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Exploring parental behavior and child interactive engagement: A study on children with a significant cognitive and motor developmental delay

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ABSTRACT

Background and aims: Parenting factors are one of the most striking gaps in the current scientific literature on the development of young children with significant cognitive and motor disabilities. We aim to explore the characteristics of, and the association between, parental behavior and children's interactive engagement within this target group.

Methods and procedures: Twenty-five parent-child dyads (with children aged 6–59 months) were video-taped during a 15-min unstructured play situation. Parents were also asked to complete the Parental Behavior Scale for toddlers. The video-taped observations were scored using the Child and Maternal Behavior Rating Scales.

Outcomes and results: Low levels of parental discipline and child initiation were found. Parental responsivity was positively related to child attention and initiation.

Conclusions and implications: Compared to children with no or other levels of disabilities, this target group exhibits large differences in frequency levels and, to a lesser extent, the concrete operationalization of parenting domains. Further, this study confirms the importance of sensitive responsivity as the primary variable in parenting research.

What this paper adds?

This paper is the first step in filling the striking gap of knowledge on family and parenting factors in the current literature on the specific target group of young children with a significant cognitive and motor developmental delay. The parents of these children, and the professionals who guide them, experience a lot of uncertainty about recognizing and stimulating the interactional abilities of these children as well as about which parental interaction style is optimal for the child's development. This paper allows parents and practitioners to gain insight into the general qualitative characteristics of parent–child interactions within this target group, compared to (parents with) children with no or other levels of disabilities. Foremost, it cautiously confirms the importance of sensitive responsivity as the primary variable in parenting research and intervention programs. Also very important, this paper sets the stage for further research focusing on disentangling the probable bidirectional influences between parent and child by analyzing reciprocal sequences of parental and child behavior and/or conducting longitudinal analyses.

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1. Introduction

A child's development is the product of continuous dynamic interactions between the child and the experiences provided by his or her social settings (Lerner, Rothbaum, Boulos, & Castellino, 2002; Sameroff, 2009). Although interrelated with nonfamilial influences and the broader context in which families live (Collins, Maccoby, Steinberg, Hetherington, & Bornstein, 2000), the most proximal and most influential social setting is the child's family (Bronfenbrenner, 1986; Lochman, 2004).

The idea that especially parents are primary agents of developmental change in their children is reflected in an extensive body of research on parenting behavior (Mahoney & Nam, 2011; Sameroff, 2010). In typically developing children, high levels of parental support (e.g. sensitive responsiveness, warmth and stimulation) are generally found to be associated with adaptive child outcomes in a variety of developmental domains (e.g. Farah et al., 2008; Landry, Smith, Swank, Assel, & Vellet, 2001; Larzelere, Morris, & Harrist, 2013; Stams, Juffer, & van IJzendoorn, 2002; Zhou et al., 2002). On the contrary, (intrusive) directiveness and negativity appear to be inversely associated with developmental outcomes (e.g. Ispa et al., 2004; Larsson, Viding, Rijsdijk, & Plomin, 2008).

Mahoney and Nam (2011) pointed out that the same general patterns of parenting associated with optimal development in typically developing children are evident in studies on children with a developmental delay. For example, maternal sensitivity and responsiveness are inversely associated with behavioral problems and positively related with appropriate behavior, communication and general development in young children with intellectual disabilities (Warren & Brady, 2007). Negative parent–child interactions are associated with child behavioral problems (Hastings, Daley, Burns, & Beck, 2006), while parental scaffolding has been demonstrated to be predictive for the social competence in children with developmental delays (Baker, Fenning, Crnic, Baker, & Blacher, 2007). A recent meta-analysis of 14 studies including 576 participants showed an association between positive parenting styles and more adaptive functioning in children with developmental disabilities (Dyches, Smith, Korth, Roper, & Mandleco, 2012). Research into the association of (different levels of) directiveness with (positive as well as negative) child outcomes has shown mixed results, although higher levels of parental directiveness are often observed in children with lower levels of cognitive functioning (Guralnick, Neville, Hammond, & Connor, 2008; Spiker, Boyce, & Boyce, 2002).

However, within the target group of children with a significant cognitive and motor developmental delay, knowledge on parenting is very scarce (Chadwick, Cuddy, Kusel, & Taylor, 2005; Van keer & Maes, 2016). The combination of significant motor and cognitive limitations, resulting in a high dependency of these children on their immediate social setting, provides parents with complex and unique challenges (Fagnart, 2011; Horn & Kang, 2012; McCollum, 2002). For example, in a study of Wilder, Axelsson, and Granlund (2004), parents perceived their children with profound and multiple disabilities to have difficulties initiating interaction and maintaining attention, and perceived themselves to be less competent in understanding the child's communication and in directing and maintaining the child's attention. However, it is unclear whether these challenges result in different patterns of parenting within this specific target group. Research on parenting young children with a significant cognitive and motor disability is especially warranted, since the early years might constitute an unique window of opportunity to influence children's developmental trajectories (Guralnick, 2005; Narvaez, 2012; Nelson, 2000).

Integrating several conceptually and empirically grounded models of child development, such as the transactional model (Sameroff, 2009), the developmental systems theory (Ford & Lerner, 1992), the ecological theory (Bronfenbrenner, 2001) and the parenting process model (Belsky, 1984), Guralnick (2011) described a framework for understanding contextual influences on early (typical and atypical) child development. This framework encompasses risk and protective factors at three levels: child's current developmental characteristics, family patterns of interaction and family resources. Overall, the importance of family patterns of interaction is greatly stressed and it is proposed that the central task of early intervention is to establish, restore and/or stimulate family patterns of interaction to the most optimal level as possible in order to optimize the child's developmental opportunities. As to how this process unfolds, Mahoney and colleagues state that parent–child interactions influence child development by stimulating pivotal developmental behaviors (Mahoney, Boyce, Fewell, Spiker, & Wheeden, 1998; Mahoney & Nam, 2011). Pivotal behaviors are "behaviors that are central to wide areas of functioning such that a change in the pivotal behavior will produce improvement across a number of behaviors" (Koegel, Koegel, & Carter, 1999, p. 577). Interactive engagement behaviors, such as attention and initiation, are considered by constructivist theories to be core processes of learning and development and thus regarded as pivotal developmental behaviors (Kim & Mahoney, 2004). Mahoney, Kim, and Lin (2007) presented data indicating that the degree to which parents engage in responsive interaction with their child is associated with the child's use of these pivotal behaviors.

Based on these previous findings, we aim to

1. provide a general characterization of parental behavior towards young children with a significant cognitive and motor developmental delay,
2. provide a general characterization of these children's interactive engagement,
3. explore the association between parental behavior and children's interactive engagement in this target group.

2. Method

2.1. Participants

Twenty-five unique parent-child dyads participated in the study. They were recruited through hospitals, diagnostic centers, early intervention teams and specialized day care centers in Flanders (Belgium; $n = 11$) and the Netherlands ($n = 14$). Professionals within these organizations were asked (by mail and/or by telephone) to inform potential participants on the study and to bring them into

contact with the researchers. Parents were free to choose whether the mother or father participated in the study. We included children between the age of 6 months and 4 years, who were at least spending their weekends and holidays with their family. We did not include children before the age of 6 months because clear indications of a significant developmental delay should be present and we wanted to respect the high emotional stress of parents in the first months after birth. A significant cognitive delay was operationalized using the 'Tandemlijst' (Stadeus, Windey, Vermeir, & Van Driessche, 1994). We included children functioning below a quarter of their chronological age, which is associated with the description of a profound intellectual disability (Grossman, 1973; Hogg, Foxen, & McBrien, 1981; Vig & Sanders, 2007). The Tandemlijst is specifically developed for young children with a developmental delay. It includes the developmental steps and milestones used in early intervention programs. By describing the cognitive developmental domain separately and in detail, the influence of the motor limitations on the estimation of cognitive functioning is minimized as much as possible. A significant motor delay was operationalized using the 'Gross Motor Function Classification System – Expanded & Revised' (GMFCS-E & R; Palisano, Rosenbaum, Bartlett, & Livingston, 2007). The GMFCS is specifically developed for and widely used in research on relatively young children with significant motor limitations and shows a good reliability and predictive value (Wood & Rosenbaum, 2000). Also, the instrument provides descriptions of motor abilities for different age bands, including 0 to 2, 2 to 4 and 4 to 6 years. We included children functioning at level IV or V (indicating a severe impairment) and, additionally, level III when the child was less than 2 years old (since combining level III, IV and V has a better predictive value at this young age; Gorter, Ketelaar, Rosenbaum, Helders, & Palisano, 2009). Detailed inclusion criteria are available from the first author upon request. The presence of a significant developmental delay in both the cognitive and the motor domain was regarded as a necessary and sufficient inclusion criterion (Nakken & Vlaskamp, 2007). Children who only showed a significant delay in one of the two domains were not included. We formulated no criteria regarding the cause of the developmental delay and the presence of additional constraints (sensory disabilities, health problems, comorbid diagnoses such as ASS, etc.). Detailed background information on the parent-child dyads is presented in Table 1.

2.2. Procedure and Instruments

Two types of parental behavior were examined: parent's interactional style (measured through observation) and parenting behavior (measured through self-report). Child's interactive engagement was simultaneously assessed through the observation procedure. This study was approved by the Social and Societal Ethics Committee (KU Leuven) and written informed consent was obtained from parents prior to data collection.

2.2.1. Child's interactive engagement and parent's interactional style

Participating parent-child dyads were video-taped during a 15-min unstructured play situation, at home or at the familiar day care facility of the child. Parents were instructed to engage with the child as they would normally do, but were not informed on the parental and child behaviors of interest to the study. Due to the significant (cognitive, motor and/or sensory) limitations and idiosyncratic needs and preferences of the children, we did not provide a standard set of toys. Two cameras were used, each directed at one interaction partner: one was placed on a tripod (usually directed at the child) and one was manually handled by the researcher in order to ensure optimal angle views. Because the observations related to this study were part of a broader project, parents and children were already familiarized with the researcher and the presence of camera equipment in the context of other test administrations.

Afterwards, the videotaped observations were scored using an adapted version of the Child Behavior Rating Scale-Revised (CBRS; Mahoney, 1998) and the Maternal Behavior Rating Scale-Revised (MBRS; Mahoney, 2008). Most dyadic interaction scales have been developed in the context of attachment research in typically developing children and are being specifically linked to attachment style as the outcome variable. In contrast, the choice for the MBRS and CBRS was inspired by its previous usage in groups of young children with developmental and intellectual disabilities as well as its aim to evaluate (aspects of parental behavior specifically related to) broader child development (Hostyn, Petry, Lambrechts, & Maes, 2011; Mahoney & Nam, 2011). Validation research on the original MBRS and CBRS showed a Cronbach's Alpha of .89 and Kaiser-Meyer-Olkin of .86 (Kim, Sung, & Hyun, 2000). The CBRS identifies two domains of children's interactive engagement: attention (*composed of the items: attention to activity, persistence, involvement and cooperation*) and initiation (*composed of the items: initiating activities, joint attention and affect*). The MBRS identifies four domains of parental interactional style: responsive/child oriented behavior (*composed of the items: sensitivity to child's interest, responsivity and effectiveness*), affect/animation (*composed of the items: acceptance, enjoyment, expressiveness, inventiveness and warmth*), achievement orientation (*composed of the items: achievement and praise*) and directive behavior (*composed of the items: directiveness and pace*). All items were rated on a 5-point Likert scale, indicating a low to high presence of the behavioral dimension. A general description of MBRS and CBRS items is presented in Table 2.

In a first stage of the coding process, the first author, a colleague researcher and two master's students explored the suitability of the CBRS and MBRS within this specific target group, by rating test observations and through conversation focused on resolving disagreements. For three items (effectiveness, directivity and cooperation), modifications to the original scoring instructions were made in order to preserve the core concept and ensure comparability of Likert ratings with previous research using the CBRS and MBRS. Regarding effectiveness, the original instructions stated that the parent's score is also dependent on the ability to gain a reciprocal exchange with the child, characterized by balanced turn taking. Since the cognitive (and communicative) limitations of the children often impede the presence of this interactional feature within this target group, it was not taken into account. Furthermore, parent's initial choice of activity and provision of materials was not regarded as directive behavior, since free play of the children in this target group is often impeded by their (cognitive and) motor limitations. Finally, (absence of) compliance was only rated when

Table 1
Sample characteristics.

Variable	*n*	%	Range	*M*	*SD*
Children	25				
Gender					
Male	7	28			
Female	18	72			
Age (months)			14–57	38.27	10.41
Sensory impairments					
Reduced vision	10	40			
Blindness	2	8			
Reduced hearing	1	4			
Deafness	2	8			
Motor impairments					
Hypotonia	18	72			
Hypertonia	10	40			
Contractures	2	8			
Scoliosis	3	12			
Others	5	20			
Health problems					
Gastro-intestinal problems	15	60			
Heart problems	0	0			
Respiratory problems	8	32			
Epilepsy	17	68			
Others	8	32			
Use of feeding tube	12	52			
Etiology					
Genetic defect	13	52			
Perinatal asphyxia	2	8			
Acquired brain injury	2	8			
Unknown	8	32			
Parity					
Firstborn	10	40			
Not firstborn	15	60			
Parents	25				
Gender					
Male	6	24			
Female	19	76			
Age (years)			26–46	33.84	4.77
Highest educational level					
Primary education	2	8			
Secondary education	8	32			
Higher education	15	60			
Activity status					
Working full-time	12	48			
Working part-time	7	28			
(Consciously) unemployed	6	24			
Families	25				
Number of family members			3–8	4.12	1.20
Parent(al figure)s			1–2	1.92	0.28
Children			1–6	2.20	1.16
One-parent households	2	8			
Reconstituted families	2	8			
Country of residence					
Belgium	11	44			
The Netherlands	14	56			

the parents' requests/suggestions were in concordance with the cognitive and motor capabilities of the child.

In a second stage, two other master's students carried out the final ratings. Several measures were taken to ensure the reliability of the rating process. First of all, raters were unfamiliar with the parent-child dyads and other test results. Further, prior to the final rating process, they completed a 30-h training until they had reached over 90% mutual agreement within one point and over 50% exact agreement. In order to reduce observation effects, the first 2 min of the video-taped observations were not taken into account during the scoring process, as this was regarded as an adjustment period. For each dyad, the subsequent 10 min were rated. The fragment was then further divided into five time sections of 2 min, since the test-runs showed that considerable fluctuations in child behavior (e.g. varying levels of attention, characteristic of this target group) and subsequent parental behavior impeded the raters' ability to reliably decide on overall scores. The test-runs also showed 2 min to be an adequate time range, since the use of a

Table 2
Description of MBRS and CBRS items.

MBRS – Responsive/child oriented behavior	
Sensitivity to child's interest	The extent to which the parent seems aware of and understands the child's activity or play interests, assessed by parent's engaging in child's activity choice, verbal comments in reference to child's interest and visual monitoring of child's behavior or activity.
Responsivity	The frequency, consistency and supportiveness of parent's responses to the child's behaviors. Responses are supportive when they match the child's actions, requests and intentions.
Effectiveness[a]	The parent's ability to engage the child in the interaction: to gain the child's attention, cooperation and participation.
MBRS – Affect/animation	
Acceptance	The extent to which the parent's behaviors and communications accept or affirm the child and what the child is doing.
Enjoyment	The parent's enjoyment of interacting with the child. There is enjoyment in child's being himself rather than the activity the child is pursuing.
Expressiveness	The tendency of the parent to communicate and react emotionally toward the child, including the frequency of verbal and nonverbal communications as well as the intensity and animation of these communications.
Inventiveness	The range of stimulation parents provide including the ability to find different things to interest the child, different ways of using materials etc.
Warmth	Positive affective expression through pats, lap-holding, caresses, kisses, hugs, tone of voice, and verbal endearments.
MBRS – Achievement orientation	
Achievement	Parent's encouragement of sensorimotor and cognitive achievement, whether through play, instruction, training, or sensory stimulation.
Praise	Quantity of verbal praise given to the child, given for compliance, achievement or for the child being himself.
MBRS – Directive behavior	
Directiveness[a]	Frequency and intensity in which the parent requests, commands, hints or attempts in other manners (e.g. physical) to direct the child's immediate behavior.
Pace	Parent's rate of behavior, assessed apart from the child's behavior.
CBRS – Attention	
Attention to activity	The extent to which the child attends to activities, whether or not the child is actively involved.
Persistence	The extent to which the child practices actions/vocalizations and continues to try solutions even though not successfully reaching his or the adult's goal.
Involvement	The intensity with which the child is motivated to engage/participate in the activities regardless of whether they are adult or child initiated.
Cooperation[a]	The degree to which the child attempts to comply with the requests or suggestions of the adult.
CBRS – Initiation	
Initiating activities	The extent to which the child initiates new or altered activities, without waiting for the adult's suggestion or guidance.
Joint attention	The extent to which the child initiates interaction and shares the attention for an object/event with the adult.
Affect	The child's general emotional state during the interaction, expressed through smiles, laughs or vocalizations.

Note: Description of items is based on Mahoney (1998, 2008).
[a] Item was adapted to the specific characteristics of this study's target group.

broader interval reduced rating reliability and the use of a smaller interval did not provide enough information to reliably decide on scores on all behavioral domains. Each time section was examined three times. During a first inspection, parent and child were simultaneously observed and raters took general notes on the participants' behaviors. During the second inspection, the raters focused on parental behavior and decided on a score for all 12 items of the MBRS. After the third inspection, focusing on the child, the seven items of the CBRS were scored. Child communication profiles were used to enhance the raters' understanding of the communicative utterances of the participating children. These profiles, filled in by parents, provided information on the ways a child usually communicates through gaze direction, facial expression, sound, posture and/or movement. Each time after rating a group of five dyads, a reliability check was performed to verify if retraining was necessary. Since overall agreement never dropped below the initial level, retraining did not occur. For each rater, the scoring process resulted in 95 section scores (19 items × 5 time sections) per dyad.

Overall, the interrater agreement within one scale point was 94%. For 56% of section scores exact agreement was obtained. Rater differences were dealt with by averaging raters' section scores that differed one scale point (38% of all scores). Section scores differing more than one scale point (6% of all scores) were discussed and decided upon through a consensus rating procedure. Interrater agreement for the 12 MBRS and 7 CBRS individual scale items ranged from 41% to 63% and 47% to 73% (exact) and from 86% to 100% and 84% to 97% (within one scale point), respectively. At last, the five section scores per item were averaged, resulting in 19 item scores for each dyad. For 1 parent-child dyad, only 3 time sections were available and consequently averaged to obtain

their item scores (i.e. due to fatigue of the child). Global domain scores relating to the four domains of parent's interactional style and two domains of child's interactive engagement were obtained by averaging item scores.

2.2.2. Parenting behavior

Parents were also asked to complete the *Parental Behavior Scale for toddlers* (PBS; Van Leeuwen, Rousseau, Hoppenbrouwers, Wiersema, & Desoete, 2011), a self-report measure of observable parenting behavior among parents of children younger than 4 years, consisting of 46 items rated on a 5-point Likert scale (ranging from occurring 'never' to 'always'). This questionnaire is a modified version of the 'Parental Behavior Scale' aimed at children between 8 and 14 years old, which is characterized by a good reliability and validity (Van Leeuwen & Vermulst, 2004). The questionnaire comprises six parenting domains: warmth/support (n = 15), autonomy support (n = 7), supervision and safety (n = 4), discipline (n = 10), rules and structure (n = 7), and rewarding (n = 3). Global ratings on these six domains of parenting behavior were obtained by averaging item scores. This instrument was developed in the context of 'JOnG!', a longitudinal study on a myriad of child and family factors in children of different ages (Van Leeuwen et al., 2011). Out of approximately 3000 participants of this longitudinal study in Belgium, we selected a control group of typically developing children (n = 25) nearest matching our own study group as a whole on gender and age of the children as well as gender and educational level of the parents. An exact match was obtained with regard to child gender and parental educational level. Further, the control group consists of 5 fathers and 20 mothers, indicating an exact match on parent's gender was obtained in all but one case. The mean age of the control parents matches closely with the study group (range = 25–44 years, M = 32.24 years), while the mean age of the control children turns out to be slightly lower (range = 12–43 months, M = 33.76 months).

2.3. Data analysis

To provide a general characterization of parental behavior and children's interactive engagement, descriptive statistics (mean, standard deviation, median and range of scores) are presented. Since the difference between mean and median scores was generally small (M = 0.09), average scores are used to describe the results in-text. Further, Mann–Whitney U tests were used for the comparison of the study group and the matched control group on the PBS. We opted for a non-parametric test because of the small sample size and the presence of non-normality in the data. Effect sizes r were calculated by dividing the Z-statistic by the square root of N (Fritz, Morris, & Richler, 2012). For interpretation of effect sizes, we followed the guidelines of Cohen (1988), regarding 0.5 as a large effect. Finally, Spearman rank correlations were used for evaluating the relation between parental behavior (i.e. the four domain scores of the MBRS and six domain scores of the PBS) and child behavior (i.e. the two domain scores of the CBRS). Here also, Cohen's guidelines were followed for evaluating the strength of the correlations. Bootstrap confidence intervals for the statistically significant correlations are presented in-text. Bootstrapping is a technique from which the sampling distribution of a statistic is estimated by taking repeated samples (with replacement) from the data set (Field, 2009), resulting in a more robust estimation in case of a small sample size (Adèr & Adèr, 2008) and the presence of non-normality in the data (Wood, 2004). In advance, scatterplots of the related variables were visually inspected for monotonicity of the relationship. A priori power-analyses (using G*Power 3.1; Faul, Erdfelder, Lang, & Buchner, 2007) with a specified power of 0.8 (Moore & McCabe, 2005) revealed that the current sample size allows for a reliable detection of large, but not small or medium correlations and group differences. Lastly, the possible influence of several child and parental characteristics on the results was evaluated by comparing the obtained values (mean scores) of the complete sample and a partial sample, based on a specified variable.

Table 3
Overview of item and domain scores on the Maternal Behavior Rating Scale.

MBRS	Mean (*SD*)	Median	Range
Responsive/child oriented behavior	**3.08 (0.54)**	**3.10**	**2.23–4.07**
Sensitivity to child's interest	3.26 (0.47)	3.20	2.50–4.10
Responsivity	3.15 (0.55)	3.10	2.10–4.00
Effectiveness	2.82 (0.70)	2.70	1.60–4.10
Affect/animation	**3.09 (0.51)**	**3.16**	**2.16–4.20**
Acceptance	3.23 (0.50)	3.20	2.50–4.20
Enjoyment	3.36 (0.54)	3.40	2.33–4.20
Expressiveness	3.11 (0.68)	3.20	1.70–4.20
Inventiveness	2.59 (0.52)	2.70	1.60–3.70
Warmth	3.16 (0.65)	3.10	1.90–4.90
Achievement orientation	**1.82 (0.54)**	**1.70**	**1.00–3.30**
Achievement	1.95 (0.67)	1.80	1.00–3.50
Praise	1.68 (0.59)	1.60	1.00–3.10
Directive behavior	**2.54 (0.45)**	**2.55**	**1.35–3.33**
Directiveness	2.37 (0.63)	2.20	1.20–3.67
Pace	2.71 (0.38)	2.80	1.50–3.30

Table 4
Overview of domain scores on the Parental Behavior Scale.

PBS	Study group			Control group			Comparison	
	Mean (*SD*)	Median	Range	Mean (*SD*)	Median	Range	*U*	*Z*
Warmth/support	3.94 (0.43)	3.93	3.13–4.53	4.14 (0.35)	4.00	3.40–4.73	230.50	−1.594
Autonomy support	3.31 (0.74)	3.29	1.86–4.43	3.97 (0.36)	3.86	3.43–4.71	156.50	−3.041**
Supervision and safety	4.61 (0.62)	4.75	2.25–5.00	4.57 (0.47)	4.75	3.50–5.00	269.50	−0.866
Discipline	1.90 (0.59)	1.80	1.00–3.20	2.88 (0.32)	2.90	2.30–3.40	45.00	−5.202**
Rules and structure	3.54 (0.46)	3.57	2.14–4.57	4.01 (0.32)	4.00	3.57–4.71	117.50	−3.814**
Rewarding	4.57 (0.58)	5.00	3.00–5.00	4.69 (0.42)	5.00	4.00–5.00	281.50	−0.665

* $p < .05$.
** $p < .01$.

3. Results

3.1. General characterization of parental behavior (Aim 1)

3.1.1. Parents' interactional style

The mean, standard deviation, median and range of item and domain scores on the Maternal Behavior Rating Scales are presented in Table 3. The average item scores related to the first two domains, responsive/child oriented behavior and affect/animation, were situated within the middle range of the 5-point Likert Scale. The other two domains showed low average item scores, with achievement orientation yielding even lower results than directive behavior.

3.1.2. Parenting behavior

The mean, standard deviation, median, range and comparison results of the domain scores on the Parental Behavior Scale for toddlers are presented in Table 4. *With regard to our study group*, scores on all but one parenting domain were situated within the higher range of the 5-point Likert Scale, with rewarding and supervision/safety yielding the highest scores. Only disciplining the child yielded a relatively low score. Relative to the other parenting domains, the scores on autonomy support indicate a high variability of this type of parenting behavior within the participant group.

At a more detailed level, the three items related to rewarding consistently received high ratings (with average item scores ranging from 4.48 to 4.64), indicating that parents often show their appreciation of children's positive behavior through verbal comments, facial expression and/or touch. Similarly, the four items related to supervision/safety were rated consistently high (with average item scores ranging from 4.24 to 4.80), indicating that parents monitor their children and the safety of their environment closely.

Furthermore, high average scores related to parental warmth/support indicate that parents play with their child, provide distraction when the child is restless, imitate the child's vocalizations, sing or hum to the child and show the child how to use a toy (with average scores ranging from 3.76 to 3.92). Even higher scores (4.12–4.72) were evident with regard to holding the child on the lap, making the child laugh (e.g. by tickling), cuddling, talking to the child during nursing, examining why the child cries or behaves differently than usual and comforting the child. Only pointing at and naming things (2.84), looking at books together with the child (2.64) and explaining the child how something works (2.52) yielded lower results.

Four items related to autonomy support yielded relatively high scores (ranging from 3.56 to 4.24), indicating the parents provide their children with the opportunity and encouragement to try new things while simultaneously supporting them when they do not succeed. Encouraging children to make their own choices as well as completely taking over when the child does not succeed, yielded relatively lower scores (2.92 and 2.88). The last item, 'I give my child small assignments that he can do on his own', yielded a very low average item score of 1.88.

The relatively high average scores on three items with regard to rules and structure (4.00–4.24) indicate that parents provide their children with a fixed daily routine. Also, parents avoid overstimulation of the child (3.56), but push through when the child does not want something, such as eating or sleeping (3.40). Relatively lower scores were given to the two items about clearly telling the child what is not allowed (2.52) and quickly allowing what the child wants or demands (2.96).

Similarly, with regard to discipline, allowing what the child wants or demands in order to avoid upsetting the child yielded a rather low average score of 2.68. The same goes for three items on disciplining the child verbally, through facial expression and body posture, with average scores between 2.56 and 2.64. However, disciplining the child by ignoring unwanted behavior, giving a time-out, providing (positive or negative) punishment, giving a light slap or handling the child in a rough manner, yielded even lower scores (1.20–1.96). Standard deviations of all average item scores ranged from 0.44 to 1.53, indicating variability in parental behavior across parent-child dyads.

In the control group, the same general pattern is recognizable: all domains yielded scores within the higher range of the scale, except for discipline. However, Mann–Whitney *U* tests showed that the control group's scores were significantly higher for autonomy support ($|r| = 0.43$), rules and structure ($|r| = 0.54$) and particularly discipline ($|r| = 0.74$); all characterized by rather large effect sizes. Also, standard deviations indicate a lower variability of scores on all domains compared to the study group.

At item level, there were no striking differences between the two groups concerning rewarding behavior and supervision of the

Table 5
Overview of item and domain scores on the Child Behavior Rating Scale.

CBRS	Mean (*SD*)	Median	Range
Attention	**2.61 (0.52)**	**2.55**	**1.60–3.57**
Attention to activity	3.12 (0.76)	3.20	1.40–4.50
Persistence	1.44 (0.63)	1.20	1.00–3.00
Involvement	2.92 (0.70)	2.80	1.60–4.20
Cooperation	2.95 (0.65)	2.90	1.67–4.60
Initiation	**2.22 (0.37)**	**2.20**	**1.60–3.33**
Initiating activities	1.29 (0.35)	1.10	1.00–2.10
Joint attention	2.06 (0.61)	2.10	1.00–3.80
Affect	3.31 (0.53)	3.30	1.80–4.20

child. With regard to the other four domains, some items showed statistically significant group differences, with *p*-values ranging from 0.000 to 0.046 and the absolute value of effect sizes ranging from 0.28 to 0.84. In the context of warm and supportive parenting, the parents in our study group more often cuddle with their child and mimic the sounds they make, but showed lower levels of pointing at and naming things, looking at books together and explaining how something works. The group difference in autonomy support is illustrated by lower scores on encouraging the child to try new things and to make own choices, compared to the control group. Also, parents in our study group less often reported letting the child discover the environment on its own or giving the child small assignments that he can do on his own. Furthermore, with regard to rules and structure, parents in our study group reported less persistence when the child does not want something, less strict bed times and lower levels of telling the child what is not allowed. At last, parents in our study group reported lower scores on almost all discipline items. Only disciplining the child by ignoring unwanted behavior or handling the child in a rough manner and allowing what the child wants or demands in order to avoid upsetting the child yielded comparable results to the control group.

3.2. General characterization of children's interactive engagement (Aim 2)

The mean, standard deviation, median and range of item and domain scores on the Child Behavior Rating Scales are presented in Table 5. Average scores on the two domains, attention and initiation were situated within the lower range of the 5-point Likert scale. Average item scores showed some diversity: persistence and initiating activities yielded very low results, while scores on attention to activity and affect were situated within the relatively higher range of the scale. Variability in participant's scores was higher for attention than initiation, with attention to activity yielding the highest variability and initiating activities yielding the lowest variability.

3.3. Associations between parental and child behavior (Aim 3)

Spearman correlations between domain scores on the MBRS, PBS and CBRS are presented in Table 6. The majority of domain scores within the different instruments were significantly and positively related. Hereafter, only the correlations with a relatively small bootstrap interval range (i.e. $\leq .50$) are discussed. Within the MBRS, responsivity and affect/animation were highly correlated (95% CI [.59, .90]). Within the PBS, warmth/support was highly correlated to autonomy support (95% CI [.55, .87]) as well as rules/

Table 6
Overview of Spearman correlations between domain scores of the MBRS, PBS and CBRS.

Domain scores	M1	M2	M3	M4	P1	P2	P3	P4	P5	P6	C1	C2
MBRS												
M1. Responsive behavior	–											
M2. Affect/animation	.80**	–										
M3. Achievement orientation	.49*	.47*	–									
M4. Directive behavior	.27	.36	.49*	–								
PBS												
P1. Warmth/support	−.05	.02	−.09	.39	–							
P2. Autonomy support	−.08	.01	.09	.45*	.76**	–						
P3. Supervision/safety	−.17	−.09	−.03	.09	.38	.46*	–					
P4. Discipline	−.20	−.04	.18	.25	.36	.40	.28	–				
P5. Rules/structure	−.29	−.17	−.04	.12	.73**	.63**	.41*	.50*	–			
P6. Rewarding	−.15	−.19	−.13	.01	.59**	.59**	.48*	.24	.50*	–		
CBRS												
C1. Attention	.51*	.20	.06	−.05	.05	.08	−.01	.11	.15	.06	–	
C2. Initiation	.56**	.39	.16	.00	−.15	.00	−.15	.14	−.09	−.16	.78**	–

* $p < .05$.
** $p < .01$.

structure (95% CI [.48, .86]). Autonomy support and rules/structure were also mutually correlated (95% CI [.33, .83]). Within the CBRS, attention and initiation of the child were highly correlated (95% CI [.45, .94]). For all other significant correlations, the lower limits of the bootstrap intervals ranged from .04 to .26 and the higher limits ranged from .66 to .86.

MBRS domain scores were not related to PBS domain scores. Only directive behavior within the interaction and reported autonomy support rendered a significant and positive correlation with a 95% CI between .05 and .76.

At last, no significant correlations were found between the PBS and CBRS domain scores. However, responsive behavior (MBRS) was significantly and positively related to attention (95% CI [.11, .79]) as well as initiation (95% CI [.12, .84]).

3.4. Child and parental characteristics

In order to evaluate the possible influence of several child and parental characteristics on the results, we compared the obtained values (mean scores and correlations) of the complete sample and partial, more homogeneous, samples. The sample was subdivided according to the specified variables (child age, child gender, child sensory impairments, parent's age, parent's gender, parent's educational level, parent's activity status, country of residence and parity) and each time, the largest part was used in the analysis in order to preserve as much sample size as possible. Within the partial samples, mean scores on parental interactional style and children's interactive engagement never differed more than .12 from the values obtained by analysis of the whole sample Mean scores on parenting behavior never differed more than .25. The largest differences ($\geq .20$) indicate that parents report less supervision when the child is younger and does not have a sensory impairment, and parents report less disciplining behavior when they are highly educated. The pattern of correlations within the partial samples (i.e. strength as well as direction) was generally comparable to the values obtained by analysis of the whole sample. Also, significance levels were comparable amongst higher ($> .60$) correlations, while correlations below .60 often did not reach the .05 significance level within the partial samples, as could be expected based on the power analysis. Further details and concrete results are available from the first author upon request.

4. Discussion

4.1. Conclusions

4.1.1. Characterization of parental behavior

The parents of young children with a significant cognitive and motor developmental delay in this study are generally sensitive and responsive towards their children within an interaction characterized by acceptance, warmth and enjoyment. Although parents ‘guide’ the interaction by offering toys and/or initiating activities, they do not often attempt to direct their child's immediate behavior. Further, since encouragement of children's sensorimotor and cognitive achievement and the use of verbal praise occurs infrequently, the interaction is more oriented at creating a mutually shared experience rather than being regarded as a teaching opportunity.

In earlier research by Mahoney et al. (2007), all four dimensions of parental behavior towards young children with mild to moderate disabilities clustered near the midpoint of the scale, with their highest ratings in directiveness and their lowest in achievement orientation. In our target group, the same midpoint tendency for responsive behavior and affect can be observed. However, levels of directiveness as well as achievement orientation are strikingly lower. These results are surprising, since earlier research indicates that parents are more directive when children have lower developmental levels, presumably in an attempt to optimize children's opportunities for developmental learning (Spiker et al., 2002). It is possible that parents’ interaction patterns are influenced by previous experiences with their child, either by knowing/assuming the child does not understand, respond to or appreciate (verbal, gestural, physical, …) directives or by having few hopes for developmental benefits resulting from a directive/achievement oriented parental approach. The objection could be made that the low ratings of directiveness are due to the modified scoring instructions regarding directiveness. However, the modification was made to ensure the representativity of the ratings in relation to the core concept, i.e. to avoid a nonsensical inflation in parental directiveness scores due to the motor limitations of the child.

Based on self-report, the parents of young children with a significant cognitive and motor developmental delay in this study are generally warm and supportive towards their child, monitor the child and his/her environment closely and often show appreciation of the child's positive behavior. They also report high levels of autonomy support and the provision of rules and structure; however significantly less than parents of typically developing children. Disciplining the child is rarely reported and yields the largest difference with the control group of parents with typically developing children.

Parenting items requiring a higher level of cognitive and/or motor functioning of the child (e.g. verbal explanations, supporting independent exploration, …) yield relatively low scores in our study group, indicating that parents adapt their behavior to the child's (dis)abilities. Also, it is highly likely that the (relatively) low levels of discipline and rules/structure are related to low levels of undesirable child behavior. Furthermore, the greater variability in parenting scores within the study group could be related to the greater variability in children's cognitive and motor functioning, compared to the control group.

In conclusion, the same conceptual domains of interactional style and parenting behavior used in research on typically developing children and children with mild disabilities are present, and thus applicable, in research on the specific target group of young children with a significant cognitive and motor developmental delay. However, there appear to be notable differences in frequency levels (e.g. with regard to discipline) as well as in the concrete operationalization of some parenting domains (e.g. parental effectiveness within the interaction).

4.1.2. Characterization of children's interactive engagement

Children within the study group are moderately attentive during an interaction with their parent, showing some involvement and cooperation, but are rarely persistent in practicing actions and/or vocalizations. The children seldom initiate new or altered activities, but show some attention to the adult (primarily through establishing eye contact). Children generally display low intensity enjoyment throughout the interaction.

In Kim and Mahoney (2004), the interactive engagement levels of young children functioning cognitively at approximately half their chronological age or lower were compared to typically developing children, matched for chronological age. Results indicated lower scores on all 7 CBRS items in children with disabilities (significantly so for attention to activity, persistence, cooperation and joint attention). The scores in our study group are generally comparable to those of children with disabilities within the study of Kim and Mahoney (2004); except for persistence (more than one scale point lower) and initiation (almost two scale points lower). A possible explanation for these strikingly lower scores is that persistence and initiation are strongly influenced by the additional motor limitations in our study group.

4.1.3. Associations between parental and child behavior

The majority of parent's behavioral dimensions showed moderate to high, but not perfect, correlations within the respective instruments. Between instruments, observed parental interactional style and reported parenting behavior were not related in all but one case. This could indicate that the MBRS and the PBS each provide unique information on different types of parental behavior. The results do suggest that parents who report higher levels of autonomy support also show more directive behavior within the interaction, but the wide bootstrap confidence interval prevents us from drawing a reliable conclusion on the strength of this relation.

Reported parenting behavior was not related to children's interactive engagement. However, parent's responsivity and child orientedness within the interaction proved to be significantly related to children's attention as well as initiation. Here also, the wide confidence interval prevents us from drawing a reliable conclusion on the strength of this relation. Nevertheless, this result confirms the importance of sensitive responsivity as the primary variable in parenting research (Mahoney & Nam, 2011), regardless of the child's developmental level, as well as in interactional research on persons with severe and multiple disabilities (Hostyn & Maes, 2009). However, it is unclear whether parental responsivity elicits child engagement, child engagement elicits parental responsivity, or both processes influence each other bidirectionally.

4.2. Strengths and limitations

Parental behavior is a broad concept, for which a variation of coding and classifying procedures has been used (Mahoney & Nam, 2011). Conclusions drawn in parenting research are thus directly dependent on the way the variables are operationalized. In this explorative study, we incorporated two types of measures of parental behavior. Through the questionnaire, information is gathered on the frequency of general parenting behavior across many contexts and over time. Using the observational method, parents' behavioral dimensions within the concrete and momentary context of a parent–child interaction are studied. While both data collection methods are possibly influenced by social desirability bias, the latter could also be influenced by the rater's subjective interpretation. By providing clear instructions (before self-report as well as observation), incorporating adjustment time (during data collection as well as coding) and keeping close track of inter-rater reliability, we attempted to constrain possible bias.

This study's results need to be interpreted with caution. For one, the sample size in this study is fairly limited, yet unavoidable due to strict eligibility criteria (e.g. combination of cognitive and motor limitations and small age range in the children), low prevalence rates and other practical and emotional barriers experienced by parents. Therefore, this study is unable to reliably detect small or medium correlations and group differences. Also, the non-randomized sampling method impedes generalizability. For example, even though we targeted children as young as 6 months, the youngest participant in our sample is 14 months. Since we primarily recruited participants indirectly through professionals, our insight into a possible selection bias and the reasons behind non-participation is fairly limited. In addition, children with severe and multiple disabilities often show fluctuations in their behavioral patterns over time (Goldbart, 1994; Munde, Vlaskamp, Ruijssenaars, & Nakken, 2011). Therefore, the momentary observation of children's interactive engagement might not be fully representative of the child's daily functioning. Furthermore, this study does not give rise to causal statements. We have merely described the co-occurring presence of parental and child behavioral dimensions. However, the correlational nature of the analyses within this study correspond to the presumed bidirectionality of parental and child behavior. We cannot rule out the possibility that different children elicit different parental responses (Collins et al., 2000) and that children with various disabilities, genotypes, and temperament styles react differently to similar parental input (Maccoby, 2001; Mahoney & Nam, 2011).

Because of the very scarce knowledge in the current research literature, we think there is great merit in providing a general characterization of parental and child behavior in the specific group of children with significant cognitive and motor limitations as such. Whenever possible, results were also compared to previously collected data (i.e. PBS) and current literature on the variable of interest (i.e. MBRS and CBRS) to gain more specific insight into how these behavioral dimensions are present in other target groups. It would have been interesting to extend the use of a control group to the observational data, in order to perform a more reliable comparison of the study group's results. However, because of the multiplicity and complexity of the participant's limitations, it is unclear which characteristics are most relevant for the selection of a control group. Therefore, the use of multiple control groups (incorporating children with no or other types and levels of disabilities) could have provided a more extensive and insightful comparison. Regrettably, both options would have compromised the feasibility of this specific study.

Finally, a rough comparison of the mean scores and correlations in the complete sample and partial samples showed relatively

small differences based on child age, child gender, child sensory impairments, parent's age, parent's gender, parent's educational level, parent's activity status, country of residence and parity. However, due to the limited sample size we are not able to formulate reliable, statistically robust, conclusions regarding this topic.

4.3. Suggestions for future research

First, we would like to point out that our target group consists of individuals with such profound intellectual disabilities that no existing standardized tests are applicable for a valid estimation of their level of intellectual capacity (Nakken & Vlaskamp, 2007). Established norms are usually based on scores of typically developing persons and are therefore not reliable for groups who achieve extremely low scores (Resing & Blok, 2002). Also, the assessment procedure is challenging, due to fluctuating attention and achievement levels as well as the additional motor limitations. Additionally, Weis (2014) reports that the measurement of intellectual functioning with standardized, individual intelligence tests is typically done in children of approximately 5 years or older. Hence, it is not common for the children in our target group to receive a definite diagnosis at this young age. Therefore, we opted to use the description 'significant cognitive (and motor) developmental delay' instead of applying the term 'profound ID'. A strong suggestion for future research is the development and (international) validation of an assessment procedure for determining the level of cognitive functioning in young children with significant cognitive and motor limitations.

Further, we would recommend studying a larger and randomized group of participants in order to increase representativeness and statistical power. However, combining the results of several small-sample replication studies and gathering data longitudinally can be a more feasible way to gather knowledge on this specific target group. In this regard, we have experienced that joining forces through (inter)national collaboration can be an ideal tool, e.g. to increase sample sizes and generalizability as well as to stimulate theoretical and methodological reflection.

In reference to this study's concrete results, further research should go beyond correlational analyses and make an attempt to disentangle the direction(s) in which child and parental behavior influence one another. Longitudinal analyses, taking into account the child's initial functioning, could provide evidence that "parenting conceivably affects – rather than simply accompanies or follows from – child adjustment" (Collins et al., 2000). Further, the questionnaire and rating scale used in this study capture qualitative aspects of parental behavior but fail to assess specific behavioral linkages (Bornstein & Manian, 2013). In-depth analysis of the observational data would allow us to explore the reciprocal sequences of parental and child behavior within this target group, taking into account the considerable fluctuations in child and parental behavior that are characteristic of this target group. Both approaches (longitudinal and sequential) could help pave the way for the development of specific parenting support interventions within the target group of young children with a significant cognitive and motor developmental delay.

As said, this study primarily focuses on qualitative aspects of parental behavior. However, children's experiences also differ in terms of the number of times parents interact with them as well as the types of activities they do together (Mahoney & Nam, 2011). Incorporating these variables into future studies, combining data on different family members (e.g. parental figures and siblings) whenever possible, would give us a more comprehensive view of the child's primary context and its influence on child behavior and development.

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