COMORBIDITIES (D DEWEY, SECTION EDITOR)



Emotion Regulation as a Transdiagnostic Feature in Children with Neurodevelopmental Disorders

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Abstract

Purpose of Review Emotion regulation is recognized as a core underlying dimension common across psychiatric disorders and could be conceptualized as a transdiagnostic feature (i.e., mechanism underlying comorbidity). This review highlights recent research examining emotion regulation in children with attention-deficit/hyperactivity disorder (ADHD) and autism spectrum disorder (ASD).

Recent Findings Difficulties with emotion regulation are increasingly recognized as common deficits in children with ADHD and ASD. Research indicates that emotion regulation transmits risk for co-occurring disorders within a diagnostic grouping (i.e., neurodevelopment disorders; homotypic comorbidity) and across different diagnostic groupings (i.e., neurodevelopmental disorder, heterotypic comorbidity) in children with ADHD and ASD.

Summary While this review provides support for the conceptualization of emotion regulation as a transdiagnostic feature in ADHD and ASD, further research examining emotion regulation in children with other neurodevelopmental disorders is needed. The field of neurodevelopmental disorders should examine emotion regulation from a developmental perspective and needs to develop valid assessments of emotion regulation.

Keywords Emotion regulation \cdot Neurodevelopmental disorders \cdot Attention-deficit/hyperactivity disorder \cdot Autism spectrum disorder \cdot Comorbidity

Introduction

In the developmental milieu of childhood, neurodevelopment involves the dynamic interplay between genetic, environmental, neurological, cognitive, emotional, and behavioral processes [1]. The term neurodevelopmental disorders has been applied to a heterogenous group of disorders that although clinically and etiologically disparate, typically manifest during childhood and involve some form of disruption to brain development [2]. In the Diagnostic and Statistical Manual of Mental Disorders-5th Edition (DSM-5), this group includes attention-deficit/hyperactivity disorder (ADHD), autism spectrum disorder (ASD), intellectual

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Gillian England-Mason gillian.englandmason@ucalgary.ca disability, communication disorders, specific learning disorders (e.g., dyslexia), and motor disorders (e.g., developmental coordination disorder) [3]. During childhood, neurodevelopmental disorders have individual prevalence rates ranging from 0.5 to 18.5%, and two of the most common neurodevelopmental disorders are ADHD and ASD [4]. Over the past decade, the prevalence of ADHD and ASD has risen, as well as their clinical co-occurrence and their co-occurrence with other disorders [5]. Accordingly, the identification of transdiagnostic features (i.e., mechanisms underlying comorbidity) is an essential step in informing novel therapeutic approaches [6]. Emotion regulation is an important feature of mental health and is well-recognized as a core underlying dimension common across psychiatric disorders [7•]. Recent research suggests that emotion regulation holds promise as a transdiagnostic feature in children with neurodevelopmental disorders, such as ADHD and ASD [8..]. The present review highlights research examining emotion regulation in children with ADHD and ASD and proposes that difficulties with emotion regulation may transmit risk for comorbidity among neurodevelopmental disorders.

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Angold et al. [9] have suggested that different types of comorbidity exist, which they refer to as homotypic and heterotypic comorbidities. Homotypic comorbidity refers to the co-occurrence of disorders within a diagnostic grouping (e.g., neurodevelopmental disorders in the DSM-5); whereas heterotypic comorbidity refers to the comorbidity of disorders from different diagnostic groups (e.g., neurodevelopmental disorder and mood disorder). Based on these definitions, many children with neurodevelopmental disorders experience homotypic comorbidity, heterotypic comorbidity, or subclinical symptoms of multiple disorders [9, 10]. However, it is important to note that these distinctions based on diagnostic groupings can be misleading, as neurodevelopmental disorders are phenotypically diverse and extend beyond a defining set of core symptoms [2]. Despite recognition that defining comorbidities based on these diagnostic distinctions warrants reconsideration, studies continue to report on co-occurring disorders based on these definitions [6]. In the field of neurodevelopmental disorders, both homotypic and heterotypic comorbidities represent challenges for clinical practice, as they often result in ambiguous clinical guidelines, fragmented service delivery, and poorer prognosis [11]. Further, they represent challenges for clinical research, as participants are often not characterized beyond a single core diagnosis and comorbidities may affect the appropriateness of patient and control populations, protocols and methodologies, and could bias study outcomes (e.g., comorbid disorders may serve as confounding variables) [2, 12]. Thus, clinical guidelines and interventions developed for the treatment of a specific neurodevelopmental disorder often neglect the problem areas associated with frequently occurring comorbid disorders [6, 11].

Difficulties with emotion regulation in children with ADHD and ASD are often overlooked; however, emerging evidence suggests that emotion regulation could serve as a mechanism underlying comorbidity in ADHD and ASD, [8••, 13•]. In order to support the conceptualization of emotion regulation as a transdiagnostic feature, this review will outline the existing literature examining emotion regulation in children with these two common neurodevelopmental disorders. In addition, it will investigate if emotion regulation is associated with homotypic (e.g., co-occurring ADHD and ASD) and heterotypic comorbidity (e.g., co-occurring ADHD and depression) in children with ADHD and ASD. This will be followed by a discussion of the clinical implications as well as future directions for the study of emotion regulation in the field of neurodevelopmental disorders.

Emotion Regulation

Emotion regulation is a multidimensional construct that refers to a broad set of processes, which are responsible for monitoring, evaluating, and modifying emotional responses and experiences [14]. Emotions may be regulated in a variety of ways, for example, through strategies considered adaptive, such as cognitive reappraisal (i.e., reframing a potentially emotion-eliciting situation to impact emotional experience), or strategies considered maladaptive, such as expressive suppression (i.e., reducing an emotional response that is already underway) [15]. Currently, research on children with ADHD and ASD considers emotion regulation based on the functionalist definition, which focuses on how the processes of emotion regulation promote adaptive, goal-directed behavior [16•, 17.]. This perspective defines difficulties with emotion regulation, or emotion dysregulation, as any pattern of emotional expression or experience that interferes with behaviors oriented towards attaining a particular goal [18, 19...]. The goals that are undermined by difficulties with emotion regulation include children's abilities to feel better during difficult situations, think clearly and remain calm during challenges, strengthen relationships with family members and peers, engage in socially appropriate interactions, and accomplish other desired activities [19••].

Children's abilities to engage in effective emotion regulation depend on their developmental status and the organization of the neural systems supporting emotion regulation [20]. Developmental theories of emotion regulation describe the differential neuromaturation of subcortical structures (e.g., nucleus accumbens, amygdala) and the prefrontal cortex. Typical prefrontal cortex maturation is thought to yield increasingly effective top-down regulation of subcortical responses [21•]. However, the development of the prefrontal cortex and the efficiency of subcortical-cortical connections important for effective emotion regulation have been reported to be altered in children with neurodevelopmental disorders [22]. Consistent with this developmental neuroscience research, observational studies have shown that children with neurodevelopmental disorders display difficulties with emotion regulation, often failing to employ adaptive emotion regulation strategies and instead reacting impulsively to emotional stimuli with tantrums, "meltdowns", aggression, or selfinjury [23, 24•].

Emotion Regulation as a Transdiagnostic Feature Emotion regulation has been labeled as "mechanistically transdiagnostic" as it is thought to reflect a functional mechanism underlying the co-occurrence of multiple disorders (i.e., puts an individual at risk for more than one disorder) and treatments targeting emotion regulation should hypothetically alleviate the specific pathological processes driving symptoms across disorders [25, 26]. In line with this mechanistic definition of transdiagnostic, some research suggests that emotion regulation transmits risk for comorbidity in children with neurodevelopmental disorders such as ADHD and ASD [8••, 13•]. Further, emotion regulation holds promise as a target for

unified treatment approaches (i.e., treatments focused on a single set of therapeutic principles rather than diverse protocols) as existing evidence-based programs targeting emotion regulation in children with ADHD and ASD have demonstrated efficacy [27–29]. However, there has been limited examination of emotion regulation in children with other neurodevelopmental disorders, such as intellectual disability, communication disorders, specific learning disorders, and motor disorders [30–33].

Emotion Regulation in Neurodevelopmental Disorders

Attention-deficit/Hyperactivity Disorder (ADHD) In North America, 9.4% of children under the age of 17 have received a diagnosis of ADHD [34]. The diagnostic criteria for ADHD include symptoms of inattention, hyperactivity/impulsivity, or a combination of inattention and hyperactivity/impulsivity [3]. Recently, research has identified difficulties with emotion regulation as a common feature of ADHD in children and adolescents, and as a significant contributor to functional impairments in social-emotional, cognitive, behavioral, and interpersonal skills [17•, 35]. Research reports that a quarter of preschool children at risk for developing ADHD experience difficulties with emotion regulation [36]. Whereas, 21-66% of older children and adolescents diagnosed with ADHD exhibit difficulties with emotion regulation [37, 38]. Meta-analytic evidence reports that children with ADHD demonstrate difficulties with recognizing emotions, understanding emotions, emotional lability (i.e., poorly controlled shifts in emotions), and regulating both positive and negative emotional responses [24•]. These difficulties typically progress from temperamental characteristics of negative emotionality (e.g., fussy, angry) during infancy, to increased emotional lability during early childhood, to impairments in emotional awareness and emotional problem-solving across childhood and adolescence [17•, 39]. In studying the biological underpinnings of emotion regulation, research has shown that children with ADHD exhibit elevated parasympathetic nervous system reactivity (i.e., respiratory sinus arrhythmia/RSA) across emotion induction and suppression tasks [40, 41...]. Further, a recent review reported that youths with ADHD displayed deficits with bottom-up emotional activation in the amygdala and difficulties tracking changes in emotional valence (i.e., emotional evaluation) in the orbitofrontal cortex [42]. Given the increasing recognition of difficulties with emotion regulation as a common feature of ADHD, it has been recommended that patient-oriented conceptual frameworks of ADHD should be re-configured to include emotion regulation as a target for treatment [41...]. In line with this recommendation, emotion regulation training components have been included in psychoeducational parenting interventions for families with

children with ADHD [43, 44] and mindfulness-based programs for children with ADHD, with promising results [29]. Nonetheless, intervention approaches targeting emotion regulation have yet to be widely applied for children with ADHD.

ADHD and Comorbid Disorders Research indicates high rates of homotypic and heterotypic comorbidity in children diagnosed with ADHD. Estimates from community and clinical samples indicate that 55-70% of children with ADHD experience homotypic comorbidity and 35-50% of children with ADHD experience heterotypic comorbidity [45, 46]. In terms of homotypic comorbidities, ADHD frequently co-occurs with learning disorders, communication disorders, motor disorders (e.g., Tourette Syndrome), and/or ASD; whereas, in terms of heterotypic comorbidities, ADHD commonly cooccurs with depression, anxiety disorders (e.g., social anxiety disorder, specific phobia), and/or oppositional defiant disorder (ODD) [46]. Given that emotion regulation deficits are a common correlate of ADHD and other disorders (e.g., ASD, depression, anxiety disorders), it has been suggested that emotion regulation may be a shared risk process that underlies comorbidity in children with ADHD [47•]. For example, greater difficulties with auditory and visual emotion recognition are predictive of increased rates of comorbid ADHD and ASD [48•]. Further, preschool children who experience difficulties with emotion regulation have increased odds of cooccurring symptoms of ADHD and anxiety [36]. Similarly, in older children with ADHD, difficulties with emotion regulation are associated with increased risk for comorbid depression and anxiety [17•, 49–51]. Although evidence suggests that emotion regulation training such as instruction on how to employ adaptive strategies (e.g., cognitive reappraisal) could improve negative emotionality and behavioral dysregulation in children with ADHD and other comorbid disorders [52], research has yet to examine the efficacy of interventions targeting emotion regulation in children with ADHD and comorbid disorders.

Autism Spectrum Disorders (ASD) Recent North American prevalence estimates indicate that 2.5–2.8% of children under the age of 17 have received a diagnosis of ASD [53, 54]. The core diagnostic criteria for ASD include impairments in social communication, peer relationships, and the presence of restrictive and repetitive behaviors [3]. In addition to these core impairments, individuals with ASD often experience emotional distress and difficulties with emotion regulation [16•], which are thought to underlie the core deficit in social communication that defines ASD [23]. The estimated percentage of children with ASD who experience difficulties with emotion regulation is difficult to ascertain, as poor emotion regulation is considered inherent in ASD [55•]. Research suggests that difficulties with emotion regulation may be a strong predictor of the severity of ASD symptomatology during childhood [56]. Further, compared to typically developing children, preschool children with ASD display less adaptive emotion regulation strategies such as social support seeking and more maladaptive strategies such as avoidance and distraction [57, 58]. Similarly, older children and adolescents with ASD employ adaptive emotion regulation strategies including cognitive reappraisal less frequently and maladaptive strategies such as expressive suppression more frequently [59•]. Moreover, they experience difficulties regulating impulsivity to emotional events and display more negative affect compared to individuals without ASD [23, 60]. Research has begun to examine the biological pathways that may explain the associations between ASD and emotion regulation deficits, and found that children with ASD display decreased cardiac vagal control (i.e., heart rate variability) and parasympathetic nervous system reactivity (i.e., RSA) [23, 61]. Further, across the lifespan, ASD is associated with decreased activity in the nucleus accumbens and amygdala during the regulation of positive and negative emotions, as well as reduced prefrontal cortex activation [23, 62]. Although existing psychosocial interventions for ASD have traditionally focused on specific impairments (e.g., cognitive-behavioral therapy for social anxiety), there is increasing support for considering emotion regulation as a treatment target in ASD [16•, 63]. Recent mindfulness-based treatments and cognitive-behavioral therapy (CBT) approaches that teach effective strategies for regulating emotions to children with ASD and sometimes their parents have demonstrated preliminary feasibility, improvements in emotion regulation skills, and potential reductions in ASD symptoms [28•, 64, 65]. However, additional research on the frequency and progression of emotion regulation deficits in children with ASD and rigorous intervention work is needed to buttress the current evidence.

ASD and Comorbid Disorders Due to the impairments associated with ASD, it is sometimes difficult to determine if the child has co-occurring conditions; however, children with ASD are thought to experience especially high rates of comorbid symptoms and disorders [66]. It is estimated that 50% of children with ASD experience homotypic comorbidity and 70-80% experience heterotypic comorbidity [67, 68]. In terms of homotypic comorbidities, ASD commonly co-occurs with intellectual disability, communication disorders, motor disorders (e.g., Tic Disorder), and/or ADHD; whereas, in terms of heterotypic comorbidities, ASD frequently co-occurs with depression, anxiety disorders (e.g., social anxiety disorder, specific phobia, obsessive compulsive disorder), and/or ODD [69, 70]. Given the ubiquity of deficits in emotion regulation in children with ASD and other comorbid disorders, emotion regulation has been proposed as a mechanism that transmits risk for homotypic and heterotypic comorbidity in individuals with ASD [71]. Recent research supports this proposition, as children with ASD and intellectual disability display higher

levels of observed emotional lability and slower recovery from emotional experiences during tasks requiring independent regulation (i.e., a "locked box" frustration task) [56]. Also, children and youth with ASD who employ less adaptive emotion regulation strategies (i.e., cognitive reappraisal) in favor of more maladaptive emotion regulation strategies (i.e., expressive suppression, rumination) display more symptoms of depression and anxiety [72-74]. In line with this evidence, emotion regulation has been recommended as a target for evidence-based treatments in order to improve social communication, promote adaptive emotion regulation, and reduce psychiatric symptoms in children with ASD [75, 76..]. Similar to the interventions developed for the treatment of ASD, emotion regulation techniques have been incorporated into CBT approaches for children with ASD and comorbid disorders, with significant improvements noted in emotion regulation abilities, adaptive behaviors, and mental health outcomes [77-79].

Discussion

This review found evidence of difficulties with emotion regulation across distinct neurodevelopmental disorders, specifically ADHD and ASD. This suggests that clinicians, researchers, and caregivers may benefit from an increased awareness that difficulties with emotion regulation are a common deficit in children with ADHD and ASD. Further, research indicates that difficulties with emotion regulation transmit risk for both homotypic and heterotypic comorbidity in children with ADHD and ASD [17•, 49-51, 72-74]. These findings are consistent with the recognition of emotion regulation as a core underlying dimension common across psychiatric disorders [7, 80] and research suggesting that emotion regulation may serve as a transdiagnostic feature in neurodevelopmental disorders [8.., 13.]. Moreover, the identification of a mechanism underlying comorbidity across neurodevelopmental and psychiatric disorders in children suggests that the present boundaries based on diagnostic groupings (i.e., homotypic versus heterotypic) warrant reconsideration [6]. Thus, although it can be clinically helpful and scientifically justified to retain diagnostic distinctions, it is important to recognize that the thresholds for defining disorders are arbitrary and to consider complexity (i.e., heterogeneity in symptoms, outcomes, and treatment responses) over reductionism [2].

Overall, this review provides support for the conceptualization of emotion regulation as a mechanism underlying comorbidity in children with ADHD and ASD and as a potential target for unified treatment approaches. Additional support for the conceptualization of emotion regulation as a transdiagnostic feature comes from the limited body of research examining emotion regulation in other neurodevelopmental disorders. In brief, research shows that compared to typically developing children, children with intellectual disability experience more difficulties in social-emotional regulation (i.e., the regulation of emotional responses during social interactions) [30, 81]. Also, other studies report that children with communication disorders exhibit high levels of emotional reactivity and negative emotional expressions [31, 33]. However, the work on other neurodevelopmental disorders is less conclusive, with research suggesting that children with specific learning disorders may suppress their emotions [82], and some children with motor disorders may display emotional lability [32]. Overall, research has only just begun to examine emotion regulation in other neurodevelopmental disorders, and it is too early to draw definitive conclusions as to whether emotion regulation represents a transdiagnostic feature across all neurodevelopmental disorders. To determine this, future research is needed, which comprehensively examines emotion regulation in children with intellectual disability, communication disorders, specific learning disorders, and motor disorders.

The findings from this review have pragmatic implications for clinical research involving children with neurodevelopmental disorders. It is important to note that within a singular neurodevelopmental disorder, there can be heterogenous manifestations of difficulties with emotion regulation. For example, children with ADHD can display different patterns of difficulties with emotion regulation (e.g., high or low emotional reactivity) during mood induction tasks [83]. These different patterns may indicate the presence of clinically and etiologically distinct groups of children within a shared diagnostic category. Given that difficulties with emotion regulation are also associated with symptom severity in neurodevelopmental disorders [17•, 56], different patterns of difficulties with emotion regulation likely impact children's performance on neuropsychological assessments and clinical prognosis. Further, difficulties with certain processes of emotion regulation may be more prominent in specific disorders. For instance, one study found that although children with ADHD, ASD, and Tourette Syndrome all display significant difficulties with self-regulation, children with Tourette syndrome demonstrate more problems in emotional control than children with ADHD or ASD [84]. Thus, although emotion regulation may represent a treatment target for multiple disorders, it will be important to consider which emotion regulation skills to incorporate into interventions (e.g., CBT, mindfulness-based programs, parent training) based on the intended population.

Future Directions

In order to better understand how emotion regulation impacts clinical assessment and treatment outcomes in children with neurodevelopmental disorders, there first needs to be a consistent conceptualization of emotion regulation across the field of neurodevelopmental disorders. Currently, the literature examining emotion regulation in children with ADHD and ASD considers the functionalist definition of emotion regulation (i.e., the processes of emotion regulation that promote goaldirected behavior) [16•, 17•]. However, research examining emotion regulation in other neurodevelopmental disorders, such as intellectual disability and communication disorders [30, 31], defines emotion regulation in the context of socialemotional competence (i.e., the processes of emotion regulation within social relationships) [85]. Historically, the term "emotion regulation" has been used to refer to a remarkable range of developmental processes, with research often diverging based on a specific definition of emotion regulation [14]. In order to integrate multiple approaches to the study of emotion regulation, the developmental perspective seeks to understand how biological and relational influences contribute interactively to the development of difficulties with emotion regulation [19., 21.]. In accordance with the developmental perspective, future research in children with neurodevelopmental disorders should adopt this multilevel approach (i.e., considering the interactions between biological and relational influences) to the study of emotion regulation. This multilevel approach will also support future research evaluating the biological pathways (e.g., neurobiological systems, epigenetic modifications) which could explain the associations between neurodevelopmental disorders and difficulties with emotion regulation.

In order to accurately assess the construct of emotion regulation in children with neurodevelopmental disorders and the efficacy of potential treatments, reliable and universally accepted measures of emotion regulation need to be developed [41••]. Experts acknowledge that there is currently a lack of ecologically valid measures available for evaluating emotion regulation in both non-clinical and clinical contexts, including in children with neurodevelopmental disorders [86, 87...]. Additionally, there are methodological issues in regard to separating the subdomains of emotional competence (i.e., emotion knowledge, emotion regulation, emotion expression) from emotion regulation in children [88, 89•]. Relatedly, a recent review suggested that although many childhood measures of emotion regulation (e.g., Emotion Regulation Checklist, Penn Emotion Differentiation Test) purportedly assess this construct, they are actually assessing broader aspects of emotional competence and not solely emotion regulation [27]. It is also important to recognize that the measurement of emotion regulation is complex in children with neurodevelopmental disorders due to complicating factors, such as the challenges in assessing children with varying levels of intellectual and verbal abilities, the unclear phenomenology of symptoms, and the limited availability of measures specifically adapted for these populations [67]. After this fundamental measurement issue is addressed, the different patterns of difficulties with emotion regulation in children with neurodevelopmental

disorders can be better delineated and targets for future intervention approaches can be identified. Following which, research will be needed to develop novel treatments targeting difficulties with emotion regulation and rigorously evaluate the effectiveness of these novel approaches for children with neurodevelopmental disorders.

Conclusions

This review found evidence that emotion regulation constitutes a transdiagnostic feature in two common neurodevelopmental disorders, ADHD and ASD. Research is needed to examine emotion regulation in other neurodevelopmental disorders, such as intellectual disability, communication disorders, specific learning disorders, and motor disorders. Further, the field of neurodevelopmental disorders needs to examine emotion regulation from a developmental perspective (i.e., considering how biological and relational influences interact to shape emotion regulation) and develop valid assessments of emotion regulation in children with neurodevelopmental disorders. Gaining a better understanding of transdiagnostic features, such as emotion regulation, and how they underlie the co-occurrence of neurodevelopmental disorders is integral for clinical assessment, the development of research protocols, treatment planning, and the organization and delivery of health care services.

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Compliance with Ethical Standards

Conflict of Interest Gillian England-Mason declares no conflicts of interest relevant to this manuscript.

Human and Animal Rights and Informed Consent This article does not contain any studies with human or animal subjects performed by any of the authors.

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