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ORIGINAL ARTICLE

Mother–child dyadic co-regulation in children with intellectual disability: A comparison among dyads with children with chromosome 14 aberrations, Down syndrome and typical development

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ABSTRACT

Background: The present study aimed at investigating mother–child dyadic co-regulation patterns in dyads with children with intellectual disability (ID).

Method: 24 children paired for developmental age and vocabulary size (8 with chromosome 14 aberrations, 8 with Down syndrome, and 8 with typical development) and their mothers participated in the study. The Revised-Relational Coding Scheme was employed to assess mother–child dyadic co-regulation.

Results: The dyads with children with ID appeared to have significantly fewer episodes of symmetric-patterns (i.e., situations in which mother and child share a mutual focus of attention) than those with typically developing children. In addition, the dyads with children with chromosome 14 aberrations showed the highest proportion of unengaged patterns (i.e., situations in which the partners do not interact with one another).

Conclusions: A severe level of ID in combination with autistic traits, as frequently found in chromosome 14 aberrations, could lead to a less optimal mother–child interaction.

KEYWORDS
Mother-child co-regulation; chromosome 14 aberrations; Down syndrome; typical development

Several studies have found that having a child with intellectual disability [ID] may negatively affect mother–child interaction (e.g., Guralnick, 2005; Warren & Brady, 2007). The present study considered two different conditions of ID: chromosome 14 aberrations, which are generally characterised by severe ID and frequently associated pathologies (e.g., epilepsy), behavioural problems, and autistic traits, and Down syndrome, which is generally characterised by mild or moderate ID and relatively preserved social skills. In the following paragraphs, we will briefly summarise the literature on these genetic conditions.

Chromosome 14 aberrations

Chromosome 14 aberrations are rare genetic conditions, which are defined by the European Union as life-threatening or chronically debilitating diseases with a prevalence of less than 5 per 10,000 (Moliner & Waligora, 2017). Accurate data on their prevalence are not available (Zampini, Zanchi, Rinaldi, Novara, & Zuffardi, 2017). Various aberrations involving chromosome 14 have been reported, such as the ring 14 syndrome, in which one of the chromosome 14 assumes a circular configuration, and the linear 14q deletions, in which a part of the chromosome 14 is lost (Van Karnebeek et al., 2002).

Although there is high individual variability, children with these genetic conditions usually show severe developmental delays and ID in addition to neurological problems and autistic traits. In fact, people with chromosome 14 aberrations frequently suffer from drug-resistant epilepsy (e.g., Giovannini et al., 2010; Morimoto et al., 2003) and they frequently show persistent deficits in social communication and interaction, and restricted, repetitive patterns of behaviour, interests or activities (Rinaldi et al., 2017; Zampini, Zanchi, & D'Odorico, 2014; Zollino et al., 2009). Their linguistic and communicative skills are usually impaired, particularly in those children who show autistic traits (Zampini et al., 2017). To date, there appear to be no studies that analyse mother–child interaction in the population of children with chromosome 14 aberrations.

Down syndrome

Down syndrome is a genetic disorder caused by an extra portion of chromosome 21, and it is the most common genetic cause of ID. These children usually show mild to moderate ID and language development delays.

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Their language impairment has often been reported to be greater than their cognitive level would suggest (Zampini & D'Odorico, 2013).

The behavioural phenotype of children with Down syndrome includes a relative strength in social functioning; in fact, they usually show a good imitative competence and ability to look toward their mothers during interaction and a high frequency of smiles to communicate positive affect (see Fidler, 2005). However, this social competence usually decreases in later development, as the demands of social functioning become more complex (Fidler, Most, & Philofsky, 2009).

Comorbidity with medical problems are quite frequent, for instance, more than 30% of children with Down syndrome have congenital heart defects, which could be related to a higher psychomotor delay (Visootsak et al., 2016). However, comorbidity with behavioural problems are less frequent, for instance in their study, Kent, Evans, Paul, and Sharp (1999) found that the comorbid occurrence of autistic traits and Down syndrome was 7%.

Few (and not recent) studies have been focussed on mother–child interaction in dyads with Down syndrome (e.g., Cielinski, Vaughn, Seifer, & Contreras, 1995; Crawley & Spiker, 1983; Fischer, 1987). Generally, children with Down syndrome appeared to be less spontaneous in initiating social communication, and their mothers appeared to be more directive and less encouraging autonomy than those of typically developing children in both linguistic input (Marfo, 1990) and physical assistance during problem-solving tasks (Gilmore, Ryan, Cuskelly, & Gavidia-Payne, 2016). Moreover, concerning attachment patterns, Atkinson et al. (1999) showed that only 40% of children with Down syndrome exhibited secure behaviour. These authors hypothesised that this result could be related to a lower level of both parental sensitivity and children's cognitive development.

Aim

The study of mother–child dyadic co-regulation patterns in children with ID is fundamental to determine the impact of mild to severe cognitive impairments and possible behavioural problems on the quality of mother–child interaction. Since no published studies have been found on mother–child interaction in children with chromosome 14 aberrations and there are only a few and dated studies on this topic in children with Down syndrome, the present study aimed to investigate mother–child dyadic co-regulation patterns in these populations. These patterns were compared with the patterns showed by dyads of mothers and typically developing children matched for developmental age and vocabulary size. In addition, the relationships among the interactive patterns and the children's competence were considered.

Methods

Participants

Participants were 24 children and their mothers. Eight of these children [C14 group] received a diagnosis of chromosome 14 aberration (one has Ring 14 syndrome, five have a linear 14q deletion, and two have a translocation) and were recruited through the Italian association "Ring14 Italia Onlus" (Reggio Emilia, Italy). Eight children [DS group] received a diagnosis of Down syndrome (free trisomy 21) and were recruited through parent–child associations in Lombardy (Italy). The remaining eight children [TD group] were typically developing children selected from a sample of participants of a longitudinal research project on early language development at the Department of Psychology of the University of Milano-Bicocca (Milan, Italy). Children's parents signed a written informed consent form before inclusion in the study.

The children in the three groups were one-to-one matched for developmental age and vocabulary size (see Table 1). We decided to control for both these variables, as they could influence the way mothers interact with their children (D'Odorico, Salerni, Cassibba, & Jacob, 1999). Participants' developmental age has been assessed by Griffiths Mental Development Scales (Griffiths & Huntley, 2007), whereas their vocabulary size has been assessed by the Italian version of the MacArthur-Bates Communicative Development Inventories (Il Primo Vocabolario del Bambino – PVB, Caselli & Casadio, 1995).

Table 1. Children's characteristics.

		Chronological age			Developmental age			Vocabulary size		
	N (females)	M	SD	Range	M	SD	Range	M	SD	Range
C14	8 (6)	73.25	25.92	30–102	13.38	2.62	10–18	3.25	2.82	0–8
DS	8 (6)	25.38	4.96	18–36	14.00	2.67	11–19	4.75	3.20	0–9
TD	8 (6)	14.63	2.97	12–18	14.88	2.96	12–19	7.00	3.72	1–12

Note. Age is measured in months.

Table 2. Maternal characteristics.

		Age at the time of the study			Level of education (*N*)		
	N	M	SD	Range	Graduate school[a]	High school[b]	Elementary/ Junior school[c]
C14	8	39	3.42	36–45	2	4	2
DS	8	41	2.77	36–44	2	4	2
TD	8	36	6.90	32–46	3	5	0

Note: [a]18 years of education; [b]13 years of education; [c]8 years of education.

No significant differences were found among groups in children's developmental age (Kruskal-Wallis' $H = 0.78$; $p > .05$) or in vocabulary size (Kruskal-Wallis' $H = 4.43$; $p > .05$). In contrast, a significant difference has been found in children's chronological age (Kruskal-Wallis' $H = 19.91$; $p < .001$); in particular, children with DS were significantly younger than children with C14 (Mann-Whitney's $U = 1.00$; $p < .001$) and TD children were significantly younger than both children with DS (Mann-Whitney's $U = 1.50$; $p < .001$) and children with C14 (Mann-Whitney's $U = 0$; $p < .001$).

Children's neuropsychiatric clinical assessment allowed to detect the presence of autistic traits (i.e., withdrawal behaviours and stereotypic movements) in six of the eight children in the C14 group. No children in the DS group and the TD group showed autistic traits.

With regard to mothers, data on their age at the time of the study and their education level are reported in Table 2. The difference among the three groups was not statistically significant for either age (Kruskal-Wallis' $H = 2.88$; $p > .05$) and education level (Fisher's exact test = 2.65; $p > .05$).

Procedure

Mother–child co-regulation has been observed during 10-minute play sessions, which were video recorded. Mothers were encouraged to play with their children as normal, using three sets of toys provided by the examiner: a toy farm set, some illustrated books and a doll with a nurturing set. The dyads were free to play with the toys they preferred. To facilitate the coding of the videos, "The Observer XT" computer program was used (Grieco, Loijens, Zimmerman, & Spink, 2007).

Coding

The Revised-Relational Coding System – R-RCS (Fogel et al., 2003) was employed to assess mother–child dyadic co-regulation. This coding scheme has been used to investigate the quality of mother–child interaction in both typically developing children (Aureli & Presaghi, 2010; Lavelli & Fogel, 2002) and preterm mother–child dyads (Sansavini et al., 2015). According to the R-RCS, the co-regulation patterns are classified as "symmetrical," "asymmetrical," "unilateral," "disruptive" and "unengaged". A "no code" category was added in case the mother or the child was away from the video. According to the R-RCS manual, coding was done continuously from the video, and the co-regulation patterns were identified segmenting the interaction into units lasting at least 3 s; a new code starts at the beginning of the movement or vocalisation that results in a change of code (Fogel et al., 2003). In the following paragraphs a description of each one of these patterns is given.

Symmetrical patterns

The interaction is characterised by mutual elaboration of the partners, they both share a mutual focus of attention and contribute to innovate a particular topic. Symmetrical patterns include "sequential-symmetrical," in which the partners share the same actions (e.g., both the mother and the child are feeding the doll) and "sequential-resonant," in which the partners share actions and emotions (e.g., both the mother and the child are laughing because the doll falls while they are feeding it).

Asymmetrical patterns

The partners share a mutual focus of attention, but only one partner produces innovation, whereas the other does not respond, although having the opportunity to intervene. Asymmetrical patterns include "asymmetrical-demonstrating," in which one partner demonstrates something to the other who is observing (e.g., the mother is showing how to feed the doll and the child is looking at her) and "asymmetrical-expecting," in which one partner tries to elicit a response from the other (e.g., the mother is asking to imitate her actions, and the child is looking at her).

Unilateral patterns

Only one partner tries to keep a mutual focus of attention, while the other is engaged in a new personal activity and does not pay attention to the partner. Unilateral patterns include "unilateral-following," in which one partner observes the other one, but the other is not attending to him/her (e.g., the mother is looking at the child who is playing alone with the doll), "unilateral-initiating," in which one partner introduces a new topic, but the other does not respond (e.g., the mother shows a book, but the child continues to play alone with the doll) and "unilateral-demanding," in which one partner actively and intrusively tries to engage the other, but the other does not respond (e.g., the mother puts a book in front of the child and tries to move the doll).

Disruptive patterns

A partner disturbs the action of the other, who shows annoyance or displeasure, and does not regulate his/her own behaviour in relation to that of the other one (e.g., the child cries because the mother has moved the doll, but she does not give it back to him or her.).

Unengaged patterns

The partners are not involved and do not interact with one another (e.g., the mother is looking at a book, and the child is playing with the doll).

In each play session, the proportion of time spent in each one of the interactive patterns and sub-patterns has been computed.

Reliability

The intercoder reliability of two independent coders was assessed in 25% of the sessions (i.e., two sessions for each group of participants). The accuracy of the coding was computed using the inter-rater reliability tool of "The Observer XT". The observations were compared record by record, and the software reported Cohen's kappa coefficient. A value of kappa = .81 demonstrated a good level of agreement.

Data analysis

To verify the existence of statistically significant differences in the co-regulation patterns among the three groups of mother–child dyads, we used the Kruskal–Wallis non-parametric test for k-samples. As a post-hoc test, we used the Mann–Whitney non-parametric test for two independent samples for those patterns that appeared to be statistically different among the three groups.

Moreover, to assess the existence of possible relationships between co-regulation patterns and children's competence, the Spearman's Rho non-parametric coefficient has been computed among the proportion of time spent in symmetric patterns and unengaged patterns and both children's developmental age and vocabulary size. These relationships have been computed considering all the dyads together since the three groups did not significantly differ for children's developmental age and vocabulary size.

Results

Dyadic co-regulation patterns in the three groups

Data analyses showed a significant difference in the dyadic co-regulation patterns of the three groups. As shown in Figure 1, the proportion of symmetrical patterns ($H = 11.90$; $p = .003$) and unengaged patterns ($H = 11.42$; $p = .003$) appeared to be statistically different among the three groups of dyads. In particular, Mann–Whitney test showed that the proportion of time spent in symmetric patterns was significantly higher in the TD group than in both the DS group ($U = 8.00$; $p = .01$) and the C14 group ($U = 3.00$; $p = .001$). Whereas, the proportion of time spent in unengaged patterns was

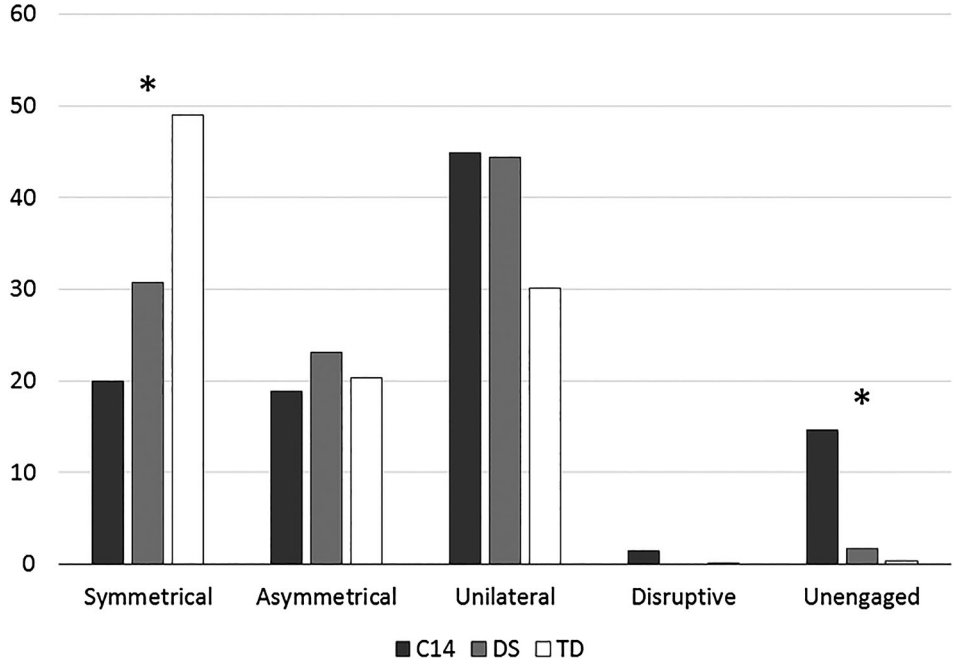

Figure 1. Percentage of dyadic co-regulation patterns in each group.

significantly higher in the C14 group than in both the DS group ($U = 8.50$; $p = .01$) and TD group ($U = 5.50$; $p = .003$).

Considering the sub-patterns of symmetric, asymmetric and unilateral patterns, data reported in Table 3 showed that there was a statistically significant difference in the proportion of time spent in symmetrical-sequential patterns, which was higher in the TD dyads than in both the DS and C14 dyads. In addition, there was a significantly higher proportion of time spent in both unilateral-initiating and unilateral-demanding patterns in the C14 dyads than in the TD dyads.

Relationships between dyadic co-regulation patterns and children's competence

Considering all the dyads together, the proportion of time spent in sequential patterns appeared to be significantly related to the children's developmental age (Rho = .46; $p = .025$), but not to their vocabulary size (Rho = .21; $p = .318$). A similar result has been found considering the proportion of time spent in unengaged patterns, which appeared to be negatively correlated with the children's developmental age (Rho = −.50; $p = .013$), but not related to their vocabulary size (Rho = −.15; $p = .491$).

Discussion

The present study aimed at comparing co-regulation patterns between dyads with typically developing children and dyads with children with ID. Two groups of children with ID have been considered: children with Down syndrome and children with chromosome 14 aberrations.

Although the children in the three groups have been paired for both developmental age and vocabulary size, the dyadic co-regulation patterns appeared to be significantly influenced by ID. The presence of symmetrical patterns (i.e., those in which both partners share a mutual focus of attention and contribute to innovate a particular topic) appeared to be significantly lower in both the groups with ID than in the group of typically developing children. Therefore, cooperation appears to be affected not only in dyads with children with chromosome 14 aberrations, who are characterised by a more severe ID and a more frequent association with autistic traits but also in dyads with children with Down syndrome, who generally have better social skills (Fidler, 2005). This data could be interpreted considering the greater difficulty for all children with ID in being involved in joint attention situations (e.g., Paparella & Kasari, 2004).

However, it should be noted that the two groups of children with ID differed in the proportion of time spent in unengaged patterns (i.e., those in which the partners do not interact with one another). In particular, the dyads with children with chromosome 14 aberrations showed a significantly higher proportion of unengaged patterns, not only compared to the children with typical development but also compared to the children with Down syndrome. These data can be interpreted on the one hand as a greater difficulty for children with autistic traits (in particular withdrawal behaviours) in getting involved in shared activities, and on the other hand, as a greater difficulty for their mothers in trying to support the interaction with these children. In fact, interacting with children who show remarkable problems in both cognitive and social skills could be very stressful for their parents (Dunn, Burbine, Bowers, & Tantleff-Dunn, 2001). Moreover, in the present study, owing to their more severe degree of ID, the children with chromosome 14 aberrations had a significantly higher chronological age than both typically developing children and children with Down syndrome. We could also hypothesise that mother–child interaction could be negatively influenced by the child's increasing age, in particular if a parent goes through many failed communicative attempts over the years. It should be noted that some of the differences in the interaction style of mothers of children with ID could be the results of modifications deriving from their children's feedback. In fact, from a transactional approach (i.e., an approach in which parent and child are viewed as reciprocally influencing their relationship in a dynamic manner), parents of children with ID seem to modify their behaviour as a function

Table 3. Percentage of dyadic co-regulation sub-patterns in each group.

	C14		DS		TD			
	M	SD	M	SD	M	SD	Kruskal-Wallis' *H*	Mann-Whitney
Sym_sequential	18.48	14.14	29.89	12.73	47.22	9.12	12.74**	C14 < TD ($p = .001$) DS < TD ($p = .010$)
Sym_resonant	1.12	0	0	0	2.35	0.79	1.50	
Asym_demonstrating	10.74	7.24	14.98	6.59	16.39	10.07	1.83	
Asym_expecting	6.60	6.50	7.53	4.13	3.57	3.27	3.08	
Uni_following	19.37	14.90	28.64	15.76	22.38	15.56	.79	
Uni_initiating	19.83	9.34	14.31	4.98	7.06	6.61	7.22*	C14 > TD ($p = .010$)
Uni_demanding	3.28	4.68	0.76	1.22	0	0	6.94*	C14 > TD ($p = .038$)

* = $p < .05$; ** = $p < .01$.

of their children's contribution to the interaction (Paparella & Kasari, 2004).

Lastly, although the proportion of time spent in unilateral-demanding patterns was higher in the dyads with chromosome 14 aberrations, the extremely reduced presence of disruptive patterns could be considered a positive sign, because it showed a limited level of intrusiveness in all the three groups. Despite the difficulties in interacting and establishing symmetric patterns with children with ID, all the mothers in the present study showed good ability in respecting their children's attention span and in re-establishing a positive interaction when their children show annoyance or displeasure after a maternal intervention.

With regard to the relationships between dyadic co-regulation patterns and children's competence, significant correlations have been found with the children's developmental age, but not with their vocabulary size. These data could be explained considering that the range of developmental ages here considered was quite wide (i.e., 10–19 months), whereas children's vocabulary size ranged from 0 to 12 words. The positive relationship between the time spent in sequential patterns and children's developmental age showed that both mothers and children increased their ability to share attention on the same activity with children's increasing cognitive competence.

Practically, these results suggest that mother–child interaction with children with ID can be enhanced by engaging the children in joint attention situations, trying to increase symmetrical and asymmetrical co-regulation patterns. In particular, in interacting with children with ID, data in the literature (Landry & Chapieski, 1990) suggested the importance to follow the children's attentional focus rather than trying to redirect their attention, because diverting the attention of a child with ID and shifting his attentional focus may overload his cognitive skills.

Limitations and future directions

Although it should be considered that chromosome 14 aberrations are rare genetic conditions, the small number of participants involved in this study does not allow to generalise to the entire populations involved. In addition, in the present study, most but not all the children with chromosome 14 aberrations showed autistic traits (i.e., six out of eight children). Due to the possible effect of autism spectrum disorder (ASD) on dyadic co-regulation patterns, in particular on those that require joint attention skills, future studies will involve a group of children with severe ID associated with ASD and another one with severe ID but without ASD comorbidity. Future studies will also investigate the possible changes that occur in mother–child interaction with children's increasing age.

As tested in other conditions (e.g., children with ASD – Poslawsky et al., 2015), we hypothesise that video feedback intervention could be useful to enhance the interaction of mothers with their children with severe ID. In fact, observing the dyadic interaction with an experienced clinician could be useful to draw a mother's attention to the patterns that are more functional to her child's development.

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