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Comfort provided by parents versus strangers after eliciting stress in children with severe or profound intellectual disabilities: does it make a difference?

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ABSTRACT

The relationships between children with severe or profound intellectual disabilities (ID) and their parents may fulfil attachment functions, such as regulating emotional responses to stress. This study examined the extent to which children with severe or profound ID differentiate between their parents and a stranger as a resource for stressregulation. A home-based experimental paradigm was conducted and video-recorded in 38 families. Children (1-8 years) were exposed to four naturalistic stressors followed by comfort, randomly provided by the parents or the stranger. Emotional behaviour (arousal and valence) and the skin conductance level were simultaneously recorded. With regard to both emotional behaviour and skin conductance, children significantly differentiated between their parents as attachment figures and the stranger during stress and comfort, despite their impairments on various developmental domains. Behavioural observation and physiology show complementary manifestations of parent-child attachment in this population.

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KEYWORDS

Parent-child attachment; severe or profound intellectual disability; significant developmental delay; behavioural observation; skin conductance

1. Introduction

One of the tenets of attachment theory is the crucial role of primary caregivers as coregulators of stress in the interest of children's adaptive social-emotional functioning and general well-being (Bowlby, 1969/1982; Cassidy, 1994, 2016; Mikulincer, Shaver, & Pereg, 2003). In children with severe or profound intellectual disabilities (ID), the role of the parents as co-regulators is considered to be even more important due to these children's limited ability to cope autonomously with the large amounts of psychological stress they tend to encounter (Chaney, 1996; Gerstein et al., 2011; Janssen, Schuengel, & Stolk, 2002; Schuengel & Janssen, 2006). Notwithstanding the importance of the parents' regulating function and the theoretical attention that has been devoted to the topic of attachment in this target group (e.g. Howe, 2006; Janssen et al., 2002; Schuengel, De Schipper, Sterkenburg, & Kef, 2013; Schuengel & Janssen, 2006; Schuengel, Kef, Damen, & Worm, 2010), the emotion regulating function of parents as attachment figures for children with severe or profound ID

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has remained an untested theoretical assumption until now. However, empirically testing this assumption is important, because parents of children with severe disabilities may lack the same kind of positive feedback and reinforcement to their sensitive caregiving compared to the feedback that parents of typically developing children receive in interaction with their children. Therefore, the current study examined the extent to which young children with a severe or profound ID differentiate between their parents, of whom we would expect an emotion-regulating function as a primary caregiver, and a stranger.

Attachment theory defines an attachment relationship as an affectional tie between children and specific other persons, which usually develops within the relationship between parents as primary caregivers and their children (Ainsworth & Wittig, 1969; Bowlby, 1969/1982; Cassidy, 2016). This relationship is described to be persistent or life long, emotionally significant, and to provide a context of comfort especially during distress (Ainsworth, 1989; Cassidy, 2016). In an optimal scenario, the parents as attachment figures fulfil the roles of both being a safe haven and a secure base for their children (Cassidy, 2016; Powell, Cooper, Hoffman, & Marvin, 2013). This implies that parents are available for their children to come back to for comfort and that parents support the children's exploratory system as well (Bowlby, 1969/ 1982; Cassidy, 2016; Powell et al., 2013; Waters & Cummings, 2000). Such a secure attachment relationship is promoted by repeated experiences of trust and sensitive, responsive parenting (Ainsworth, Blehar, Waters, & Wall, 2015; Atkinson et al., 1999; Powell et al., 2013). Through experience, children build trust in the availability of their parents: they have confidence that their parents will act as a source of support in times of distress (Ainsworth, 1979). In turn, children's responses to sensitive caregiving act as a reinforcement for parents' future sensitive behaviour (Dix, Gershoff, Meunier, & Miller, 2004). Secure attachment is shown in a specific set of secure attachment behaviours from children (e.g. seeking proximity upon reunion, differential smiling, preference for support; Ainsworth, 1964; Ainsworth et al., 2015). These behaviours are aimed at establishing a dynamic equilibrium between proximity to the parents and exploring the environment (Bowlby, 1969/1982; Cassidy, 2016). The attachment behavioural system organises itself on the basis of interactions with regular caregivers, who become attachment figures. Thus, attachment involves a specific person who is not merely interchangeable (Cassidy, 2016). This gives way to selective attachment behaviours and at the same time wariness towards strangers (Bowlby, 1969/1982). From this perspective, it could be hypothesised that children would prefer to be comforted by their parents, with whom children may have built a specific, comforting relationship, compared to strangers during distress.

However, parents, professionals as well as researchers have less behavioural evidence (compared to typically developing children) on which they may assume that attachment develops and that parents have a special salience in the emotional experience of children with severe or profound ID. The limited behavioural evidence can be explained by these children's characteristics on two developmental domains. First, on the cognitive domain, children with severe or profound ID have a significant life-long cognitive delay (in adults an IQ below 40 is provided as a guideline; American Association on Intellectual and Developmental Disabilities, 2010; American Psychiatric Association, 2013; Kraijer & Plas, 2007). Because attachment reflects not only emotional but also cognitive processes (Bretherton & Munholland, 2016; Cassidy, 2016; De Winter, Bosmans, & Salemink, 2017; De Winter, Salemink, & Bosmans, 2018) and because the (severity of the) ID itself also influences the attachment (Bretherton & Munholland, 2016), it remains to be clarified whether these children have learned from

previous care experiences with their parents in stressful situations and therefore, differentiate between their parents and a stranger. Trust in the care of parents is an expectation of the parents' availability derived from different learning processes (Bowlby, 1969/1982; Cassidy & Shaver, 2016; De Winter et al., 2017). Indeed, when children repeatedly and consistently experience sensitive and prompt caregiving when in distress, they build the expectation that their parents will be accessible in future stressful situations. These specific meanings of care that children can attribute to their parents are organised in an internal working model which is a cognitive-emotional representation, based on children's past experiences (Bowlby, 1969/1982; Bretherton & Munholland, 2016; Cassidy & Shaver, 2016; De Winter et al., 2017). This internal working model guides children through the anticipation of social situations and the interpretation and reaction on their social environment (Bretherton & Munholland, 2016). However, because children with severe or profound ID possibly lack object/person permanence, the ability to differentiate means from ends and/or contingency awareness (Berk, 2018; Blain-Moraes & Chau, 2012; Bruce & Muhammad, 2009; Cassidy, 2016; Dykas & Cassidy, 2011; Sherman, Rice, & Cassidy, 2015), one could wonder whether children have (yet) learned from previous care experiences with their parents as primary caregivers and whether they are able to profit from the storage of these past experiences into mental representations of their parents as a valuable resource of support.

Second, on the domain of behavioural expression and communication, children with severe or profound ID often display attachment-related behaviours in an extremely subtle and idiosyncratic way, which makes it very hard to notice and to correctly interpret (Blacher & Meyers, 1983; De Schipper & Schuengel, 2010; Moran, Pederson, Pettit, & Krupka, 1992; Schuengel & Janssen, 2006; Schuengel et al., 2010; Vandesande, Bosmans, Schuengel, & Maes, 2019a). In addition, these children often have difficulties understanding verbal communication and communicate at a pre- or protosymbolic level (i.e. non-symbolic communication level) (Goldbart, Chadwick, & Buell, 2014; Grove, Bunning, Porter, & Olsson, 1999; Hostyn, Daelman, Janssen, & Maes, 2010; Nakken & Vlaskamp, 2007). This implies that communication occurs through exchange of affective behavioural signals and through idiosyncratic, context bound and mainly bodily expressions (Hostyn & Maes, 2009). These specific characteristics influence the expression of children's attachment behaviour.

In a comparable but slightly higher functioning group of youth with a moderate-to-severe ID, De Schipper and colleagues (De Schipper & Schuengel, 2010; De Schipper, Stolk, & Schuengel, 2006) reported on clear inter-individual differences in differentiated attachment behaviour, which could not be fully explained by children's diagnosis. Sterkenburg's (Sterkenburg, Janssen, & Schuengel, 2008) research demonstrated the ability of youth with profound ID and visual impairments to develop an attachment relationship with their psychotherapist. Blacher (1984) reported on a Strange Situation observation in 50 young children (between the age of three and eight years old) with severe or profound ID. Based on her observations, she concluded that attachment did develop among these children, albeit that responses were less complex, less intense and less differentiated compared to typically developing children. Therefore, she deemed, for example, that it was not possible to classify attachment using the well-known Ainsworth categories (corroborating other researchers, e.g. Cicchetti & Serafica, 1981; Thompson, Cicchetti, Lamb, & Malkin, 1985). Similar reflections were made by Vandesande et al. (2019a) who conducted an exploratory home-based observation in 20 children with severe or profound ID, which was inspired by the Strange Situation Procedure (Ainsworth et al., 2015). Vandesande et al.'s (2019a) procedure contained two

separations from the parent and an interaction with a stranger. In that study, the authors reported more intense and persistent proximity- and contact seeking, contact-maintaining and resistant behaviour towards parents compared to strangers. However, for children with severe or profound ID who additionally had a significant motor impairment and limited mobility, the coding scheme for attachment behaviour was not fine-meshed enough to differentiate subtle behavioural differences (Vandesande et al., 2019a).

Hence, the limited behavioural expression and the lack of clear symbolic, social communication of children with severe disabilities (Goldbart et al., 2014) – or the difficulties parents may experience to notice all attachment-related behaviours, needs and communicative attempts in these children – might impede parents to notice, interpret and react promptly and consistently in a sensitive manner (Atkinson et al., 1999; Moran et al., 1992; Schuengel et al., 2010). In its turn, the reduced opportunities for parents to be sensitive to the children's emotional cues could diminish the amount of learning experiences children have with regard to their parents' social communication and care. In particular, children with severe or profound ID experience difficulties to process social information and to adequately assess and learn from the motives behind their parents' behaviour (Benson, Abbeduto, Short, Nuccio, & Maes, 1993; Schuengel & Janssen, 2006).

Attachment theory leads to the theoretical proposition that parents would have an emotion co-regulating function as attachment figures for their children that strangers would not have. In the current study, the extent to which children with severe or profound ID differentiate between their parents as attachment figures and a stranger, in reaction to stress and comfort, is investigated. Thus, the current study focused on intra-individual (i.e. within-child) processes. Based on general attachment theory (Blacher & Meyers, 1983; Bowlby, 1969/1982; Cassidy, 2016), it was hypothesised that children would prefer proximity to their parents during distress and regulate their stress levels more successfully than in the proximity of a stranger. If children with severe or profound ID would have learned from previous care experiences with their parents, this should show first in their differential responses to comfort by their parent versus a stranger (Ainsworth, 1979; Dujardin et al., 2016).

To study this proposition, a home-based experimental observation was done, combining behavioural observation and physiological measures (similar to Lima, Silva, Amaral, Magalhães, & Sousa, 2011; Vos, De Cock, Petry, Van Den Noortgate, & Maes, 2013). Psychophysiology affords a proximal indicator of emotion regulation processes. It is the scientific discipline that studies psychological phenomena (e.g. emotion, motivation, alertness) by unravelling physiological (autonomous) changes in the body as related to the physical and social environment in which one is located (Cacioppo, Tassinary, & Berntson, 2007; Dawson, 1990). The current study was limited to the measurement of electrodermal activity (EDA). EDA is an overarching concept to indicate all autonomic, physiological changes in the electrical properties of a person's skin (Braithwaite, Watson, Jones, & Rowe, 2015). Throughout history, EDA is a much-used process because of its value to be an objective indicator of (implicit or explicit) emotional states (Braithwaite et al., 2015; Dawson, Schell, & Filion, 2007). Given the fact that children from this population have restricted behavioural repertoires to communicate (Goldbart et al., 2014; Nakken & Vlaskamp, 2007), psychophysiology can provide parents with valuable insights into their role as co-regulators of emotions, which indirectly contributes to the children's psychological well-being (Cassidy, 2016; Shaver, Mikulincer, Gross, Stern, & Cassidy, 2016; Thompson, 2016).

2. Method

2.1. Participants

To recruit participants for the current study, care organisations (e.g. specialised day care centres and home support services) sent flyers and invitational letters to parents whose children met the following inclusion criteria: (1) The children's chronological age ranged from one to eight years; (2) the children had a severe or profound intellectual disability (ID) or functioned at that level according to their professional caregivers in case an official diagnosis was lacking; and (3) the children primarily lived at home at the time of the participation (i.e. children sleep at home for a minimum of four nights a week on average). To respect the families' privacy, professional caregivers always functioned as intermediates in the participant recruitment. Therefore, they provided a first check of the inclusion criteria to the researchers. In this study, children who were adopted or were living in foster families were excluded to rule out any deviations in attachment development due to these specific life circumstances (Carlson, Hostinar, Mliner, & Gunnar, 2014). In addition, children with open wounds on their foot sole at the time of participation were excluded due to hygienic reasons in relation to the use of the sensor sock (see 2.3.3). Other medical problems (e.g. epilepsy) or additional disabilities apart from the cognitive delay (e.g. sensory or motor impairments) were no reason for exclusion.

After several contact rounds by e-mail and by phone, a total of 49 families (contacted by 25 care organisations) were interested to participate in the current study. Of that number, one family dropped out before the home visit and two children were excluded later on because they did not tolerate the sensor sock. Another eight children were excluded because their cognitive delay with respect to their chronological age and/or their clinical phenotype was not associated with the description of a severe or profound ID. This judgement was based on previous (mainly cognitive) test results from professional organisations and clinical observation of at least three researchers, combined with the results of the Tandemlijst (Stadeus, Windey, Vermeir, & Van Driessche, 1994). With regard to the latter instrument, children were excluded from this study when their developmental age exceeded half of their chronological age.¹

A sample of 38 children participated in this study (13 girls, 34.2%; 25 boys, 65.8%) aged $2y_2m$ (27 months) to $8y_7m$ (103 months; M = 72.07, SD = 20.40). Recent data on intellectual functioning (i.e. less than one year before participation) were only available for 15 children and showed a mean developmental age of 9.60 months (SD = 4.85).² Compared to their chronological age, this resulted in an average cognitive delay of 50.53 months (SD = 21.33). Results of the Tandemlijst showed that most children functioned below two years of age (see Table 1). Most of the children had one or more diagnosis (n = 33, 86.8%) and for the majority of children the aetiology of the disability was known (n = 25, 65.8%). Additional motor impairments were frequently reported (n = 34, 89.5%), especially hypotonia (n = 24, 63.2%) or a general delay in motor development (n = 13, 34.2%). A limited (independent) mobility was reported by parents for half of the children (n = 19), as well as limited fine motor skills in 28 children (73.7%).³ Almost a quarter of the children were visually impaired (n = 6; 15.8%) or blind (n = 2; 5.3%), and one child was deaf (2.6%). More than half of the children struggled with medical problems (n = 22, 57.9%) of various kinds (e.g. epilepsy, reflux). In general, the children's communicative abilities were low, but there were a lot of individual

	Less than 50% of the items achieved	More than 50% of the items achieved	More than 90% of the items achieved		
Category A (0–4 months)	4 (10.53%)	22 (57.89%)	12 (31.58%)		
Category B (4 months- 1 year)	17 (44.74%)	18 (47.37%)	3 (7.89%)		
Category C (1-2 years)	32 (84.21%)	6 (15.79%)	0 (0%)		
Category D-F (2–5 years)	38 (100%)	0 (0%)	0 (0%)		

Table 1. Descriptive results of the Tandemlijst (Stadeus et al., 1994), n (%) per category.

differences on that domain (see Table 2). All children attended specialised day care or special education schools; most of them even each weekday (n = 31, 81.6%). All children (n = 38) cohabited with their biological parent(s) and had the Belgian nationality. With regard to siblings, almost half of the children had one or more brother(s) (n = 19, 50%) and one or more sister(s) (n = 17, 44.7%). Almost half of the children were their mothers' firstborn (n = 18, 47.4%). Around 45% of the mothers worked full-time (n = 17), compared to almost 85% of the fathers (n = 32). Mothers had a mean age of 37.09 years (SD = 4.95), slightly below the mean age of the fathers (M = 39.68; SD = 3.93).

2.2. Design and procedure

The first author conducted a two-hour home visit in each family. After the parents received a detailed explanation of the study's aims and signed the informed consent form, the sensor sock was put on (see 2.3.3). In doing so, children grew accustomed to the sock, which is an instrument to measure the skin conductance level, and a correct baseline of the children's arousal level could be estimated. Next, the general background guestionnaires which were already sent to parents and were completed before the start of the home visit, were discussed to avoid missing data. Subsequently, parents were asked to identify four stressors to which their children typically respond with high negative intensity and which could be used during the home visit. A home-based experimental observation was conducted during which children were confronted with four stressors and four instances of comfort by the parent or by the stranger. In a play episode to end the home visit, parents played freely with their children for 20 minutes using a standardised box of toys. Both the observation and the play episode were video-recorded from two perspectives. However, the play interaction will not be discussed in the current paper, because associating children's reaction patterns to parenting behaviours is beyond the scope of this paper. The Social and Societal Ethics Committee (SMEC, KU Leuven, Belgium) granted ethical approval for the protocol (G- 2016 12 708).

 Table 2. Descriptive statistics of communication and symbolic behavior scales (Wetherby & Prizant, 2002).

	Min	Max	Potential Max	М	SD
Social composite ^a	4.00	43.00	48	21.20	10.65
Speech composite b	1.00	39.00	40	8.00	9.20
Symbolic composite ^c	0.00	48.00	51	11.30	11.27
Total composite	5.00	126.00	139	40.20	29.18

^aEmotion and gaze, communication and gestures ^b Sounds and words ^c Understanding and object use

2.3. Measures and instruments

2.3.1. Background questionnaire

Parents completed a general questionnaire on background variables (constructed by the researchers), both regarding the family characteristics (e.g. siblings, parents' employment), the children's characteristics (e.g. developmental age, medical problems) and the children's daytime activities. The other questionnaires concerned the children's communicative abilities (Communication and Symbolic Behaviors Scales; CSBS; Wetherby & Prizant, 2002), motor functioning (Mini-Manual Ability Classification System; MACS; Eliasson, Ullenhag, Wahlström, & Krumlinde-Sundholm, 2017; Eliasson et al., 2006, and Gross Motor Function Classification System; GMFCS; Palisano et al., 1997) and general cognitive development (Tandemlijst; Stadeus et al., 1994).

2.3.2. Home-based experimental paradigm with stress and comfort

During the home-based experimental paradigm (see Table 3), we confronted children with four stressors and instances of comfort, randomly provided by the parent or a stranger. For all four dyads of stressor and comfort, the following issues were assessed: (1) the effect of the stressor and comfort on children's arousal and valence; (2) the differential person effect (i.e. whether the parent or the stranger performed the action); (3) the differential order effect of the stressor (i.e. whether it was the first, second, third or fourth stressor) and (4) the differential effect of immediate (1–15 seconds) or delayed (15–30 and 30–180 seconds) comfort.

Children were confronted with four naturalistic stressors, which were identified by parents to have an intense negative effect on the children's emotions. In the current study four different stressors were included to avoid habituation and adaptation effects (Frederiks, Croes, Chen, Bambang Oetomo, & Sterkenburg, 2015a), unless parents were not able to identify four different stressors during the home visit. Parents were not restricted with regard to the nature of the stressor, because this seemed highly child specific (similar to the procedure of Vos et al., 2013). Thus, stressors could include both temporarily depriving children from something they regard as positive (e.g. taking the pacifier away) or exposing them to something they regard as negative (e.g. brushing teeth). To determine the length of the stressor, parents indicated when the peak in the children's stress level was reached, based on their previous experiences and observation of their children's behaviour. Because the purpose was only to induce mild stress, i.e. a mild increase in the arousal level in

	Situation A	Situation B
Phase 1	Baseline: Low-level interaction (min. 5 minutes)	Baseline: Low-level interaction (min. 5 minutes)
Phase 2	Stranger confronts the child with a stressor	Parent confronts child with a stressor
Phase 3	Parent indicates the peak in child's arousal level	Parent indicates the peak in child's arousal level
Phase 4	Parent comforts the child & stranger keeps distance (min. 5 minutes)	Stranger comforts the child & parent keeps distance (min. 5 minutes)
Phase 5	Break (min. 5 minutes)	Break (min. 5 minutes)

Table 3. The protocol of the home-based experimental observation.

reaction to the stressor compared to the baseline, comfort to the children was provided immediately after parents indicated the peak.

Additionally, the person inducing the stress and the one comforting children were randomly determined. While in "situation A" the stranger (which was the researcher in the current article, who was unfamiliar to the child) 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). To determine the order of the conditions, parents drew four cards (depicting the letters A and B) during the baseline measurement from a closed envelope, containing two A's and two B's.⁴

During stressor and comfort, the person not doing an action was asked to keep distance (and if possible even to stay outside the children's range of vision to control for social referencing; Ainsworth, 1992; Braarud & Stormark, 2006). To diminish the influence of different manners of comforting, the researcher asked the parents about their habits on beforehand (e.g. "How do you comfort your child normally? Do you normally say or do something specific?") and adapted her own style of comforting to be like the parents' style of comforting. Comfort provided after the stressor lasted at least five minutes, after which a five-minute break was taken. During the baseline measurements and the breaks, there was a low-level interaction between parents and children in which the parents did not deliberately seek contact with the children, but were however not insensitive or unresponsive to their emotional cues. The entire home-based experimental paradigm was video-recorded with hand-held cameras on tripods, to the extent possible recording from two different angles. Psychophysiological arousal was measured continuously with the sensor sock (see 2.3.3).

2.3.3. Skin conductance level

The children's psychophysiological fluctuations in emotional arousal were measured with the sensor sock (Frederiks et al., 2015b; Sterkenburg et al., 2017), which was developed by the Eindhoven University of Technology (TUE), the Vrije Universiteit Amsterdam (VU) and Bartiméus. This sock, with two embedded textile electrodes on the sole of the children's foot, is a non-invasive, wireless and wearable tool to monitor and register Galvanic skin response (GSR; Frederiks et al., 2015b). GSR reflects the activity of the sympathetic nervous system, more particular the alterations in skin conductance or the skin's electrical properties (Ogorevc, Geršak, Novak, & Drnovšek, 2013). The latter are influenced by sweat secretion, which in their turn are affected by the emotions one experiences (Mokhayeri, Akbarzadeh-T, & Toosizadeh, 2011; Ogorevc et al., 2013). More specifically, the sensor sock measures skin resistance between the two electrodes (in Kilo Ohm, $k\Omega$), which is the inverse of skin conductance (in the current study reported in micro Siemens, μ S). The Shimmer[™]3 GSR module (Shimmer, 2018), which is the technical software used to register GSR, sends the physiological data to an Android tablet via Bluetooth[®]. By converting the raw skin resistance data to skin conductance (1000/1 k Ω = 1 μ S), the sensor sock gives insight into the emotional arousal and stress level of the children wearing it.

2.3.4. Emotional behaviour (Arousal and valence)

To assess the children's emotional arousal state during the different episodes of the home-based experimental paradigm, the self-developed coding scheme for behaviour observation of arousal and valence of emotions was used (Sterkenburg & Frederiks, 2017). This coding scheme departs from the operational definition of arousal described by Pfaff, Ribeiro, Matthews, and Kow (2008) and consists of two scales: 1) Arousal; and 2) Valence. Arousal is defined as the overall amount of both positive and negative tension/ emotions one experiences, scored 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. freaking out, no control over behaviour, yelling, being aggressive). The second scale reflects the value of emotions, which can be neutral, positive (e.g. delighted) or negative (e.g. frustrated). Valence was judged 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 was done continuously by the first author (event sampling; Haynes & O'Brien, 2000; Thompson, Symons, & Felce, 2000). Hence, each behaviour that forms an indication of a specific arousal level is recorded whenever it occurs. However, when events belonging to the current score and to the score below alternate quickly within five seconds after the score was assigned, the current score was maintained for the whole period. In case children were outside of the camera's range, a code "inadequate visibility" was listed. All registered codes (accurate to the millisecond) were saved in the software program "Noldus Observer XT 7.0". Two independent coders double-coded 12 video's, which results in double coding of more than 30% of the video recordings. Interrater reliability was calculated with a linear weighted kappa which takes into account the degree of disagreement between observers (Cohen, 1968; Landis & Koch, 1977). This proofed 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.

2.4. Data preparation and analysis

2.4.1. Data preparation

With respect to the video scoring, first, the perspectives of the two cameras were synchronised. The composed video was watched closely in order to define the exact time indications for certain time windows around each stressor. The four time windows for each stressor were: (1) a baseline of three minutes before the start of the stressor (e.g. StressorA1_pre); (2) the action of the stressor itself (e.g. StressorA1); (3) the window between the stop of the stressor and the start of comfort (e.g. StressorA1_between); and (4) three minutes of comfort (e.g. StressorA1_comfort). The second (i.e. stressor) and third (i.e. between-phase) time window both had child-specific time durations (depending on the kind of stressor and the time it took to change the interaction partner). The first and fourth time window had a fixed time, analogous to previous research in the domain of psychological recovery after pain (e.g. in skin conductance research in newborns; Tristão, Garcia, Lacerda de Jesus, & Tomaz, 2013). 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 & Frederiks,

2017) using the software "Noldus Observer XT 7.0". Though behavioural coding was done continuously, data were aggregated within one-second intervals, resembling a partial interval coding system.

With respect to the skin conductance data, Matlab programming was applied to aggregate raw data (consisting of approximately 51 data points per second) within onesecond intervals, similar to the video scoring. The first and the last 20% of values within each time interval (of 1 s) were omitted to reduce dependency in the time series data. For the remaining 60% of data points in the middle of the one-second interval, the median was used as final data point for that particular second. The median is less sensitive to outlying values than the mean (which could be, for example, caused by movement). Finally, the skin conductance data were synchronised with the video scoring, during which we checked thoroughly and controlled for inconsistencies and errors in the synchronisation of the two measurements (e.g. delays due to technical hitches). The final stacked data file consisted of both skin conductance data and synchronised video scoring in one-second intervals together with a label of the corresponding time window per participant.

2.4.2. Statistical analysis

Data from seven out of the total of 152 stressors (4 stressors \times 38 children), were excluded from analyses on the behavioural data due to problems with the procedure (e.g. the parent comforted the child while the stranger should have done this) or with the video recordings (e.g. the stressor was done outside the range of the camera). These values were randomly missing. The remaining 145 stressors across children were subjected to multilevel analyses on observed arousal and valence. An additional 34 stressors were excluded from the multilevel analysis on the skin conductance data (distributed across 11 children), because these skin conductance data were atypical (i.e. extreme outliers and non-fluctuating, most probably due to measurement errors of the instrument), resulting in a total of 111 stressors across 30 children. In this study, we opted for hierarchical linear two-level models (Raudenbush & Bryk, 2002; Snijders & Bosker, 2012), because our data were repeated measures in one-second intervals (either being video scorings or skin conductance data; level 1) clustered within participants (level 2). Three separate univariate multilevel analyses were performed for the three outcome variables: 1) Arousal (behavioural coding), 2) Valence (behavioural coding), and 3) Skin conductance data (physiological measure). The phase within each cycle was included as a categorical predictor, more specifically by including dummy variables (0 and 1; Kugler, Trail, Dziak, & Collins, n.d.) for the phases with a stressor (Stressor), between a stressor and comfort (Stressor_Between) and with comforting (Stressor_Comfort). The coefficients of these variables therefore refer to the change in the outcome variable, compared to the reference phase, i.e. the three-minute baseline before the stressor. In order to investigate the evolution in the outcome variable during the comfort phase, two additional dummy variables were used that refer to seconds 15-30 and seconds 30–180 during the comfort phase (analogous to Tristão et al., 2013). Their coefficients refer to the change in the outcome variable in this timeframe, compared to the first 15 seconds of the comfort phase. The person performing the action (i.e. stressing the child or comforting the child) was effect coded (Kugler et al., n.d.) and included as an additional predictor. This implies that a value of 0.5 was assigned when parents performed the action, compared to -0.5 when the stranger performed the action. The coefficient of the predictor refers to the difference between these types (a positive coefficient refers to a higher expected outcome for parents). The order of the stressor (i.e. whether it was the first, second, third or fourth stressor) was as well included as a predictor. Time in seconds was included as a covariate in the analyses. Due to the extensiveness of the data files, and the number of parameters, it was not feasible to include all interaction effects, to include random effects for the predictors, to perform a multivariate multilevel analysis (including the three outcome variables in one model), or to model a possible autocorrelation. However, because there was already a minimal correction for time dependency in the phase of data preparation, we believe that this has not changed our results. For all analyses, SPSS software package (version 25.0) was used.

3. Results

Results of the multilevel analyses are organised by outcome variable (i.e. behavioural arousal, behavioural valence and skin conductance), describing the empty and the full model, respectively. For the full model, (1) the effect of the stressor and comfort; (2) the differential person effect; (3) the differential order effect of the stressor; and (4) the differential effect of immediate or delayed comfort, are discussed in that order.

3.1. Behavioural observation of arousal

The empty multilevel model with arousal as outcome variable showed a mean arousal of $\beta = 2.90(SE = 0.08)$, with the largest amount of unexplained variance situated within persons (75.0%, $\sigma^2 = 0.75$, SE = 0.00) compared to the variability between persons (25.0%, $\sigma^2 = 0.25$, SE = 0.06). However, variability between children was still large, with the 95% prediction interval of expected behavioural arousal varying between 1.92 and 3.88. Table 4 presents the estimates, standard errors, and the significance levels of the multilevel model with arousal as outcome variable (full model). With regard to the various phases of the experimental paradigm, there was a significant increase of arousal from the baseline (with an expected level of arousal before the fourth stressor of 2.71) to the stressor ($\beta = 0.82$). The increased behavioural arousal remained after the stressor ended ($\hat{\beta} = 0.94$) and when comfort was provided ($\hat{\beta} = 0.94$). Thus, the behavioural arousal level during comfort did not go back to the baseline level. As expected, there was a significant effect of the person (parent vs. stranger) performing the action. The amount of arousal observed was significantly lower when the parent elicited stress $(\beta = -0.15)$, compared to the stranger. Moreover, there was a higher decrease of arousal when the parent comforted the child $(\hat{\beta} = -0.20)$, compared to the stranger. The order of the phase had a significant effect on arousal, with arousal during the comfort after the stressor gradually building up from the first (expected arousal = 2.36, this is 2.71 minus 0.35) to the fourth comfort phase (expected arousal = 2.71). With regard to the order effect of the stressor itself on arousal, only the difference between the third (expected arousal = 2.78) and the fourth stressor (expected arousal = 2.71) was

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		Estimate $(\widehat{\beta})$	Std. Error (SE)	Df	t	<i>p</i> -value
Intercept		2.71	0.12	181.49	22.84	.000
Time window						
	Stressor	0.82	0.02	63424.13	42.84	.000
	Stressor Between	0.94	0.06	63395.15	15.19	.000
	Stressor Comfort	0.94	0.02	63416.81	42.11	.000
	Comfort 15–30 s	-0.03	0.02	63392.19	-1.29	.198
	Comfort 30–180 s	-0.01	0.02	63404.52	- 0.46	.643
Person						
	Stressor person	-0.15	0.02	63398.96	-9.99	.000
	Comfort person	-0.20	0.02	63398.44	-14.06	.000
Order stressor (F(3610	(073.81) = 10.14; p < .000)					
	Order = 1	-0.19	0.06	56830.81	-2.92	.003
	Order = 2	-0.15	0.04	57485.56	-3.54	.000
	Order = 3	-0.03	0.02	60039.98	-1.07	.287
Order*Stressor						
	[Order = 1]*Stressor	0.01	0.03	63401.60	0.35	.723
	[Order = 2]*Stressor	0.03	0.03	63400.35	1.31	.190
	[Order = 3]*Stressor	0.07	0.03	63396.64	2.77	.006
Order*Stressor Betwee	en					
	[Order = 1]*Stressor Between	-0.27	0.09	63392.11	-3.14	.002
	[Order = 2]*Stressor Between	-0.13	0.08	63392.26	-1.53	.126
	[Order = 3]*Stressor Between	-0.29	0.08	63392.17	-3.41	.001
Order*Stressor Comfo	rt					
	[Order = 1]*Stressor Comfort	-0.35	0.02	63392.45	-16.29	.000
	[Order = 2]*Stressor Comfort	-0.29	0.02	63392.31	-14.25	.000
	[Order = 3]*Stressor Comfort	-0.25	0.02	63392.35	-12.66	.000
Variances						
	Between persons	0.24	0.06			
	Within persons (residual)	0.61	0.01			

significant. There was no significant difference between the immediate comfort (1–-15 seconds) and the delayed comfort, both at 15–30 seconds, and 30–180 seconds. As per these including predictors in the full multilevel model, 18.67% of the variance within persons was explained.

3.2. Behavioural observation of valence

The empty multilevel model with valence as outcome variable ($\hat{\beta} = 0.36, SE = 0.14$) showed that the largest amount of unexplained variance was again situated within persons (83.69%, $\sigma^2 = 3.90, SE = 0.02$) compared to the variability between persons (16.31%, $\sigma^2 = 0.76, SE = 0.18$). However, the 95% prediction interval of expected behavioural valence ranged between -1.35 and 2.07, which reflects substantial interindividual variability. Table 5 displays the estimates, standard errors and the significance levels of the multilevel model with valence as outcome variable (full model). Conforming to expectations, the valence of the child's emotions became more negative when the stressor was presented ($\hat{\beta} = -1.83$). Immediately after the stressor ended, this trend continued (Stressor between, $\hat{\beta} = -2.71$) and became slightly less negative during the comfort phase ($\hat{\beta} = -1.67$). In general, parents provoked less negative valence ($\hat{\beta} = 0.65$) when presenting the stressor, compared to the stranger. During comfort, parents provoked more positive valence ($\hat{\beta} = 0.60$), in comparison with the stranger. As shown

		Estimate $(\widehat{\beta})$	Std. Error (SE)	Df	t	<i>p</i> -value
Intercept		1.95	0.24	270.77	8.00	.000
Time window		1.25	0.27	2/0.//	0.00	.000
	Stressor	-1.83	0.04	63412.69	-43.50	.000
	Stressor Between	-2.71	0.14	63380.45	-19.86	.000
	Stressor Comfort	-1.67	0.05	63407.12	-33.92	.000
	Comfort 15–30 s	0.61	0.05	63376.27	11.43	.000
	Comfort 30–180 s	1.40	0.04	63393.02	35.20	.000
Person						
	Stressor person	0.65	0.03	63385.76	19.21	.000
	Comfort person	0.60	0.03	63385.06	18.88	.000
Order stressor (F(35863	(9.35) = 76.21; p < .000)					
	Order = 1	-0.77	0.14	50884.52	-5.44	.000
	Order = 2	-0.61	0.10	51995.44	-6.40	.000
	Order = 3	0.06	0.05	56536.84	1.11	.267
Order*Stressor						
	[Order = 1]*Stressor	-0.53	0.06	63389.29	-9.18	.000
	[Order = 2]*Stressor	-0.28	0.06	63387.64	-5.09	.000
	[Order = 3]*Stressor	-0.71	0.06	63382.51	-12.47	.000
Order*Stressor Betweer	1					
	[Order = 1]*Stressor Between	0.57	0.19	63376.18	3.03	.002
	[Order = 2]*Stressor Between	0.63	0.18	63376.39	3.43	.001
	[Order = 3]*Stressor Between	0.13	0.19	63376.25	0.69	.489
Order*Stressor Comfort						
	[Order = 1]*Stressor Comfort	0.83	0.05	63376.69	17.36	.000
	[Order = 2]*Stressor Comfort	0.60	0.04	63376.48	13.60	.000
	[Order = 3]*Stressor Comfort	0.37	0.04	63376.49	8.43	.000
Variances						
	Between persons	0.81	0.20			
	Within persons (residual)	3.00	0.02			

Table 5. Univariate multilevel linear analysis with valence as outcome variable (full model).

in Table 5, there was a significant order effect, in which the observed valence in reaction to the stressor was significantly higher in the fourth stressor (*expected valence* = 1.95), compared to the first (*expected valence* = 1.42, that is 1.95 minus 0.53), second (*expected valence* = 1.67), and third stressor (*expected valence* = 1.24). There was a significant difference between the immediate comfort (1–15 seconds) and the delayed comfort, with valence becoming more positive after 15–30 seconds of comfort ($\hat{\beta} = 0.61$), and 30–180 seconds of comfort ($\hat{\beta} = 1.40$). In the full multilevel model, 23.08% of the variance within persons was explained

3.3. Skin conductance measures of emotion

The empty multilevel model with skin conductance (in μ S) as outcome variable ($\hat{\beta} = 7.35, SE = 1.52$) showed a large amount of between-person variability (74.85%, $\sigma^2 = 69.65, SE = 18.30$). The 95% prediction interval of expected skin conductance varied between -9.01 and 23.71. The amount of unexplained within-person variance was smaller (25.15%, $\sigma^2 = 23.40, SE = 0.15$). In Table 6, the estimates, standard errors and the significance levels of the multilevel model with the skin conductance data (in μ S) as outcome variable are reported (full model). In relation to the baseline, skin conductance increased significantly in general when presenting the stressor ($\hat{\beta}$ = 2.16). The arousal continued to increase in the between phase, immediately after the stress peak was

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		Estimate $(\widehat{\beta})$	Std. Error (SE)	Df	t	Sig.
Intercept		7.37	1.68	41.66	4.40	.000
Time window						
	Stressor	2.16	0.12	50636.20	17.73	.000
	Stressor Between	3.97	0.40	50627.86	9.98	.000
	Stressor Comfort	1.31	0.15	50633.15	9.04	.000
	Comfort 15–30 s	-0.13	0.16	50627.57	-0.84	.402
	Comfort 30–180 s	-0.08	0.12	50629.45	-0.71	.480
Person						
	Stressor person	-0.71	0.10	50627.82	-7.15	.000
	Comfort person	0.69	0.10	50627.57	7.43	.000
Order stressor (F.350602	(2.39) = 307.90; p < .000)					
	Order = 1	-2.49	0.49	50550.63	-5.08	.000
	Order = 2	-2.42	0.33	50560.98	-7.26	.000
	Order = 3	1.10	0.18	50609.76	6.08	.000
Order*Stressor						
	[Order = 1]*Stressor	-0.75	0.17	50627.75	-4.42	.000
	[Order = 2]*Stressor	-1.62	0.16	50628.57	-10.16	.000
	[Order = 3]*Stressor	-1.93	0.16	50627.52	-11.71	.000
Order*Stressor Between						
	[Order = 1]*Stressor Between	-3.08	0.56	50627.01	-5.46	.000
	[Order = 2]*Stressor Between	-2.24	0.54	50627.02	-4.11	.000
	[Order = 3]*Stressor Between	-3.21	0.55	50627.02	-5.89	.000
Order*Stressor Comfort						
	[Order = 1]*Stressor Comfort	0.33	0.14	50627.15	2.35	.020
	[Order = 2]*Stressor Comfort	1.17	0.13	50627.09	9.10	.000
	[Order = 3]*Stressor Comfort	-0.67	0.13	50627.02	-5.27	.000
Variances	-					
	Between persons	70.31	18.47			
	Within persons (residual)	20.72	0.13			

indicated ($\hat{\beta} = 3.97$) and dropped during the comfort phase ($\hat{\beta} = 1.31$). Consistent with the findings on the behavioural observation of arousal, the skin conductance level did not reach the baseline level during comfort. Parents elicited less arousal during the stressor phase ($\hat{\beta} = -0.71$) compared to the stranger. However, arousal levels tended to be significantly higher when parents comforted the child, compared to the stranger ($\hat{\beta} =$ 0.69). With regard to the order of the stressors, the expected skin conductance (ESC) was significantly higher in the fourth stressor (*ESC* = 7.37), compared to the first (*ESC* =6.62, that is 7.37 minus 0.75), second (*ESC* = 5.75) and third stressor (*ESC* = 5.44). There was no significant difference between the immediate comfort (1–15 seconds) and the delayed comfort, both at 15–30 seconds, and 30–180 seconds. In the full multilevel model, 11.45% of the variance between persons was explained.

4. Discussion

The current study demonstrated that children with severe or profound intellectual disabilities (ID) differentiated between parents as attachment figures and the stranger in a home-based experimental paradigm. This differentiation occurred both when stress was elicited and comfort was randomly provided by either the parents or a stranger. Moreover, the results of both behavioural observations and psychophysiological measurements were in the same line and, thus, provided valuable insights into the children's emotional responses towards their attachment figures.

The study's results indicated that the stressors did indeed significantly increase arousal (during the stressor and in the between phase, immediately after the peak of arousal). During the three minutes of comfort, behavioural arousal did not (yet) decrease to baseline level, though the valence of emotions seemed to become slightly more positive compared to the stressor phase, regardless of the person doing the action. However, in line with Bowlby's (1956) and Cassidy's (2016) hypotheses on parent-child attachment, children in this study differentiated significantly between their parents and the stranger, both during stress and comfort. The stranger both elicited more (negative) arousal (i.e. stress) when performing the stressor compared to the parents and she evoked more behavioural arousal during comfort, which was less positive compared to the parents' evoked arousal. Despite this difference, the stranger was, however, still able to comfort the child and to shift the valence of the children's emotions in a positive direction over time during comfort. These findings strengthen the view of parents that, whereas they are their children's preference persons, other persons could as well function as safe haven (Vandesande, Bosmans, & Maes, 2019b). Ainsworth, Blehar, Waters, and Wall (1978) corroborates this view by stating that comfort provided by a stranger may not be as satisfying to children as comfort of their parents, though it could still be efficient.

In general, the psychophysiological results supported these behavioural findings. However, there were some (slightly) different or contradicting findings in comparison with the behavioural observations (this lack of consistency is also described in previous research; Koolhaas et al., 2011; Lima et al., 2011; Parihar, Hattiangady, Kuruba, Shuai, & Shetty, 2011). A first difference between behaviour and physiology to be noted is situated in the arousal during comfort. Whereas behavioural arousal did not (yet) decrease in the first three minutes of comfort, there was already a decreasing trend noticeable in arousal reflected by skin conductance during the comfort phase. This indicates that physiology may reflect changes in the body that have not (yet) been shown in behaviour (in line with Vos et al., 2012). Arousal could be viewed as a physiological response that activates and potentiates behaviour (Koolhaas et al., 2011; Pfaff et al., 2008). In that respect, physiological arousal is clearly to be discerned from behavioural reactions and may provide complementary information in this target group. A second difference is that behavioural arousal levels were significantly lower during comfort by the parents compared to the stranger, whereas for skin conductance the opposite was true. Though this might seem contradictory, this may hypothetically be explained by the amount of positive emotions the parents elicited during comfort. The valence of emotions is, indeed, not reflected in skin conductance (Dawson et al., 2007; Frederiks et al., 2015b; Picard, Fedor, Ayzenberg, 2016; Picard, Vyzas, & Healey, 2001). Both positive and negative emotions, like agitation, alertness, frustration, being happy or being upset increase skin conductance. To evaluate the extent to which arousal increases are linked with positive or negative emotions, behavioural observation is needed (Koolhaas et al., 2011; Petry & Maes, 2006; Picard et al., 2016, 2001). Yet, it remains a difficult task to match the skin conductance data with the behavioural observations to interpret the results. Future research would benefit from exploring especially those situations where skin conductance and behaviour seem to contradict. Picard et al. (2001) used machine learning to explore physiological correlates for certain emotional states, but also concluded that there is still a lot of (interpretation) work left on this domain. A third difference is the ratio of within-person versus between-person variability in behavioural observation versus physiological measures. Whereas the largest amount of unexplained variance was situated at a within-person level in the behavioural measures, the between-person variance stood out in the measures of the sensor sock. However, this is inherent to the instruments. In the behavioural coding the range within which one can fluctuate is much smaller (restricted by a fixed Likert-scale), compared to the potential measuring range of skin conductance. Every person is different regarding the baseline of electrodermal activity depending on, for example, their typical amount of sweat secretion (Braithwaite et al., 2015; El-Sheikh, 2007). Moreover, the general skin conductance level within one person only fluctuates over longer periods (Braithwaite et al., 2015; Dawson et al., 2007), whereas during behavioural coding the child's behaviour is constantly compared with previous behaviour and changes are more likely to occur.

There are two main limitations in this study. The first limitation is that the protocol of the home-based experimental paradigm evoked uncertainty or raised questions, both in the researcher and the parents, in some families. Questions arose relating to the baseline (e.g. the researcher allowed the use of activating toys during baseline in certain specific situations, but doubted whether they influenced the protocol in other situations), the stressor (e.g. parents found it difficult to identify stressors or to elicit stress in their child when actually normally not needed) and the comfort (e.g. some parents started to comfort their child when the protocol instructed that they needed to keep distance; the stranger could not always imitate the comfort of parents when it was inappropriate, like kissing the children; different styles of comforting could possibly have demanded a different level of energy and arousal of the children). Furthermore, for some children the procedure took too long and was (emotionally) fatiguing. Where these deviations from the protocol during a part of the observation raised the concern that the data may not have been accurate or complete, the respective data were excluded from data analyses (this occurred in seven stressors across seven children, see 2.4.2 for more details on exclusion). Additionally, the use of a fixed protocol, may have caused an interpretation bias during behavioural coding. It was, indeed, not possible to code blindly with regard to the phase of the observation that was displayed. Because the observer always saw the action that was going on in the video recording, she could easily derive the position of the video excerpt in the whole video recording. To confine the influence of possible observer biases, the coding manual included detailed coding guidelines, which were consistently applied by all the coders. Due to the randomisation of stressors (situation A or B), there was no coding effect with regard to the order effect (because coding was always done in the following order: A1, A2, B1, B2). In addition, the corroborating psychophysiological results exclude the possibility that the study's results were merely observer-induced.

The second limitation is that some technical problems and interpretation difficulties using the sensor sock were experienced. In a few homes the Bluetooth[®] connection between the Shimmer and the tablet that registered the data got interrupted once or twice. In addition, also other factors (such as sleep or metabolic demand; Vos et al., 2012) influence the changes in the skin's electrical properties. Although the researcher asked the parents about the children's medication use, meals and hours of sleep that day, it was not possible to identify the exact effect of these factors on an individual level, partially because it lacks normative data on the range and fluctuations in skin conductance of children with severe or profound ID. The current study has excluded certain participants when the data did not resemble a reliable or valid measure of skin

conductance (e.g. when there was a disproportional amount of constant values or when there were many extreme outliers outside of the typical range of skin conductance due to technical hitches or motion artefacts, Dawson et al., 2007; see 2.4.2 for more details on exclusion). Including these incorrect measures of skin conductance in data analyses would have biased the study's results to a large extent. It is, however, interesting to note that from the eight children who were excluded from analyses for this particular reason, five children had extremely limited mobility comorbid with a profound cognitive delay. It can be speculated that deviations in their central nervous system (e.g. in premotor cortex or cerebellum in case of motor impairments) may have had an impact on their electrodermal activity (Dawson et al., 2007). The other three were, on the other hand, completely mobile and were moving heavily (e.g. walking, running) during the observation. Distortion in their data can possibly be identified as motion artefacts, which is a common hindrance in the analysis of skin conductance measures (Lee & Choi, 2007; Novak, 2014).

First evidence of differential responses towards the parents, with whom children are expected to have previous care experiences, and a stranger, with whom the children do not have previous care experiences, was provided. However, besides the amount of and the nature of these care experiences, the parents and the stranger also differ in the extent to which they are familiar to the children. Not only are the children more accustomed to the way in which actions are performed by the parents, but they may also feel the hesitance of the stranger when performing certain distressing activities (e.g. brushing the teeth). Though the paradigm of the current study was similar to Ainsworth et al.'s Strange Situation Procedure (2015), in which support effects from parents and strangers are as well compared, further research including comparison with other familiar persons who are not regarded as attachment figures, is warranted in an attempt to separate attachment-related and mere familiarity-effects from each other.

The current study has extended our understanding of the unique role parents fulfil for their children with severe or profound ID. In a structured and systematic way, the study has demonstrated that children do differentiate between their parents as attachment figures and the stranger after experiencing mild stress. In that way, this study is a first valuable step to point towards the special meaning of their parents' comfort for children and, hence, the special role parents fulfil in their children's emotional life. These findings may contribute towards meaning-making in parents of children with severe or profound ID, as they are trying to overcome the significant barriers in communication and behavioural expression (Nakken & Vlaskamp, 2007; Schuengel & Janssen, 2006). Parents might get a sense of self-efficacy when they experience the unique meaning of parent-child interaction, from which they can derive pleasure, satisfaction and motivation to interact with their child. Further scientific inquiry is warranted in the future to distinguish attachment-related differentiation between caregivers from mere familiarityeffects and to explore inter-individual differences. The current study has drawn attention to new approaches, such as combining behavioural and psychophysiological measures, to study attachment-related questions in this population in order to achieve a comprehensive understanding of the unique parent-child attachment relationships in children with limited expressive abilities.

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Notes

- 1. To the best of our knowledge, there are no clear guidelines to identify a severe ID at young age on the basis of a comparison between children's developmental and chronological age. Hogg, Foxen, and McBrien (1981) associate profound ID with a developmental age lower than a quarter of children's chronological age. In line with this reasoning, half of the children's chronological age was used as an upper limit. Because the current study did not include children older than nine years, the children's developmental age did never exceed four and a half years. This is in accordance with the maximum age associated with severe ID in adults with ID according to Kraijer and Plas (2007). However, for each child multiple sources of information were triangulated to warrant a valid and reliable inclusion of children.
- 2. Especially for the youngest children with very low cognitive abilities, test results were lacking because reliable IQ-tests and established norms are lacking at the very low end of the spectrum of intellectual functioning (Resing & Blok, 2002; Weis, 2014). Hence, the average cognitive level of 9.60 months is possibly an underestimation.
- 3. Level 4 and 5 of the Gross Motor Function Classification Scale (GMFCS; Palisano et al., 1997) reflect a severe motor impairment (Palisano, Rosenbaum, Bartlett, & Livingston, 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 reached level 4 and 26.3% reached level 5 on the Manual Ability Classification System (MACS; Eliasson et al., 2006, 2017).
- 4. A number is added to these letters in the remaining text to identify whether the first (A1) or the second (A2) performance of "situation A" was implied. The same applies for "situation B".

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