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Variation in differential reactions to comfort by parents versus strangers in children with severe or profound intellectual disabilities: the role of parental sensitivity and motor competence

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Abstract
Displaying selective attachment behaviours is an important developmental milestone for children with severe or profound intellectual disabilities (SPID). In the current study, between-child differences in their selective emotional responses to comfort provided by parents versus strangers were observed. We explored links between these differences and parental sensitivity and motor competence. A home-based experimental observation was conducted in 38 parent-child dyads, exposing children to four naturalistic stressors and to comfort provided by either their parents or a stranger. Emotional behaviour (arousal and valence) was micro-coded and differentiation variables were constructed, reflecting the children's level of differentiation between the parent and the stranger. Parental sensitivity was coded using the Emotional Availability Scales. Results showed that these children's differentiated responses to comfort were related to children's motor competencies (particularly their fine motor skills), but not to parental sensitivity. This study shows the need to go beyond sensitivity to understand individual differences in the most basal aspects of attachment for children with SPID.

Keywords Parent-child attachment · Severe or profound intellectual disability · Comfort · Emotional behaviour · Parental sensitivity · Motor competence

Introduction

Secure attachment relationships with primary caregivers are conducive to resilient development, as secure attachment experiences foster adaptive social-emotional functioning and general emotional well-being (Bowlby, 1969/1982; Cassidy, 2016). For children with significant cognitive disabilities, attachment relationships are even more important to counterbalance the difficulties linked to their impairment(s) (Janssen et al., 2002; Schuengel et al., 2013). It could be argued that fostering secure attachments is one strategy to prevent mental health and behavioural problems in children with significant cognitive disabilities on the long term, often caused or sustained by maladaptive affect regulatory skills (Bradley, 2000).

However, to understand differences in attachment security (which are referred to in literature as differences in '*the quality of attachment*'), children must first develop consolidated, selective attachment relationships with their principal caregivers (Ainsworth et al., 1978; Marvin et al., 2016). Developing consolidated attachment relationships is a developmental milestone, typically acquired around the age of six to nine months (Ainsworth et al., 1978; Marvin et al., 2016). Not all children with significant cognitive disabilities will reach this developmental age and the cognitive, communicative and (loco)motor prerequisites that come with that age (AAIDD, 2021; Marvin et al., 2016; Nakken & Vlaskamp, 2007). Previous research indeed found indications for interindividual differences in (the development and/

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All authors have contributed to, seen, and approved of the manuscript and agree to the order of authors as listed on the title page.

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or expression of) selective attachment (behaviours) in children with severe or profound intellectual disabilities (SPID) (Vandesande et al., 2019, 2020). However, these differences were not the explicit focus of these studies and were, thus, not yet explained with regard to explanatory factors.

Children with SPID have significant deficits in intellectual functioning (IQ < 35–40) and adaptive functioning (AAIDD, 2021). They often have additional (neuro)motor impairments (Arvio & Sillanpää, 2003). In literature, a subgroup within the group of children with SPID is referred to as children with '*profound intellectual and multiple disabilities*' (PIMD; Maes et al., 2020; Nakken & Vlaskamp, 2007). Children with PIMD have the profoundest intellectual disabilities (corresponding with an estimated IQ < 20 or developmental age below two years, in adults) combined with a significant motor impairment. Both children with SPID (and the subgroup of children with PIMD)[1] require intensive support in every aspect of daily life (AAIDD; 2021; Nakken & Vlaskamp, 2007) and often communicate at pre- or protosymbolic level (Dhondt et al., 2019). This implies communication through affective behavioural signals and idiosyncratic, bodily expressions (often person- and context-bound).

With this target group, Vandesande et al. (2020) conducted a home-based experimental observation in which 38 children with SPID (of which 15 could also be classified as children with PIMD) were confronted with four naturalistic stressors until the parents indicated a negative effect on their children's stress levels. Immediately after, the children received comfort, either provided by their parents or a stranger in random order. The main goal of the latter study was to examine the extent to which children with SPID differentiated between their parents and a stranger providing comfort after eliciting stress. From an intra-individual (within-child) perspective, the study of Vandesande et al. (2020) investigated the theoretical proposition that parents would have an emotion co-regulating function as attachment figures for their children with SPID that strangers would not have. Therefore, Vandesande et al. (2020) used emotional behaviours as indicators for attachment-relevant processes, more specifically: (1) *arousal*: the overall amount of both positive and negative tension/emotions, and (2) *valence*: the extent to which these emotions are positive, negative or neutral. They complemented these behavioural indicators with psychophysiological measures of arousal (in the latter study skin conductance). Psychophysiology offers an additional indicator of a person's emotional states (Braithwaite et al., 2015), and can be used to investigate arousal in persons with PIMD (Vos et al., 2013). Vandesande et al. (2020) demonstrated that children with SPID, on a group level, showed selective attachment by differentiating their psychophysiological and behavioural emotional responses between their parents as attachment figures and a stranger after experiencing mild distress. However, the study also reported that a substantial amount of unexplained variance was situated at the interindividual level, which was not yet examined with regard to explanatory factors (e.g., approximately 25% of the variance for arousal and 16% for valence was situated between children).

The professional support we provide to the parents of these children would benefit from in depth understanding of the nature of these interindividual differences in selective attachment and their associated factors. The study of individual differences in attachment is an extensive tradition of research, especially in children with typical abilities. However, these studies have almost exclusively focused on understanding differences in attachment quality (Ainsworth et al., 1978; De Wolff & Van IJzendoorn, 1997; Fearon & Belsky, 2016). In children with SPID, the study of individual differences in both attachment quality and – on a more basal level- selective attachment is challenging and, thus, yet scarce. With regard to attachment quality, there is some research in related target groups. For instance, older research often focused on Down's syndrome (see Janssen & Schuengel, 2006 and Schuengel et al., 2013, for a review), one of the most prevalent genetic birth defects causing intellectual disability (ID; Presson et al., 2013). These studies found significantly different distributions in attachment quality categories compared to children with typical abilities (e.g., Vaughn et al., 1994). Similarly, in children with non-specific ID syndromes, resulting in mild-to-moderate cognitive impairments, Feniger-Schaal and Joels (2018) found individual differences in attachment quality, with an underrepresentation of secure attachment. On the more basal level, De Schipper et al. (2006) studied selective attachment behaviours in their descriptive study using the Attachment Q-sort (Waters, 1987). The latter study included six children with a moderate-to-severe ID (varying in chronological age, developmental age and DSM diagnosis). They found reliable variation in attachment behaviours towards the children's professional caregivers both within and across children, irrespective of their level of functioning.

Though the latter studies demonstrated meaningful interindividual differences in related target groups, they focused mostly on attachment quality and/or in a slightly higher functioning group than the group of children with SPID. For the latter children, two reflections must be made related to the study of interindividual attachment-related differences. First, in this group, the study of attachment quality is even

[1] In what follows, there will be referred to the broader group of '*children with SPID*', of which children with PIMD are part, and are thus also implied. When only the subgroup of children with a profound intellectual disability and the additional neuromotor impairment are implied, the term '*children with PIMD*' will be explicitly used.

more challenging (methodologically) due to the severity and complexity of their impairments. Most instruments are not adept to reliably and validly assess attachment quality, irrespective of the children's varying motor and communicative abilities. Second, it could be argued that researchers should first address the demonstrated interindividual differences on the most basal level of attachment development, namely on the domain of selective attachment (Vandesande et al., 2020). Indeed, given their developmental age, the question of attachment quality is not necessarily yet an issue for all children with SPID (Ainsworth et al., 1978; Marvin et al., 2016). These reflections are exemplified when one takes a closer look at the application of the well-known instrument to assess attachment quality, '*Scoring System of Interactive Behaviors*' (SSIB; Ainsworth et al., 1978) in children with SPID (as was done in Vandesande et al., 2019). The behavioural indicators of the SSIB are not only described in a very physical manner (which leads to unequal chances to exhibit these behaviours for children with SPID and varying motor competencies), but the indicators are also not fine-grained enough to differentiate subtle behavioural differences within and between these children (Vandesande et al., 2019). For example, children who orient towards and glance at their mother upon reunion are scored equally low as children who do not react at all. Such subtle behavioural differences may still be meaningful among children with SPID, given that differentiation and selective attachment may already be an important developmental achievement on a more basal level (i.e., before one can speak of differences in attachment quality, which already requires the development of selective attachment; Marvin et al., 2016). In addition, the question remains which parental and/or child factors are potentially linked with meaningful interindividual differences in selective attachment among children with SPID.

To address this question, the current study builds on the research of Vandesande et al. (2020) by exploring the link between interindividual differences in the children's responses to comfort by parents versus strangers and associated factors at the level of the parent and at the level of the child. In the current exploratory study, we included parental sensitivity and children's motor competences as associated factors, inspired by the study of individual differences in attachment quality. One of the basic tenets of attachment theory is that parent factors contribute more decisively to individual differences in attachment quality compared to child characteristics, as was also found in the meta-analysis of Van IJzendoorn et al. (1992) and previous research (e.g., Feniger-Schaal & Joels, 2018). At the level of parental factors, parental sensitivity, defined as the ability of parents to read their children's signals and respond to them in an appropriate, prompt and effective way, has proven to be one of the most consistent, robust predictors of attachment quality (Ainsworth et al., 1978; Atkinson et al., 1999; De Wolff & Van IJzendoorn, 1997). It remains, however, the question whether this could also be a contributing factor to explain individual differences at the most basal aspects of (selective) attachment development. Dykas and Cassidy (2011) reported that individual differences in attachment quality might lead to defensive strategies (such as ignoring the parents' attachment-related care behaviours or looking less at the parent). These behavioural indicators might be related to these children's discriminative behaviours.

However, it is hypothesised that variation in expressed emotional differentiation, which is the index the current study adopts to identify selective attachment, is determined by both parent factors and child factors (e.g., ineffective affective signalling), especially in children with intellectual disabilities (Schuengel & Janssen, 2006). Specifically for children with SPID, who have a complex combination of impairments and a specific set of needs, the possibility is raised that attachment-related individual differences might not be a simple function of parental sensitivity but rather reflect an interplay of factors. For example, children with PIMD may have a significant motor impairment, physically impeding them to show certain attachment behaviours which require (minimal) motor competence (like crawling towards the parent). Tessier et al. (2002) reported that children with physical disabilities use alternative (subtle) expressions of attachment. From a theoretical viewpoint, children's locomotion is regarded as important in the ontogeny of attachment (Bowlby, 1969/1982; Marvin et al., 2016). Motor functioning shapes the expression of attachment behaviour (Marvin et al., 2016) and provides crucial building blocks of attachment development, such as understanding cause-effect and (uttering) person permanence (Bigelow et al., 1995; Trawick-Smith, 2019). Children's motor skills are furthermore strongly intertwined with their cognitive, social and perceptional development (Libertus & Hauf, 2017). In children who are also limited in their cognitive and communicative abilities to appeal to their parents (Nakken & Vlaskamp, 2007), motor skills might even be more of influence on the expression of attachment than for children who do have these abilities. In addition, it could be argued that children's motor competencies and their parents' sensitivity level are influencing factors that are possibly intertwined. Children's motor competencies indeed influence not only attachment, but also the interactive behaviours (e.g., parental sensitivity) shown by their interaction partners (Libertus & Hauf, 2017). Vandesande et al. (2019) reported for example extremely subtle and hard-to-notice expressions of contact-seeking behaviour in children with PIMD (such as subtly orienting the face towards the parent). These subtle expressions might require extraordinary sensitivity from the caregivers (Schuengel et al., 2010), leading to parenting behaviours which are usually not described in rating scales for the general population. The question is still outstanding whether these factors are also helpful in explaining interindividual differences in children's selective responses (that were first reported in Vandesande et al., 2020).

The current study reports on a first exploration of the links between interindividual differences in children's selective emotional reactions to comfort provided by their parents versus a stranger and parental sensitivity and the children's motor skills among children with SPID. Measures of both behavioural arousal and valence were included in the current study as a derivative of selective attachment of children with SPID and are considered as a proxy for their attachment development (at the most basal level). The research questions addressed were threefold:

(1) Are interindividual differences in selective reactions of children with SPID to the parents' or the stranger's comfort associated with parental sensitivity?
(2) Are these differences associated with the children's (fine and gross) motor competences?
(3) Does the association between these interindividual differences and sensitivity vary depending on children's motor competences?

Methods

The current study builds on the study of Vandesande et al. (2020) from an interindividual perspective, so that parts of the data set from the former study were used as a basis for the construction of differentiation measures. These measures reflect the extent to which children behaviourally differentiated between their parents and a stranger after experiencing mild distress. The Social and Societal Ethics Committee (SMEC, KU Leuven) ethically approved the research design and protocol (G- 201612708).

Participants

Participants were recruited via care organisations as intermediate partners, based on the following criteria: (1) Children had a chronological age from one up to and including eight years; (2) Children had a severe or profound intellectual disability (SPID) or in case an official diagnosis was lacking, they functioned at that level as judged by their professional caregivers; (3) Children primarily lived at home with their biological parent(s) at the time of the participation (i.e., the child slept at home for a minimum of four nights a week on average). Living in adoptive or foster care was an exclusion criterion, because these special life circumstances may have had an impact on attachment development (Carlson et al., 2014). Children varied in the presence and nature of additional medical problems (e.g., epilepsy) or comorbid sensory and/or motor impairments.

A sample of 38 children was included, of which 13 girls (34.2%) and 25 boys (65.8%), aged 2y2m (27 months) to 8y7m (103 months; $M = 72.07$, $SD = 20.40$) with a mean developmental age of 9.60 months ($SD = 4.85$, $n = 15$).[2] Children had an average cognitive delay of 50.53 months compared to their chronological age ($SD = 21.33$, $n = 15$). Following the definition of Nakken and Vlaskamp (2007) and Maes et al. (2020), 15 children could be classified as having PIMD . These children had a cognitive developmental age, corresponding with a profound intellectual disability (i.e., developmental age below a quarter of the chronological age or estimated IQ < 20) and a profound motor impairment (i.e., GMFCS levels equal to or larger than four). Nineteen children were classified as not having PIMD: they had no significant motor impairment which was combined with a profound intellectual disability. For four children, crucial (diagnostic) information was lacking so that they could not be reliably classified as having PIMD or not. Table 1 provides a description of general child and family characteristics.

For half of the children ($n = 19$) parents reported limited (independent) mobility, as well as limited fine motor skills in 28 children (73.7%). A severe motor impairment is reflected by level 4 and 5 of the Gross Motor Function Classification Scale (GMFCS; Palisano et al., 2007). In the current study, 15.8% of the children scored level 4 and 34.2% scored level 5. With regard to fine motor skills (e.g., handling objects), 47.4% of the children had level 4 and 26.3% had level 5 on the Manual Ability Classification System (MACS; Eliasson et al., 2017; Eliasson et al., 2006).

Procedure

A two-hour home visit was conducted by the first author, consisting of 1) a full explanation of the study's design and aims; 2) signing of the informed consent; 3) the further completion of general background questionnaires (which were already mostly completed by parents before the home visit); 4) the execution of a home-based experimental paradigm with stress and comfort; 5) a 20-min free interaction between the parent and the child.

The home-based experimental paradigm is described in Table 2. Children were confronted with four different, naturalistic stressors (i.e., daily stressors that have a negative

[2] Recent data on cognitive functioning (i.e., less than one year ago) were only available for 15 children. The large number of missing data with regard to cognitive functioning can partially be explained by the lack of reliable IQ-tests and established norms for young children with very low cognitive abilities (Maes et al., 2020; Weis, 2014). The estimation of 9.60 months may be an overestimation, because it were often the children with the profoundest disabilities for whom intellectual testing was missing.

Table 1 Description of the sample (n = 38)

Child characteristics			n (%)	M (SD)	Min - Max
Gender	Boy		25 (65.8)		
	Girl		13 (34.2)		
Chronological age (months)			38 (100.0)	72.07 (20.40)	27.00–103.00
Developmental age (months)[a]			15 (39.5)	9.60 (4.85)	2.00–20.00
Average cognitive delay compared to chronological age (months)			15 (39.5)	50.53 (21.33)	16.57–83.37
One or more official diagnosis[b]			33 (86.8)		
Aetiology known of the disability			25 (65.8)		
Additional impairments/problems	Motor		34 (89.5)		
		Hypotonic	24 (63.2)		
		Spasticity	8 (21.1)		
		Contractures	2 (5.3)		
		Scoliosis	3 (7.9)		
		Other (general delay)	13 (34.2)		
	Visual		8 (21.1)		
	Auditory		1 (2.6)		
	Medical		22 (57.9)		
GMFCS level	Level 1		8 (21.1)		
	Level 2		11 (28.9)		
	Level 3		0 (0.0)		
	Level 4		6 (15.8)		
	Level 5		13 (34.2)		
MACS level	Level 1		3 (7.9)		
	Level 2		1(2.6)		
	Level 3		6 (15.8)		
	Level 4		18 (47.4)		
	Level 5		10 (26.3)		
Family and living situation					
Children attend day care or special education school	Part-time		7 (18.4)		
	Full-time		31 (81.6)		
Living situation	With both biological parents		37 (97.4)		
	With one biological parent		1 (2.6)		
Mother's firstborn			18 (47.4)		
Siblings (≥ 1)	Brother(s)		19 (50.0)		
	Sister(s)		17 (44.7)		
Age parents	Mother		38 (100.0)	37.09 (4.95)	22.39–46.66
	Father		36 (94.7)	39.68 (3.93)	32.78–48.74
Full-time work parents	Mother		17 (45.0)		
	Father		32 (85.0)		

[a]Only reported in the current study if the test for intellectual functioning was administered recently (i.e., less than one year ago)

[b]This can either be a specific, established diagnosis such as Down Syndrome, Rett Syndrome, Angelman or ASD, or a broader diagnosis such as severe intellectual disability. Note that whereas all children functioned at the level of SPID, not all of them (yet) received an official, established diagnosis of intellectual disability (e.g., because intellectual testing was not possible due to age or complex disabilities)

effect on the children's emotions as identified by parents). These stressors were randomly provided by either the parent or a stranger (which was the unfamiliar researcher in the current study). During each stressor, parents observed their children and indicated the peak in their children's stress level (based on their previous experiences). Immediately after parents indicated the stress peak, comfort was provided to the children. The person providing the comfort was as well

Table 2 The protocol of the structured observation

	Situation A	Situation B
Phase 1	Baseline: Low-level interaction (min. 5 min)	Baseline: Low-level interaction (min. 5 min)
Phase 2	Researcher confronts the child with a stressor (e.g., wiping nose, washing face…)	Parent confronts child with a stressor (e.g., wiping nose, washing face…)
Phase 3	Parent indicates the peak in child's arousal level (parent keeps distance)	Parent indicates the peak in child's arousal level (researcher keeps distance)
Phase 4	Parent comforts the child & researcher keeps distance to control for social referencing (min. 5 min)	Researcher comforts the child & parent keeps distance to control for social referencing (min. 5 min)
Phase 5	Break (min. 5 min)	Break (min. 5 min)

Note. Copied with little adaptations from Vandesande et al. (2020)

randomly determined. The stranger adapted her own style of comforting as much as possible to resemble the comfort of parents. While in 'situation A' the stranger applied the stressor and parents comforted children, in 'situation B' the roles interchanged. In total, 'situation A' and 'situation B' each occurred two times in random order (e.g., AABB, ABAB, BABA), separated from each other by five-minute breaks. The entire home-based experimental observation was video-recorded with two hand-held cameras on tripods, to the extent possible recording from different angles. Psychophysiological arousal (skin conductance) was measured continuously with the sensor sock, to which the children got familiarised at the start of the home visit (Sterkenburg et al., 2017). These data were however not included in the current study. In the study of Vandesande et al. (2020) an intraindividual (within-child) perspective was adopted, so that comparing children's own skin conductance levels across conditions (situation A and B) was meaningful. However, in the current study, interindividual (between-child) differences are explored. Comparing fluctuations in skin conductance levels across children is not meaningful, because physiologically speaking, each child can have a different range wherein (s)he can potentially fluctuate (Dawson et al., 2007).

Measures and Instruments

Background Questionnaires

Information on various background variables was provided by parents by means of a general background questionnaire, constructed by the researchers. This questionnaire included questions on the child's characteristics (e.g., birth date, developmental age, medical problems) and the family characteristics or living situation of the child (e.g., siblings, parity, parents' employment). The Communication and Symbolic Behaviors Scales (CSBS; Wetherby & Prizant, 2002) was filled out to assess the children's communicative abilities and the Tandem-list (in Dutch: Tandemlijst) provided information on the children's general cognitive development (Stadeus et al., 1994). The CSBS and Tandem-list were solely used to estimate whether children met the inclusion criteria for the current study and did not offer a fine-grained estimate of the children's cognitive development.

Emotional Behaviour (Arousal and Valence)

Children's emotional behaviour (arousal and valence) was assessed using the self-developed coding scheme for behaviour observation of arousal and valence of emotions (Sterkenburg et al., 2017, unpublished work). Conform the definition of arousal by Pfaff et al. (2008), arousal was defined as the overall amount of positive and negative emotions children experience. This amount was judged by an observer on a six-point scale, ranging from one (*very low arousal*, i.e., being passive, no response, drowsy, asleep or absorbed) to six (*very high arousal*, i.e., being highly aggravated, no control over behaviour, yelling, being aggressive). In addition, the valence of emotions was scored as neutral, positive (e.g., delighted) or negative (e.g., frustrated) on a 13-point scale ranging from −6 (*very high negativity*, i.e., extremely frustrated, raging, no control, crying out) to +6 (*very high positivity*, i.e., being excited, cannot control enthusiasm). Coding on these two subscales was continuously done by the first author within each phase of the home-based experimental paradigm. All registered codes (accurate to the millisecond) were saved in the software program 'Noldus Observer XT 7.0'. Two independent coders (master degree students) double-coded more than 30% of the video recordings (12 video's). Interrater reliability was substantial for the current study with a linear weighted kappa of .61 and .65 on arousal, and of .61 and .64 on valence between the first author and the first and second independent double coder, respectively (Cohen, 1968).

Parental Sensitivity

Parental sensitivity was coded for each parent-child dyad using the video-recordings of the 20-min free (play) interaction that ended the home visit. To install similar circumstances for all families, parents received a standardised box of toys (e.g., cracker booklet, music box, hand puppet). These were adapted to the level of functioning of most children and included a variety of toys stimulating various senses. They were instructed to interact or play freely with their child, as they normally would (either using the researchers' toys or their own toys/materials when they felt these were more suitable). The whole interaction was video-recorded using two hand-held cameras on tripods from different angles (if possible, one orienting at the parent and one orienting at the child).

Parental sensitivity was coded using the Emotional Availability Scales 4th Edition (EAS; Biringen, 2008), in which the first author was trained and certified as reliable. Emotional Availability refers to an integration of the attachment conception of sensitivity with emotional perspectives. The EAS aims to measure the quality of parent-child interactions and their levels of emotional connection, by rating four adult dimensions and two child dimensions. For the current study, only the first adult dimension of the EAS, '*Adult Sensitivity'*, was used. This dimension reflects the ability of the parent to be warm and emotionally connected to the child, as well as to respond in an accurate, prompt and efficient way to communication of the child. Adult sensitivity is scored globally on a seven-point scale ranging from 1 (*highly insensitive*) to 7 (*highly sensitive*), and on seven components of sensitivity. These components include (1) *Affect* (seven-point scale), (2) *Clarity of perceptions* and appropriate adult responsiveness (seven-point scale), (3) Awareness of *timing* (three-point scale), (4) *Flexibility, variety and creativity* (three-point scale), (5) Adult *acceptance* (three-point scale), (6) Amount of *interaction* (three-point scale), and (7) *Conflict* situations (three-point scale). As was described in Biringen et al. (2005), the EAS can be applied in the target group of children with (intellectual) disabilities by building some leniency into the system so that parents are not fully downgraded because of the children's disability. For example, normally the dimension of sensitivity is scored dyadically. This implies that parents will score lower on this subscale in case their children do not reciprocate their bids. However, for children with disabilities (which possibly limit their responsiveness to parental overtures) the parents' skills to overcome these impairments are taken into account and the system allows more non-dyadic scoring. For parents of children with disabilities, the use of mid-points (.5) is recommended. Hostyn et al. (2011) used the EAS in the target group of persons with PIMD and showed substantial inter-rater agreement ($\kappa = .72$).

For the current study, inter-rater reliability was established after an intensive training course by the first author of the second coder. The latter obtained a doctoral degree in Educational Sciences and has extensive experience in coding parent-child interactions and parental sensitivity. The training process consisted of an elaborate discussion of the EAS manual (with a focus on the adult sensitivity dimension), providing clarifications in the manual for its application on the target group of children with SPID and discussing four video recordings that were independently coded as an exercise until sufficient inter-rater agreement was reached. After the training phase, 20 video recordings out of 38 parent-child dyads (53%) were randomly chosen to be double-coded by two observers. For each parent-child dyad a background information vignette was constructed by the first author including information on relevant child characteristics (e.g., information on the diagnosis). The linear weighted kappa (Cohen, 1968) was substantial for the global score ($\kappa = .74$), with 95% of the scores within one-point and 80% within half a point, and was moderate to perfect for the subscale dimensions (κ ranging from .56 to 1.00).

Motor Competence

Motor functioning was assessed in the current study using the Gross Motor Function Classification System – Expanded and Revised (GMFCS- E&R; Palisano et al., 2007) for gross motor functioning, and using the (Mini-)Manual Ability Classification System; (Mini-)MACS; Eliasson et al., 2017; Eliasson et al., 2006) for fine motor skills. The GMFCS-E&R is a standardized 5-level classification system, originally developed for children (in different age bands) with cerebral palsy (CP) and widely used in children with severe motor impairments. The GMFCS levels reflect meaningful differences in the gross motor functioning (e.g., sitting and walking) of children, ranging from I (mild impairment) to V (severe impairment). Whereas level I, II and III indicate the ability to walk (with or without a hand-held mobility device), level IV and V refer to very limited mobility of children. In the current study, parents chose the level which reflected their children's abilities in daily life the most. Wood and Rosenbaum (2000) described the instrument's good reliability (especially above an age of 2 years) and predictive value over time ($r = .79$) when rated by a professional caretaker. The studies of Morris et al. (2004 and 2006) indicated that parents' ratings of the GMFCS were highly consistent with those of professionals (ICC ranging from .87 to .94) for the age band of 6 to 12 years. The MACS (for children >4 years) and Mini-MACS (for children <4 years) was similarly assessed by parents to estimate the children's current fine motor skills. In particular, the MACS is used to assess the typical manual performance (instead of their maximal capacity) of both hands when handling objects in daily life. Whereas children in level I handle objects without any problem, children in level V do not handle objects and require full assistance. Eliasson et al. (2006) described that the MACS was based on a valid construct (defined and

Table 3 Meaning of the differentiation measure of arousal and valence, based on the mean

Arousal	Valence
< 0: larger decrease of arousal during comfort compared to stressor phase when the parent provides comfort compared to the stranger (or a smaller increase of arousal when the parent comforts in case there was a general increase of arousal during comfort compared to stressor phase)	**< 0:** Smaller increase in valence during comfort compared to stressor phase when the parent provides comfort compared to the stranger (or a larger decrease of valence during comfort compared to stressor in case there was a general decrease of valence during comfort compared to stressor phase)
= 0: No difference between the changes in arousal during comfort compared to stressor phase when the parent or the stranger provides comfort	**= 0:** No difference between the changes in valence during comfort compared to stressor phase when the parent or the stranger provides comfort
> 0: smaller decrease of arousal during comfort compared to stressor phase when the parent provides comfort compared to the stranger (or a larger increase of arousal when the parent comforts in case there was a general increase of arousal during comfort compared to stressor phase)	**> 0:** Larger increase in valence during comfort compared to stressor phase when the parent provides comfort compared to the stranger (or a smaller decrease of valence during comfort compared to stressor in case there was a general decrease of valence during comfort compared to stressor phase)

discussed until consensus was reached by an expert group) and reported excellent inter-rater agreement, both between parents and therapists (ICC = .96) and between therapists (ICC = .97). This was also demonstrated by Eliasson et al. (2017) for the Mini-MACS (under the age of four years), with an ICC between parents and therapists of .90 and between therapists of .97.

Data Preparation and Analysis

Data Preparation

First, the data of the video scoring was prepared before conducting statistical analyses. Both micro-coded behavioural measures (arousal and valence) were reduced to obtain one-second intervals and were synchronised to each other. For each of the four stressors per child, four time windows were included: (1) a baseline of three minutes before the start of the stressor (e.g., StressorA1_pre), (2) the enacting of the stressor itself (e.g., StressorA1), (3) the window between the end of the stressor and the start of comfort (e.g., StressorA1_between), and (4) three minutes of comfort (e.g., StressorA1_comfort). These four time periods were coded for all four cycles, so in total, 16 time windows were micro-coded with the coding scheme for behaviour observation of arousal and valence (Sterkenburg et al., 2017, unpublished work). In the current study, only the behavioural data for the stressor (phase 2) and the comfort phase (phase 4) were included.

Second, data from seven out of the total of 152 stressors (4 stressors × 38 children) were excluded from all analyses due to problems with the procedure (e.g., the child was partially comforted by the parent, whereas it was the stranger's turn) or with the video recordings (e.g., there was inadequate visibility of the stressor). The remaining 145 stressors across 38 children were included in behavioural analyses on valence and arousal.

Third, for the included data, a differentiation measure was calculated as an outcome variable (separately for behavioural arousal and valence) to express the extent to which children differentiated between the parents' and the stranger's comfort. The measure is based on subtracting behavioural arousal and valence during the comfort phase from arousal and valence during the stressor phase. In order to subtract these values the means for each phase of the observation were used. Both repetitions of the 'situation A' and the 'situation B' were then averaged to obtain one differentiation score for A (i.e., parent provides comfort) and one for B (i.e., stranger provides comfort). The final differentiation measures of arousal and of valence were separately calculated (based on the mean), by subtracting the average differentiation measure of the 'situation B' (i.e., stranger provides comfort) from 'situation A' (i.e., parent provides comfort). The final differentiation measures provide a straightforward, intuitive way to gain insight into the (behavioural) difference in responses to comfort by the parent versus the stranger. The meaning of the final differentiation measure for arousal and for valence is described in Table 3. The closer the value of the differentiation measure is to zero, the less difference there was between the children's reactions to the parents' or the stranger's comfort. The sign of the differentiation measure determines the meaning (see Table 3).

Statistical Analyses

First, preliminary analyses were conducted to validate the differentiation measures of arousal and valence, i.e. to ensure that they reflected the research data well, despite the summarising nature of the measures. Therefore, descriptives (*M*, *SD*, *range*) were calculated for both differentiation measures, as well as a one sample t-test with test value zero.

The differentiation measures were assumed to be a valuable summary of the research data, when they were in line with the study's results of Vandesande et al. (2020) in which all data points in one-second intervals were included.[3]

Second, concerning the first and second research question, the differentiation measures of arousal and valence were, respectively, regressed on parental sensitivity and (gross and fine) motor functioning, respectively, using simple linear regressions. Third, concerning the third research question, multiple linear regressions were performed to associate the differentiation measures of arousal and valence, respectively, with both the global score of parental sensitivity and motor competence (fine and gross motor competence separately). In addition, scatter plots were created to visualise the association between parental sensitivity and the differentiation measures, depicting the participants by their level of motor competence. The assumptions of linearity, normality, and homoscedasticity were checked by scatter plots, normal Predicted Probability plots, and plotting the predicted values and the residuals, respectively. All assumptions were met for the described simple and multiple linear regressions.

Fourth, post-hoc analyses were performed in relation to the third research question. These post-hoc analyses were driven by the aforementioned multiple linear regressions and visual inspection of the scatterplot, to compare two subsets of participants: those having PIMD ($n=15$) and those definitely having no PIMD ($n=19$). Four children were excluded as they did not (fully) met the criteria of PIMD by Nakken and Vlaskamp (2007) (or because information was lacking to confirm the criteria; see Participants). All above-mentioned analyses (in step 1, 2 and 3) were conducted for both subgroups separately by way of exploratory post-hoc analyses. In addition, moderation analyses (model 1) using PROCESS macro by Hayes (2013) for SPSS were carried out to predict the differentiation measures using parental sensitivity as predictor and with the dummy variable 'having PIMD' as moderator (with value 1 being defined as having PIMD).

For all analyses, effects were judged as significant when the probability level was below .05 and the corresponding effect sizes were reported. In addition, bootstrap confidence intervals (using 1000 samples and the bias corrected accelerated method) were reported to provide a more robust estimation of the effect. Bootstrap is a statistical technique, often used in studies with small sample sizes (Adèr & Adèr, 2008) and/or when the normality assumption is (possibly) violated in the data (Wood, 2004), to estimate the sampling distribution by taking repeated samples from the existing data set with replacement (Field, 2009). A sensitivity power analysis using G*Power 3.1 (Faul et al., 2007) with a specified power of 0.8 showed a minimal detectable effect ρ of 0.44 in this study for simple linear regressions and f^2 of 0.22 for multiple regressions. This implies that only large effects and some medium effects could be reliably detected given the current sample size (following Cohen's (1988) effect size benchmarks of ρ is 0.10 for small, 0.30 for medium and 0.50 for large effects, and of f^2 is 0.02 for small, 0.15 for medium and 0.35 for large effects). For all analyses, SPSS software package (version 26.0) was used.

[3] In the study of Vandesande et al. (2020) statistical analyses were conducted that took into account all research data in one-second intervals, instead of collapsing/summarizing the research data per observation period. Therefore, before doing statistical analyses for the current study using the differentiation measures, the authors made sure that the differentiation measures were a good reflection of the research data.

Results

Preliminary Results

In line with Vandesande et al. (2020), the differentiation measures overall reflected a larger decrease of arousal ($M=-0.20$, $SD=0.65$, $Min=-1.95$, $Max=0.64$) and a larger increase of valence ($M=0.81$, $SD=1.88$, $Min=-2.72$, $Max=5.06$) when the parent provided comfort compared to the stranger (situation A). A one sample t-test was carried out to check whether the differentiation measures diverged significantly from test value zero. Test results were marginally significant for arousal, $t(37)=-1.93$, $p=.06$, 95% CI [−0.40, −0.03], $d=0.31$, and statistically significant at the .05 level for valence, $t(37)=2.67$, $p=.01$, 95% CI [0.26, 1.43], $d=0.43$.

The Link between the Interindividual Differences in Selective Responses and Parental Sensitivity

(Research Question 1)

Parental sensitivity was globally scored on average 5.25 ($SD=1.43$) on the seven-point scale of the EAS (Biringen, 2008), ranging from 1.50 to 7.00. The differentiation measures of arousal and valence, respectively, were regressed on parental sensitivity. Sensitivity was not significantly associated with the differentiation measure of arousal, $F(1,36)=0.25$, $p=.62$, 95% CI [−0.16, 0.09], $R^2=.01$, nor valence, $F(1,36)=0.32$, $p=.58$, 95% CI [−0.31, 0.58], $R^2=.01$.

The Link between the Interindividual Differences in Selective Responses and Motor Competence

(Research Question 2)

Based on simple linear regressions, children's gross motor functioning (GMFCS level) was not significantly associated with the differentiation measure of arousal, $F(1,36)=0.36$, $p=.56$, 95% CI [−0.09, 0.17], $R^2=.01$, nor valence, $F(1,36)=1.82$, $p=.19$, 95% CI [−0.60, 0.08], $R^2=.05$. Higher fine motor skills (corresponding with low levels of the MACS, see 2.3.4) were significantly associated with differential arousal response to comfort provided by parents compared to comfort provided by the stranger (i.e., arousal decreased more during the parents' comfort after distress compared to the stranger's comfort), $F(1,36)=4.19$, $p<.05$, 95% CI [−0.02, 0.37], $R^2=.10$. The differentiation measure of valence was not significantly associated with children's fine motor skills, $F(1,36)=0.63$, $p=.43$, 95% CI [−0.81, 0.54], $R^2=.02$.

The Link between the Individual Differences in Selective Responses, Parental Sensitivity and Motor Competence

(Research Question 3)

Parental sensitivity and gross motor competence were not significantly associated with the differentiation measure of arousal in a multiple regression, $F(2,35)=0.41$, $p=.66$, $R^2=.02$ (with 95% CI [−0.21,0.09] and [−0.08, 0.18] for sensitivity and gross motor competence, respectively, as predictors) nor with valence, $F(2,35)=1.41$, $p=.26$, $R^2=.08$ (with 95% CI [−0.35, 0.74] and [−0.74, 0.07] for sensitivity and gross motor competence, respectively, as predictors).

Parental sensitivity and fine motor competence were significantly associated with the differentiation measure of arousal, $F(2,35)=3.78$, $p=.03$ $R^2=.18$ (with 95% CI [−0.29, 0.03] and [0.05, 0.49] for sensitivity and fine motor competence, respectively, as predictors), but not with valence, $F(2,35)=0.90$, $p=.42$, $R^2=.05$ (with 95% CI [−0.27, 0.71] and [−1.04, 0.59] for sensitivity and fine motor competence, respectively, as predictors). Figure 1 displays a grouped scatter plot for the association between parental sensitivity and the differentiation measure of arousal, organised by the level of fine motor competence with a reference line at value zero for the differentiation measure (full line). Visual inspection of the scatter plot revealed that the children with the highest MACS level (which reflected the poorest fine motor skills), were generally located in the upper right quadrant.

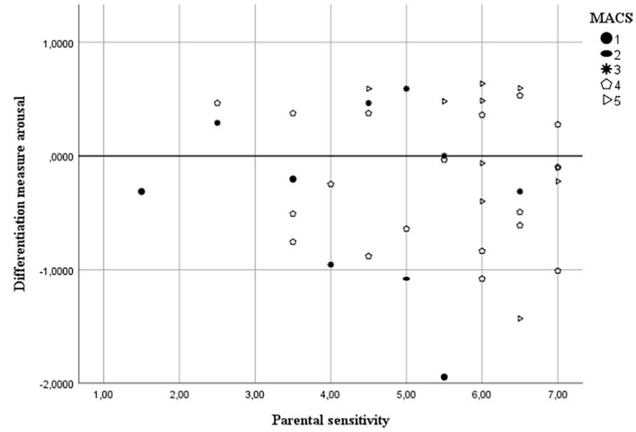

Fig. 1 Grouped scatter plot for the association between parental sensitivity and the differentiation measure of arousal, organised by level of fine motor competence. *Note*. Reference line at differentiation measure of arousal value zero. Fine motor skills are most limited with rising MACS levels

Post-hoc Analyses Comparing Children with and without PIMD

(Research Question 3)

Both the multiple linear regression and visual inspection of the scatter plot, pointed towards (fine) motor skills as a possible moderator of the association between parental sensitivity and the differentiation measure of arousal. Post-hoc visual inspection of the data set learned that the children with the highest MACS levels were often children who could be classified as having PIMD (according to the definition of Nakken & Vlaskamp, 2007). Therefore, post-hoc analyses further explored the differentiation measures for two subsets of participants: children having PIMD ($n=15$) and those definitely having no PIMD ($n=19$). This is a way of operationalising the children's motor competence in an alternative manner (since children with PIMD have a significant motor impairment in addition to the profound intellectual disability). It appeared that children with PIMD had a differentiation measure of arousal ($M=-0.15$; $SD=0.66$) that was not significantly diverging from zero, $t(14)=-0.91$, $p=.38$, and as well not diverging from zero for valence ($M=0.51$, $SD=1{,}43$), $t(14)=1.39$, $p=.19$. However, for the children who did not have a profound motor impairment (without PIMD), both the differentiation measure of arousal, $t(18)=-2.78$, $p=.01$, and of valence, $t(18)=2.32$, $p=.03$, diverged significantly from the test value zero in a one sample t-test. Conducting the simple and multiple linear regressions separately for both subsets of children did not show significant effects, except for the positive association between the differentiation measure of valence and the global score of parental sensitivity for children with PIMD ($r=.54$), $F(1,13)=5.36$, $p=.04$, $R^2=.29$. Moderation analysis showed no significant interaction effect between parental sensitivity and the dummy variable PIMD as moderator (model 1), $F(1,30)=0.01$, $p=.91$.

Discussion

The current study explored the links between interindividual differences in expressed selective attachment during comfort among children with severe or profound intellectual disabilities (SPID) and associated factors. The results indicated that interindividual differences were significantly associated with children's fine motor skills, but not with their gross motor competence and parental sensitivity as such. Children with the highest fine motor skills showed more signs of differentiation between the parents' and the stranger's comfort as was reflected in their general behavioural arousal levels, in the way that their arousal levels decreased more during parents' comfort. In the current study, parental sensitivity was not significantly associated with interindividual differences in the selective behavioural reactions of children with SPID, except for the statistical model that also included the children's level of fine motor skills. The grouped scatter plot indicated that, although their parents were highly sensitive, children with the lowest fine motor skills generally differentiated less between the parent and the stranger (i.e., the differentiation measure of arousal was situated around value zero) or that their differentiation measure was slightly positive (i.e., that the decrease of arousal was greater during the stranger's comfort). In the current sample, children with the poorest fine motor skills, could often be classified as children having profound intellectual and multiple disabilities (PIMD), according to the definition of Nakken and Vlaskamp (2007) and Maes et al. (2020). Exploratory post-hoc analyses showed that children with PIMD did not significantly differentiate between the parents' and the stranger's comfort when the general arousal level and the valence of their emotions is taken into account. These results are in line with the behavioural observation study of Vandesande et al. (2019), in which children with comorbid disabilities (in addition to the cognitive delay) generally seemed to differentiate less with regard to observed attachment behaviour.

The lack of significant associations between differential child responses to their parents, which are their putative attachment figures, and the sensitivity of their attachment figures requires further discussion. On the one hand, a possible explanation for the non-significant results is that the power of our statistical tests, given the relatively small sample size (which is common in research with this target group, Maes et al., 2020), is too limited to detect small or even some medium effects (following the conventional benchmarks of Cohen, 1988). Moreover, meta-analyses from research in the attachment-field showed that effect sizes are often even much smaller in attachment research and, thus, that field-specific benchmarks should be lower than the conventional benchmarks (e.g., $r = 0.10$ for small, $r = 0.20$ for medium and $r = 0.30$ for large effects; Schuengel et al., 2020). Given these considerations, the relatively small sample size might have resulted in insufficient power to detect these effects (especially in the multiple regression and moderator analyses; Field, 2009). It is, however, interesting to note that - although no statistical significance was found – there were few cases who differentiated in favour of their parent during comfort (i.e., who showed a greater decrease of arousal during comfort by the parent compared to comfort by the stranger) while their parent scored low on sensitivity (see Fig. 1). In any case, the current study confirmed that there were no large effects of parental sensitivity. Future research on this particular target group would benefit from (international) multi-centre research, in which participant samples can be pooled (Maes et al., 2020).

On the other hand, in retrospect, it is not that surprising that no significant associations were found between interindividual differences in selective attachment and parental sensitivity, because the question of selective attachment is a different one than the question of attachment quality (Ainsworth et al., 1978; Marvin et al., 2016). Rajecki et al. (1978), for instance, reported the development of strong, consolidated attachment bonds in children who were maltreated. This implies that children, who are most likely insecurely attached, also show selective, discriminate attachment behaviours. The ability to attach is indeed biological, embedded in the genetic make-up of children (Bosmans et al., 2020). The current study confirmed that differences in the quality of care are not related to differences in selective attachment, as they are related to differences in quality of attachment (Atkinson et al., 1999; Behrens et al., 2011; De Wolff & Van IJzendoorn, 1997; Zeegers et al., 2017). The differentiation measures based on children's selective emotional responses were used in the current study as an attempt to overcome the limited informativeness of the conventional indicators of attachment quality for children with SPID (e.g., due to their possible motor impairment obscuring the expression of attachment behaviours). In that way, the differentiation measures functioned as a proxy for selective attachment development. Despite the fact that these measures inform on individual differences in one of the most basal aspects of attachment development (*To what extent and how does a child differentiate between attachment figures and others?*), the search continues for more behavioural indicators of attachment quality which may be predicted by parents' sensitive care. In the pursuit of this theoretical understanding of attachment in the group of persons with SPID/PIMD, finding the balance between applying general theories versus developing specific theoretical models will be crucial (Maes et al., 2020).

The study's results did point towards a possible important role of fine motor skills, more so than the children's gross motor skills. One the one hand, this might be explained by the design of the protocol. To ensure that the interaction was going on inside the range of the video cameras on tripods, parents were instructed to keep their children as much as possible in a delineated space. For that reason, a lot of parents organised the situation in a way that children were limited in their freedom of movement (e.g., by placing them in their wheelchair or in their dining chair). Possibly, the limited freedom of movement would have restricted children in their ways to show attachment behaviours for which gross motor functioning is relevant (e.g., crawling towards their mother). On the other hand, children's fine motor skills (but not gross motor skills) might also be a proxy for children's cognitive development (Martzog et al., 2019). Developing attachment indeed requires certain basic cognitive conditions (such as object/person permanence; Bell, 1970). Differences in these cognitive conditions, such as differences in cognitive information processing, can lead to differences in the quality of their emotional responses (as was shown for children with Down Syndrome by Thompson et al., 1985).

Exploratory post-hoc analyses examined the subgroup of children with PIMD, who – on a group level - had the most limited fine motor skills and also the profoundest cognitive disability in the current sample. Although the post-hoc linear regressions and moderation analyses were not statistically significant (presumably also due to limited power; Field, 2009), results of the post-hoc analyses showed that children with PIMD might be a special group within the larger group of children with SPID when it comes to attachment-related interindividual differences. Visual inspection of the scatter plot (Fig. 1) pointed towards a possible differential susceptibility effect (Fearon & Belsky, 2016). This means that there were possibly not only interindividual differences in the amount of differentiation but also in the extent to which sensitivity could have an impact thereon. Although the parents of children with PIMD were generally scored as highly sensitive in this study, their children differentiated less in emotional behaviour during comfort.

The current study took a step forward in the road ahead of us to broaden the scientific knowledge of individual differences – and eventually attachment quality - in the target group of children with SPID, by elucidating interindividual differences in selective attachment and their associated factors. The findings of the current study imply that a full understanding of the interplay of parenting and developmental factors to grasp differences in attachment, is not yet achieved for the particular group of children with SPID. This means that one cannot naturally draw on knowledge gained from attachment research in children without or with less severe disabilities when supporting or advising parents of these children on the domain of attachment. Future research on this topic would, for example, benefit from elucidating other potential associating factors (e.g., visual impairment and epilepsy) that may play a role in explaining interindividual attachment-related differences in children with SPID. Furthermore, creating a reliable and valid individual image of children's cognitive functions, that are known to be relevant for attachment development (such as person permanence, understanding cause-effect and intersubjectivity), is a valuable future research path in that respect.

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Availability of Data and Material The datasets generated during and/or analysed during the current study are not publicly available due to privacy reasons but are available from the corresponding author on reasonable request.

Authors' Contributions All authors contributed to the study conception and design. Material preparation, data collection and analysis were performed by Sien Vandesande and (for a specific part) Ines Van keer. The first draft of the manuscript was written by Sien Vandesande and all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

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Declarations

Conflict of Interest The authors have no conflicts of interest to declare that are relevant to the content of this article.

Code Availability Not applicable.

Ethics Approval The Social and Societal Ethics Committee (KU Leuven, Belgium) ethically approved the research design and protocol (G-201612708).

This study met the ethical procedures and standards of the KU Leuven, Belgium.

Consent to Participate Written informed consent was obtained from the parents.

Consent for Publication Parents signed informed consent regarding publishing their (anonymised) data.

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