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

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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 to 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.

Keywords

Communication impairment; Fragile X syndrome; Home videos; Socio-communicative development; Speech-language development; Video analysis

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

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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 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; Baranek et al., 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, Bartl-Pokorny, et al., 2013; Marschik, Kaufmann, et al., 2012; Marschik, Kaufmann, et al., 2013; Marschik, Pini, et al., 2012; Marschik, Sigafoos, et al., 2012). 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

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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, Bartl-Pokorny, et al., 2013; Marschik, Kaufmann, et al., 2012). We analyzed audio-video footage of 224 minutes 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 to 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; Sigafoos, 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 action' (e.g., help with walking/dressing), (f) 'Request information' (e.g., requesting the name of an object, requesting

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clarification), (g) ‘Comment’ (e.g., happy, sad), (h) ‘Choice making’ (e.g., choosing between two objects/activities), (i) ‘Answer’ (e.g., indicating yes/no in response to a question), (j) ‘Imitate’ (e.g., imitating another person’s speech/manual routine). In case of disagreement, the respective sequences were discussed within the team until consensus was achieved.

3 Results

3.1 Socio-communicative forms

Table 1 shows the communicative forms coded for each child. Fourteen different communicative forms (range: 2 to 11) were observed. They consisted of five different body movements, two facial expressions/eye movements, five types of non-linguistic vocalizations, one type of (pre-)linguistic vocalizations, and one gesture. In addition, one participant (Child 7) was observed to use hand flapping, which was coded as serving the functions of directing attention to herself and requesting an object.

3.2 Socio-communicative functions

Table 2 shows the communicative functions coded for each child. The number of functions per participant ranged from three (Children 3 and 7) to six (Child 5). Whereas all participants used at least one communicative form to express ‘Attention to self’ and ‘Answering’, none of them showed any behaviors indicating ‘Requesting action’, ‘Requesting information’, ‘Choice making’ or ‘Imitating’.

3.3 Non-verbal behavior versus non-linguistic or (pre-)linguistic vocalizations

Table 3 shows the coding of non-verbal behaviors versus non-linguistic or (pre-)linguistic vocalizations for each child. For ‘Social convention’, ‘Attention to self’ and ‘Answering’ more participants were observed to use non-verbal behaviors over verbal behaviors (i.e., non-linguistic or (pre-)linguistic vocalizations). Only for ‘Commenting’ did non-linguistic vocalizations dominate over non-verbal behaviors. (Pre-)linguistic vocalizations were observed in one participant each with respect to the functions of ‘Attention to self’ (Child 1), ‘Commenting’ (Child 5) and ‘Answering’ (Child 5).

4 Discussion

Coding behaviors in terms of their perceived communicative functions, based on the IPCA, has been used to identify the socio-communicative skills of children and adults with RTT mainly by interviewing caregivers and teachers (e.g., Didden et al., 2010; Sigafoos et al., 2000; Sigafoos, Woodyatt, Tucker, Roberts-Pennell, & Pittendreigh, 2000), ASD (Braddock et al., 2013; Keen, Woodyatt, & Sigafoos, 2001; Keen, Woodyatt, & Sigafoos, 2002; Keen, Woodyatt, & Sigafoos, 2005) and Angelman syndrome (Didden et al., 2009). This approach has also been applied to a more direct assessment, involving the retrospective analysis of videotapes (Bartl-Pokorny et al., 2013; Marschik, Bartl-Pokorny, et al., 2013; Marschik, Kaufmann, et al., 2012). Having used this latter approach to identify early socio-communicative forms and functions in RTT, we have here extended the literature by applying our retrospective analysis of videotapes and IPCA methodology to examine the early socio-communicative development in individuals with FXS.

The results of our study indicate that these seven children with FXS were observed to use a varying number of communicative forms (range: 2 to 11) when they were between 9 and 12 months of age. In a previous study, we found a comparatively limited set of communicative forms used by individuals with classic RTT during the same age period (Bartl-Pokorny et al., 2013), whereas a normally developing 9- to 12-month-old infant was observed to use more communicative forms (Marschik, Bartl-Pokorny, et al., 2013). At the end of the first year of life, typically developing children acquire a decent number of communicative gestures and use them for various purposes and the use of such gestures has been found to be closely related to vocabulary development (Rowe, Özçalıskan, & Goldin-Meadow, 2008; Tomasello, Carpenter, & Liszkowski, 2007). In the present study, however, we observed only one child who used a gesture (i.e., demonstrating an object). Restricted gestural repertoires in young individuals with FXS were already reported by Roberts et al. (2002). From the results of our study, it remains open whether the absence of communicative gestures in the given data set is due to a developmental delay or rather represents a set of forms never to develop.

Communicative forms (Table 1) were assigned to three to six different communicative functions based on the categories of the IPCA (see Table 2). Whereas all participants were coded as expressing the functions of ‘Attention to self’ and ‘Answering’, none of them was observed to ‘Imitate’, ‘Request information’, ‘Request action’ or ‘Make choices’. This is an interesting finding as it resembles the findings of early communicative functions in individuals with classic RTT, who showed the same pattern except for ‘Imitating’ (three out of six individuals with RTT had imitation behavior; Bartl-Pokorny et al., 2013). Also Rogers et al. (2003) reported less imitation behavior for toddlers with FXS. Fewer participants with FXS (only one) exhibited ‘Rejecting/protesting’ as compared to individuals with classic RTT (four out of six; Bartl-Pokorny et al., 2013).

In general, non-verbal behaviors were more commonly observed than non-linguistic vocalizations to cover communicative functions, especially that is for ‘Social convention’, ‘Attention to self’ and ‘Answering’. However, non-verbal behaviors and non-linguistic vocalizations were observed equally often for the functions of ‘Rejecting/protesting’ and ‘Requesting an object’; whereas non-linguistic vocalizations were more commonly observed than non-verbal behaviors for ‘Commenting’ (Table 3). (Pre-)linguistic vocalizations were coded as indicating three communicative functions. This picture is slightly different from what we found for individuals with classic RTT at the same age. In RTT non-verbal behaviors dominated over non-linguistic vocalizations in seven out of eight observed functional categories; whereas (pre-)linguistic vocalizations were not observed at all (Bartl-Pokorny et al., 2013).

In this study, there was only one individual who was later diagnosed with FXS and ASD (Child 2). Interestingly, his early socio-communicative forms and functions showed a similar profile to the other six participants. This is in contrast to findings of Rogers et al. (2003) who reported an influence on the imitation behavior by the presence of autistic features. As mentioned above, in our study none of the participants showed imitation behaviors; however, our observations were in a younger group.

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Although intriguing, the results of the present study must be interpreted with caution due to the well known methodological limitations of retrospective video analysis (e.g., Marschik & Einspieler, 2011; Palomo, Belinchón, & Ozonoff, 2006), the small sample size, and the lack of a control group or available norms for the IPCA protocol. One should also keep in mind the age specificity of various communicative functions; for example, 'Requesting information' might not occur during this age period, not even in typically developing toddlers (Chapman 2000). Despite these shortcomings, this study is the first to investigate the repertoire of socio-communicative forms and functions by means of a standardized protocol in 9- to 12-month-old individuals with FXS using retrospective video analysis. Our findings 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, but further research is necessary to gain a more detailed picture of their socio-communicative development.

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Highlights

- This is the first study to assess communicative forms and functions in 9- to 12-month-old infants later diagnosed with FXS.
- All children later diagnosed with FXS had a limited gestural repertoire.
- The overall repertoire of communicative forms (e.g., babbling) appears to be limited.
- All participants were observed to express ‘Attention to self’ and ‘Answering’.
- The functions ‘Requesting action’, ‘Requesting information’, ‘Choice making’, and ‘Imitating’ were never observed.

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Table 1

Socio-communicative forms observed for the seven participants between 9 and 12 months of age who were later diagnosed with FXS (number of participants showing a particular behavior is given in brackets)

Body movements	Facial expression/eye movements	Non-linguistic vocalizations	(Pre-)linguistic vocalizations	Gestures
Reaching (n=5)	Eye contact (n=7)	Unspecified vocalizations (n=5)	Babbling (n=2)	Demonstrating an object (n=1)
Touching person (n=2)	Smiling (n=6)	Fussing (n=5)		
Turning away (n=1)		Crying (n=3)		
Retaining object (n=1)		Pleasure vocalizations (n=4)		
Manual routine (n=1)		Laughing (n=4)		

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Table 2

Socio-communicative functions observed (■) or not observed (□) for each participant between 9 and 12 months of age who was later diagnosed with FXS. Categories according to the Inventory of Potential Communicative Acts (IPCA; Sigafoos et al., 2006)

	Social convention	Attention to self	Reject/protest	Request object	Request action	Request information	Comment	Choice making	Answer	Imitate	Total
Child 1	□	■	□	■	□	□	■	□	■	□	4
Child 2*	□	■	□	■	□	□	■	□	■	□	4
Child 3#	■	■	□	□	□	□	□	□	■	□	3
Child 4#	■	■	□	□	□	□	■	□	■	□	4
Child 5	■	■	■	■	□	□	■	□	■	□	6
Child 6	■	■	□	■	□	□	■	□	■	□	5
Child 7	□	■	□	■	□	□	□	□	■	□	3
Total	4	7	1	5	0	0	5	0	7	0	

*FXS+ASD

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Table 3

(a) Non-verbal behavior (●) versus non-linguistic (▲) or (pre-)linguistic (♦) vocalizations to express the socio-communicative functions according to the Inventory of Potential Communicative Acts (IPCA; Sigafoos et al., 2006); **(b)** number of participants exhibiting relevant behaviors within the ten IPCA-categories

	Social convention	Attention to self	Reject/protest	Request object	Request action	Request information	Comment	Choice making	Answer	Imitate
(a)										
Child 1		●▲♦		●▲			●▲		●▲	
Child 2*		●▲		●▲			●▲		●▲	
Child 3#	●	●							●	
Child 4#	●	●▲					▲		●	
Child 5	●	●▲	●▲	●▲			●▲♦		●▲♦	
Child 6	●▲	●▲		●▲			●▲		●▲	
Child 7		●▲		●▲					●	
(b)										
Non-verbal behavior	4	7	1	5	0	0	4	0	7	0
Non-linguistic vocalizations	1	6	1	5	0	0	5	0	4	0
(Pre-)linguistic vocalizations	0	1	0	0	0	0	1	0	1	0

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