



Emotion regulation and development in children with autism and 22q13 Deletion Syndrome: Evidence for group differences[☆]

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ABSTRACT

Emotion regulation (ER) abilities and developmental differences were investigated among 19 children with autism and 18 children with 22q13 Deletion Syndrome (a rare chromosomal disorder with certain autistic symptoms). The purpose of this study was to examine the phenotypic similarities between the two disorders. ER was measured by the Temperament and Atypical Behavior Scale (TABs) and development was measured by the Developmental Profile – Third Edition (DP-3). It was hypothesized that children with autism would score higher on the Detached domain of the TABs because the domain is associated with autism spectrum disorders. It was also hypothesized that because of similarly well established cognitive and communicative deficits between the two populations, no significant differences in development would be found. Results indicated that the autism group was significantly more impaired on the Detached domain of the TABs and scored higher on every domain of the DP-3 except in social-emotional skills. These findings differentiate the phenotypes of the two disorders and suggest that detached emotional behavior is more salient among children with autism. Early intervention programs that target the improvement of ER and social skills may ensure healthier behavioral outcomes for these individuals.

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1. Introduction

Emotion regulation (ER) involves the regulation of emotions by monitoring expressive behavior in order to achieve one's goals (Carlson & Wang, 2007; Thompson, 1991). In addition, ER refers to the behaviors involved in coping emotionally with both positive and negative feelings, such as pleasure, fear, and anxiety (Kopp, 1989). ER is a crucial developmental task for children (Stifter, Spinrad, & Braungart-Rieker, 1999), and the acquisition of effective regulatory strategies plays an important role in many aspects of behavior, such as in guiding early social relationships (Butler, Lee, & Gross, 2007; Dennis, Malone, & Chen, 2009).

Not all children learn effective methods of ER. Some children become emotionally dysregulated, a pattern of unhealthy regulatory skills that results in an inability to be soothed, difficulties returning to homeostasis, and problems maintaining positive affect (Keenan, 2000). According to the Diagnostic and Statistical Manual for Mental Disorders, Fourth Edition, Text Revision (DSM-IV-TR), emotion dysregulation is implicated in over half of all psychiatric disorders (American Psychiatric

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Association [APA 2000; Gross, 1998]). Identifying unhealthy patterns of ER can provide information regarding risk factors for later psychopathology, as well as aid in the creation of specialized interventions for emotion dysregulation.

1.1. *The importance of studying emotion regulation in atypical populations*

Although the main focus of ER research has been conducted with typically developing populations, some studies have recently addressed these skills in children with developmental disabilities (e.g., Jahromi, Gulsrud, & Kasari, 2008; Konstantareas & Stewart, 2006). In particular, children with autism have been of special interest to researchers who study emotional development because of the severe social–emotional deficits associated with the disorder (Loveland, 2005). The DSM-IV-TR lists the emotion-related deficits among this population as a lack of awareness of others' feelings, absence of facial expressiveness, intolerance to transitions and environmental change, and abnormal comfort-seeking during times of stress (APA, 2000; Cole, Michel, & Teti, 1994).

Although the deficits in ER among individuals with autism are fairly well established, it is of clinical and diagnostic interest to compare ER abilities between children with autism and children with other types of developmental disabilities. For example, various genetic disorders have been uncovered recently that result in behavioral symptoms similar to those in autism, such as severe cognitive impairment, social–emotional difficulties, and language delays (Abrahams & Geschwind, 2008). A rare population of children with 22q13 Deletion Syndrome (also known as Phelan McDermid Syndrome) has been identified through improved cytogenetic techniques such as fluorescence *in situ* hybridization (FISH) technology (Havens, Visootsak, Phelan, & Graham, 2004). 22q13 Deletion Syndrome results from partial microdeletion of the SHANK3 gene on chromosome 22q13.3 (Phelan et al., 2001). Symptoms of this syndrome include hypotonia (i.e., poor muscle tone), moderate to severe intellectual disability, repetitive behaviors, severely impaired speech, accelerated physical growth, and increased tolerance to pain, among other physical anomalies (Cohen et al., 2005; Phelan et al., 2001). Examining ER abilities among children with autism and 22q13 Deletion Syndrome can help researchers differentiate between the phenotypes of the two conditions. Studies that highlight the phenotypic overlaps between autism and 22q13 Deletion Syndrome most often focus on the severe language and communication deficits that are present in both disorders (e.g., Durand et al., 2006; Manning et al., 2004). However, the social–emotional overlaps between autism and 22q13 Deletion Syndrome remain largely unknown.

1.2. *Emotion regulation in children with autism*

ER impairments among children with autism are well documented (Loveland, 2005). Often, emotion dysregulation starts in infancy. As Gomez and Baird (2005) suggest, these ER difficulties may represent the earliest risk factor for autism, even before language and cognitive delays are apparent. The disorder is rarely diagnosed before the age of 2 or 3, despite many parents' earlier concerns. Identification of ER deficits may be one way that clinicians can identify autism much earlier in children, at just 1 or 2 years of age (Gomez & Baird, 2005). For example, in a case study of a child with autism, ER impairments marked the earliest symptom of the disorder and were evident before the child was 12 months of age (Dawson, Osterling, Meltzoff, & Kuhl, 2000). Another study involved identifying markers that may represent early indicators for autism (see Adrien et al., 1991). In order to characterize these behaviors, Adrien et al. (1991) analyzed home movies of children with autism from the ages of birth to two years. Results showed that severe emotion-related deficits represented the early impairment that best predicted the development of autism.

There are several hypotheses regarding the cause of ER deficits in children with autism. Gomez and Baird (2005) hypothesize that impairments in ER may be sensory related. In typically developing children, ER is a skill that develops in infancy and is learned through sensory interactions with the environment, becoming more sophisticated as they grow older to include tactics such as delay of gratification (Gomez & Baird, 2005). In contrast, infants with autism experience severe sensory deficits and hypersensitivities (see Baranek et al., 2007; Leekam, Nieto, Libby, Wing, & Gould, 2007). They often have difficulty regulating sleep–wake cycles, are extremely sensitive to touch, and lack the desire to be comforted by a caregiver (Gomez & Baird, 2005). As infants with autism grow older, they use poorer ER strategies compared to their typically developing peers and find it particularly difficult to re-establish a state of equilibrium once they are emotionally upset by a stressful event (Loveland, 2005). For example, a study by Konstantareas and Stewart (2006) investigated ER in children with autism by exposing them to a frustrating situation. Results indicated that compared to typically developing children, individuals with autism showed poorer ER strategies and demonstrated a greater inability to be soothed.

From a developmental perspective, emotion dysregulation may be related to a lack of language proficiency. Many individuals with autism experience severe language delays or are non-verbal. According to Kopp (1989), the understanding of emotions may be contingent upon the development of language, as it provides children with a multipurpose vehicle to manage strong emotions and employ more sophisticated ER strategies. Furthermore, with spoken words, children are able to vocalize their needs and gain feedback on their performance from others. In addition to language deficits, poor social skills common to children with autism may also be related to their emotion dysregulation. For example, as ER methods become increasingly more effective and complex, peer interactions and social abilities also improve, and vice versa (Kopp, 1989). Social impairments and poor ER skills have a reciprocal relationship among children with autism. Deficits in social abilities contribute to their poor ER (Kopp, 1989; Loveland, 2005), and their ER impairments lead to peer difficulties such as hostility, irritability, and non-compliance during play (Bieberich & Morgan, 2004).

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