



Development of socio-communicative skills in 9- to 12-month-old individuals with fragile X syndrome



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ARTICLE INFO

Article history:

Received 31 December 2013

Accepted 3 January 2014

Available online 27 January 2014

Keywords:

Communication impairment

Fragile X syndrome

Home videos

Socio-communicative development

Speech-language development

Video analysis

ABSTRACT

We investigated the early socio-communicative development of individuals with fragile X syndrome (FXS) by undertaking a retrospective analysis of family videos. Videos were analyzed to identify existing communicative forms and functions. Analyses were undertaken on seven children who were later diagnosed with FXS. The children were filmed when they were 9–12 months old and before being diagnosed. Fourteen different communicative forms and six different communicative functions were observed. All participants were observed to express the functions of 'Attention to self' and 'Answering', but none indicated 'Requesting action', 'Requesting information', 'Choice making', or 'Imitating'. Results suggest that children with FXS may have a limited range of communicative forms and functions when they are from 9 to 12 months of age. However, further research is necessary to gain a specific developmental profile of socio-communicative forms and functions in FXS.

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1. Introduction

Fragile X syndrome (FXS) is the most prevalent form of inherited intellectual disability and among the most prevalent genetic causes of autism spectrum disorder (ASD; Cohen et al., 2005). The molecular basis of FXS is an unstable expansion of a triplet repeat polymorphism in the regulatory region of the *FMR1* gene (Kaufmann & Reiss, 1999). As is typical of X-linked disorders, the most severe manifestations are seen among males. The core neurobehavioral features of FXS include variable cognitive impairment and a diverse group of behavioral abnormalities, including attention-deficit/hyperactivity disorder symptoms, anxious behavior, autistic behavior, and prominent stereotypies that, in a high proportion of individuals reach diagnostic threshold (Boyle & Kaufmann, 2010). Up to 90% of males with FXS display autistic-like features, such as perseveration, repetitive speech, and poor eye contact (Hagerman, 2002), while as many as 30–60% of FXS males meet

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diagnostic criteria for comorbid diagnosis of ASD (Harris et al., 2008; Kaufmann et al., 2004; Loesch et al., 2007). Anxiety can also lead to social interaction impairment, which can be further exacerbated by co-morbidity with ASD (Boyle & Kaufmann, 2010). Thus deficit in socio-communicative abilities, more pronounced in subjects with FXS and ASD (FXS + ASD), is central to FXS' psychopathology (Kaufmann et al., 2004; Losh, Martin, Klusek, Hogan-Brown, & Sideris, 2012; Martin, Losh, Estigarribia, Sideris, & Roberts, 2013; Roberts, Hatton, & Bailey, 2001; Roberts, Mirrett, Anderson, Burchinal, & Neebe, 2002; Roberts, Mirrett, & Burchinal, 2001; Rogers, Hepburn, Stackhouse, & Wehner, 2003). Commonly observed atypical behaviors in the socio-communicative domain include eye gaze avoidance, social withdrawal, attentional deficits, atypical play, and atypical imitation behaviors (Bailey et al., 1998; Baranek et al., 2005; Hessl, Glaser, Dyer-Friedman, & Reiss, 2006; Kaufmann et al., 2004; Rogers et al., 2003). Young children with a diagnosis of FXS are also reported to have delays in gestural development and speech-language acquisition (Abbeduto, Brady, & Kover, 2007; Finestack, Richmond, & Abbeduto, 2009; Roberts et al., 2002; Roberts, Mirrett et al., 2001).

A number of studies have focused on socio-communicative and speech-language development in individuals with FXS during the first year of life (Baranek et al., 2005, 2008; Hatton et al., 2009; Hinton et al., 2013; Mirrett, Bailey, Roberts, & Hatton, 2004; Prouty et al., 1988; Roberts et al., 2009; Roberts, Hatton et al., 2001). Most of the current knowledge in these domains is however based on retrospective questionnaires and interviews. Retrospective video analysis is another method that has been used to investigate pre-diagnostic socio-communicative and speech-language development in neurodevelopmental disorders, which are recognized during the toddler period or even later. Most such studies have focused on individuals with ASD (Baranek, 1999; Colgan et al., 2006; Ozonoff et al., 2011; Poon, Watson, Baranek, & Poe, 2012; Thorsen, Goldberg, Osann, & Spence, 2008; Watson, Crais, Baranek, Dykstra, & Wilson, 2013) and Rett syndrome (RTT; Bartl-Pokorny et al., 2013; Einspieler et al., 2013; Marschik, Kaufmann, et al., 2012; Marschik, Pini, et al., 2012; Marschik, Sigafos, et al., 2012; Marschik, Bartl-Pokorny, et al., 2013; Marschik, Kaufmann, et al., 2013). Although retrospective video analysis has proven to be a valuable tool to delineate early atypical behavioral patterns in ASD and RTT, to the best of our knowledge only one study used this approach in FXS during the first year of life (Baranek et al., 2005). They found *inter alia* delays in object play and unusual motor patterns such as repetitive leg movements or spinning of objects. Apart from a few variables, socio-communicative forms and functions were not in the focus of this study.

The present study aimed to investigate the early socio-communicative development of individuals with FXS using retrospective analysis of family videos. Videos were analyzed to identify communicative forms (e.g., body movements, vocalizations, gestures) and functions (e.g., imitation, requesting an object, commenting) in 9- to 12-month-old infants who were later diagnosed with FXS. We aimed to (a) describe socio-communicative forms, (b) delineate their socio-communicative functions, and (c) determine the difference between verbal and non-verbal communication strategies.

2. Methods

2.1. Participants

Seven children with an FXS diagnosis were included in the present study. Child 1 to Child 5 were male and Child 6 and Child 7 were female. We retrospectively analyzed their socio-communicative behaviors between 9 and 12 months of age, prior to diagnosis. Five participants, all singletons, came from monolingual German-speaking families; a twin pair (Child 3 and Child 4) had a bilingual family background (German-Spanish). Pregnancies and deliveries were uneventful in all individuals. Birth weights, birth lengths, and occipitofrontal circumferences were within the normal ranges. Clinical diagnosis revealed the following co-morbidities: Child 2 was reported to have an additional ASD diagnosis; Child 1, Child 2 and Child 6 had anxiety disorder; attention-deficit/hyperactivity disorder (ADHD) did not occur in our participants. The study was approved by the research ethics committee of the Medical University of Graz. All parents gave their informed consent for participation in the study and dissemination of the results.

2.2. Procedure

The procedures applied in this study were in accordance with those of our previous studies on socio-communicative forms and functions in children with classic RTT, the preserved speech variant (PSV) of RTT or normal development (Bartl-Pokorny et al., 2013; Marschik, Kaufmann, et al., 2012; Marschik, Bartl-Pokorny, et al., 2013). We analyzed audio-video footage of 224 min in communicative settings recorded during typical family routines (e.g., play situations, bathing, feeding) and special events (e.g., family gatherings) when the participants were 9–12 months old. At that time, none of the parents were aware of their children's medical condition. A research assistant, who was blind to the purpose of the study, checked the recordings for sufficient length and quality standards, copied the relevant recordings, and prepared them for coding.

All socio-communicative forms observed including body movements (e.g., reaching), facial expressions/eye movements (e.g., smiling, eye contact), non-linguistic vocalizations (e.g., laughing), (pre-)linguistic vocalizations (e.g., babbling) and gestures (e.g., demonstrating an object) were coded by the second and fourth authors using the Noldus Observer-XT (www.noldus.com). Based on the classification system of the Inventory of Potential Communicative Acts (IPCA; Sigafos, Arthur-Kelly, & Butterfield, 2006), we assigned the socio-communicative forms to one of 10 different socio-communicative functions: (a) 'Social convention' (e.g., greeting, orienting to name), (b) 'Attention to self' (e.g., getting attention, seeking comfort), (c) 'Reject/protest' (e.g., rejecting objects/activities), (d) 'Request object' (e.g., requesting a toy/food), (e) 'Request

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