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**Parent-child interaction as a dynamic contributor to learning and cognitive development in typical and atypical development**

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**Abstract:** Converging evidence suggests that parent-infant interaction is one of the most crucial formative influences on child development. In neurodevelopmental disorders, however, different timings and trajectories of development may add a layer of difficulty to the existing challenges of dyadic interaction. The current study therefore set out to compare the specific aspects of dyadic interaction (i.e., responsiveness, directiveness, attentiveness, positive affect, liveliness, mutuality, and engagement) between parent-infant dyads with Down syndrome, Williams syndrome, and typical development. Video-clips of parent-infant play interaction were rated using a validated tool, namely, the Social Interaction Measure for Parents and Infants. Significant effects emerged with respect to infant group on the quality of dyadic interaction, with the multiple comparison tests revealing differences between atypically and typically developing infant-parent dyads. The findings are discussed in relation to the effects of dyadic interaction on the linguistic and socio-cognitive development of atypical children.

**Keywords:** parent-child interaction; Down syndrome; Williams syndrome; typical development

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Central to the discipline of child development is the role of parent-infant interaction (also referred to as parent-child interaction, PCI). The past two decades have seen increasingly rapid advances in research on this topic, with studies examining the effect of various qualities of PCI on different domains of child development (e.g., cognitive, social, linguistic, and emotional). Converging evidence reveals that parent-infant dyads rich in mutual engagement and filled with a caregiver's sensitive responsiveness and low directive behaviours (i.e. focus on the infant's experience and agenda as opposed to a caregiver-directed focus) are the most productive form of interaction (Feldman, 2007; Feldman & Greenbaum, 1998; Landry, Smith, Swank, Assel, & Vellet, 2001; National Institute of Child Health and Human Development Early Child Care Research Network, 1999, 2001, 2003; Owen, Ware, & Barfoot, 2000; Pastor, 1981). There is a general understanding that these early interactions between infants and their primary caregiver are one of the most crucial formative influences on subsequent development, in particular since evidence suggests an intricate interplay between the quality PCIs and the developmental timing of different cognitive milestones (Elsabbagh et al., 2013; Hohenberger et al., 2012; Karmiloff-Smith et al., 2010). The quality of the PCI can either foster or delay the developmental progress (Carpenter, Nagell, & Tomassello, 1998; Dyches, Smith, Korth, Roper, & Mandleco, 2012; Hodapp, 2004; Karmiloff-Smith, 1998; Karmiloff-Smith et al., 2010; Oates, Karmiloff-Smith, & Johnson, 2012; Pastor, 1981; Siller & Sigman, 2008; Tomassello & Farrar, 1986; Wan et al., 2012). Parents are not intrinsically sensitive or directive, they also react to their child's own personality traits (maladaptive vs. adaptive behaviour) as well as to the child's cognitive and linguistic characteristics (Hodapp, 2004). This can be particularly obvious in parent interaction when dealing with twins discordant for a neurodevelopmental disorder. These characteristics obviously differ between genetic disorders and typical development (TD), thereby shaping the quality of dyadic interaction in quite specific ways.

When it comes to understanding the quality of PCIs in Down syndrome (DS) and Williams syndrome (WS), there are considerable knowledge gaps in the scientific literature, in particular with respect to WS. One reason for this may be that WS is much rarer, occurring only in one in 7,500 to 20,000 births (Donnai & Karmiloff-Smith, 2000; Stromme, Bjornstand, & Ramstad, 2002), arising as a result of a microdeletion of some 28 genes on the long arm of one copy of chromosome 7 (Karmiloff-Smith, 1998). In contrast, DS is the most common chromosomal abnormality, with an incidence of about one in 700 births, caused by an extra copy of chromosome 21 (Morris & Alberman, 2009). While both syndromes encompass complex medical problems and characteristic facial dysmorphologies, they also present with developmental delay and atypical cognitive and social profiles, with some specific cross-syndrome differences. Taken collectively, it is reasonable to postulate that these atypicalities are likely to yield different parental responses within the dyad in comparison to the parents of TD infants, thereby affecting the interaction itself (Hodapp, 2004).

In terms of cognitive development, different timings and developmental trajectories are evident in DS and WS. While children with DS have some strengths in joint attention skills, in children with WS these skills are weak and may arguably have a knock-on effect not only on their early language development, but also on the quality of their social interactions (Laing et al., 2002). In DS, sustained attention tends to be short and less frequent (Brown et al., 2003), while in WS it is a relative strength, although there is an indication that they lack the ability to disengage from a stimulus (Brown et al., 2003; Lense, Key, & Dykens, 2011). Increased interest in face-to-face-interaction is evident in both syndromes. However, in DS it is described as coy friendliness towards others (Doyle et al., 2004; Mundy, Kassari, Sigman, & Ruskin, 1995; Wetherby, Yonclas, & Bryan, 1989) while in WS it manifests as an uninhibited tendency to approach and engage others, including strangers, in an interaction, in spite of a limited

understanding of social norms and difficulty making and maintaining friendships (Doyle et al., 2004).

Cross-syndrome differences in WS and DS are also evident with respect to language, one of the important mediators of dyadic interaction and something dyadic interaction contributes to in important ways. The onset of first words is considerably delayed in both WS and DS but in different ways (Paterson, Brown, Gsodl, Johnson, & Karmiloff-Smith, 1999). Children with WS are able to produce many words they do not fully understand, while children with DS understand many words, but cannot produce them (Bellugi et al., 2000). In the preverbal stage, requesting behaviour, such as referential pointing, emerges at the expected time in DS (Moore et al., 2000), although it is far less frequent than in TD development (Mundy et al., 1995). In contrast to DS and TD, referential language precedes referential pointing in WS, with toddlers producing less pointing behaviours and showing difficulties in understanding the reference purpose of human pointing (Karmiloff-Smith, 1997; Laing et al., 2002).

Social environments and parental responses in DS and WS differ too, especially in comparison to TD. It is thus plausible that different social inputs may be required at different developmental time points, and that disruptions in these inputs may add to the difficulties these infants are already experiencing. Toddlers with DS show an atypical pattern of social interaction within the dyad and exhibit fewer non-verbal requests for objects, use less vocalisations to initiate conversation in a dialogue, and show a reduced ability to learn through dyadic interactions (Moore et al., 2000; Mundy et al., 1995). As far as WS is concerned, there is a substantial gap in the literature regarding the quality of PCIs in this group. Nonetheless, the existing evidence demonstrates that children with WS show an uninhibited drive towards social interactions and spend more time looking exclusively at the parent rather than at a toy to which the parent is trying to draw the child's attention (Bellugi et al., 2007; Jones et al., 2000; Losh,

Bellugi, & Anderson, 2001). In terms of parental responses in both syndromes, the evidence that does exist suggests that they exhibit more intrusive, attention-directive behaviours (Cielinski, Vaughn, Seifer, & Contreras, 1995; Hodapp, 2004).

In the face of subtle cross-syndrome differences of various aspects of dyadic interactive behaviours between genetic disorders, a question that remains concerns the quality (rather than the quantity) of interaction within the parent-infant dyads in DS and WS, and how it compares to TD. This is an important question to address since evidence concurs that a child's cognitive development depends upon two specific factors. First, it depends on endogenous factors such as gene expression and the infant's developmental trajectory. Second, it relies on exogenous factors such as environmental input and the quality of PCI. And, of course, gene expression is influenced by exogenous factors, too. These various factors point to the very fundamental nature of dyadic interaction, namely *bi-directionality*, which implies that the behaviours of both infant and parent influence and shape each other's future responses, expectations, and conceptions, as well as the nature of the dyadic interaction itself. It is therefore reasonable to hypothesize that the early atypicalities in DS and WS infant development will disrupt the caregiver's responses, as well as dyadic synchrony and engagement. On the other hand, in their attempts to encourage desirable behaviour from their atypically developing infant, parents may resort to a highly directive interactional style, which is counterproductive in TD and could turn out to be counterproductive in atypical development, i.e., have a negative impact on the infant's engagement and developmental progress (Wan et al., 2012).

Furthermore, a recent meta-analysis of the relationship between positive parenting and outcomes in atypically developing children showed a moderate, positive association between the two, encouraging further, more in-depth studies of PCI across typical and atypical populations (Dyches et al., 2012). In turn, these studies can inform more successful intervention through promoting effective parenting skills, reducing interactive difficulties, and

helping children achieve their developmental milestones with greater ease. For example, parent-mediated intervention that helps improve the quality of dyadic interaction, with particular focus on parental communicative synchrony and emotional involvement, also helps improve child communication in autism (Green et al., 2010) and cognitive functioning in DS (Venuti, de Falco, Esposito, Zaninelli, and Bornstein, 2012; Venuti, de Falco, Giusti, & Bornstein, 2008). Moreover, a better understanding of the quality of parent-infant interaction in the genetic disorders of WS and DS may also yield a deeper understanding of typical development. This is because of the subtle differences between the two disorders which, when disentangled, may be particularly informative about the effects of the specific infant behavioural characteristics on parental responses, as well as vice versa (Hodapp, 2004). The main objective of the current study is therefore to explore and develop a more in-depth understanding of the quality of the parent-infant interaction in the neurodevelopmental disorders of DS and WS and how they both overlap and differ from one another, by examining specific qualities of both parent and infant behaviours, i.e., the dyad itself. We will analyse the findings in terms of multiple facets of dyadic interaction: sensitivity, directedness, attentiveness, affect, liveliness, mutuality, and engagement, using a validated observational tool, namely, Social Interaction Measure for Parents and Infants (SIM-PI; Wan et al., 2012).

## Method

### *Participants*

Twenty-four families participated in total, comprising 8 families in which a child had DS, 8 in which a child had WS, and 8 in which a child was developing typically. Families of children with DS and WS (16 in total) who volunteered for the research and are currently on the active database at the Centre for Brain and Cognitive Development participated in the study. In addition, 8 typically developing control children were recruited through a local playgroup. Overall, the mean chronological age of the infants was 2,0 years, with no statistical

differences emerging between groups (see Table 1). Likewise, gender was distributed roughly equally across groups, with no statistical differences (see Table 2). Informed consent was obtained from one or both parents.

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Tables 1 and 2 here  
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### *Measures*

Eighteen of the 24 PCIs were recorded during home visits using a laptop camera. The remaining 6 PCIs were obtained from the families who visited the baby lab where a video camera was used to record the play session. The interactions were recorded on Digital Audio Tapes and imported onto a computer using iMovie software. Overall, each PCI was recorded for approximately 7 minutes, which is the length of time used in similar studies. The behaviour was scored on a 7-point Likert scale using a validated tool, the SIM-PI, devised by Wan et al. (2012), who initially used it to investigate PCI in infants at risk of autism. The measure encompasses eight scales, namely, parent sensitive responsiveness, parent directiveness, infant attentiveness to parent, infant attentiveness to joint activity, infant positive affect, infant liveliness, dyadic mutuality of the play experiences, and intensity of the mutual engagement. In the original SIM-PI, attentiveness to parent encompassed both attentiveness to parent and attentiveness to joint activity; however, in the current study, these two behaviours were analysed separately in order to capture subtle cross-syndrome behavioural differences in this aspect of PCI. For instance, studies have shown that infants with WS tend to spend more time looking at the parent, while those with DS have relatively good joint attention skills, so clearly the two criteria should not be collapsed into a single category in this cross-syndrome comparative study. For further details on the scale, see Appendix.

### *Procedure*

A home visit was arranged with the families in which a child had either DS or WS. During the home visit, the researcher recorded a 7-minute unstructured play interaction between the parent and their child. Upon arrival at the home of the participant, the researcher presented the parent with a set of toys – similar across all children in the study – and instructed the parent to engage in play with the toys as they normally would and in their usual play area. As per the Wan et al. (2012) study that used and developed the SIM-PI instrument, the 7-minute episode used for rating began after the camera was set up and the experimenter left the room. Out of 24 dyads, only 2 were with the father (one DS and one WS, respectively), the remaining 22 being with the mother. Once the recording was completed, the family was verbally debriefed about the study. Data were coded minute-by-minute and averaged over each 7-minute episode for each individual scale on the SIM-PI. Inter-rater reliability was assessed on 35% of the tapes. A single measure interclass correlation (two-way-mixed effects model with an absolute agreement definition) analysis was conducted. A high agreement (all  $p < .001$ ) emerged between the two raters on all the PCI scales; parental sensitivity:  $r = 0.94$ ; parental non-directiveness:  $r = .95$ ; infant attentiveness to parent:  $r = .90$ ; infant attentiveness to joint attention:  $r = .95$ ; infant positive affect:  $r = .90$ ; infant liveliness:  $r = .96$ ; mutuality:  $r = .98$ ; engagement intensity:  $r = .94$ .

### *Data analysis*

The internal characteristics of the PCI scales between TD, DS, and WS groups were explored using correlation analyses. The interaction differences between the groups were analysed using one-way between subjects ANOVAs, followed up with a set of multiple comparison tests. Chronological age was entered as a covariate. Finally, chi-square analyses were conducted to explore prevalence of scores across the groups.

## Results

### *Internal characteristics of the PCI scales*

High internal consistency was found within each scale with the average Cronbach's alpha of 0.79. Table 3 shows the pattern of inter-correlations between the scales across all three participant groups.

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 Table 3 here  
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### *Interaction differences between groups*

A major research question addressed in the current study concerned whether parent-infant interaction varied across syndrome status. The scores on the SIM-PI scales were normally distributed as assessed with the Kolmogorov-Smirnov test, so one-way between-subjects ANOVAs were used. The factor included was infant status, with three levels, namely, DS, WS, and TD, while the dependent variables were the eight scales of the SIM-PI, namely, parent responsiveness, parent directiveness, infant attentiveness to parent, infant attentiveness to joint activity, infant mood, infant liveliness, mutuality, and engagement intensity. The mean ratings on the SIM-PI scales across participant groups are displayed in Table 4. Group differences in parent-infant interaction were statistically significant in six scales, namely, infant status on parent responsiveness, parent directiveness, infant attentiveness to parent, infant attentiveness to joint activity, dyadic mutuality, and dyadic engagement. The significance of this finding remained after analyses were adjusted for chronological age (see Table 4).

Moreover, application of multiple comparison tests with a Bonferroni adjusted alpha level of .002 revealed that the parents of TD infants showed significantly higher responsiveness and lower directiveness in comparison to the parents of infants with DS or WS (both  $ps < .001$ ). It also revealed that the TD infants were more attentive to the parent than the

infants with DS and WS (both  $ps < .001$ ). Dyads with a TD infant were higher in mutuality and intensity of engagement than the dyads with a child with DS or WS (both  $ps < .001$ ). No significant difference was found between the groups on the scale of infant attentiveness to joint activity, infant positive affect, and infant liveliness, or between the DS and WS groups (all  $ps > .002$ ).

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 Table 4 here  
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Chi-square tests revealed that the low ratings (1-3 on a scale of 1-7) were more highly represented in the DS and WS groups as opposed to the TD group, which tended towards higher scores (6-7). In the DS group, 87.5%, and in the WS group, 75% of parents showed particularly high directiveness (1-3 ratings; Fisher exact test:  $p = .009$ ). Low responsiveness (1-3) was evident in 75% of parents in the DS, and 50% of parents in the WS groups (Fisher exact test:  $p = .020$ ). Low attentiveness to parent (1-3 scores) was observed in 87.5% of infants with WS, and in 75% of infants with DS (Fisher exact test:  $p = .034$ ). Moreover, 75% of WS dyads and 62.5% of DS dyads were low (1-3 scores) in mutuality (Fisher exact test:  $p = .019$ ), and engagement intensity (Fisher exact test:  $p = .008$ ).

#### Discussion

Our analyses addressed the issue of whether the quality of PCI varied across the genetic syndromes of DS and WS, as well as in comparison to TD. Contrary to our predictions, however, there were no syndrome-specific differences on the PCI scales between the neurodevelopmental disorders themselves. However, correlation analysis revealed different patterns of relationships between scales in WS and DS. There was a significant effect of infant status suggesting that the genetic syndromes led to lower parent responsiveness, higher parent directiveness, and lower infant attentiveness to parent, infant attentiveness to joint activity,

dyadic mutuality, and dyadic intensity of engagement (lower SIM-PI ratings) than did TD.

However, the effect of infant status on the scales of infant positive affect and infant liveliness was not significant. The effect size was moderate for all the scales apart from the attentiveness to joint activity, for which it was weak. On the question of cross-syndrome differences, the multiple comparison tests revealed significant differences between TD, on the one hand, and both DS and WS, on the other, on the parent scales, infant attentiveness to parent, and dyadic scales.

The current study corroborates and extends the findings of previous work into PCIs in DS, which show that parent-infant dyads are low in mutuality and engagement, while parent responses are directive and infants less attentive (e.g., Berger & Cunningham, 1983; Cielinski et al., 1995; Landry & Chapieski, 1989; Roach et al., 1998; Slomins & McConachie, 2006). For instance, Berger and Cunningham (1983) showed an increased number of vocal clashes between the parent and their infant with DS. Parents have also been found to frequently physically orient their infant towards the object of interest and seldomly gave them a toy, while the infants themselves were more passive, showing low sustained engagement (Cielinski et al., 1995; Landry & Chapieski, 1989). Those findings therefore are compatible with the results of the current study.

In terms of WS, studies into PCIs have been scarce, and the current study provides the first direct evidence of the quality of PCIs within dyads with an infant with WS. It shows that not only are parents of infants with WS more directive and less responsive than parents of TD infants, but their infants are less attentive to joint activity. Furthermore, like the dyads with a DS infant, those with a WS infant tend to lack mutuality and engagement. Previous studies have hitherto only provided a description of behavioural tendencies separately of parents and infants with WS during interactions, and have rarely focused on infants with WS. For instance, children with WS have been reported to show heightened sociability, in particular a tendency

to spend more time looking at the dyadic partner as opposed to focusing on the activity at hand, which can disrupt dyadic mutuality and engagement, as well as parental responses (Bellugi et al., 2007; Jones et al., 2000; Losh et al., 2001). Using a puzzle task, Hodapp (2004) showed that in comparison to the mothers of children with Prader-Willi syndrome, the mothers of children with WS were more intrusive and goal-directed. These observations of interactions with somewhat older children, together with the present findings on infant-parent dyads, further strengthen the hypothesis for the presence in WS of high parental directiveness, and low parental responsiveness, as well as low infant attentiveness to joint activity, dyadic mutuality, and engagement, but surprisingly little difference between WS and DS.

Moreover, our study revealed lower attentiveness to parent in WS compared to TD, which is somewhat in contrast with previous findings. For instance, Mervis et al.'s (2000) case study of a 10-month-old girl with WS indicated higher attentiveness to parent in WS than in TD during a dyadic interaction. There are, however, a couple of potential explanations for this discrepancy. On the one hand, it is possible that these findings are due to the fact that the infants with WS in the current study did not play with a stranger, but with a person with whom they had interacted frequently before, i.e., their mother. There is some evidence to suggest that infants with WS exhibit a less extreme form of hypersociability in interaction with a person they know well compared to a stranger (Mervis et al., 2003). The results of the current study in terms of infant attentiveness to parent are indeed consistent with that finding. On the other hand, group data versus findings from individual case studies are often not entirely consistent.

Interestingly and contrary to our hypotheses, the present study failed to yield cross-syndrome differences between the DS and WS groups on the entirety of the PCI scales. The reasons for this are unclear. However, the correlation analyses of the internal characteristics of the PCI scales revealed a different pattern of relationships between the scales across the two syndromes (see Table 3). For instance, the WS group showed a very distinctive pattern of

relationships between the scales: the higher the parent sensitive responsiveness, the lower was infant liveliness; and the lower the parent directiveness, the stronger was the infant's attentiveness to joint activity. In addition, as the WS infants became more attentive to their parent, the stronger were dyadic engagement and mutuality, and as infants became livelier, dyadic mutuality and engagement decreased. Interestingly, these relationships did not obtain for the DS group.

Nonetheless, in both the WS and DS groups, intensity of engagement increased with attentiveness to parent. Moreover, all the groups showed one similar pattern in that the more non-directive the parent responses were towards the infant, the more sensitively responsive they were to the infant's immediate developmental needs, while the more attentive the infant was to their parent, the stronger the dyadic mutuality. Noteworthy is the fact that only the TD group demonstrated that the higher the parent sensitive responsiveness and non-directiveness, the stronger was dyadic engagement. Overall, these correlational findings point to the very specific relationships of a certain pattern of behaviours within a PCI and the ways in which these relationships differ between the genetic syndromes.

Importantly, it is evident that a subtle mismatch in social behaviours and cognitive profiles between the syndromes has a different impact on parent responses and the dynamics of the dyadic interaction (e.g., it is only in WS group that the infant liveliness and parent responsiveness are negatively correlated).

#### *Theoretical and clinical implications of the study*

Taken together, the current findings add substantially to our understanding of the quality of PCIs in DS and WS. Our study shows that the parent-infant dyads with these syndromes are characterised by a specific pattern of interactive characteristics: dyads are low in mutuality and engagement, parent responses are highly directive and low in sensitive responsive, and infants are less attentive to parent and joint activity. At the same time, the

study also highlights syndrome-specific patterns of relationships between the scales. This combination of findings provides support for the conceptual premise that atypicalities in infants' development influence parents' responses, as well as dyadic mutuality and engagement (Berger & Cunningham, 1983; Campos & Sotillo, 2012; Cielinski et al., 1995; Hodapp, 2004; Hodapp et al., 2003; John & Mervis, 2010; Karmiloff-Smith et al., 2002; Landry & Chapieski, 1989; Wan et al., 2012). This applies even for the children with DS, who had previously been found to elicit positive responses from their parents due to their social and positive personality and infantile facial characteristics (Fidler, 2003; Gilmore & Cuskelly, 2012; Hodapp, 2004; Hodapp et al., 2003; Roach et al., 1999; Slomins & McConachie, 2006).

One explanation might be that once parents are informed that their child has a syndrome, their behavioural responses change and become more directive, thereby providing fewer opportunities for spontaneous learning. In this respect it is worth noting that parents of children with DS are aware of the atypicality of their baby already during foetal life and immediately postnatally. By contrast, because WS is much less frequent and less known, the parents have less time to become aware of the atypicality of their baby. Future research should factor in the time at which a firm diagnosis was made. For instance, parents of atypically developing children tend to restrict infants' exploration of the environment due to fear of accidents (Karmiloff-Smith et al., 2002), and discourage them from overgeneralization when starting to name objects, which is important for vocabulary learning (John & Mervis, 2010). Moreover, syndromic labels can have a huge impact on parental perception of their infant's potential abilities. The parents tend to lower their expectations of their infants, and so attribute their successes to effort but their failures to lack of skill. In contrast, parents of children with higher ability tend to attribute their successes to their skills, and failures to their lack of effort (Hodapp et al., 2003). These atypical parental responses have a knock-on effect on the developmental progress of the infants with genetic disorders, who are highly sensitive to the

environmental input received within the social interactions. As evidence concurs, the quality of the interactive experience thus has a more profound impact on shaping the child's developmental trajectories in these populations, since high quality parent-infant interactions can to some extent bridge the gaps and aid cognitive development where it is at risk of delay due to genetic factors (Siller & Sigman, 2008; Slomins & McConachie, 2006; Wan et al., 2012). So, our understanding of atypical development must of necessity include consideration of parent-infant interaction as a contributing factor.

The findings of the current study also have important practical implications for future clinical practice. Service providers will need to re-consider their approach and suggestions when advising parents on the most productive and stimulating interactive styles with their child. A large, consistent and converging body of evidence across typical and atypical development, including the current study, shows that directive parenting style is counterproductive and negatively affects infant attentiveness and dyadic synchrony, and that the most optimal child development can be achieved with an approach that is non-directive and infant-initiated (Dyches et al. 2012; Siller & Sigman, 2008; Slomins & McConachie, 2006; Wan et al., 2012).

Following on from this, if the understanding of the quality of PCI in the genetic syndromes of WS and DS is to be moved forward, a better understanding of parent directiveness needs to be developed. The current study has identified a very important factor in this respect, i.e., that parents of children who are in therapy are instructed by health professionals to be more directive in their interactions. Thus, therapy in this study could be a confounding variable leading to fewer cross-syndrome differences. Moreover, we have seen that a more directive style of interaction can in fact give rise to increased passivity. Future studies should further investigate these issues by measuring the differences in parent responses between the dyads where a child has been diagnosed early, and those where a child has been

diagnosed later, or alternatively, between those children who are in therapy and those who are not. The latter option may be more difficult to achieve for practical reasons: children who have a diagnosis tend to already be in therapy. More broadly, research is also needed to determine the effects of parent non-directiveness and responsiveness on the development of children with WS and DS. A study that can employ a parent-mediated intervention over a period of time would be a particularly useful tool in addressing such a multifaceted problem.

It would also be interesting to ascertain how the *same* parents behave with the TD siblings of the atypically developing child. A current study is underway (Karmiloff-Smith et al., in prep.) with twins discordant for Down syndrome, in order to address the important question of whether parents are intrinsically more directive and less responsive to all their children, or whether – as the current study strongly suggests – they adapt their style to the status of the children.

### *Limitations*

Finally, our findings are subject to at least four limitations. First, assessing the quality of the PCI is a challenging task because such behaviours are not easily measured. In particular, the SIM-PI scales can be somewhat subjective, and open to interpretation (although we achieved high inter-rater agreement). Second is the observer effect. Although the researcher left the room during the recording, the parents were aware that they were being filmed, and that subsequently the recording of their interaction would be viewed and analysed. As a result, the parent's behaviour may not have been captured in its entirely natural state. Nonetheless, the present study yielded more robust results than simple case studies. Third is the short time of interaction recorded (i.e., 7-minutes). Future research should aim to capture longer periods of play while controlling for different environments (home/laboratory), for instance, to clarify our findings. Finally, although the sample size is small, this is the first study, to our knowledge, to examine multiple elements of the dyadic interaction in infants with DS and WS using a

quantitative tool, and as such it provides a range of valuable insights – including, for example, likely outcomes, adequacy of the instrument, feasibility of a larger scale study using the same methodology and establishing a workable protocol, upon which further research can be built and current findings clarified.

### *Conclusions*

This study set out to determine the quality of parent-infant interaction in the genetic disorders of Down syndrome and Williams syndrome. We clearly demonstrated that in comparison to TD, parents of children with these two neurodevelopmental conditions are more directive and less responsive, while their infants are less attentive to parent and to joint activity, with dyads being low in mutuality and synchrony. Understanding the details of the quality of the PCI in genetic syndromes is clearly important, since converging evidence across typical and atypical populations suggests that early social interactions are one of the most critical formative influences on a child's subsequent development. The dynamics of parent-child interaction are often neglected in studies of cognitive variability, learning and cognitive development, yet they play a crucial role alongside many other genetic and environmental factors.

Table 1

*Age of children across the groups*

	<b>DS</b>	<b>WS</b>	<b>TD</b>	<b>F</b>	<i>df</i>	<i>p</i>
Mean	2;1	2;2	1;10	0.63	2	.55
SD	1	0;9	1			
<i>N</i>	8	8	8			

*Note.* Age stated in years and months.

Table 2

*Gender of children across the groups*

	<b>DS</b>	<b>WS</b>	<b>TD</b>	<b>Total</b>	<i>Chi-square</i>	<i>df</i>	<i>p</i>
Male	2	4	4	10	1.37	2	.50
Female	6	4	4	14			
Total <i>N</i>	8	8	8	24			

Table 3

*Correlation matrix of individual items of SIM-PI in DS, WS and TD groups*

	1	2	3	4	5	6	7
<b>DOWN SYNDROME</b>							
1 Parent Responsiveness							
2 Parent Non-Directiveness	<b>.90**</b>						
3 Infant Attentiveness to Parent	-.38	-.09					
4 Infant Attentive to Joint Activity	.14	.28	.59				
5 Infant Positive affect	.48	.52	.00	.52			
6 Infant Liveliness	.26	.32	.00	.28	.42		
7 Dyadic Mutuality	-.11	.13	<b>.89**</b>	<b>.84**</b>	.21	.31	
8 Dyadic Engagement	-.11	.13	<b>.89**</b>	<b>.84**</b>	.21	.31	<b>1**</b>
<b>WILLIAMS SYNDROME</b>							
1 Parent Responsiveness							
2 Parent Non-Directiveness	.65						
3 Infant Attentiveness to Parent	.23	.54					
4 Infant Attentive to Joint Activity	.41	<b>.73*</b>	<b>.77*</b>				
5 Infant Positive affect	-.43	-.25	-.34	-.43			
6 Infant Liveliness	<b>-.72*</b>	-.37	-.52	-.61	<b>0.72*</b>		
7 Dyadic Mutuality	.32	.47	<b>.79*</b>	<b>.92**</b>	-.48	<b>-.73*</b>	
8 Dyadic Engagement	.32	.47	<b>.79*</b>	<b>.92**</b>	-.48	<b>-.73*</b>	<b>1**</b>
<b>TYPICAL DEVELOPMENT</b>							
1 Parent Responsiveness							
2 Parent Non-Directiveness	<b>1**</b>						
3 Infant Attentiveness to Parent	.30	.30					

4 Infant Attentive to Joint Activity	-.14	-.14	.55				
5 Infant Positive affect	-.31	-.31	-.33	.31			
6 Infant Liveliness	.05	.05	-.26	-.14	.67		
7 Dyadic Mutuality	.27	.27	<b>.90**</b>	.54	-.43	-.36	
8 Dyadic Engagement	<b>.71*</b>	<b>.71*</b>	.06	.14	.31	.24	.00

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*Note.* \* $p < 0.05$ . \*\* $p < 0.01$ . Two-tailed.  $N = 24$ .

Table 4

*Ratings of SIM-PI scales by participant groups*

SIM – PI scales	DS	WS	TD	Unadjusted F (p value)	Adjusted F* (p value)	$\omega$
	M (SD)	M (SD)	M (SD)			
<i>Parent</i>						
Responsiveness	3 (1.07)	3.88 (1.36)	6.25 (0.71)	19.49 (.001)	20.52 (.001)	.59
Non-Directiveness	2.63 (1.19)	2.63 (1.41)	6.25 (0.71)	27.01 (.001)	20.91 (.001)	.68
<i>Infant</i>						
Attentiveness to parent	3.00 (1.41)	2.50 (1.60)	5.88 (.84)	15.12 (.001)	15.37 (.001)	.54
Attentiveness to joint activity	4.13 (1.89)	3.88 (2.03)	6.25 (0.25)	5.00 (.02)	4.06 (.03)	.25
Positive affect	4.38 (1.41)	4.13 (1.46)	4.50 (1.31)	0.15 (.86)	0.96 (.91)	.08
Liveliness	4.25 (1.04)	4.37 (2.13)	4.38 (0.38)	0.02 (.98)	0.27 (.77)	.09
<i>Dyadic</i>						
Mutuality	3.13 (1.25)	2.75 (1.58)	6.00(0.76)	16.41 (.001)	16.77 (.001)	.56
Engagement	3.13 (1.25)	2.75 (1.58)	6.25(0.71)	19.46 (.001)	17.11 (.001)	.60

Note.\*Co-varying for infant age.  $\omega$  = Effect size (0.2 < small, 0.5 < medium, 0.8 < large).  $N = 24$ .

## Appendix

## Social Interaction Measure for Parent and Infants (SIM-PI; Wan et al., 2012)

Domain	Description	Scale extremes
Caregiver		
1. Sensitive responsive-ness	The identification of, and behavioural response to, infant behaviour and signals that are contingent and appropriate to meet the infant's immediate and developmental needs. An attentive attitude, appropriate engagement, support and structuring in response to infant behaviour (and lack of behaviour).	1=Minimally sensitively responsive 7=Very sensitively responsive
2. Non-directiveness	A focus on the infant's experience and agenda as opposed to a caregiver-directed focus. High 'non-directiveness' includes accepting and encouraging non-intrusive behaviour, and positive comments reflecting the infant's experience. Low 'non-directiveness' includes demanding, intrusive, and negative behaviours and comments directed at the infant not at the service of promoting infant-initiated behaviour.	1=Highly directive 7=Highly non-directive
Infant		
4. Attentiveness to parent	The amount of visual contact with and amount and quality of interest in the parent directly (particularly in younger infants) and/or through mutual focus in a joint activity (particularly in older infants) as opposed to focus on other environmental stimuli or self-absorption. Considerations include infant body/face orientation toward the caregiver and interest in and acceptance of objects demonstrated by the parent, imitation and social referencing.	1=Inattentive 7=Very highly attentive
5. Positive affect	The amount and extent of positive mood, which includes positive expression and vocalisation, and enthusiasm, weighed against negative affect and behaviour, including negative expression, vocalisation and bodily gestures.	1=Highly negative affect 7=Highly positive affect

6. Liveliness	The level of physical activity, independent of the nature of the activity, weighting particularly behaviour initiated by the infant spontaneously over that which is in response to the mother's actions. Reflex movements and those controlled by the parent (e.g. by manipulating limbs) are not included.	1=Unlively 7=Extremely lively
Dyadic		
7. Mutuality	The degree of dyadic togetherness, 'tunefulness', and sharedness of the play experience, including shared attention, infant acceptance of maternal involvement, playing together, interactive flow, and shared body orientation.	1=Very low mutuality 7=Very high mutuality
8. Intensity of engagement	The intensity (not quantity) of mutual engagement at its most optimal point, either directly or through mutual focus on a third object. Intensity rates higher with level of interest and positivity, and includes smiles, vocalisations, deepening of interest, and peaks of infant excitement, with laughter or mirroring.	1=Almost no engagement 7=Very intense engagement