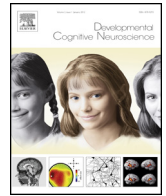




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Attention to novelty versus repetition: Contrasting habituation profiles in Autism and Williams syndrome

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ABSTRACT

Background: Abnormalities in habituation have been documented in Autism Spectrum Disorder (ASD) and Williams syndrome (WS). Such abnormalities have been proposed to underlie the distinctive social and non-social difficulties that define ASD, including sensory features and repetitive behaviours, and the distinctive social phenotype characterizing WS.

Methods: We measured habituation in 39 preschoolers with ASD, 20 peers with WS and 19 typically developing (TD) children using an eye-tracking protocol that measured participants' duration of attention in response to a repeating stimulus and a novel stimulus presented side by side across multiple trials.

Results: Participants in the TD group and the WS group decreased their attention toward the repeating stimulus and increased their attention to the novel stimulus over time. Conversely, the ASD group showed a similar attentional response to the novel and repeating stimuli. Habituation was correlated with social functioning in the WS but not in the ASD group. Contrary to predictions, slower habituation in ASD was associated with lower severity of repetitive behaviours.

Conclusions: Habituation appears to be intact in WS and impaired in ASD. More research is needed to clarify the nature of the syndrome-specific patterns of correlations between habituation and social and non-social functioning in these neurodevelopmental disorders.

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1. Introduction

Individuals with Autism Spectrum Disorder (ASD) often show prolonged attention to visual repetition, such as watching the spin cycle of a washing machine (Kanner, 1943; Baron-Cohen, 2006), and reduced attention to social stimuli (Klin et al., 2015). Both phenomena, which reflect the pathognomonic social impairments and

behavioural rigidity that defines ASD, have been linked to potential abnormalities in habituation (Green et al., 2015; Guiraud et al., 2011; Ramaswami, 2014).

Habituation is defined by the progressive decrement in response to a stimulus when it is repeated (Thorpe, 1966; Schmid et al., 2015). From infancy onwards, repeated exposure to an event make children less interested and responsive to it over time. This process reflects a strategic allocation of processing resources away from what is “already known” in favour of “what is not known yet”, thus facilitating learning and adaptive response to changes in the environment (Groves and Thompson, 1970; Lloyd et al., 2014). Habituation in infancy is thought to reflect information processing efficiency and it is predictive of cognitive functioning later in life (Bornstein and Sigman, 1986; Colombo, 1993).

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It has been proposed that lack of habituation to sensory inputs might result in an exaggerated perception of changes in the environment in children with ASD. This in turn would lead to sensory overstimulation, distress and the perception of the environment as highly unpredictable, with repetitive behaviours serving as a way of constraining and controlling this unpredictability by imposing structure (Dawson and Lewy, 1989; Uljarevic, 2013). This account is consistent with the original descriptions of Leo Kanner (1943), and more detailed theoretical accounts put forward by other scholars (e.g. Hutt et al., 1964; Ornitz and Rivto, 1968; Kinsbourne, 1980), who suggested the potential role of repetitive behaviours as a means of warding off anxiety caused by inefficient habituation to sensory stimuli (see also Gomot et al., 2008; Leekam et al., 2011).

Some empirical research appears to be consistent with this framework. For example, James and Barry (1980) documented lack of habituation in response to the repeated observation of visual stimuli (white circles on a black background) in ASD. More recently, Perry et al. (2007) documented that adults with ASD were slower compared to typical peers in dampening their response to repeated stimuli in a startle response paradigm, and Guiraud et al. (2011) reported reduced habituation to repeated sounds in infants at high risk for autism. However, counterevidence exists, including reports of normative habituation in ASD, and a lack of clear associations between habituation and repetitive behaviours (Baranek et al., 2006; Rogers and Ozonoff, 2005; McCormick et al., 2014). Some of the inconsistent findings might reflect heterogeneity in habituation profiles within the ASD population. Indeed, research has suggested the existence of subgroups, characterized by different habituation response to sensory stimuli within the ASD population. For example, Schoen et al. (2008) identified two subgroups within a sample of 40 children with ASD (age range: 5–15 years), one characterized by slower latency and faster habituation and the other by faster latency and slower habituation in response to sensory stimuli. Similar findings were reported by Hirstein et al. (2001). Additionally, Green et al. (2015) reported decreased neural habituation in sensory cortices and the amygdala in a subgroup of individuals with ASD who were characterized by sensory over-responsiveness at the behavioural level.

Abnormalities in habituation have been also linked to the social deficits characterizing ASD. Webb et al. (2010) documented slowed visual habituation to faces in toddlers with severe symptoms of ASD, but not in those whose symptoms were mild. Moreover, the slower habituation rate was correlated with poor social and communication skills. Similarly, other studies reported reduced neural habituation (lack of decrease in amygdala responsiveness) in response to faces in children and adults with ASD (Kleinhans et al., 2009; Swartz et al., 2013; Wiggins et al., 2014). Atypical habituation was associated with severity of social impairment, and modulated by both the properties of the stimuli (emotions displayed in the face stimuli) and the participants' characteristics. Based on this body of research, it has been proposed that lack of habituation is causally related to the core social and non-social symptoms of autism, with social symptoms reflecting failure to habituate and consequent hyper-responsivity to social stimuli, and repetitive behaviours emerging as a coping strategy in response to over-responsivity to sensory inputs (Guiraud et al., 2011; Ramaswami, 2014; Sinha et al., 2014; Wiggins et al., 2014).

Somewhat paradoxically, a lack of habituation has also been proposed to be causally related to the behavioural features of Williams syndrome (WS), a condition characterized by symptoms that are remarkably different from those that define ASD. WS is a rare neurodevelopmental disorder (estimated prevalence of 1:7500 to 1:20,000; Stromme et al., 2002) presenting with impaired visuospatial abilities and social-pragmatic skills alongside an increased drive for social engagement. While WS and ASD present with similar difficulties in "reading" the meaning of people's gaze and facial

expressions (Tager-Flusberg and Skwerer, 2013), individuals with WS, in sharp contrast with those with ASD, show unusually intense interest toward social stimuli, in particular toward faces and emotional displays, as well as increased motivation for social interaction (Hocking, 2017; Riby and Hancock, 2008, 2009; Riby et al., 2013; Dodd and Porter, 2010; Doherty-Sneddon et al., 2009). Recently, it has been proposed that the increased social motivation characterizing WS might reflect a failure to habituate to faces, which causes social stimuli to appear unusually novel and interesting (Järvinen et al., 2012, 2013). Empirical findings supporting this notion include evidence of slow habituation in electrodermal activity response to social (and non-social) stimuli in adults with WS (Järvinen et al., 2012). However, no other studies have examined habituation in young children with WS.

In summary, a lack of habituation has been linked to both the sensory abnormalities and impaired social abilities in ASD, and the increased social motivation that characterizes the behavioural profile in WS. To resolve the logical inconsistency of a specific factor (reduced habituation) leading to opposing behavioural profiles (decreased social orienting/motivation in ASD and increased social orienting/motivation in WS), cross-syndrome research focused on habituation in ASD and WS is needed.

The current study addresses this issue by examining habituation in preschoolers with ASD, age- and IQ-matched children with WS, and a comparison group of typically developing (TD) children. In particular, we investigated whether (a) visual habituation to repeated stimuli differed across children with ASD, WS and TD; and (b) whether habituation rate was associated with social and non-social features within each group. On the basis of the literature discussed above, we predicted abnormal habituation across both the ASD and the WS groups. We also expected associations between habituation and both social features and repetitive behaviours in ASD, in keeping with previous research. Due to the lack of previous research, no specific hypotheses were proposed regarding the associations between habituation and social features in young children with WS.

2. Methods

2.1. Participants

The participants were 39 preschoolers with Autism Spectrum Disorder (ASD; mean age = 44.1 months), 20 children with Williams syndrome (WS; mean age = 50.8 months) and 19 typically developing (TD) children (mean age = 49.4 months). Participants with ASD were recruited through the Victorian Autism Specific Early Learning and Care Centre, an autism intervention program located at the La Trobe University Community Children's Centre. Participants in the WS group were recruited through the Williams Syndrome Family Support Group (Victoria) and the Williams Syndrome Association Australia. The TD children were recruited through a childcare setting affiliated with Macquarie University.

The diagnoses of ASD were previously made by community-based health care professionals and confirmed for the study using the Autism Diagnostic Observation Schedule-Second edition (ADOS-2) (Lord et al., 2012) administered by a clinician with demonstrated reliability in the use of this measure. Exclusion criteria for the ASD group included the presence of uncorrected hearing or vision impairment, and the presence of a major medical problem. All participants with WS had their diagnosis confirmed with the positive fluorescent in situ hybridisation (FISH) test and displayed the typical ~1.6 Mb heterozygous microdeletion at 7q11.23. The TD children had no reported neurodevelopmental disorders. The ASD and WS groups did not differ on language, cognitive level, motor skills or overall adaptive behaviour (Table 1). However, as expected,

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