinics and school settings. Before introducing a new method, curriculum or product, schools have to answer a practical question: does the new approach leads to better outcomes for their students than *whatever* intervention strategies they currently have in place? In translating research from the laboratory to classrooms, we have found that most school administrators and curriculum directors are only willing to make important decisions for their school after they have conducted their own, internal, independent study. As a result, hundreds of independent school-based studies, some of them RTC, of one or more levels of *Fast ForWord*® Language, Literacy and/or Reading, have been conducted by educators and clinicians in their own schools or clinics. Unfortunately, these professionals rarely share these data or publish them in the form of peer-reviewed academic publications, nor do they routinely read academic journals. Conversely, research scientists interested in topics such as dyslexia generally do not work in schools or K-12 education settings, nor are they willing to accept data that have not been published in peer-reviewed academic journals. This lack of communication between research scientists focused on topics of importance to education (such as dyslexia), and the educators themselves who could make most use of these data, makes translation from the laboratory to the classroom exceedingly difficult. In an attempt to bridge this gap, several independent agencies have been created, such as the US Department of Education’s Office of Special Education Programs (OSEP), the National Center on Response to Intervention (NCRTI) and The What Works Clearing House (WWCH). These agencies have developed stringent and consistent metrics for evaluating both study quality and designs and outcome data from education research studies, based on the quality of a study rather than whether or not it has been published in a peer-reviewed academic journal.

The National Center on Intensive Intervention (NCII) is funded by the US Department of Education at the American Institutes for Research. As part of their mission to help educators implement data based individualized instruction, NCII reviews studies on various educational interventions used with struggling students, and publishes their analyses. NCII reviews focus on the degree to which intervention studies meet the following criteria: Participants: at-risk students in Grades K-12; Study design: two group study, preferably with random assignment, comparable initial skills and demographics between the two groups, and no attrition bias; Fidelity of implementation: data showing the program was used as designed; Study measures: accurate (psychometrically reliable) and important (relevant to the program’s instructional content). Targeted measures assess skills targeted by the intervention. Broader measures assess related aspects of competence. NCII reviews also report the effect size found in each study. The effect size quantifies the impact of the intervention by comparing the post-intervention skills of the two groups (a

medium effect size is around 0.5, while a large effect size is around 0.8). NCII reviewed three studies on the *Fast ForWord*[®] Language products: (1) Miller, Linn, Tallal, Merzenich, & Jenkins (1999). This RTC study included 388 students. Result demonstrated positive efficacy with a Medium (0.59) Effect size; (2) Scientific Learning Corporation, 2004. This study used a matched group design and included 50 students. Result demonstrated positive efficacy with a Medium (0.44) Effect size; and (3) Slattery (2003). This RTC included 60 students. Result demonstrated highly positive improvements with a Large (1.44) Effect size. For full report see <http://www.intensiveintervention.org/chart/instructional-intervention-tools>.

Using funding from the US Department of Education's Office of Special Education Programs (OSEP), the National Center on Response to Intervention (NCRTI) was established by the American Institutes for Research and researchers from Vanderbilt University and the University of Kansas. The Center provides guidance to educators on implementing proven models for Response to Intervention (RTI) and Early Intervening Services (EIS). NCRTI reviews studies evaluating the impact of various products on struggling students. The reviews focus on the following components of the study: Participants: students in 5th grade and below; students below the 30th percentile or groups that average below the 25th percentile; Study design: two group, preferably random assignment. Analysis showing comparable initial skills between the two groups, demographic breakdown showing similar demographics between the two groups; Fidelity of implementation: data showing the product was used as designed; Study measures: accurate and relevant (psychometrically valid). Proximal measures assess skills directly targeted by the intervention; distal measures assess aspects of competence that are related to the targeted skills. NCRTI also reports effect size. NCRTI reviewed the same three studies on the *Fast ForWord*[®] Language products as cited above (N = 498 students), breaking out results further into Proximal and Distal effect sizes. The two RCT studies (Miller et al. 1999; Slattery 2003) showed large proximal effect sizes (7.45 and 1.46, respectively), while the Slattery study also showed a Large Distal effect size (1.05).

Nevada Senate Bill 185 (SB 185) funded districts to purchase and implement innovative and remedial educational programs, materials, and strategies specific to their academic needs. The Nevada Department of Education commissioned the Colorado-based Leadership and Learning Center (LLC) to conduct an in-depth, independent evaluation of the programs that were purchased with SB 185 grants. LLC used multivariate analysis to determine the impact of programs on student achievement: "Emphasis was placed on measuring student growth toward academic proficiency and mastery using state and local assessments . . . The analyzes were completed as a result of extensive site visits, phone interviews, and an examination of two-year sets of school cohort achievement data for Criterion-Referenced Tests (CRT) for grades three through eight and High School Proficiency Exams (HSPE) for grades nine through twelve." The advantage of this report is that it compares many of the currently available commercial products used across several schools. *Fast ForWord*[®] products were used at three schools. Efficacy results showed that across the schools that used these products, *Fast ForWord*[®] products increased

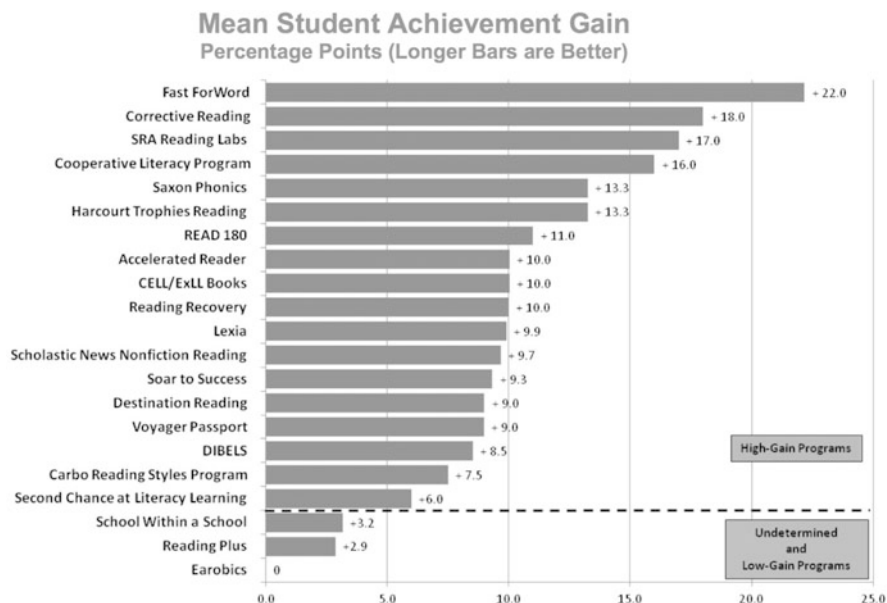


Fig. 15.3 Nevada Department of Education and The Leadership and Learning Center Innovation and Remediation Interim Report. The Leadership and Learning Center used multivariate analysis to determine the impact of programs on student achievement: “Emphasis was placed on measuring student growth toward academic proficiency and mastery using state and local assessments. The analyses were completed as a result of extensive site visits, phone interviews, and an examination of two-year sets of school cohort achievement data for Criterion-Referenced Tests (CRT) for grades three through eight and High School Proficiency Exams (HSPE) for grades nine through twelve.” Their report concludes that Fast ForWord products increased student reading achievement by an average of 22.2% points. This was the largest average impact of all programs reviewed in the report, and it qualified Fast ForWord as a “High-Gain Program.” The percentage gain scores shown in the graph represent an analysis of data from one to multiple schools using the specified product. In the case of Fast ForWord products, data from three schools were included in the analysis

student reading achievement scores by an average of 22.2% points. Fast ForWord® was found to have the **largest average impact** of all programs reviewed in this comprehensive report, classifying *Fast ForWord*® as a “High-Gain Program.” Figure 15.3 shows the results from the Nevada study.

The What Works Clearinghouse (WWC) was created by the NSF Institute for Education Sciences (IES) to review and give ratings to products and programs aimed at teaching and improving academic skills. WWC has developed and standardized a stringent rating scale both for the quality of a research study as well as the effectiveness of a product or program. WWC selects specific topics of most concern to educators, strictly defines the scope for each topic area, and specifies the grade range that each review will include. *Fast ForWord*® products have been reviewed and received positive rating in three areas: Early Reading K-3rd grade, Adolescent Literacy 3rd-10th grade, and English Language Development K-6th grade.

WWC-Early Reading Effectiveness Rating K-3. The What Works Clearinghouse identified nine studies of *Fast ForWord*[®] that both fell within the scope of the Beginning Reading topic area and met WWC evidence standards. Seven studies met standards without reservations and two studies meet WWC evidence standards with reservations. Together these studies included 1,390 students from several areas of the United States and Western Australia. Results show that WWC considers the extent of evidence for *Fast ForWord*[®] on the reading skills of beginning readers to be medium to large for two outcome domains – alphabets and comprehension – and small for one outcome domain – reading fluency.

WWC-Adolescent Literacy Effectiveness Rating Grades 3–10. WWC identified two studies of *Fast ForWord*[®] that fell within the scope of the Adolescent Literacy review protocol that met evidence standards, and six studies that met WWC evidence standards with reservations. The eight studies included approximately 2,000 students. Based on these studies, WWC considered the extent of evidence for *Fast ForWord*[®] on adolescent learners to be small for the alphabets and reading fluency domains, and medium to large for the comprehension and general literacy achievement domains, the domains most important for students in this age range.

WWC-English Language Development K-6. The What Works Clearinghouse (WWC) identified one study of *Fast ForWord*[®] Language that met evidence standards, and a second study that met standards with reservations. The two studies included a total of 250 K – 6th grade English language learners from 16 school districts. *Fast ForWord*[®] Language received a positive rating for improvement of English language development, raising the English language scores of ELL students by an average of +31 percentile points. This was one of the highest ratings given by WWC for English Language Development for ELL K-6 students.

The studies also examined reading achievement. For some unspecified reason, for this topic area phonological and phoneme awareness, which are the most important early reading skills for young English Language Learners (ELL), and those that *Fast ForWord*[®] Language has been shown to most significantly improve in this age range, were considered to be outside the scope of this review and were not included in measures of reading achievement. Furthermore, none of the *Fast ForWord*[®] Reading products were used in the evaluated studies. As such, significant improvement in the higher levels of reading, that were the only reading measures included in this review, would not be expected and were not found.

15.9 Cognitive Neurotherapeutics: The Challenges of Translation

The biggest challenge we have faced along our journey to translate our laboratory research into real world settings has been negotiating the torturous path between the world of our scientific colleagues, as compared to the very different world

of K-12 educators and clinicians who make the decisions about whether our products will be offered to the children who could benefit from them. Nowhere have these different worlds collided more directly than when it comes to assessing and reporting the efficacy of *Fast ForWord*[®] products. Our University-based colleagues have primarily used a combination of behavioral, physiological and neuroimaging technologies to address questions about neuroplasticity-based training that have the potential to advance scientific knowledge and theory. Scientists are experts at designing elegant studies in which we can manipulate one variable at a time, within a well-controlled environment. These studies generally include “active control” methods designed to assess not only efficacy, but also specificity. Many “active controls”, designed only for this purpose, are not scalable and would be prohibitively expensive to implement in a real-world settings. For example, the NIH-funded *Fast ForWord*[®] trial used 50 h of one-on-one speech therapy provided by a licensed SLP as an “active control” in order to match the 50 h training protocol for *Fast ForWord*[®] (Gillam et al. 2008). Finding that these two methods were highly successful in increasing language scores, albeit equivalent, was interpreted as a failure to demonstrate specificity for *ForWord*[®]. However, providing the intensity of therapy used as the active control in this study to the majority of students who need it is cost prohibitive in the real world. What schools, clinics and parents need to know is how a new method, like *Fast ForWord*[®], compares to the actual alternatives that are available to their students, not hypothetical ones that are not. As such, these same results have an entirely different valence to educators, clinicians and parents searching for efficient, cost effective and enduring methods for improving the outcomes of their students. They view these same data as strongly positive scientific evidence supporting their own experience using *Fast ForWord*[®].

Regardless of whether research scientists studying the science of learning, including reading, are involved directly in translating research from their lab to clinics and/or classrooms, most state in their grant applications that a primary goal of their research is to improve educational and/or clinical outcomes. However, the reality is that we face considerable challenges and roadblocks should we actually attempt to make good on this promise. The catch 22 is that many scientists are eager to translate our research ideas and innovations into practical clinical and educational applications. However, once these innovations are translated, they take on a life of their own to meet the needs of the intended end users. Despite the best of intentions, scientists, clinicians and educators continue to be akin to the proverbial ships passing in the night. The work being done by independent agencies such as the WWC, NCRTI and NCII to bridge this gap is an invaluable step in the right direction. If we as scientists are serious about translating our research into practical applications, that have the potential to improve educational outcomes on a broad scale, we need to develop more effective, **bi-directional** ways to collaborate, communicate and value educator’s and clinician’s real-world experience using our applications. As the intended consumers of our research we need to engage educators and clinicians as equal partners throughout the ongoing, iterative, translational process.

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Chapter 16

Effects of the Computer-Based Training Program *Lautarium* on Phonological Awareness and Reading and Spelling Abilities in German Second-Graders



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Abstract Intact phonological processing abilities are of major importance for successful acquisition of literacy skills. Training studies confirmed that programs which combine phonological training with systematic instruction on letter-sound-relationships are effective in fostering reading and spelling skills. Based on this evidence, we developed the computer-based training program *Lautarium* for German-speaking primary school children experiencing reading and spelling difficulties. This chapter provides an overview of the structure and contents of *Lautarium*, and summarizes the empirical evidence concerning the effectiveness of *Lautarium*-training in children with poor literacy skills. Additionally, we describe a study on the effects of *Lautarium*-training in two groups of second-graders with relatively low class-level reading skills. Group 1 performed *Lautarium*-training for a period of 8 weeks at the beginning of second grade, while Group 2 received regular classroom instruction. A significant training effect was found for spelling, but not for phonological awareness or reading. Since only a few children finished the training within the 8-week period, *Lautarium* was modified in order to allow faster completion of the exercises. Group 2 trained with the modified version at the end of second grade. Subsequent tests revealed stronger improvements in reading, spelling, and phonological awareness in Group 2 when compared to Group 1.

Keywords Dyslexia · Intervention · Reading · Spelling · Phonological awareness · Speech perception · Children

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16.1 Introduction

In today's society, literacy skills are a precondition for knowledge access and social participation. Instructing children in reading and spelling is thus a major task in primary schools. However, according to a recent German study, more than 18% of the children have severe difficulties in acquiring these essential skills, despite average intelligence and adequate schooling (Fischbach et al. 2013). It has been shown that literacy disorders are associated with psychiatric disorders and lower educational and professional attainment (Carroll, Maughan, Goodman, & Meltzer 2005; Esser, Wyschkon, & Schmidt 2002). In order to circumvent these outcomes, teachers and educators need effective and feasible methods for prevention and remediation of reading and spelling disorders.

The effectiveness of treatment approaches for children with reading and spelling disabilities has been evaluated in several systematic reviews and meta-analyses (Ehri, Nunes, Willows, et al. 2001; Galuschka, Ise, Krick, & Schulte-Körne 2014; McArthur et al. 2012). In these studies, phonics-based interventions consistently proved effective in fostering children's literacy skills. In a strict sense, phonics instruction means training of grapheme-phoneme-correspondences and their application in reading and spelling of syllables and words. However, most phonics-based programs go beyond "pure" phonics, in that they also include auditory-phonological training (McArthur et al. 2012). Such combined programs foster phonological processing abilities that are essential for reading and spelling acquisition, and explicitly teach the children how to use these phonological skills when dealing with written language. In this way, phonics-based interventions help the children to grasp the alphabetic principle, i.e., to understand how letters in written words map onto phonemes in spoken words (Hatcher, Hulme, & Snowling 2004; Snowling & Hulme 2012).

Based on this evidence, we developed the computer-based training program *Lautarium* for German-speaking children with reading and/or spelling difficulties (Klatte, Steinbrink, Bergström, & Lachmann 2017). Computerized training was chosen because, especially with respect to reading and spelling, computer programs provide several advantages over conventional teacher-based instruction (Macaruso, Hook, & McCabe 2006; Torgesen & Barker 1995). First, the combined presentation of high-quality speech recordings and the respective orthographic units foster the acquisition of distinct phonological representations and letter-sound-correspondences. Second, adaptive learning algorithms ensure training schedules optimized for each individual child. Thus, extremes of either excessive demands or too few demands are avoided, and the training time is most efficiently used. Third, children with reading or spelling disorders need much more learning time and practice in order to acquire basic literacy skills (Torgesen 2002). Such intense training is difficult to achieve with conventional, teacher-based instruction, but easy to accomplish with computer-based training, since the latter can be supervised by parents or other family members, without support of a professional. Finally, working with computer programs is motivating for children. Computer programs are capable of providing lively graphics, immediate feedback, and attractive reinforcement.

The following section provides an overview of the structure and contents of *Lautarium*, and its theoretical base. Subsequently, the available evidence on the adequacy and efficacy of *Lautarium* is summarized. Finally, details are provided for a current study on the effects of *Lautarium*-training on phonological awareness and reading and spelling skills in second-graders from German primary schools.

16.2 Training Components in *Lautarium*

Based on the findings concerning the efficacy of phonics instruction, *Lautarium* combines auditory-phonological training with training of grapheme-phoneme-relationships and reading and spelling of transparent words. In addition, rapid access from written words to meaning is included by means of word-to-picture matching with short word presentation times.

16.2.1 Phonological Training

Phonological training in *Lautarium* comprises phoneme perception and phonological awareness. Phoneme perception is included because several studies found speech perception deficits in children with reading disorders (Manis et al. 1997; McBride-Chang 1995), and some authors argue that these deficits are causally involved in disordered reading acquisition (Hornickel, Skoe, Nicol, Zecker, & Kraus 2009; Noordenbos & Serniclaes 2015; Ziegler, Pech-Georgel, George, & Lorenzi 2009). Poor phoneme perception may result in underspecified or noisy phonological representations in the mental lexicon. Inaccurate phonological representations, in turn, affect processing, storage, and access to phonological information, and hamper the building-up of phoneme-grapheme-correspondences (Elbro & Jensen 2005; Swan & Goswami 1997). Concerning intervention, studies proved that phoneme perception is ameliorated by training (Bischof et al. 2002; McArthur, Ellis, Atkinson, & Coltheart 2008; Strehlow et al. 2006), and that phoneme perception training transfers to phonological awareness (Moore, Rosenberg, & Coleman 2005; Thomson, Leong, & Goswami 2013). Furthermore, training that combines phoneme perception with phonological awareness or letter-sound-matching showed transfer effects to reading (e.g., Ecalle, Magnan, Bouchafa, & Gombert 2009; Gonzalez, Espinel, & Rosquete 2002).

In *Lautarium*, phoneme perception training focuses on discrimination and identification of plosive consonants and vowel lengths. Regarding consonant perception, discrimination between plosives is especially difficult when they differ only with respect to voicing (e.g., /b/ vs. /p/) or place of articulation (e.g., /b/ vs. /d/). The acoustic information relevant for discrimination between these phonemes is located in a time window of about 50 ms, making fine-grained temporal resolution of the speech signal necessary (Bishop 1997). In a recent study with German children (Klatte, Steinbrink, Bergström, & Lachmann 2013), plosive perception proved still

more difficult when the critical consonants were embedded in consonant clusters, e.g., /bla:/ vs. /pla:/. The performance drop in trials with clusters was stronger in children with reading disabilities when compared to normal readers. Based on this evidence, *Lautarium* includes discrimination and identification of plosives with and without consonant clusters.

Vowel length perception is included in *Lautarium* since, in German language, vowel length is phonemic and orthographically marked. For example, the spoken German words *kann* (/kan/ [can]) and *Kahn* (/ka:n/ [barge]) differ only in vowel length. In orthography, vowel length is marked by the letters following the vowel according to specific rules. Short vowels are usually followed by two consonants, e.g., *Mund* [mouth], *Wand* [wall]. When a short vowel is followed by only one consonant phoneme, the latter is doubled in spelling, e.g., *Ball* (/bal/ [ball]), *Kamm* (/kam/ [comb]). Long vowels, on the contrary, are either not marked at all, e.g., *Hut* (/hu:t/ [hat]), *Wal* (/va:l/ [whale]), or are followed by a “lengthening h”, e.g., *Kahn* (/ka:n/ [barge]), *Huhn* (/hu:n/ [chicken]). Thus, in order to learn these orthographic rules, children must be able to identify whether a vowel is long or short. Studies with German-speaking children revealed impaired vowel length perception in children with literacy disorders (Landerl 2003; Steinbrink, Klatte, & Lachmann 2014).

Phonological awareness, i.e., the ability to consciously access and manipulate the sound units of language, has proved fundamental for reading and spelling acquisition in numerous studies since the 1980s (e.g., Bradley & Bryant 1983; Wagner & Torgesen 1987). Phonological awareness in kindergarten predicts later literacy skills (for reviews, see Melby-Lervag, Lyster, & Hulme 2012; Pfof 2015), and children with literacy disorders exhibit severe difficulties in phonological awareness tasks when compared to typically developing children matched for chronological age, or reading age (Melby-Lervag et al. 2012). Furthermore, meta-analyses of intervention studies with English- and German-speaking children confirmed that training phonological awareness in preschoolers fosters reading and spelling acquisition in the early grades (Ehri, Nunes, Stahl, & Willows 2001; Fischer & Pfof 2015). However, meta-analyses also revealed that, in older children with literacy disorders, isolated trainings of phonological awareness do not show beneficial effects on reading (Galuschka et al. 2014; Ise, Engel, & Schulte-Körne 2012) or spelling (Ehri, Nunes, Stahl, & Willows 2001; Galuschka et al. 2014; Ise et al. 2012). As stated in the introduction, for these children, programs that combine phonological training with training of letter-sound-correspondences and decoding skills seem more effective. In *Lautarium*, phonological awareness is trained through exercises requiring sound-to-word matching, matching of initial or final sounds in words, and segmentation and blending.

16.2.2 Grapheme-Phoneme Correspondences

In order to master the alphabetic principle, children have to understand the relationships between phonemes in spoken words and graphemes in written words. To accomplish this, both phonemically structured representations of spoken words and

letter-sound-knowledge is needed. In line with this, longitudinal studies identified letter knowledge as a further pillar of early literacy acquisition, over and above phonemic awareness (Fricke, Szczerbinski, Fox-Boyer, & Stackhouse 2016; Hulme, Bowyer-Crane, Carroll, Duff, & Snowling 2012; Leppänen, Aunola, Niemi, & Nurmi 2008; Näslund & Schneider 1996), and intervention studies showed that training programs combining both components are most effective in fostering reading and spelling skills (Ehri, Nunes, Willows, et al. 2001; Galuschka et al. 2014; Ise et al. 2012; McArthur et al. 2012). Accordingly, in *Lautarium*, training of grapheme-phoneme-mappings and phonological training are interlinked. For example, when the correct phonemes have been selected in phoneme identification or segmentation tasks, the corresponding graphemes have to be assigned.

16.2.3 *Reading and Spelling*

In *Lautarium*, newly acquired phonological skills and letter knowledge are immediately applied in reading and spelling tasks. Reading tasks comprise matching printed words to spoken words (and vice versa), and matching pictures to printed words. The latter task focuses on reading speed, aiming to foster direct access from print to meaning and thus enlarge the child's sight word vocabulary. Spelling tasks require segmentation of target words presented auditorily or pictorially into their constituent graphemes.

16.3 *Materials and Training Procedure in Lautarium*

Lautarium uses two types of building blocks representing phonemes and the corresponding graphemes, respectively. Phonemes are represented by blocks with pictures of easy-to-name objects. The phoneme represented by a specific block is the initial sound of the picture's verbal label (e.g., the picture of a ball represents the phoneme /b/). Graphemes are represented by blocks with a single letter or a letter combination that is usually used for a specific phoneme in German orthography (e.g., the phoneme /aI/ is usually represented by the grapheme *ei*, Thomé 2000).

The speech material comprises about 1,300 and 1,400 pseudowords (CV, VCV, CVC, CCV, VCCV), and German nouns, respectively. Each speech item is implemented through high-resolution recordings produced by professional speakers (a male and a female). For about 40% of the nouns, pictorial presentation is also available.

Tasks belonging to different training domains are intermixed, and within each domain, children start with simple tasks and proceed to more difficult ones. For example, training of consonant perception proceeds from discrimination to identification, and from speech targets without consonant clusters to targets with clusters. Due to the combinations of tasks and materials, *Lautarium* consists of 58 different exercises. Each exercise comprises 10 to 30 trials, depending on task

complexity. Responses are followed by immediate feedback (correct, incorrect, time-out). In case of errors or time-outs, the respective trial is repeated until the correct answer is provided in time. Depending on the percentage of trials correctly solved in the first attempt, the child either has to repeat the respective exercise, or proceeds to the following one.

Before starting a new task, children have to work through an interactive instruction that provides explanation, practice with examples, and informative feedback. In addition, *Lautarium* is equipped with a token system fostering concentrated, intensive training. For correct answers, the children receive virtual money. After completion of an exercise, the children get access to a virtual “aquarium shop”, where they can buy fishes, shells, plants, and other objects for stepwise construction of an animated, individual aquarium.

Lautarium aims to improve reading and spelling by fostering phonological processing and the acquisition and automatization of phoneme-grapheme correspondences. Prior studies in non-German language areas have shown that daily practice over several weeks is effective for achieving these aims (e.g., Eden et al. 2004; Hintikka, Aro, & Lyytinen 2005; Moore et al. 2005). Based on this evidence, *Lautarium*-training is performed through sessions of 20–30 min, 5 times per week, for a period of about 8 weeks.

16.4 *Lautarium*: Empirical Findings

During the development process, the appropriateness and effectiveness of *Lautarium* was tested in several empirical studies, aiming to uncover potential shortcomings and generate suggestions for improvements (“formative evaluation”, see Nieven & Folmer 2013). In the first step, the validity of the training tasks implemented in *Lautarium* was evaluated. Most of the tasks are adaptations of tasks that proved successful in international training studies. Thus, the tasks were originally designed for the phonology of other languages (e.g., English or French), and had to be adjusted to the specifics of German phonology. In addition, tasks that were originally used in the context of face-to-face-instruction had to be transformed into a computer-based training format. In order to test the appropriateness of these adaptations, primary school children with reading disorders ($n = 35$) and age-matched controls ($n = 75$) completed a total of 17 phoneme perception, phonological awareness, and phoneme-grapheme mapping tasks designated for implementation in *Lautarium* (Klatte et al. 2013). We hypothesized that, if the implemented tasks really tap into the poor reader’s phonological deficits, significant performance differences in favor of the normal readers should emerge. The analyzes confirmed that, for each of the 17 tasks, the poor readers were outperformed by the controls (all $ps < 0.05$), with group differences of medium to large effect sizes ($d = 0.50 - 1.49$). Thus, the tasks proved appropriate, since they differentiate between children with and without reading disorders.

Based on this evidence, we conducted two studies evaluating the effectiveness of the *Lautarium*-training with primary school children using a pilot version of

the training program. In both studies, the children's phonological processing and reading and spelling abilities were assessed at three measurement times: Pretest (before start of training), posttest 1 (immediately after training) and posttest 2 (two months later). After the pretest, the training group performed *Lautarium* during school lessons, 5 times per week, for 20–30 min, over a period of 8 weeks, using laptops and circumaural headphones provided by the research institute. The control group continued to receive standard classroom instruction. Potential group differences in test scores were analyzed at posttest 1 and posttest 2 by means of analyzes of covariance (ANCOVAs), with the pretest scores from the respective test treated as covariate (Rausch, Maxwell, & Kelley 2003). In case of significance, effect sizes corrected for pretest differences were calculated as proposed by Klauer (1989).

Participants in the first study (Klatte et al. 2014) were German third-graders with developmental dyslexia who attended special dyslexia classes in two primary schools in Leipzig, Germany. The training and control group consisted of 20 (13 boys, mean age 9;0) and 21 (14 boys, mean age 9;1) children, respectively. During the training period, the control group continued to receive regular face-to-face remedial instruction provided in the dyslexia classes.

The test battery comprised phoneme perception (consonant discrimination and vowel length classification), phonological awareness (deletion and substitution of sounds in words), reading (reading aloud of words and pseudowords, sentence comprehension), and spelling of words and pseudowords. Analyzes of the posttest data confirmed significant group differences of small to moderate effect size in favor of the *Lautarium*-training group. Concerning phonological processing, significant and enduring training effects were found for one out of two subtests of phoneme perception, and for one out of two subtests of phonological awareness. Concerning reading, significant training effects were found for word and pseudoword reading at posttest 2, but not at posttest 1. This finding indicates that *Lautarium* initiates phonological development, the positive effects of which, with respect to reading, unfold with time. Concerning spelling, a stronger decrease in the number of errors reflecting violations of the alphabetic principle in the training group was found when compared to the controls. To conclude, these results indicate that *Lautarium*-training is effective for German dyslexic children, even though some of the effects were small in magnitude and, in some subtests, no group differences were found. It must be kept in mind that, in this study, the control children were not “untreated”, but received intensive remedial teaching in the context of the dyslexia classes. Thus, the effects evoked by *Lautarium*-training exceeded those of school-based remedial instruction.

As stated in the introduction, *Lautarium* aims to foster letter knowledge and phonological abilities that proved essential for literacy acquisition. Thus, *Lautarium*-training should be effective not only in children with manifest reading and spelling disorders, but also in younger children at risk of such disorders. The second study (Klatte, Steinbrink, Bergström, & Lachmann 2016) aimed to test this assumption. The effects of *Lautarium*-training were assessed in a sample of 102 first-graders from two primary schools in Bavaria. The children from one school performed *Lautarium*-training ($n = 53$, 24 boys, mean age 6;10), the children from the other school served as controls ($n = 49$, 31 boys, mean age 6;11).

The test battery was comprised of phoneme perception (consonant discrimination and vowel length classification), phonological awareness (rapid classification of initial sounds in words, comparison of initial and final sounds in words, deletion and substitution of sounds in words), reading (reading aloud of words and pseudowords, word-to-picture matching, sentence comprehension), and spelling (cloze test). Based on pretest performance in the word reading test, each child was classified as “poor reader” (percentage rank ≤ 22) or “non-poor reader” (percentage rank > 22), yielding 18 and 17 poor and 35 and 32 non-poor readers in the training and control groups, respectively. For both the poor and the non-poor readers, analyzes of the pretest data indicated good comparability between the training and control groups with respect to reading, spelling, and nonverbal intelligence.

Concerning the poor readers, ANCOVAs of the posttest scores, with pretest scores as covariate, confirmed significant advantages of the *Lautarium*-training group with respect to phoneme perception, phonological awareness, reading, and spelling. For phoneme perception, significant and enduring effects of strong effect size were found for consonant discrimination, but not for vowel length classification. Concerning phonological awareness, significant training effects of medium to strong effect size were found for two out of three subtests in posttest 1, and for each of the three subtests in posttest 2. For reading, analyzes confirmed significant training effects of medium to strong effect size for three out of four subtests in posttest 1, and for each of the four subtests in posttest 2. Concerning spelling, analyzes proved significant training effects of strong effect size in both posttests.

For the non-poor readers, analyzes confirmed significant beneficial effects of *Lautarium*-training for subtests of phoneme perception, phonological awareness, and reading. Concerning phoneme perception, significant effects were found for consonant discrimination in posttests 1 and 2, and for vowel length classification in posttest 1. Concerning phonological awareness, analyzes revealed significant and enduring effects in one of the three subtests. For reading, beneficial effects of medium to strong effect size were found for three measures in posttest 1, and for one measure at posttest 2.

To summarize, in this study, *Lautarium*-training remarkably improved phonological processing and reading and spelling in struggling beginning readers. In addition, *Lautarium* proved beneficial also for non-poor beginning readers. In both reading level groups, children who performed *Lautarium*-training either outperformed the controls, or performed on a par with the controls. None of the analyzes revealed an advantage of the control group receiving regular classroom instruction.

Based on the individual training data obtained in these studies, *Lautarium* was optimized through modification of existing tasks, and addition of new tasks. Specifically, tasks that proved too easy or too demanding were modified or discarded, and new tasks focusing on reading and spelling of transparent words were implemented. In addition, aiming to foster rapid access from print to meaning, a word-to-picture matching task was included, in which the presentation times of the target words are more and more reduced according to an adaptive learning algorithm (Kaernbach 1991). In this task, both transparent and non-transparent words were used as target items.

The effectiveness of the resulting program was assessed in a further study, which is reported in the following section.

16.5 Effects of *Lautarium*-Training on Phonological Awareness and Reading and Spelling Skills in Second-Graders from German Primary Schools

16.5.1 Design and Participants

This study used a waiting group design in order to analyze the effects of *Lautarium*-training on children's phonological awareness and literacy skills. Participants included 122 children from two primary schools located in socially disadvantaged catchment areas in a major city of Rhineland-Palatine, Germany. Due to long-standing illness, change of school, or repetition of a school year, 23 children were unavailable at one or more measurement times. Thus, analyzes are based on a sample of 99 children. According to parents' reports, 20 children had a non-German first language or grew up bilingually with German and a further language. As these children did not differ from the others with respect to pretest performance in reading and spelling, first language was not included as variable in the analyzes.

The children from one school served as training group ($n = 53$; 24 boys, mean age 7;5). The children from the other school acted as waiting control group ($n = 46$, 21 boys, mean age 7;4). *Lautarium*-training was performed during the first and last two months of second grade in the training and waiting control group, respectively. As in prior studies, the training took place during school lessons, 5 times per week, for 20–30 min, over a period of about 8 weeks. While one group performed the training, the other group continued to receive regular classroom instruction.

Children's phonological awareness and reading and spelling abilities were assessed in groups of whole classes at three measurement times: end of first grade (pretest), middle of second grade (posttest 1), and end of second grade (posttest 2). Thus, at posttest 1, the children from the waiting group served as untreated controls, as *Lautarium*-training was finished in the training group, whereas the waiting group had not yet started. At posttest 2, both groups had received the training.

16.5.2 Tests

Reading was assessed by means of two subtests from a standardized German reading test (Lenhard & Schneider 2006), measuring reading speed and comprehension on the level of single words and sentences, respectively. For word reading, the children had to select, out of four alternatives, the word that matched a target picture. For sentence reading, the children had to select, out of five alternatives, the word that

fit into the target sentence. For both subtests, children had to complete as many items as possible within 3 min. For spelling, a standardized German spelling test was used, in which the children had to write down words and sentences according to dictation (May 2012). Three raw scores were derived from this test, one representing the number of correct graphemes, and the other two representing success in application of letter-sound mappings (“alphabetic strategy”) and orthographic rules (“orthographic strategy”), respectively. Since different versions of the test had to be applied over the time course of the study, percent correct scores were calculated from each measure and used as dependent variables in the analyzes.

As no standardized group tests measuring phonological awareness are available for German-speaking children above grade 1, a new test was constructed for use in this study (Klatte, Bergström, Konerding, & Lachmann [manuscript in preparation](#)). This test consisted of three subtests requiring identification, deletion, and substitution of sounds in words presented pictorially. For each subtest, time limit was 3 min. Concerning identification, the children had to decide whether or not a specific speech sound was present in a number of target words. In case of “yes”-answers, the children also had to indicate the position of the sound in the word (beginning, middle, end). For sound deletion and substitution, the children had to indicate, for each target word, which word emerged when the second phoneme is eliminated or substituted, respectively.

16.5.3 Statistical Analyzes

Potential group differences in learning gains between pretest and posttest 1 were analyzed by means of ANCOVAs, with the pretest scores from the respective test treated as covariate. In the case of unequal regression slopes, t-tests on gain scores were performed instead of ANCOVAs. In addition, in order to evaluate the effects of *Lautarium*-training in the waiting group, we tested for group differences in learning gains between posttest 1 and posttest 2. For this purpose, the differences between test scores in posttest 2 and posttest 1 were calculated for each individual child, and compared between groups with the pretest score of the respective test included as covariate (Rausch et al. 2003).

16.5.4 Results and Discussion

Analyzes of the pretest reading and spelling scores showed that, on average, performance in this sample was comparably low. Mean scores were 0.5 and 0.3 standard deviations (SDs) below the norm (mean of the reference sample) for reading and spelling, respectively. Further analyzes confirmed that 40 and 30% of the children performed one or more SDs below the norm in the reading and spelling tests, respectively. We attributed these results to the social characteristics of the

schools' catchment areas (concerning the association between socioeconomic status and early literacy skills, see Aikens & Barbarin 2008; Bos 2007). Reading, spelling, and phonological awareness scores at pretest did not differ significantly between treatment groups.

At posttest 1, analyzes of the spelling measures revealed a significant effect of moderate effect size in favor of the training group with respect to application of letter-sound-mappings ("alphabetic strategy"), $t(95)=3.23$, $p < 0.01$, $d=0.66$. Concerning percentage of correct graphemes, the advantage of the training group was marginally significant, $F(1, 94)=3.73$, $p=0.06$, $d_{\text{corr}}=0.43$. No effect was found for application of orthographic rules, $F(1, 94) < 1$. With respect to reading and phonological awareness, none of the analyzes yielded a significant difference between groups (all $ps > 0.24$).

The unexpected lack of training effects on reading and phonological awareness at posttest 1 was attributed to insufficient use of the training time. Observations of researchers who occasionally monitored the training sessions indicated that the children spent much time selecting objects for their aquarium and interacting with peers sitting next to them. Analyzes of the individual training data confirmed that the majority of the children did not finish the *Lautarium*-training within the predetermined 8-week period. In fact, 24 children completed only half or less than half of the exercises. Error rates often just exceeded the criterion, resulting in time-consuming – and sometimes frustrating – repetitions of the exercises. Based on these findings, *Lautarium* was again modified. For example, the criterion for passing an exercise was released from 80 to 75% of correct trials, the maximum number of repetitions per exercise was reduced to 5, and access to the aquarium was only allowed when all trials of the current exercise were solved.

Prior to posttest 2, the waiting group performed *Lautarium*-training with the modified version. From the 45 children of the waiting group, 22 completed the training within the predetermined 8-week period. Only one child completed less than half of the program. Observations during the training sessions confirmed that the children were more attentive and concentrated when compared to the children from the training group.

Analyzes of the children's learning gains between posttest 1 and posttest 2 confirmed significant effects in favor of the waiting group with respect to phonological awareness, reading, and spelling (see Fig. 16.1). For phonological awareness, significant effects of strong and medium effect size were found for sound identification, $F(1, 96)=17.24$, $p < 0.001$, $d=0.82$, and sound deletion, $F(1, 94)=5.3$, $p < 0.05$, $d=0.48$. For reading comprehension, a significant effect of strong effect size was found for words, $F(1, 96)=16.96$, $p < 0.001$, $d=0.77$, but not for sentences, $F(1, 96)=1.29$, $p=0.26$. For spelling, analyzes revealed significant advantages of the waiting group for percentage of correct graphemes, $F(1, 94)=6.48$, $p < 0.05$, $d=0.47$, and for application of letter-sound mappings ("alphabetic strategy"), $t(95)=3.97$, $p < 0.001$, $d=0.81$. No effect was found for application of orthographic rules, $F(1, 94) < 1$.

Thus, the beneficial effects of *Lautarium*-training were more pronounced in the waiting group when compared to the training group. This is presumably due to the

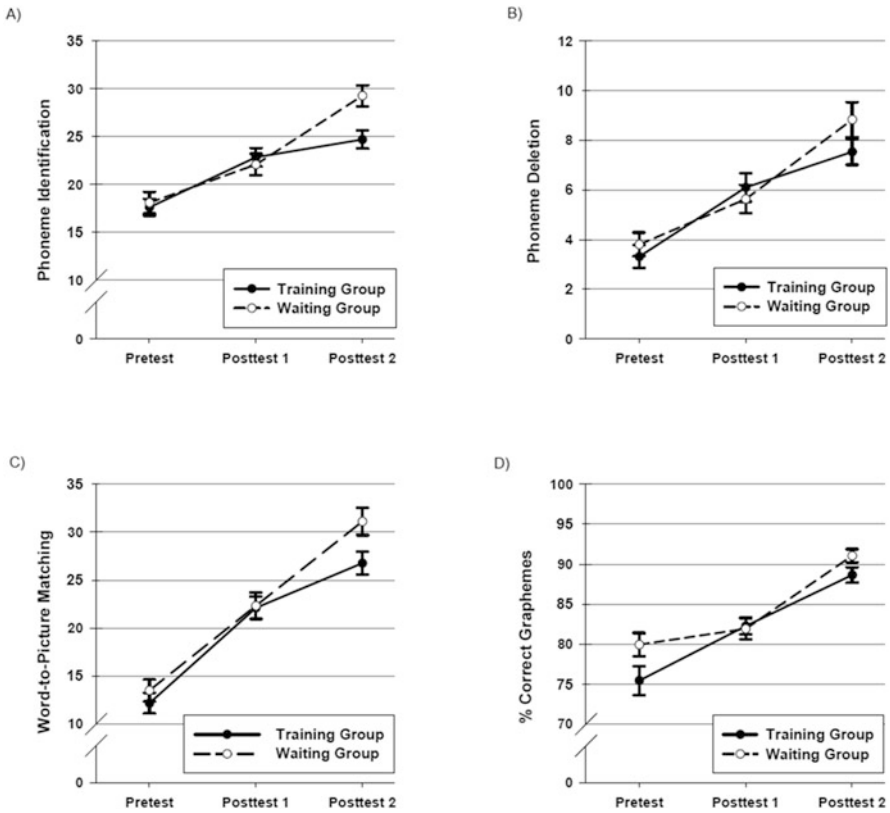


Fig. 16.1 Average performance scores in the training and control groups at pretest, posttest 1, and posttest 2. **(a)** Phonological awareness: Phoneme identification, **(b)** Phonological awareness: Phoneme deletion, **(c)** Reading: Word-to-picture matching, **(d)** Spelling: % graphemes correctly spelled. Error bars represent standard errors of the mean. Note: Training took place between pretest and posttest 1 in the training group and between posttest 1 and 2 in the waiting group

modifications implemented in the program between the training periods. However, since the children from the waiting group were at the end of year 2 at the time of training, developmental progress in attention control and self-regulation may also have contributed to the comparably higher training efficacy in this group.

To summarize, this study provided further evidence for the efficacy of *Lautarium*-training in fostering phonological processing and literacy skills in German-speaking primary school children. In addition, the study showed that, when *Lautarium* is performed in groups of whole classes, teachers and educators should take care to ensure efficient use of the training time. In addition to usage of circumaural, noise-reducing headphones, the distance between work places should be maximized in

order to avoid distraction due to peers. With these considerations, it is expected that most children would be able to complete the training within the designated 8-weeks period.

16.6 Summary and Conclusions

Based on the available evidence concerning the effectiveness of treatment approaches for children with reading and spelling disorders, we developed the computer-based training program *Lautarium* for primary school children instructed in German language. In *Lautarium*, auditory-phonological training is interlinked with training of letter-sound-correspondences and word-level reading and spelling. Exercises are selected according to the individual performance level, and interactive instructions and immediate feedback enable the children to work through the program largely without adult help. Training studies confirmed beneficial effects of *Lautarium*-training on phonological processing and reading and spelling abilities in samples of struggling beginning readers, second-graders with relative low class-level literacy skills, and third-graders with developmental dyslexia. These studies provide further evidence for the efficacy of computer-based instruction in fostering literacy skills in primary school children (Macaruso et al. 2006).

However, these studies also bear some limitations. First, due to the complexity of the program, it is not clear whether the learning gains in reading and spelling result from a specific training component (e.g., letter-sound-mappings), a combination of specific components (e.g., phonological awareness and letter-sound-mappings), or the integration of all components in the entire program. Second, training was performed during school lessons in groups of whole classes. Thus, the results confirm the efficacy of *Lautarium*-training as a supplement to regular classroom instruction. Further studies are needed in order to assess the effectiveness of *Lautarium*-training in other settings, e.g., the home environment. Third, even though the studies confirmed that, overall, children receiving *Lautarium*-training showed advantages in literacy skills relative to controls, there were considerable differences in learning gains between children. Each of the samples included some non-responders to *Lautarium*-training, i.e., children whose spelling scores (raw scores) even declined over the training period (first-graders: 1/53 children, second-graders: 13/99 children; third-graders with dyslexia: 3/20 children). Future research should address predictors of children's responsiveness in order to ensure effective training for each individual child.

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Publisher's Correction to: Introduction to Reading and Dyslexia



Thomas Lachmann and Tina Weis

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This book was published inadvertently with the “Introduction to Reading and Dyslexia” by Thomas Lachmann included in the front matter of the book. This has now been included as Chapter 1 and a separate DOI has been assigned in order to make it citable and searchable. The subsequent chapters have been re-numbered and the page numbers have been updated. The content and the DOI of the existing chapters remain unchanged.

The updated online version of these chapters can be found at

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