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Abnormal processing of emotional prosody in Williams syndrome: An event-related potentials study

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ABSTRACT

Williams syndrome (WS), a neurodevelopmental genetic disorder due to a microdeletion in chromosome 7, is described as displaying an intriguing socio-cognitive phenotype. Deficits in prosody production and comprehension have been consistently reported in behavioral studies. It remains, however, to be clarified the neurobiological processes underlying prosody processing in WS.

This study aimed at characterizing the electrophysiological response to neutral, happy, and angry prosody in WS, and examining if this response was dependent on the semantic content of the utterance. A group of 12 participants (5 female and 7 male), diagnosed with WS, with age range between 9 and 31 years, was compared with a group of typically developing participants, individually matched for chronological age, gender and laterality. After inspection of EEG artifacts, data from 9 participants with WS and 10 controls were included in ERP analyses.

Participants were presented with neutral, positive and negative sentences, in two conditions: (1) with intelligible semantic and syntactic information; (2) with unintelligible semantic and syntactic information ('pure prosody' condition). They were asked to decide which emotion was underlying the auditory sentence.

Atypical event-related potentials (ERP) components were related with prosodic processing (N100, P200, N300) in WS. In particular, reduced N100 was observed for prosody sentences with semantic content; more positive P200 for sentences with semantic content, in particular for happy and angry intonations; and reduced N300 for both types of sentence conditions.

These findings suggest abnormalities in early auditory processing, indicating a bottom-up contribution to the impairment in emotional prosody processing and comprehension. Also, at least for N100 and P200, they suggest the top-down contributions of semantic processes in the sensory processing of speech. This study showed, for the first time, that abnormalities in ERP measures of early auditory processing in WS are also present during the processing of emotional vocal information. This may represent a physiological signature of underlying impaired on-line language and socio-emotional processing.

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1. Introduction

Williams syndrome (WS), a genetic neurodevelopmental disorder due to microdeletion in chromosome 7, has been described a syndrome with an intriguing socio-cognitive phenotype (Bellugi, Bihrle, Neville, Jernigan, & Doherty, 1992; Bellugi, Lichtenberger, Mills, Galaburda, & Korenberg, 1999; Bellugi, Marks, Bihrle, & Sabo, 1988; see Martens, Wilson, & Reutens, 2008 for a review).

The initial descriptions of individuals with WS made reference to their apparently preserved abilities of linguistic expression, as exemplified by complex and elaborated narratives along with an intense interest in being engaged in social communication (e.g., von Arnim & Engel, 1964). However, this keen interest in engaging in social interactions (coupled with an overfriendly personality and empathic behavior) tends to coexist with severe pragmatic impairments (Laws & Bishop, 2004), such as the difficulty to adjust the amount of speech production to the listener's interests and attitudes. For example, some narrative studies suggest that participants with WS use significantly more affective expressive prosody than individuals with Down syndrome and typically developing children (Gonçalves et al., 2004, 2010; Jones et al., 2000; Reilly, Klima, & Bellugi, 1991; Reilly, Losh, Bellugi, & Wulfeck, 2004), and that this pattern seems to be independent of the audience and on how many times they tell the story. In other words, the frequent use of dramatic devices and social hookers, used to capture the attention of the audience, may have been masking WS individuals' deficits in understanding social cues (Skwerer, Schofield, Verbalis, Faja, & Tager-Flusberg, 2007). This is corroborated by studies showing difficulties in effective deployment and interpretation of paralinguistic devices as illustrated by difficulties in theory-of-mind tasks (e.g., Sullivan & Tager-Flusberg, 1999; Tager-Flusberg & Sullivan, 2000), and deficits in the identification and discrimination of emotions (e.g., Catterall, Howard, Stojanovik, Szczerbinski, & Wells, 2006; Plesa-Skwerer, Faja, Schofield, Verbalis, & Tager-Flusberg, 2006; Tager-Flusberg & Sullivan, 2000), particularly negative emotions (Plesa-Skwerer et al., 2006).

One of the powerful paralinguistic cues routinely employed in verbal communication is prosody. Emotional prosody represents a paralinguistic device that allows human beings to represent and convey affect (Scherer, 1986). It relies on language suprasegmental features such as fundamental frequency (F0), sound intensity and duration (Hesling, Clément, Bordessoules, & Allard, 2005; Kotz & Paulmann, 2007; Wildgruber, Ackermann, Kreifelts, & Ethofer, 2006). The perception of emotional prosody is a multi-stage process that consists of (1) the analysis of acoustic features of spoken words, (2) deriving emotional significance from acoustic cues, (3) applying it in higher cognition operations, and (4) integrating emotional prosody in language processing (Hoekert, Bais, Kahn, & Aleman, 2008; Schirmer & Kotz, 2006; Wildgruber et al., 2006). Thus, the study of prosody processing may provide us with information on how individuals recognize and interpret sensory input (e.g., voice inflection), an ability that is crucial to social interactions and, in particular, to social reciprocity.

Behavioral studies on prosody processing in WS have found deficits in prosody comprehension (Catterall et al., 2006; Plesa-Skwerer et al., 2006; Skwerer et al., 2007), suggesting that, in spite of an easy sociability, these individuals may be impaired in their ability to use vocal cues to interpret emotional states particularly in the presence of a semantic conflict such as sarcasm or irony (Skwerer et al., 2007; Sullivan, Winner, & Tager-Flusberg, 2003). However, in spite of their difficulties in using prosody for semantic processing, individuals with WS still seem to perform better than participants with learning or intellectual disabilities on the recognition of emotional tone of voice in filtered speech (Plesa-Skwerer et al., 2006), suggesting that sensitivity for non-linguistic affective information may be relatively spared in WS. This seems to be consistent with the unusual profile of auditory processing that characterizes WS, which includes a keen interest in music and musical activities (Hopyan, Dennis, Weksberg, & Cytrynbaum, 2001; Levitin & Bellugi, 1998; Udwin, Yule, & Martin, 1987).

In spite of the few studies devoted to prosody processing in WS reviewed above, there is a dearth of data on this issue, in contrast to the number of studies focusing on the morphosyntactic and semantic aspects of language processing in WS individuals.

Importantly, electrophysiological studies of prosody processing in WS are, to the best of our knowledge, nonexistent. Due to their temporal resolution, Event Related Potentials (ERPs) (Coles & Rugg, 1995; Münte, Urbach, Düzel, & Kutas, 2000) provide valuable information on the order of msec about cognitive processes under consideration. As such, they afford a window of enquiry into the neural underpinnings of sensory and cognitive processes associated with prosody processing in WS.

ERP studies in normal individuals show that prosody comprehension has distinct electrophysiological signatures (e.g., Kotz & Paulmann, 2007; Paulmann & Kotz, 2008a; Paulmann & Kotz, 2008b; Paulmann, Seifert, & Kotz, 2010). Most studies examined interactions between semantics and prosody and these studies often used a 'prosody violation' approach where juxtapositions between two intonational patterns, or between semantics and prosody, were investigated. These studies reported late occurring negativities and positivities that indexed processing incongruities between the prosody in the initial and the final part of the sentence (e.g., initial happy prosody ending with sad prosody) or between semantics and prosody. For example, expectancy violations of integrative emotional prosodic and semantic information elicited a more negative-going component in the time window between 450 and 650 ms, while expectancy violations of emotional prosodic information were linked to a more positive-going component in the time window between 700 and 1000 ms, in a task using a cross splicing technique (Paulmann & Kotz, 2008a).

However, studies using naturalistic designs (i.e., in sentences delivered with either neutral or emotional intonation without artificially introducing discrepancy between sentence fragments, or between message and the tone with which it was delivered) are few. The existing ones (Paulmann & Kotz, 2008b; Paulmann et al., 2010) suggest that the differentiation of basic vocal emotional expressions from prosodically neutral sentences occurs around 200 ms, with emotional sentences eliciting less positive P200 amplitudes, irrespective of valence (positive vs. negative) (Paulmann & Kotz, 2008b). According to

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