

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

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28 **Abstract**

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

42

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

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45

46 ***Introduction***

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

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

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

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

78

79 **1. Method**

80 *1.1 Participants*

81 Forty parents who participated in an early intervention study eight years previously were
82 approached. Of those approached 19 mothers agreed to be involved, 20 did not respond and one
83 responded saying they would like to help but could not at this time as they had been unwell.

84 Children in the early intervention study (a controlled evaluation of an early parent-mediated social
85 communication intervention) had a clear diagnosis of autism or ASD (as assessed by local clinical
86 teams and confirmed by the senior authors) (McConachie et al 2005).

87

88 *1.2 Measures*

89 *1.2.1 Family History Interview-Subject version (FHI-S)*

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

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

103

104 | 1.3 *Outcome Measures used at the time of the early intervention study (McConachie et al 2005)*

105 | 1.3.1 *Joy and Fun Assessment (JAFA; a measure of parent-child interaction style)*

106 The JAFA is an observational checklist developed for the previous intervention study to
107 measure nine facilitative interaction strategies taught in the early social communication group
108 course. The parent interaction strategies include: use of fun words, simplified language, musicality of
109 speech, praise, pretend games, fun physical contact, smiles and laughter, turn-taking routines, and
110 imitations and expansions. The ratings were made on a 5 minute recorded observation of parent-
111 child play with toys, and were found to have good inter-rater reliability ($r=.88$) (McConachie et al.
112 2005). The JAFA total maximum score is 36 and participants' scores at baseline ranged from 4 to 18.

113 | 1.3.2 *MacArthur Communicative Development Inventory (MCDI)*

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

119 | 1.3.3 *Vineland Adaptive Behaviour Scales (VABS)*

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

123 1.3.4 Autism Diagnostic Interview (ADI-R; Lord et al 1994)

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

126 1.3.5 Autism Diagnostic Observation Schedule (ADOS; Lord et al 2000).

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

129 1.4 Procedure

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

136 1.5 Analysis plan

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

146

147 **2. Results**

148 | One parent withdrew, leaving 18 participants with BAP data. The children of the mothers
149 | participating included 14 males and 4 females. At baseline, 14 children had a diagnosis of autism and
150 | 4 of ASD, and mean age was 33.5 months (SD = 6.04). The mothers' educational status was: 8 with
151 | basic school-leaving qualifications (16 years); 4 with higher qualifications at 18 years or vocational
152 | education; 6 educated to degree level. Other baseline characteristics are shown in Table 1.

153 | Table 1 about here

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

163 | Table 2 about here

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

166 | Figure 1 about here.

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

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

175 The 14 parents for whom there were complete data were split into a lower BAP group
176 (including those participants scoring 2 or below on the FHI-S factor total score, n=9) and a higher
177 BAP group (those with scores of 3 or above on the BAP factor total, n=5). Change across time in
178 MCDI words understood was significantly greater in children of mothers scoring in the lower BAP
179 group (mean = 39.4; SD =27.46) than of those in the higher BAP group (mean = 5.4; SD = 8.96; p
180 =.042, z = -2.06, r = -.55 indicating a large effect size. Change across time in MCDI words said tended
181 also to be greater in children of mothers in the lower BAP group (mean = 22.6; SD=23.31) than of
182 those in the higher BAP group (mean = 9.4; SD = 10.06) though the difference did not reach
183 significance (see Figures 2 & 3) because of one outlier.

184 Finally, JAFA data (n=18) were compared for the higher (n=6) and lower BAP (n=12) groups.
185 There was also less change across time in mother-child interaction (Figure 4) for mothers in the
186 higher BAP group (mean = -.5; SD=4.23) compared to those in the lower BAP group (mean = 3.42; SD
187 = 5.87), though the difference was not significant.

188 (Figures 2, 3 and 4 about here)

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

191 Table 3 about here

192 **3. Discussion**

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

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

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

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

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

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

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

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

238 3.1 Conclusion

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

242

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246

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328
329

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
JAFAs 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

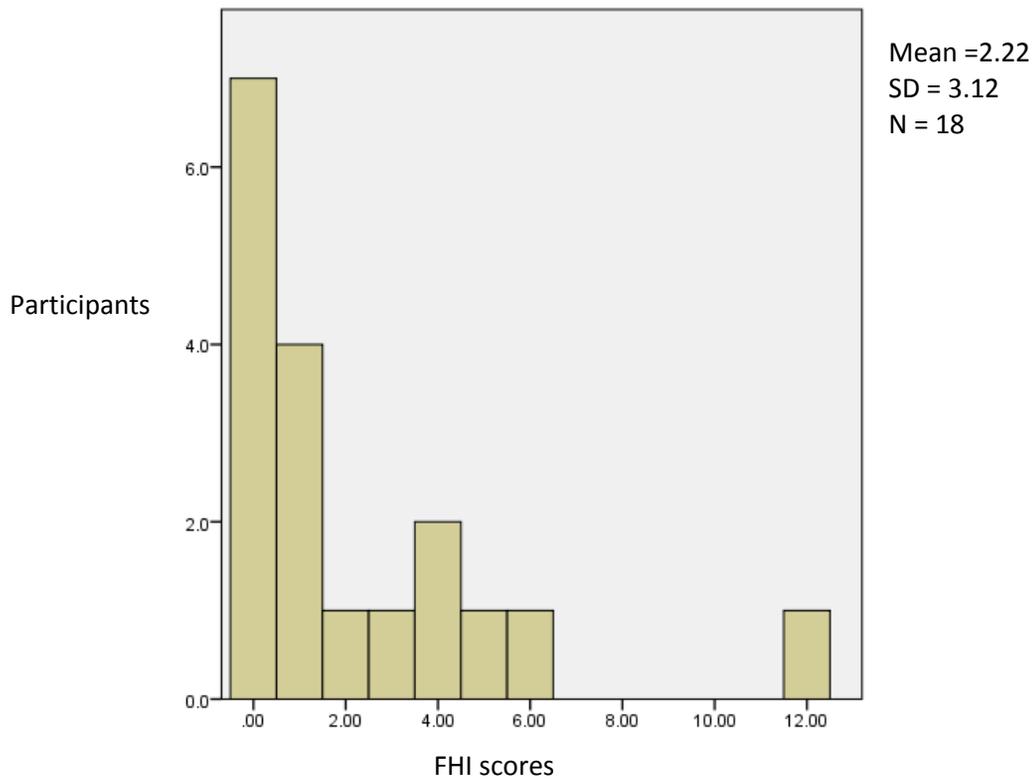


Figure 1. Distribution of Family History Interview (FHI) scores in the sample

SD: standard deviation

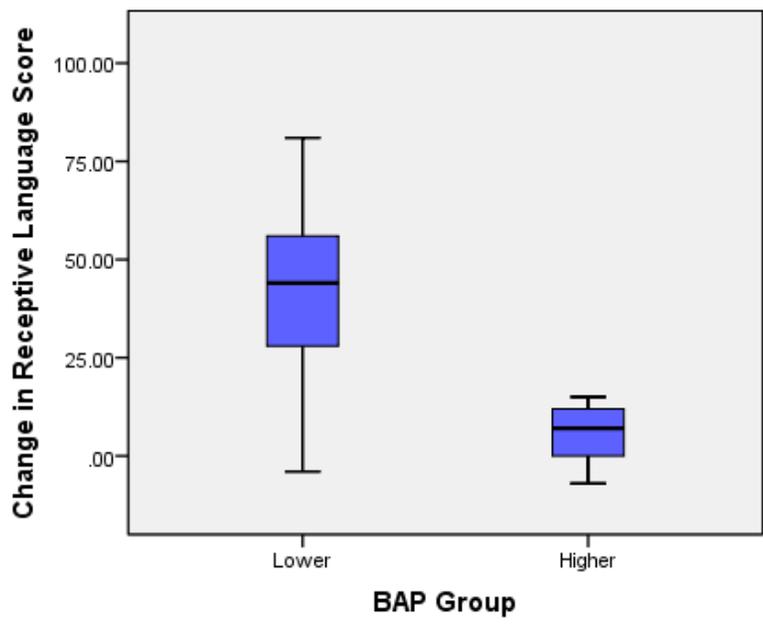


Figure 2 Comparison of change in children's words understood over time between the lower and higher BAP groups

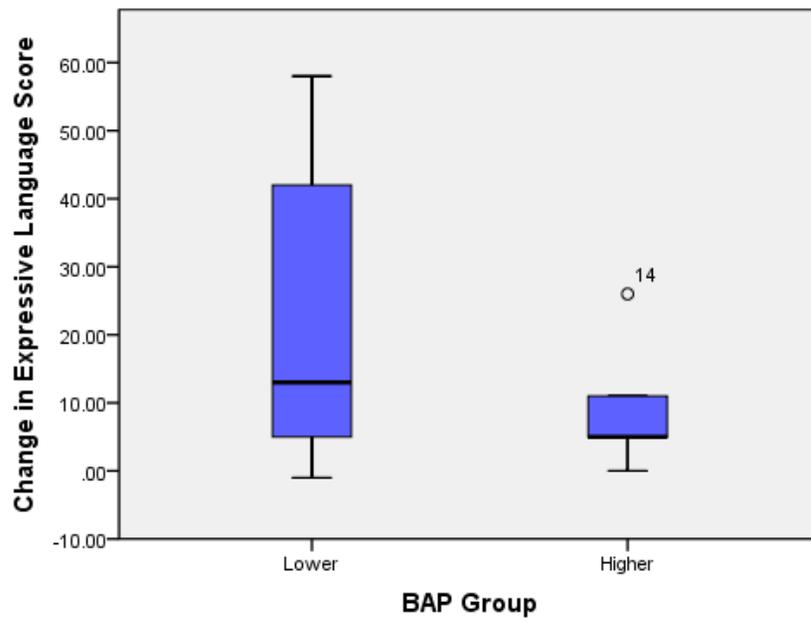


Figure 3 Comparison of change in children's words said over time between the lower and higher BAP groups

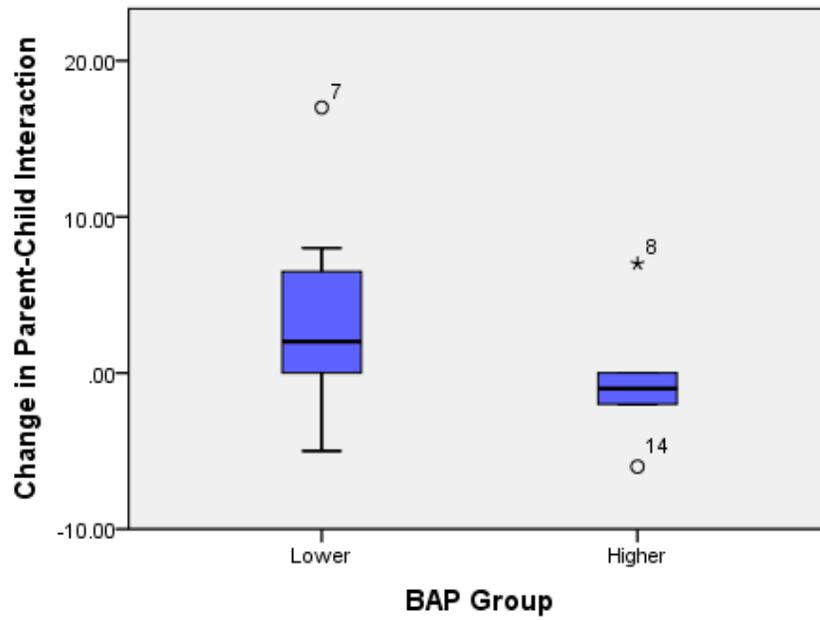


Figure 4 Comparison of change in mother-child interaction over time between the lower and higher BAP groups