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Measuring the relationship between the parental Broader Autism Phenotype, parent-child interaction, and children's progress following parent mediated intervention

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Abstract

Parents of children with ASD may show ASD type behaviours including particular social communication interaction styles - the Broader Autism Phenotype (BAP). Understanding the potential impact of defined parent characteristics may be relevant when designing and evaluating the efficacy and effectiveness of parent-mediated interventions. In this proof of principle analysis, 18 mothers who had taken part in an early parent-mediated intervention later completed Family History Interviews. Parent data were split into lower and higher BAP groups. There was a significant negative correlation between BAP factor total scores and mother-child interaction total and post-intervention change scores ~~total scores~~ and mother-child interaction ~~mother-child total and change~~ post intervention scores at follow up. Change in number of words understood was significantly greater in children of mothers scoring in the lower BAP group compared with children of mothers in the higher BAP group. These preliminary findings provide some support for further investigation of parent BAP status as a potential moderator of the impact of early parent-mediated psychosocial interventions.

Keywords: Broader Autism Phenotype; Child progress; Parent-child interaction; Early intervention

Introduction

Twin and family studies have identified that genetic factors are important in the development of ASD and the broader autism phenotype (BAP - milder ASD related behaviours and traits seen in some relatives of people with ASD) (Bolton et al. 1994). The BAP is associated with impairments in language use and conversation (Losh and Piven 2007; Whitehouse et al. 2010), face processing (Ingersoll 2010; Dawson et al. 2005), emotion recognition and theory of mind skills (Sasson et al. 2012), social difficulties, and rigidity (Losh and Piven 2007; Pickles et al. 2013; Wainer et al. 2013). Most studies report that fewer than half of relatives of a child with ASD have BAP traits; however, BAP traits are more common in multi-incidence ASD families (Bernier et al. 2012). In keeping with the gender difference in ASD, males are more likely to show BAP behaviours or traits than females (Parr et al., 2015).

There is increasing evidence that parent-mediated early interventions can improve outcomes for some children with ASD (Green et al. 2010; Carter et al. 2011; Oono et al. 2013). Parent-mediated interventions have also been shown to enhance the impact of nursery-delivered intervention (Roberts et al 2011). Further, parent-mediated interventions improve confidence in parenting abilities as well as impacting upon the child's targeted behaviour (Keen et al. 2010). However, mothers with depression or ADHD are often less able to administer interventions or engage in training aimed at improving outcomes for their children, as their own difficulties can have a mediating effect on successful outcomes (Hutchings et al. 2012; Chronis-Tuscano et al. 2011). It is possible that the presence of parents' BAP traits may affect how they engage with young children. Parents with BAP traits may find it difficult to vary their own social communication style, which could potentially affect the delivery and effectiveness of parent-mediated interventions. On the other hand, parental rigidity as part of the BAP may make parents more likely to deliver the intervention with the suggested frequency and regularity (Parr et al., 2011). However, if parental BAP were associated with less good outcomes for children, clinicians and researchers should investigate what

might be appropriate support and/or individual modifications for the delivery of parent-mediated interventions.

This preliminary study aimed to investigate whether, following attendance at a parent-group social communication parent- intervention, the degree of observed change in parent-child interaction and the progress of children would be lower for mothers showing BAP characteristics than for mothers without BAP traits. The aim was to gather preliminary evidence as proof of concept that might warrant evaluation in a larger study.

1. Method

1.1 Participants

Forty parents who participated in an early intervention study eight years previously were approached. Of those approached 19 mothers agreed to be involved, 20 did not respond and one responded saying they would like to help but could not at this time as they had been unwell. Children in the early intervention study (a controlled evaluation of an early parent-mediated social communication intervention) had a clear diagnosis of autism or ASD (as assessed by local clinical teams and confirmed by the senior authors) (McConachie et al 2005).

1.2 Measures

1.2.1 Family History Interview-Subject version (FHI-S)

The FHI-S is a semi-structured interview designed to identify the Broader Autism Phenotype. It takes 30-60 minutes to complete, shows good inter-rater and test retest reliability and consists of items relating to the interviewee's childhood and current adult functioning. The questions focus on aspects of language and communication, social skills and behaviour, rigidity, hobbies, circumscribed interests, academic achievements, and occupational functioning. BAP behaviours are scored as '0' (behaviour does not reach scoring threshold); '1' (difficulties of the type specified, but not associated with impairment); or '2' (associated with impairment). Evidence of validity includes the

finding that the interview differentiates between parents of children with ASD and those with Down syndrome (de Jonge et al 2014). Internal reliability in this study was good ($\alpha = .85$). A BAP total factor score was derived using 11 items: lack of interest in conversation; quality of reciprocal conversation; pragmatics; aloofness; friendships; affection; intimacy; responsiveness to emotional cues; demonstrativeness; social behaviour; and rigidity. The full range of possible total scores across these items is 0-22 (Parr et al., 2015).

1.3 *Outcome Measures used at the time of the early intervention study (McConachie et al 2005)*

1.3.1 *Joy and Fun Assessment (JAFA; a measure of parent-child interaction style)*

The JAFA is an observational checklist developed for the previous intervention study to measure nine facilitative interaction strategies taught in the early social communication group course. The parent interaction strategies include: use of fun words, simplified language, musicality of speech, praise, pretend games, fun physical contact, smiles and laughter, turn-taking routines, and imitations and expansions. The ratings were made on a 5 minute recorded observation of parent-child play with toys, and were found to have good inter-rater reliability ($r=.88$) (McConachie et al. 2005). The JAFA total maximum score is 36 and participants' scores at baseline ranged from 4 to 18.

1.3.2 *MacArthur Communicative Development Inventory (MCDI)*

The MCDI is a parent-report checklist of words and gestures, to determine the number of words understood, and the number of words (understood and said) by the child (Fenson et al. 1993). Parent-child interaction style (JAFA) and child language measures (MCDI) were taken at a 7 month interval, before and after parents attended the early social communication group course. Change scores on measures were calculated by subtracting baseline scores from scores at follow up.

1.3.3 *Vineland Adaptive Behaviour Scales (VABS)*

The VABS (Sparrow et al 1984) is a parent interview regarding a child's communication, social, motor and daily living skills from which a standardised composite score of adaptive functioning is calculated.

1.3.4 Autism Diagnostic Interview (ADI-R; Lord et al 1994)

The ADI-R is a semi-standardised diagnostic interview completed with caregivers and focuses on ASD related behaviours during childhood.

1.3.5 Autism Diagnostic Observation Schedule (ADOS; Lord et al 2000).

The ADOS is a standardised observational assessment of ASD related social communication and behaviours.

1.4 Procedure

A positive ethical opinion was received from North East - County Durham & Tees Valley NRES Committee (11/NE/0023; March 2011) and informed consent obtained from participants. Two interviewers (LG, SW) were trained and supervised by JP in the administration of the FHI-S. Using the previous contact details, mothers were invited to participate by letter; once informed consent was obtained they were interviewed using the FHI-S in their homes. The FHI-S interviewers were blind to the outcome of the early intervention at the time of the interview and coding.

1.5 Analysis plan

Non parametric tests were used because the data was non normal in distribution, with Spearman's rho to look at correlations between BAP scores and mother-child interaction and child language. Independent samples Mann Whitney U tests were used to investigate: between group differences for baseline participant characteristics and scores on key measures for responders/non-responders and low/high BAP factor groupings; low/high BAP between-group difference in change across time for words said and understood. Tests were one tailed where the direction of the relationships between variables was predicted. Effect sizes are represented by the *r* value in the output from correlations, and were calculated from the *z* score in the test of difference where small effect = .10; medium effect = .3; large effect = .5 (Cohen 1992; Field 2005).

2. Results

One parent withdrew, leaving 18 participants with BAP data. The children of the mothers participating included 14 males and 4 females. At baseline, 14 children had a diagnosis of autism and 4 of ASD, and mean age was 33.5 months (SD = 6.04). The mothers' educational status was: 8 with basic school-leaving qualifications (16 years); 4 with higher qualifications at 18 years or vocational education; 6 educated to degree level. Other baseline characteristics are shown in Table 1.

Table 1 about here

There were no significant differences at baseline between parents participating in the present study, and those who were invited but declined or did not respond, on the following variables: parent education, head of family social class, Townsend socioeconomic status, and child baseline characteristics (at time of the early intervention study) including: age, adaptive behaviour as measured by the Vineland composite (Sparrow et al., 1984), communication, social and repetitive behaviours ~~algorithm domain scores~~ as measured by the Autism Diagnostic Interview (ADI-R; Lord et al 1994) and Autism Diagnostic Observation Schedule (ADOS; Lord et al 2000). There were also no significant between-group differences in the key baseline measures and the children's social and communication abilities at baseline (Table 2).

Table 2 about here

The distribution of scores (mean = 2.2; SD = 3.12) on the Family History Interview is shown in Figure 1.

Figure 1 about here.

JAJA data were available for all 18 parents. As we hypothesised, there was a significant negative correlation between BAP factor total scores and mother-child interaction scores at post intervention follow-up ($r_s = -.472$, $p = .024$), and a significant negative correlation between BAP factor total scores and mother-child interaction change ($r_s = -.473$, $p = .024$). MCDI follow-up data post-intervention were available for 14/18 children. There was a significant negative correlation between BAP factor total scores and change across time in MCDI words understood ($r_s = -.58$; $p =$

.01); however there was no significant correlation between BAP factor total scores and change across time in MCDI words said.

The 14 parents for whom there were complete data were split into a lower BAP group (including those participants scoring 2 or below on the FHI-S factor total score, $n=9$) and a higher BAP group (those with scores of 3 or above on the BAP factor total, $n=5$). Change across time in MCDI words understood was significantly greater in children of mothers scoring in the lower BAP group (mean = 39.4; SD =27.46) than of those in the higher BAP group (mean = 5.4; SD = 8.96; $p = .042$, $z = -2.06$, $r = -.55$ indicating a large effect size. Change across time in MCDI words said tended also to be greater in children of mothers in the lower BAP group (mean = 22.6; SD=23.31) than of those in the higher BAP group (mean = 9.4; SD = 10.06) though the difference did not reach significance (see Figures 2 & 3) because of one outlier.

Finally, JAFA data ($n=18$) were compared for the higher ($n=6$) and lower BAP ($n=12$) groups. There was ~~also~~ less change across time in mother-child interaction (Figure 4) for mothers in the higher BAP group (mean = -.5; SD=4.23) compared to those in the lower BAP group (mean = 3.42; SD = 5.87), though the difference was not significant.

(Figures 2, 3 and 4 about here)

The numbers of individuals scoring above and below the mean change score for the low BAP group on the dependent variables are shown in Table 3.

Table 3 about here

3. Discussion

This is the first study to investigate the relationship between BAP traits in mother's parent interaction style and child's communication skills. The negative correlation between BAP factor total scores and mother-child interaction change across time suggests possible limited impact of the intervention. The lack of change on average in mother-child interaction scores for mothers in the higher BAP group suggests that the mothers with higher rates of BAP traits may have been less able to vary their social communication style following training in parent-mediated intervention, than

those with less evidence of BAP behaviour/traits. If this finding is replicated, there are potentially important implications for the design and delivery of parent-mediated intervention for children with ASD, since this might indicate that parental BAP may be a significant moderator or mediator of the impact of the intervention.

Since there were no significant differences in the child baseline scores on key measures, the differences between BAP groupings in degree of change in children's reported and receptive vocabulary suggest that these skills may be more difficult for mothers with BAP traits to facilitate in their children. It is also possible that since the MCDI is a parent report measure, the findings may reflect less good observation of child language behaviour by mothers with higher rates of BAP traits. This possibility could be investigated further through comparison of parent report measures with directly measured language skills.

Helping parents develop an increased understanding and gain new strategies to facilitate their child's effective communication can in turn, through positive parent-child interaction, enable children to learn and develop new skills (Rogoff 1990; Kim and Mahoney 2004; Childress 2011). Gains in skills such as receptive and expressive language in children with ASD have been shown to have wide-ranging beneficial effects; for example, decreasing restricted and repetitive behaviours (Ray-Subramanian and Weismer 2012). If a parent has a limited capacity to use new social communication strategies, this may impact on their ability to adopt a facilitative style of interaction, potentially leading to reduced learning opportunities for more effective communication, with knock-on effects for the child's social communicative and other progress.

Further research is needed to understand how best to facilitate parent-child interaction taking into account the profiles of strengths and vulnerabilities of both the child with ASD and of their parent(s)/carers. Whether parents with BAP traits require specific types of additional support to assist them to achieve the best possible outcomes for their children with ASD and/or whether specialist training could facilitate the successful delivery of parent-mediated interventions are important research questions (Parr et al. 2011).

The findings from this study suggest that further studies with larger sample sizes are needed to investigate whether or not it might be helpful to take into account parental BAP characteristics when designing appropriate intervention approaches.

Limitations of this proof of principle study include the small sample size and the time lag between the two data collection points, although there is some emerging evidence of [short term](#) stability of the BAP construct e.g. across 6-12 months (Parr et al., 2015). As mothers were the usual primary caregiver who attended the group parent-mediated intervention course in the previous study, only mothers were interviewed using the FHI-S in this study. It was mothers who were the usual primary caregiver who attended the group parent-mediated intervention course in the previous study, this meant that in this study only mothers were interviewed using the FHI-S.

Investigating fathers' BAP status is also of importance. Indeed BAP traits are reported more frequently in males and thus perhaps are of more relevance when considering facilitating effective father-child communications in young children with ASD.

3.1 Conclusion

These preliminary findings require replication, but provide some support for the further investigation of parent BAP status as a potential moderator of the impact of early parent-mediated psychosocial interventions.

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Table 1 Child scores on key baseline measures

	ADI-R social interaction	ADI-R communication	ADI-R repetitive behaviours	ADOS social communication	VABS composite
Mean (SD)	16.1 (5.77)	10.5 (3.17)	5.44 (1.19)	15.33 (6.59)	66.28 (8.34)

ADI-R: Autism Diagnostic Interview Revised (Rutter et al 2003); ADOS: Autism Diagnostic Observation Schedule (Lord et al 2000); VABS: Vineland Adaptive Behaviour Scales (Sparrow et al 1984)

Table 2 Baseline scores on key measures: comparison of lower and higher BAP groups

	Child MCDI words said	Child MCDI words understood	JAFa mother-child interaction	ADOS social communication	VABS composite
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)
Lower BAP group	62.17 (98.85)	107.3 (98.4)	11.67 (4.44)	14.67 (6.71)	66.67 (9.92)
Higher BAP group	77.5 (142.81)	129.8 (135.58)	11.83 (4.54)	16.67 (6.74)	65.5 (4.32)
Significance of difference	$p = .75$ (ns)	$p = .82$ (ns)	$p = .89$ (ns)	$p = .34$ (ns)	$p = .82$ (ns)

ADOS: Autism Diagnostic Observation Schedule

MCDI: MacArthur Communicative Development Inventory

JAFa: Joy and Fun Assessment

VABS: Vineland Adaptive Behaviour Scales

ns: not significant

Table 3 Number of individuals scoring above the mean change score in the low BAP group

	Mean change score in low BAP group (SD)	Proportion of children scoring above the mean change score	
		Low BAP group	High BAP group
JABA mother-child interaction (n=18)	3.42 (5.87)	5/12	1/6
Child MCDI words understood (n=14)	39.4 (27.46)	5/9	0/5
Child MCDI words said (n=14)	22.6 (23.31)	3/9	1/5

Figure 1. Distribution of Family History Interview (FHI) scores in the sample

SD: standard deviation

Figure 2 Comparison of change in children's words understood over time between the lower and higher BAP groups

Figure 3 Comparison of change in children's words said over time between the lower and higher BAP groups

Figure 4 Comparison of change in mother-child interaction over time between the lower and higher BAP groups