Diagnostic Assessment of Attachment Related Disorders in Children with Intellectual Disability

Bartiméus reeks

Deze publicatie is mogelijk gemaakt door de Vereniging Bartiméus Sonneheerdt.

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Lay out en design
WEBEDU

ISBN/EAN: 978-94-91838-60-6
First edition

VRIJE UNIVERSITEIT

Diagnostic Assessment of Attachment Related Disorders in Children with Intellectual Disability

ACADEMISCH PROEFSCHRIFT

ter verkrijging van de graad Doctor aan
de Vrije Universiteit Amsterdam,
op gezag van de rector magnificus
prof.dr. V. Subramaniam,
in het openbaar te verdedigen
ten overstaan van de promotiecommissie
van de Faculteit der Gedrags- en Bewegingswetenschappen
op donderdag 23 november 2017 om 11.45 uur
in de aula van de universiteit,
De Boelelaan 1105

door
Hans Peter Giltaij
geboren te Arnhem

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Paranimfen

Marieke Weijburg-Melis

Thijs Jan van der Leij

Special thanks to:

kinder- en jeugdpsychiatrie karakter

de jutters

de Bascule
Academisch centrum voor kinder- en jeugdpsychiatrie

GGZ Rivierduinen Kristal

accare
kinder- en jeugdpsychiatrie

dichterbij
We dagen ieder mens uit!

ggz Drenthe
geestelijke gezondheidszorg

BARTIMEUS

intermetzo

Voor Yvonne, Willemijn en Maarten

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Chapter 1

General introduction

Introduction

The fall of the Berlin Wall in 1989 and the opening of the borders to Eastern Europe revealed distressing images of huge numbers of institutionalised children in Eastern European countries. In Romania alone there were a reported 170,000 orphaned or abandoned children in 700 children's homes (Greenwell, 2003). These children were living in situations of serious neglect (poor nutrition, improper care, no educational materials) and a lack of selective attention on the part of the caregivers. The children displayed extremely abnormal behaviour: self-harm, self-stimulation, aggression, a vacant gaze, lack of an urge to explore, substantially impaired development, and no initiative to interact or play with others.

However, these images and reports were nothing new. In 1945, psychoanalyst René Spitz released film footage and published an article on his observations of children from the institution where he worked, a shelter for foundlings in the USA. Spitz compared the children from the children's home with those raised in normal families with warmth and individual attention. He discovered that the psychological and physical development of the children in the children's home was delayed and that they were more apathetic (Spitz referred to this as 'anaclitic depression'). Despite good physical care (nutrition, clothing), a third of these children died within one year. This prompted Spitz to conclude that maternal affection is a vital necessity for the child. The findings of René Spitz, and the poignant images that accompanied them, have had significant consequences for policy relating to children's homes, hospitals and institutions.

In the 1940s and 50s, psychologist John Bowlby started to work on his attachment theory. Publishing in 1969 his first volume of his trilogy explaining this theory (Bowlby, 1969/1984), he wrote that all children have an innate drive to preferentially seek out familiar adults, as this increases the child's chances of survival. He based this assertion on, among other things, experiments with infant monkeys conducted by Harry Harlow and experiments with geese conducted by Konrad Lorenz. Bowlby defined attachment as the tendency to seek the proximity, comfort and support of a specific caregiver in frightening situations (stress), when tired, or when ill. This tendency develops in infancy, but is a phenomenon that characterizes human behaviour from the cradle to the grave. In a report to the World Health Organization (WHO, 1951), Bowlby compared the need

for an attachment relationship with the primary need for nutrition, suggesting that having a relationship of attachment is a basic vital necessity for every child.

Spitz (in the case of institutionalised children) and Harlow (in the case of monkeys) demonstrated that growing up in the first years of development without a caring, protective maternal bond leads to serious maladaptive behaviour. Bowlby referred to this as the attachment relationship with a mother (although this may be another specific adult) as the attachment figure. Due to the limited number of available case studies, this maladaptive behaviour was only clinically recognized as Reactive Attachment Disorder (RAD) in 1980 with the third edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-III, American Psychiatric Association, APA). In this initial version of the RAD diagnosis, the definition centred on lack of development and social responsiveness. From the revised version, DSM-III-R (1987, APA), followed by DSM-IV (1994, APA), RAD was redefined as defective social behaviour, in which two mutually exclusive subtypes were distinguished: the emotionally withdrawn, inhibited type (RAD inhibited type) and the socially undifferentiated, indiscriminate type (RAD disinhibited type). The inhibited subtype is characterised by excessive inhibition, hypervigilance, and highly ambivalent and contradictory responses in the social behaviour vis-à-vis caregivers. The disinhibited subtype is characterized by diffuse attachment behaviour with indiscriminate sociability and excessive friendliness, even towards relative unfamiliar adults. The maladaptive behaviour must originate before the child's fifth birthday and be the result of pathogenic care (neglect, maltreatment, institutionalisation). This definition also states that the disordered behaviour may not be diagnosed as RAD if the behaviour can also be explained on the basis of a developmental delay (as in mental retardation) or a pervasive developmental disorder (PDD).

DSM-5 criteria

Over the past 20 years, the definition of the Reactive Attachment Disorder (RAD), as described in DSM-IV, has guided studies of this disorder. However, the DSM-IV definition has been the subject of increasing criticism, not only from the perspective of clinical practice, but also following the outcomes of scientific research (Zeanah & Gleason, 2010). In the first place it was asserted that the DSM definition of RAD describing the

relational component as 'disrupted social relatedness' and not as 'disrupted attachment behaviour', is confusing. This makes it more complicated to differentiate, in the case of disrupted social behaviour, between autistic-like behaviour and disrupted attachment behaviour. Another point of criticism relates to the criterion that the child's disrupted social behaviour could not be explained by a delay in development (as in the case of children with an intellectual disability) and does not meet the criteria of a pervasive developmental disorder (PDD). While this formulation does not explicitly exclude children with an intellectual disability or PDD from the RAD diagnosis, this unintentional effect is inherent in the formulation of this criterion. With regard to cognitive development, it is important for the RAD diagnosis that the child must at least have reached a certain cognitive milestone in order to form a selective attachment relationship (corresponding to normal cognitive functioning at 9 months of age). The presence of PDD is assumed to be an exclusion criterion for the RAD diagnosis. This is a rather bizarre statement when comprehensive research has demonstrated that children with PDD clearly have the wherewithal to enter into a (secure) attachment relationship (Rutgers, Bakermans-Kranenburg, Van lJzendoorn, & Van Berckelaer-Onnes, 2004). This would imply that a PDD diagnosis constitutes a 'protective factor' for a RAD diagnosis. The background to this formulated criterion is that distinctions are made in diagnosis between the origins of the disrupted social behaviour. In the case of PDD, defective social behaviour is explained on the basis of a neurological development disorder, while in the case of RAD the explanation is found in the 'reaction' to serious neglect and lack of emotional support, comfort, and attention. Incidentally, both clinical and scientific research has shown that the criterion of 'pathogenic care' is a complicating factor (Zeanah & Gleason, 2010). However, a key reason for maintaining this criterion in the diagnosis is the fact that the indiscriminate, disinhibited behaviour is also observed in children with Williams syndrome (Dykens, 2003) or fetal alcohol syndrome (Streissguth, Barr, Kogan, & Bookstein, 1997), even when pathogenic care is clearly absent. Williams syndrome is attributable to a chromosomal mutation while fetal alcohol syndrome is caused by excessive alcohol usage by the mother during pregnancy. These children are also often found to have an intellectual disability.

A number of studies of young children growing up in institutions or in circumstances of serious neglect found children who met the description of the inhibited subtype as well as the disinhibited subtype of RAD, and children who matched the behavioural characteristics of both subtypes (Smyke, Dumitrescu, & Zeanah, 2002; Zeanah, Scheeringa, Boris, Heller, Smyke, & Trapani, 2004; Oosterman, & Schuengel, 2007). This is inconsistent with the DSM-IV definition of RAD. In response to these criticisms of the definition of the DSM-IV diagnosis of Reactive Attachment Disorder, which received extensive scientific underpinning, a revision was made for DSM-5 (Zeanah & Gleason, 2010). In doing so it was decided to view the two subtypes of the DSM-IV Reactive Attachment Disorder as two distinct disorders in DSM-5. The inhibited subtype was redefined under the name Reactive Attachment Disorder (RAD, Table 1), while the disinhibited subtype was redefined under the name Disinhibited Social Engagement Disorder (DSED, Table 2). On the one hand, the two disorders are assumed to arise in similar conditions of risk: experiences of extremes of insufficient care that limit the child's ability to form selective attachment relationships. On the other hand, they differ with regard to phenomenology, correlates, course, response to treatment, and vulnerability factors (Zeanah & Gleason, 2015). While RAD involves disordered attachment behaviours, the core abnormality in DSED concerns social disinhibition across relationships, including but not limited to attachment relationships. Because DSED may occur in the absence of attachment, in a maladaptive attachment relationship, or in an adaptive attachment relationship to a subsequent foster or adoptive parent, it is a reasonable question whether DSED is an attachment disorder at all (Zeanah et al., 2016). Because of the similarity in aetiology and context of child-caregiver relationship, researchers and clinical practitioners focus mostly on both disorders in their study or diagnostic procedure. For this reason we use the term 'attachment related disorders' for the collective of the disorders RAD and DSED.

Table 1 DSM-5 Criteria for Reactive Attachment Disorder (APA, 2013)

313.89 Reactive Attachment Disorder

A) A consistent pattern of inhibited, emotionally withdrawn behaviour toward adult caregivers, manifested by both of the following:
   1) The child rarely or minimally seeks comfort when distressed.
   2) The child rarely or minimally responds to comfort when distressed.

B) A persistent social and emotional disturbance characterized by at least two of the following:
   1) Minimal social and emotional responsiveness to others.
   2) Limited positive affect.
   3) Episodes of unexplained irritability, sadness, or fearfulness that are evident even during nonthreatening interactions with adult caregivers.

C) The child has experienced a pattern of extremes of insufficient care as evidenced by at least one of the following:
   1) Social neglect or deprivation in the form of persistent lack of having basic emotional needs for comfort, stimulation, and affection met by caregiving adults.
   2) Repeated changes of primary caregivers that limit opportunities to form stable attachments (e.g., frequent changes in foster care).
   3) Rearing in unusual settings that severely limit opportunities to form selective attachments (e.g., institutions with high child-to-caregiver ratios).

D) The care in Criterion C is presumed to be responsible for the disturbed behaviour in Criterion A (e.g., the disturbances in Criterion A began following the lack of adequate care in Criterion C).

E) The criteria are not met for autism spectrum disorder.

F) The disturbance is evident before the age 5 years.

G) The child has a developmental age of at least 9 months.

Table 2 DSM-5 Criteria for Disinhibited Social Engagement Disorder (APA, 2103)

313.89 Disinhibited Social Engagement Disorder

A) A pattern of behaviour in which a child actively approaches and interacts with unfamiliar adults and exhibits at least two of the following::
   1) Reduced or absent reticence in approaching and interacting with unfamiliar adults.
   2) Overly familiar verbal or physical behaviour (that is not consistent with culturally sanctioned and with age-appropriate social boundaries).
   3) Diminished or absent checking back with adult caregiver after venturing away, even in unfamiliar settings.
   4) Willingness to go off with an unfamiliar adult with minimal or no hesitation.

B) The behaviours in Criterion A are not limited to impulsivity (as in attention-deficit/hyperactivity disorder) but include socially disinhibited behaviour.

C) The child has experienced a pattern of extremes of insufficient care as evidenced by at least one of the following:
   1) Social neglect or deprivation in the form of persistent lack of having basic emotional needs for comfort, stimulation, and affection met by caregiving adults.
   2) Repeated changes of primary caregivers that limit opportunities to form stable attachments (e.g., frequent changes in foster care).
   3) Rearing in unusual settings that severely limit opportunities to form selective attachments (e.g., institutions with high child-to-caregiver ratios).

D) The care in Criterion C is presumed to be responsible for the disturbed behaviour in Criterion A (e.g., the disturbances in Criterion A began following the lack of adequate care in Criterion C).

E) The child has a developmental age of at least 9 months.

Confusing criteria

A study by Rutter et al. (1999) describes a number of children with a background of serious insufficient care and institutionalization who initially met the criteria for autism (ASD), but whose social behaviour improved after being placed with adoptive families and who no longer fulfilled the criteria for the autism diagnosis after two years. This implies that the deviant social behaviour is probably related to the background of insufficient care and not to a disorder in neurological development. Unfortunately, very little research has been conducted to date into the possibility of whether RAD and ASD can occur alongside each other, or whether they are mutually exclusive. This is why the definition of RAD in DSM-5 still includes the autism spectrum disorder (ASD) as an exclusion criterion. Recently however, the results of an investigation into the correlation between disrupted attachment behaviour and ASD was published. In an investigation involving two groups of children, one subgroup who had been diagnosed with an attachment related disorder (RAD or DSED) and one subgroup who had been diagnosed with ASD, Davidson et al. (2015) demonstrated that problematic social behaviour between both groups can be clearly distinguished by means of structured observation procedures. Because of the implications of this exclusion criterion for the clinical practice of diagnostic decision-making and use of an adequate intervention, especially for children with intellectual disability, a study of the overlap and exclusion elements between RAD and ASD is required (Zeanah & Gleason, 2010).

On the basis of their randomised controlled trial with socially deprived children from the Bucharest Early Intervention Project (BEIP), Nelson and colleagues (2007; Nelson, Furtado, Fox, & Zeanah, 2009) reported that young institutionalised children have significantly greater developmental delay (cognitive, language) than children growing up with their biological parents. On average, 4.5-years-old children institutionalised at a very young age scored 36 points lower in an IQ test than children raised by their biological parents. This study also demonstrated that developmental delay increases with more prolonged institutionalised care before becoming eligible for foster care. Evidence suggests that social deprivation in the early years negatively impacts neurobiological brain development, leading to suboptimal cognitive, language and socioemotional development. In the BEIP-study the participants were all socially deprived children. Within a population with more diverse experiences, we will examine the associations between an attachment related disorder (RAD / DSED) and the level of

functional development and comorbid psychopathology in children with low intellectual functioning.

Procedural barriers

In DSM-5, the diagnosis of attachment related disorders is established by determining the combination of specific behavioural characteristics, patterns of extremes of insufficient care (social neglect or deprivation), and age/level of development. The guidelines on the basis of which this must be determined are set out in the *Practice Parameter for the Assessment and Treatment of Children and Adolescents with Reactive Attachment Disorder of Infancy and Early Childhood* (American Academy for Child and Adolescent Psychiatry, AACAP, 2005), in 2016 revised in the *Practice Parameter for the Assessment and Treatment of Children and Adolescents with Reactive Attachment Disorder and Disinhibited Social Engagement Disorder Development and Attribution* (Zeanah et al.). The specific behavioural characteristics must be determined within a structured observational setting by observing the interaction patterns of the child with the primary caregiver, which is compared with interactions with an unfamiliar adult. The development history of the attachment behaviour of the child should also be identified, together with the primary caregiver where possible. The child's behaviour in both the observation setting and in the description of the development history is analysed and assessed for the presence of maladaptive behaviour, which is characteristic of disordered attachment. Here, the AACAP recommends using the *List of Behavioral Signs of Disturbed Attachment in Young Children* (BSDA; Zeanah, Mammen, & Lieberman, 1993), in which the behavioural pattern of the child is assessed on the basis of eight dimensions of (disturbed) attachment behaviour: displaying affection, seeking comfort, showing confidence in another individual, collaboration, explorative behaviour, controlling behaviour, reaction on reunion, reaction to strangers. Studies using multi-informant diagnoses of attachment related disorders are rare. This is the first multi-informant study using the Practice Parameter recommended elements for the assessment of attachment related disorders in children with low intellectual functioning.

In clinical practice the establishment of insufficient care requires a retrospective approach. This is often difficult, unless for example, a report from the Dutch Child Care

and Protection Board (*Raad voor de Kinderbescherming*) is available. In the event of institutionalisation, foster care or adoption, information about the development circumstances of the child's initial years is often unavailable. But also home-reared children, living with their biological parents and referred to mental health care for emotional or behavioural problems, may experience extreme insufficient care (Walrath, Yabarra, Sheehan, Wayne, & Burns, 2006) and may therefore be at risk for attachment related disorders. As a rule, children are unable to give a reliable description of the first few years of their life, and the primary caregivers of the young child are unlikely to be open about any maltreatment or neglect they may have inflicted on their child owing to social or legal consequences. Reliable identification of extreme insufficient care is complicated, all the more so because there is a lack of clear empirical foundation for the choice of indicators for insufficient care. Interesting steps have been made in this regard, however. Levendosky, Bogat and Huth-Bocks (2011) showed an association between experiencing domestic violence and disturbances in the attachment relationship between mother and young child. Children whose mothers (caregivers) had a history of psychiatric disturbances were more likely to be diagnosed with indiscriminate attachment behaviour (Lyons-Ruth, Bureau, Riley, & Atlas-Corbett, 2009; Zeanah et al., 2004). Scheper et al. (2016) reported an association between parenting stress and disinhibited social engagement behaviour in their study with home-reared children, referred to mental health care for emotional or behavioural problems. Multiple studies (Breidenstine, Bailey, Zeanah, & Larrieu, 2011; Kay, Green , & Sharma, 2016) show associations between a diversity of complex circumstances during the early developmental years of the child and the manifestation of disordered attachment behaviour. Furthermore, the DSM-5 prescribes that for the diagnosis of an attachment related disorder evidence of patterns of extreme insufficient care is experienced by the child. However, it is a challenge to collect reliable information about possible insufficient care (abuse or neglect) in an ethical way without disturbing the relationship with the child and the caregiver (Minnis et al., 2013).

Prevalence

The number of studies into the prevalence of attachment related disorders is extremely limited. This is caused on the one hand by the controversy about the correct definition of the diagnosis of attachment related disorders, in which there is a lack of consensus on diagnostic procedures and diagnostic measurement instruments. On the other hand, attachment related disorder among the general population is assumed to be rare. Richters and Volkmar (1994) estimated the prevalence among the overall population to be less than 1%. This is why attachment related disorders were only studied in specific groups of children with an increased risk of RAD (so-called high-risk populations), such as children from orphanages (Smyke, Dumitrescu, & Zeanah, 2002; Zeanah, Smyke, Koga, & Carlson, 2005), children from Romanian children's homes (Rutter et al., 2007), children in women's refuge centres (Boris et al., 2004), and children in foster care (Oosterman & Schuengel, 2007). In these study groups, disturbed attachment behaviour rates varying from 18 to 56% were measured. Green and Goldwyn (2002) stated that children with an intellectual disability may also constitute a high-risk group, given that the likelihood of out-of-home placement (institutionalisation) and maltreatment in this target group is higher than in the general population. Hardly any studies of the prevalence of attachment related disorders among people with an intellectual disability have been conducted. Minnis, Fleming and Cooper (2010) conducted a study into RAD among institutionalised adults with an intellectual disability. They found disordered attachment behaviour among this group, which appeared to be associated with problems during early childhood development. It should be noted that the display of disordered attachment behaviour does not always lead to a clinical diagnosis of an attachment related disorder. According to the Practice Parameter, for diagnosis multi-informant and multi-measurement data are needed to call the diagnosis. Minnis and colleagues (2013) did a large multi-informant study to estimate the prevalence of RAD and DSED in children (age 6-8 years) in a deprived urban area and found a prevalence of 1.4%. Assuming that children with an intellectual disability and psychological or behavioural problems are a high-risk group for developing attachment related disorders, we hypothesize a higher prevalence of RAD and DSED in this group of children.

Causes

In line with the DSM-5 definition of the two attachment related disorders (RAD and DSED), extremes of insufficient care during the first few years are assumed to be the cause of disordered attachment behaviour. This is understood to include neglect of the emotional and/or physical needs of the child, unstable circumstances for care and parenting as a result of inability to provide adequate support among caregivers, multiple changes of primary caregivers and institutionalisation (with a high child/caregiver ratio), which are an obstacle to the development of an attachment relationship. However, this assumption is still hardly tested because of a lack of epidemiological and experimental research. The few studies for causal factors, like the long-term studies of the Bucharest Early Intervention Project (BEIP) with children from the Romanian orphanages (Nelson, Fox, & Zeanah, 2014) show confusing findings as symptoms of RAD, but not of DSED, decline when the extremes of insufficient care are ended by placement of the children in adoption or foster care.

Risk factors and consequences

Schuengel and Janssen (2006) developed an explanatory model for the high prevalence of psychopathology and behavioural problems among people with a mild intellectual disability, in which the elements for attachment problems and stress regulation have been given a leading role. The characteristic cognitive limitations typical for this group of people mean that everyday situations are experienced as problematic and stressful sooner and more frequently. People with a mild intellectual disability may more often lack overview, perspective, reflection and verbal strategies for understanding new or more complex social situations and for resolving them adequately. In addition, the lack of a support network, and having negative social experiences make these people vulnerable in social situations. The repeated confrontation with stressful situations not resulting in socially adequate solutions strengthens the negative self-image of the person and the perception of a threatening environment. A secure attachment development may be a protective factor; however, Janssen, Schuengel and Stolk (2002) documented that people with an intellectual disability actually run a higher risk of developing attachment problems or disorders. The potential causes they listed are: reduced sensitivity among the parents, coping and parenting problems among the

parents of a child with an intellectual disability, potential (intellectual) disabilities among the parents themselves, increased likelihood of out-of-home placement (institutionalisation) of the child with an intellectual disability. All these factors are likely to accelerate the downward cyclical process from emotional deregulation towards defective behaviour and psychopathology.

Various studies confirm the overriding risk associated with disrupted attachment of the development of emotional and behavioural disorders (MacLean, 2003). Bos et al. (2011) established a clear relationship between severe early deprivation and neglect on the health and development of the child. They found high levels of internalising disorders (anxiety, depression) and externalising disorders (ADHD, ODD and behavioural disorder) among these children. In the Romania Project, Zeanah et al. (2009) found a prevalence of psychiatric disorders of 53.2% among children with a background of institutionalisation, compared with 22% among children without such a background.

Dekker and Koot (2004) demonstrated that the presence of an intellectual disability also markedly increased the risk of developing severe psychopathology. They showed that the risk of mental or behavioural problems (psychopathology) in intellectually disabled youth increased 3 to 4-fold as compared to youth without an intellectual disability. Without a clear assessment procedure with diagnostic measuring instruments to diagnose attachment related disorders, it is not possible in clinical practice to adequately explain the diagnosed psychiatric disorders from the perspective of impaired attachment development. The introduction of attachment disorder diagnostics could change our views on the socio-relational inadequate behaviours as in the diagnosis of Pervasive Development Disorders (PDD) or Behavioural Disorders. Moreover, the diagnosis of RAD or DSED (whether comorbid or not) may be vital to the (therapeutic) strategy (complementing or replacing regular treatment of the comorbid disorder) and consequently the developmental opportunities of the youth. While no specific treatments exist that have been shown effective to reduce RAD or DSED, these diagnoses may be used to justify interventions aimed at improving the conditions for adaptive attachment relationships to develop (Zeanah et al., 2016).

Aims of this study

The aim of this study project was to contribute to the description and improvement of the clinical mental health practice of diagnostic assessment and intervention of the diagnosis of attachment related disorders, as described in DSM-IV (RAD inhibited type and RAD disinhibited type) and the DSM-5 (RAD and DSED). This study included children aged 5-11 years at baseline, mentally subnormal or with a mild intellectual disability, who had been referred for child psychiatric consultation. The unacceptable gap between needed and available research and mental health care facilities for children with intellectual disability (Dekker & Koot, 2004), in combination with the assumption that this is a high-risk group for development of disordered attachment (Green & Goldwyn, 2002; Schuengel & Janssen, 2006) is the primary motivation and justification for choosing this specific group of children in this study. In the Netherlands, the majority of people with intellectual disability, referred for psychiatric consultation because of mental health or behavioural problems, are in the age range of 5 to 22 years (Woittiez, Ras, & Oudijk, 2012). Expert diagnostic assessment of attachment related disorders in young and school-aged children is of great importance, for unidentified disordered attachment and therefore deprivation of necessary appropriate care and treatment, will increase the risk for serious psychopathology, criminality, and addiction. This explains the focus on the age-group of 5-12 in this study.

The second aim of this project was to collect and disseminate knowledge, contribute to guidelines, and training of professionals to lift the knowledge and clinical expertise of attachment, attachment related disorders and assessment practice, in particular concerning caregivers and clinicians of children with intellectual disability.

The first part of the study addressed the prevalence of behavioural symptoms of impaired attachment in children with a mild intellectual disability, and whether these symptoms overlapped with symptoms of ASD. This study objective is relevant in view of the exclusion criterion of ASD for the diagnosis of RAD (DSM-5).

In the second part of the study, we tested a protocol to diagnose attachment related disorders according to the guidelines formulated in the internationally recognized Practice parameter for the assessment and treatment of children and adolescents with attachment related disorders (AACAP, 2005, 2016). The objective was to evaluate the

associations among the various protocolled measurement instruments for the diagnostics of impaired attachment. Associations were analysed between the screening instrument (Disturbances of Attachment Interview, DAI, Smyke & Zeanah, 1999) for behavioural symptoms of disturbed attachment, the observation instrument (Clinical Observation of Attachment 6-12, COA and List Behavioral Signs of Disturbed Attachment, BSDA) for behavioural characteristics of disordered attachment, and information from the child's development history that might suggest insufficient care in the first years of life. With this information, it should be possible to diagnose attachment related disorder and thereby establish the prevalence for this specific target group.

Additionally, this study described the comorbidity with other psychiatric disorders (as per DSM-IV) in children with (and without) the diagnosis of RAD and/or DSED. We also compared the functional level of children with the diagnosis of RAD and/or DSED with children who did not receive this diagnosis.

The final chapter summarizes and integrates the findings of this study project and discusses the clinical implications and recommends a stepped-care model for diagnosing attachment related disorders.

Method

This is an observational study, where the parents of children aged 5 to 11 years were invited to participate in this study project. The parents have contacted one of the nine centres for child and adolescent psychiatry specialised in the diagnosis and treatment of children with an intellectual disability. Parents requested child psychiatric diagnostics in view of mental and/or behavioural problems in the child. These centres did not preselect parents or children for participation. After the first intake interview, parents were invited to participate in the study project separately from the regular diagnostics for their child. This participation consisted of a telephone interview (DAI) with one of the parents and the completion of a questionnaire by the child's current teacher (AUTI-R). The diagnostician of the diagnostic centre tested the child's intelligence level and communicated the results with the study researcher. Exclusion criteria for participation were a total intelligence test score outside the IQ 50-85 range, or if the two parents or the child did not speak Dutch. Collected data were used for the first study.

After data collection for the first part of the study was completed, the parents of this study group were approached again after 1 - 1.5 years to participate in the second part of the study. Participation consisted of another telephone DAI with one of the parents, the administration of a structured interview (Diagnostic Interview Schedule for Children, version IV, DISC-IV) with one of the parents by a trained research assistant in the home, completion of a paper questionnaire (the Dutch version of the Developmental Behaviour Checklist, DBC) by both the parent (DBC-P) and the child's teacher (DBC-T), the completion of a paper questionnaire by the parent (Vineland Screener), and a protocolled clinical observation (Clinical Observation of Attachment, COA) in a child therapeutic room with one of the parents and the child. The parents also gave the researcher permission to retrieve information on the child's personal living situation and developmental history from the child psychiatric centre's medical file.

The researchers administering and scoring the DAI and COA have been trained extensively by Neil Boris, co-author of the Practice Parameter (AACAP, 2005) and developer of the COA. The researchers have demonstrated their reliability. The BSDA has been coded by five coders, trained by the two DAI/COA-trained researchers and subsequently proven reliable. They observed and made meticulously detailed descriptions of the behaviours, conversations and interactions between child, caregiver and stranger in the video-taped COA's, in total 110 hours, and used the BSDA for coding the COA's. The research assistant who abstracted information from the case files of the children was instructed by the author of this dissertation. Two researchers coded the information on insufficient care; the interrater reliability between these two coders was high. The interviewer administering the DISC-IV has been trained by trainers of Robert Ferdinand, co-author of the DISC-IV-P.

Medical ethical approval was obtained from the VU University Medical Centre Medical-Ethical Review Board. This board is licensed to approve research by the Central Committee on Research Involving Human Subjects, which monitors compliance with Dutch legislation on medical research.

Thesis outline

The studies of this project have been conducted over a period of 12 years. During this period the meaning and the clinical definitions of the attachment related disorders have been subject to change. At the start of the study the DSM-IV-TR (APA, 2000) was the leading reference for the classification and diagnostic criteria of psychiatric disorders, and the AACAP Practice Parameter for the assessment and treatment of Reactive Attachment Disorder (AACAP, 2005) as the leading guideline for assessment, was recently published. And now, finishing the study, the DSM-5 (APA, 2013) has replaced the DSM-IV-TR, and the AACAP Practice Parameter has been revised, bolstered with new research findings relevant to the attachment related disorders (Zeanah et al., 2016). As a result of these changes the terminology and the reference to the DSM-5 in this chapter differ from the other chapters. In the Discussion chapter we will come back to these changes.

Chapter 2 examines the prevalence of the behavioural characteristics of disordered attachment behaviour in children with a mild intellectual disability who have been referred for child psychiatric assessment, and analyses the potential overlap between behavioural signs of disordered attachment and of PDD.

Chapter 3 examines the extent to which a diagnostic assessment consisting of structured observations, clinical interviews and file research of extremes of insufficient care are interrelated and contribute to the diagnosis of attachment disorder.

Chapter 4 addresses the prevalence of comorbid psychopathology in children from the study group with diagnosed attachment disorder and compares the level of adaptive functioning in children with an attachment disorder versus children without attachment disorder.

Chapter 5 contains the summary and discussion of the results of the studies, all in light of the main aim of the study: to contribute to the description and improvement of the clinical mental health practice of diagnostic assessment and intervention of the diagnosis of attachment related disorders. Limitations of this study and implications for further research and practice are discussed. This chapter ends with the description of a

stepped-care guideline for the clinical assessment of attachment related disorders of children with intellectual disability and psychological or behavioural problems.

The Protocol of the Clinical Observation of Attachment 6-12 (COA 6-12, Giltaij & Sterkenburg), used in this study as a diagnostic measurement instrument, is included in this manuscript as appendix, in English and Dutch version.

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Chapter 2

Psychiatric diagnostic screening of social maladaptive behaviour in children with mild intellectual disability: differentiating disordered attachment and pervasive developmental disorder behaviour

Giltaij, H.P., Sterkenburg, P.S., & Schuengel, C. (2015). Psychiatric diagnostic screening of social maladaptive behaviour in children with mild intellectual disability: differentiating disordered attachment and pervasive developmental disorder behaviour. *Journal of Intellectual Disability Research, 59, 138-149.*

Abstract

Background Children with intellectual disability (ID) are at risk for maladaptive development of social relatedness. Controversy exists whether Pervasive Developmental Disorder (PDD) takes precedence over disordered attachment for describing maladaptive social behaviour. The aim of this study was to assess the prevalence of disordered attachment symptoms in children with ID referred for mental health diagnosis, and to study the potential diagnostic overlap between symptoms of disordered attachment and PDD symptoms.

Method Children (*n* = 102) in the age of 5 to 11 with intellectual disabilities (borderline or mild; IQ 50-85) referred for psychiatric consultation were recruited. Parents were administered a screening interview for disturbances of attachment and teachers were administered a screening questionnaire for PDD.

Results Behavioural symptoms of disordered attachment were reported for 38% of the children, with 20% showing symptoms of inhibited and 29% showing symptoms of disinhibited attachment disorder. In 11% both types of symptoms were noted. Attachment disorder symptoms were not significantly associated with gender, ethnic background or age. Positive screening for PDD occurred for 27%. Positive screening for PDD was not significantly associated with symptoms of attachment disorder.

Conclusions Among children with ID referred for psychiatric consultation, Reactive Attachment Disorder (RAD) and PDD symptoms were both highly prevalent. RAD and PDD symptoms appear as distinct, but sometimes comorbid, forms of aberrant social relatedness.

Keywords children with intellectual disability, disordered attachment, Disturbances of Attachment Interview, Pervasive Developmental Disorders (PDD), Reactive Attachment Disorder (RAD)

Introduction

Children with intellectual disability (ID) are at a three to four-fold higher risk of mental health disorders than children without disabilities (Einfeld *et al.* 2011). More than other children, problem behaviour of children with ID manifests itself in social problems (Dekker *et al.* 2002; Bielecki & Swender 2004). In addition, socially withdrawn and autistic-like behaviours are also highly prevalent (Dekker *et al.* 2006). Psychiatric nosology offers at least two categories that can be used to diagnose the problems of these children, namely Pervasive Developmental Disorders (or Autism Spectrum Disorder) and Reactive Attachment Disorder (RAD) (World Health Organization 1992; American Psychiatric Association 1994). Currently, both categories can only be used exclusively, meaning that if a diagnosis of Pervasive Developmental Disorder (PDD) is made, no diagnosis of RAD should be made. This requirement is controversial. O'Connor & Zeanah (2003) state that there are insufficient data to determine whether RAD and PDD can co-occur or whether it is reasonable that PDD excludes RAD. In reviewing the recommendations for updating diagnostic criteria for RAD, Zeanah & Gleason (2010) noted that children with PDD and intellectual disabilities can develop normal attachment relationships, and therefore may develop comorbid disorders of attachment if they are exposed to pathogenic environments such as disruptions of attachment relationships, abuse, or neglect. In fact, children with ID (many of whom are diagnosed with PDD) may be at heightened risk for experiencing such environments and be more vulnerable for the effects (Schuengel & Janssen 2006; Minnis *et al.* 2010). Therefore, the need exists for effective and research-based screening instruments and diagnostic rules, because both disorders are in the domain of social functioning while having highly different probable aetiological pathways and highly different treatment implications. The current study investigated the prevalence of symptoms of disordered attachment in clinical practice, relative to symptoms of PDD, and studied the extent to which symptoms could be discriminated using behavioural screening.

Attachment is conceptualised as the affective bond or tie that develops between persons as a result of regular caretaking and provision of emotional security (Bowlby 1969/1982; Ainsworth *et al.* 1978; Cassidy & Shaver 1999; Rutgers *et al.* 2004). Infants have inborn tendencies to cry, cling to, and orient towards caregivers when they are distressed, afraid, or unwell. Insofar as these attachment behaviours lead to soothing caregiver responses, supporting the infants' regulation of emotions, and supporting

infants' return towards exploration, infants develop an open style of affective signalling with this caregiver. If this is the case, their relationship is called secure (Ainsworth *et al* 1978). Consistent low sensitivity or rejection on the part of the caregiver is associated with an avoidant quality of relationships, in which infants show less active attachment behaviour and limit the expression of attachment-related affect. Inconsistent sensitivity is associated with infant hypervigilance as to the caregiver's whereabouts and overexpression of attachment needs, leading to a resistant attachment pattern. In attachment relationships tainted with fear, disorganisation and disorientation of attachment behaviour can be observed (Main & Solomon 1986). Children may develop multiple attachment relationships with the people around them, with differing attachment qualities. Children in so-called insecure attachment relationships appear to adapt to a given extent to less than optimal caregiving, so that anxiety increasing interactions with their attachment figures are minimised. The insecure and even the disorganised patterns should not, therefore, be seen as inherently pathological, although they are modestly associated with elevated risks for the development of internalising and externalising behaviour problems (Fearon *et al.* 2010; Groh *et al.* 2012).

A small number of children show highly aberrant attachment behaviours or appear to ward off caregiving interactions altogether, behaviours which do not seem confined to single relationships (Zeanah *et al.* 2011). Clinical descriptions of a disturbing lack of affective responses to familiar caregivers were based on early work on children growing up in institutions or exposed to severe neglect (Spitz 1945; Tizard 1977; George & Main 1979). Tizard & Rees (1975) described two patterns. One was an emotionally withdrawn pattern, in which no comfort is sought nor accepted, especially when the child appeared to be in pain or distress. Another was an indiscriminate, disinhibited sociable pattern, in which children readily sought intimate, often physical contact with strangers, and would venture away from their caregivers without checking. These patterns were not only observed in institutionalised children, but later also in maltreated children (Boris *et al.* 1998; Albus & Dozier 1999; Zeanah & Boris 2000; Zeanah *et al.* 2001). These observations formed the basis for defining RAD of Infancy and early childhood as a disorder in the third edition of the *Diagnostic and Statistical Manual of Mental Disorders* (DSM-III) (American Psychiatric Association 1980), later revised for the DSM-III-R (American Psychiatric Association 1987) and DSM-IV (American Psychiatric Association 1994). In the DSM-IV (pp. 116-118) and DSM-IV-TR (American Psychiatric Association

2000, pp. 127-130) Reactive Attachment Disorder is defined as 'markedly disturbed and developmentally inappropriate social relatedness in most contexts, beginning before 5 years of age', classified as either a social withdrawn/inhibited type or an indiscriminately social/disinhibited type. In the International Statistical Classification of Diseases and Related Health Problems, 10th Revision (ICD-10) (World Health Organization 1992, p. 280) the two patterns of attachment disordered behaviour are divided into two distinctive disorders, the RAD, similar to the DSM-IV emotionally withdrawn/inhibited subtype of RAD and the Disinhibited Attachment Disorder (DAD), similar to the DSM-IV indiscriminately social/disinhibited subtype of RAD.

The presumption that the disturbed social behaviour is 'reactive' to exposure from pathogenic care led to the stipulation that the disordered behaviour should not be 'accounted for solely by developmental delay (as in mental retardation)' (DSM-IV; p. 118) and 'does not meet criteria for a pervasive developmental disorder" (DSM-IV; p.118, ICD-10; p. 280). The ICD-10 has a clear description of how RAD can be distinguished from PDD. Five main features differentiate this condition from pervasive developmental disorders. 'First, children with a reactive attachment disorder have a normal capacity for social reciprocity and responsiveness, whereas those with a pervasive developmental disorder do not. Second, although the abnormal patterns of social responses in a reactive attachment disorder are initially a general feature of the child's behaviour in a variety of situations, they remit to a major degree if the child is placed in a normal rearing environment that provides continuity in responsive care-giving. This does not occur with pervasive developmental disorders. Third, although children with reactive attachment disorders may show impaired language development (of the type described under F80.1), they do not exhibit the abnormal qualities of communication characteristic of autism. Fourth, unlike autism, reactive attachment disorder is not associated with persistent and severe cognitive deficits that do not respond appreciably to environmental change. Fifth, persistently restricted, repetitive, and stereotyped patterns of behaviour, interests and activities are not a feature of reactive attachment disorders.' (World Health Organization 1992; p. 280). Although the ICD-10 describes the differences in behaviour and development between RAD and PDD, it is unknown to what extent differentiation is possible at the level of symptoms. Therefore the question remains whether a child could be comorbid for both PDD and RAD (Zeanah & Gleason 2010).

Studies of children growing up with a heightened likelihood of exposure to pathogenic environments have focused on children in orphanages (Smyke *et al.* 2002; Zeanah *et al.* 2005), children adopted from institutions (Rutter *et al.* 2007a), children in women's shelters (Boris *et al.* 2004), and foster children (Zeanah *et al.* 2004; Oosterman & Schuengel 2007). Percentages of children showing symptoms of disorders of attachment ranged from 18 to 56%. Green & Goldwyn (2002) noted that individuals with an ID may be a high risk group as well, because people with ID are more likely to experience institutionalised care and maltreatment than the general population. Furthermore, depending on the severity of the disability and the levels of care and support, parents and professional caregivers may have more difficulty identifying and meeting the affective, social, cognitive, and physical needs of people with ID, increasing the risk of inadvertent neglect of these basic needs (Janssen *et al.* 2002). Minnis *et al.* (2010) administered the Relationship Problems Questionnaire (RPQ) to the professional caregivers of 50 institutionalised adults (mean age 48 years; 56% profound ID). Symptoms of RAD were negatively associated with increasing age and positively associated with childhood adversity (childhood experience of abuse, severe neglect, or other serious adverse experience). No association was found between RAD symptoms and level of ID, or with years of institutional living prior to age 18. No direct comparison with other populations was undertaken, but the mean rate of disordered attachment behaviour was considerably higher than the rate found with the same instrument in a large general population sample of twin children (Minnis *et al.* 2007). Studies on prevalence of disordered attachment behaviour of children with ID have not been reported, however.

Autism, also known as Autistic Spectrum Disorder (ASD) or Pervasive Developmental Disorder (PDD), is a severe neurodevelopmental disorder involving abnormalities and deficits in social interactions, communication, conduct, and cognition. Main diagnostic criteria describe (1) qualitative impairments in social interaction, (2) qualitative impairments in verbal and non-verbal communication and (3) restricted and stereotyped patterns of behaviour, interests and activities (American Psychiatric Association 1994). The prevalence of PDD in people with ID has been estimated by Kraijer (1997) to be 38% and by Morgan *et al.* (2002) to be 30%, with an inverse association between prevalence and level of intellectual functioning.

Aetiological research on PDD has shown associations between PDD and genetic factors, prenatal and perinatal complications (Gupta & State 2007; Ploeger *et al.* 2010; Rommelse *et al.* 2011). Male gender appears to be a strong risk factor (male/female ratio of 4.3:1) (Fombonne 2003; Newschaffer *et al.* 2007). There is no evidence of associations between PDD and socioeconomic status, parental wealth, or education (Muhle *et al.* 2004; Newschaffer *et al.* 2007). Despite the differences in aetiological background between PDD and disorders of attachment, early studies have described phenotypic similarities between disordered behaviours observable in institutionalised children and children with PDD. Rutter *et al.* (1999), and Rutter *et al.* (2001) examined the behaviour of institutionalised children from Eastern European countries, who were later adopted. Rutter *et al.* (1999) referred to a complex of observed behavioural disturbances as 'quasi-autistic patterns'. Rutter *et al.* (2007b) found quasi-autistic patterns in over 11% of the children who experienced profound institutional deprivation. Rutter *et al.* (2001) showed that in the population of the Romanian children adopted out of institutional privation, there was an association between the domains attachment problems, inattention/overactivity, quasi-autistic problems and cognitive impairment. This association was not seen in the comparison group of non-deprived within-UK adoptees. The maladaptive social behaviours observed by Rutter *et al.* (1999) and Rutter *et al.* (2001) occurred across the spectrum of PDD-behaviour (disturbed behaviour in social interaction, disturbed behaviour in verbal and non-verbal communication and restricted and stereotyped patterns of behaviour, interests and activities). Improvement of children's environmental circumstances, such as placement in a well-functioning adoptive family or treatment setting, reduced autistic-like symptoms (Rutter *et al.* 1999, 2001, 2007b; Hoksbergen *et al.* 2005). If disturbed social behaviour of children exposed to pathogenic environments shows similarity to the behaviour of children with PDD, it is important to ask whether diagnostic screening in clinical practice may distinguish between symptom patterns for PDD and disorders of attachment.

This study focused on children with a mild or borderline ID who were referred for psychiatric assessment. High prevalence was expected of PDD (Morgan *et al.* 2002; Fombonne 2003), while given the possibility of neglect, abuse, and relationship disruptions (Stalker & McArthur 2012), symptoms of RAD were expected as well. First, the prevalence of symptoms of both types of disorders was investigated. Second, the associations between symptoms of both types of disorders were studied, given concerns that symptoms of PDD may be confused with symptoms of disordered attachment.

Whether inhibited and disinhibited symptoms of disordered attachment reflect subtypes of one disorder of attachment, or rather reflect reactive attachment disorder proper (indicated by severe inhibition of expectable attachment behaviour) or a separate disorder of disinhibited social engagement, remains controversial (Zeanah & Gleason 2010; Gleason *et al.* 2011)[1]. Therefore, the associations with PDD symptoms were examined separately for inhibited and disinhibited symptoms.

Method

Participants

The sample included 102 children recruited from nine Dutch centres for mental health care, specialised in psychiatric assessment and treatment of children with IDs. These centres were scattered over eight of the 11 provinces of The Netherlands. Children referred for (psychiatric) diagnostic consultation because of psychological, learning and/or behavioural dysfunctioning to one of these specialised centres were invited to participate. The centres' psychiatrists and psychologists were instructed not to preselect cases. Informed consent letters were returned by 146 parents. Inclusion criteria were: child's age (between 5 and 11 years at the moment of assessment), lowered intellectual functioning (measured IQ between 50 and 85, borderline and mild ID), and ability to communicate in Dutch. IQ testing after recruitment led to exclusion of 31 children. Failure to respond to either one of both screening instruments led to loss of another 13 participants, giving a total of 102 participants.

The mean age of the participants was 8.8 year (*SD* = 1.7); 72 children were boys. The mean level of cognitive functioning (IQ) was 71.7 (*SD* = 9.7); 41 (40%) children had a mild ID (IQ 50-70) and 61 (60%) children showed borderline intellectual functioning (IQ 71-85). Table 1 shows that 83 of the 102 participating children had Dutch parents, 19 children had at least one non-Dutch parent (Morocco 6, Turkey 2, Surinam 4).

1 Shortly after publication of this chapter the American Psychiatric Association (APA, 2013) decided to designate Reactive Attachment Disorder (DSM-IV) as two distinct disorders in the DSM-5: Reactive Attachment Disorder and Disinhibited Social Engagement Disorder.

Table 1 Demographic Characteristics and IQ-scores of Participants (*n* = 102)

Categories	Characteristics	*n*	%	*IQ score M (SD)*
Ethnicity	Dutch	83	81.4	72.2 (9.8)
	Non-Dutch, western	3	2.9	68.7 (10.0)
	Non-Dutch, non-western	16	15.7	69.3 (9.8)
Gender	Girl	30	29.4	71.0 (10.0)
	Boy	72	70.6	72.0 (9.7)

Note. M = Mean, *SD* = standard deviation

Medical ethical approval was obtained from the VU University Medical Centre Medical-Ethical Review Board. This board is licensed to approve research by the Central Committee on Research Involving Human Subjects, which monitors compliance with Dutch legislation on medical research. The participants received a small present (voucher) after completing the interview and returning the questionnaires.

Instruments

Symptoms of disordered attachment.

Disordered attachment symptoms were measured using the Disturbances of Attachment Interview (DAI; Smyke & Zeanah 1999, Smyke *et al.* 2002; Gleason *et al*. 2011). The DAI is a 12-item semi-structured interview. The first five items rate behaviours relevant to the RAD inhibited/emotionally withdrawn type: (1) absence of a discriminated, preferred adult, (2) lack of comfort seeking for distress, (3) failure to respond to comfort when offered, (4) lack of social and emotional reciprocity, and (5) emotion regulation difficulties. The next three items rate signs of the RAD disinhibited/indiscriminate type: (6) not checking back after venturing away from the caregiver, (7) lack of reticence with unfamiliar adults, and (8) a willingness to go off with relative strangers. The final four items assess distortions of secure base behaviour not currently described in psychiatric classifications, including (9) self-endangering behaviours, (10) excessive clinging behaviour, (11) hypervigilance and (12) role-reversal. Trained interviewers probed the respondent to acquire enough information on the children's behaviour so that they could rate each item as '0' = 'no sign of RAD', '1'= 'somewhat or sometimes a sign of RAD' and '2'= 'considerable or frequently a sign of RAD'. Previous studies have shown good internal consistency and inter-rater reliability (Smyke *et al.* 2002; Zeanah *et al.* 2002, 2004, 2005; Oosterman & Schuengel 2007). The DAI scores converged with similar

measures used in other studies of signs of RAD (Chisholm 1998; O'Connor *et al.* 2000) and diverged from measures of aggression, stereotypies, and language development (Smyke *et al.* 2002). Although the DAI was not developed specifically for children with ID, Smyke *et al.* (2002) and Zeanah *et al.* (2005) reported good reliability in their study with Romanian orphanage children, samples in which IQ ranged between the moderate ID to normal intelligence. DAI scores were found to be highly predictive of RAD diagnosis using an independent psychiatric assessment (Boris *et al.* 2004; Gleason *et al.* 2011). Gleason *et al.* (2011) examined the reliability ($\kappa = 0.88$) and validity (Cronbach $\alpha = 0.83$ and 0.80, respectively) of the DAI criteria for RAD Inhibited and RAD Disinhibited. Criterion validity, using the standardisation criteria of Gleason *et al.* (2011), was demonstrated by convergence of diagnoses with a psychiatric diagnostic interview (Preschool Age Psychiatric Assessment, PAPA) and an observational measurement (Stranger-at-the-Door Procedure). DAI and PAPA showed concordance in 86% for RAD Disinhibited and 98% for RAD Inhibited (Fisher exact test, $p \leq 0.001$); DAI and the Stranger-at-the-Door test showed concordance in 85% for RAD Disinhibited (Fisher exact test, $p \leq 0.001$). Thus, the reliability and validity of the DAI are empirically supported.

The DAI is not normed, and therefore criteria for positive and negative screen for disorders of attachment are not fixed. Findings by Oosterman & Schuengel (2007) suggested that only clear symptoms should be considered indicative of a disorder of attachment, whereas Gleason *et al.* (2011) used the sum of minor and clear symptoms to determine the screening. Given the validity evidence provided by Gleason *et al.* (2011), we used their coding criteria.

In the current study, internal consistency analyses yielded Cronbach's alpha of 0.90 for DAI items 1 to 8. Inter-rater reliability was analysed in 10 randomly selected DAI transcripts coded by the first two authors. The RAD inhibited/emotional withdrawn inter-rater reliability analyses reached an intraclass correlation of 0.88 and RAD disinhibited/indiscriminate an intraclass correlation of 0.98.

Symptoms of Pervasive Developmental Disorders.

The AUTI-Revised scale (AUTI-R; Van Berckelaer-Onnes & Hoekman 1991) was used to measure symptoms of PDD. The AUTI-R is a questionnaire to be filled in by professionals such as teachers, containing 51 questions with six possible replies in ranging order: "no,

never; only once or twice; sometimes; regularly; often; very frequently". The AUTI-R has a version for children aged 1-6 years and a version for children aged 6-12 years, and can be used for children with and without ID. The items cover seven areas: relationship disturbance (12 questions), language disturbance (15 questions), striking motor phenomena (5 questions), striking sensory phenomena (9 questions), resistance to change (6 questions), acute, illogical fears (2 questions), and two other questions. The children were classified in the non-autistic, the no-classification (borderline autistic) or the autistic group. AUTI-R-results were compared with the psychiatric diagnosis according to the DSM-classification. Inter-rater reliability of the AUTI-R was reported as good (r = 0.97; n = 73) and the criterion validity excellent (92% of the survey group, n = 220, was correctly classified) (Van Berckelaer-Onnes & Hoekman 1991; Evers *et al.* 2000). The AUTI-R has been used in a follow-up study with Romanian adoptees with a background of severe neglect (Hoksbergen *et al.* 2005). In that study 16% of the adopted children were classified in the autistic range (autistic or borderline autistic), exhibiting symptoms of autistic-like behaviour.

Procedure

After informed consent was obtained from parents or caregivers of the children, a telephone appointment with the parent or other primary caregiver of the child was made for the DAI. The DAI was audiorecorded and verbatim transcribed. The transcriptions were then independently scored by two trained coders (the first and second author); discrepancies were resolved by discussion leading to a consensus code for each item. Parents were asked which teacher would know their child best, and this teacher received the AUTI-R by postal mail.

Data-Analysis

First, the frequencies of RAD and PDD classification were determined according to the DAI and AUTI-R, respectively. Second, the association between the demographic characteristics (gender, age, ethnicity and IQ) and classification of RAD symptoms and PDD symptoms were tested. For the continuous variables age and IQ the t-test for independent samples was used; for the categorical variables gender and ethnicity the chi-squared test was used. Third, the association between the RAD (total scale, RAD inhibited and RAD disinhibited) and PDD (non-PDD, PDD, Borderline PDD) classifications were computed with a Pearson chi-squared test. In order to comprehensively test for clusters of symptoms that would lack discriminant validity, an additional set of analyses

was done on item cluster level. A set of t-tests compared the average scores on AUTI-R item clusters (relationship disturbance, language disturbance, striking motor phenomena, striking sensory phenomena, resistance to change, acute, illogical fears) between children with a classification of RAD Inhibited and children without a classification of RAD Inhibited, followed by the same set of t-tests comparing children with a classification of RAD Disinhibited and children without a classification of RAD Disinhibited. Cohen's *d* effect sizes were computed.

Results

Prevalence of disordered attachment symptoms

Based on the cut-off criteria of Gleason *et al.* (2011), probable RAD was found in 43 out of the 102 children (42%). Among these 43, 16 children had at least three minor or clear symptoms (score 1 or 2) of RAD Inhibited and 38 children at least two minor or clear symptoms (score 1 or 2) of RAD Disinhibited; 11 showed enough symptoms for classification of RAD Inhibited as well as RAD Disinhibited (Table 2). There were no significant associations between RAD classifications and gender, ethnic background, IQ or age.

Table 2 Frequency of the RAD Classifications (DAI) and IQ level (*n* = 102)

		IQ 50-70	IQ 71-85
RAD Classification		***n***	
No RAD	59	26	33
RAD Inhibited	5	1	4
RAD Disinhibited	27	10	17
RAD Mixed (Inhib + Disinh)	11	4	7

RAD, reactive attachment disorder; DAI, disturbances of attachment interview.

Prevalence of pervasive developmental disorder symptoms

Based on the AUTI-R, 28 (27%) of the 102 children showed enough symptoms of PDD for a possible or definite positive screen. A definite screen was found for four children; a possible screen for 24 children (Table 3). Boys showed significant more PDD symptoms than girls (X^2 (1, n = 102) = 4.253, p = 0.039), similar to epidemiological surveys by Fombonne (2003). The odds ratio for a possible or definite positive screen for boys was

3.25 (95% confidence interval = 1.02-10.38) in this sample. There were no significant associations between PDD classification and IQ, ethnic background, nor for age.

Table 3 Frequencies of the RAD and PDD Classifications (*n* = 102)

		PDD Classification	
RAD Classification	***n***	**No PDD (*n* = 74)**	**(Borderline) PDD (*n* = 28)**
No RAD	59	45	14
RAD Inhibited	5	2	3
RAD Disinhited	27	18	9
RAD Mixed (Inhib + Disinh)	11	9	2

RAD, reactive attachment disorder; PDD, pervasive developmental disorder.

Associations between symptoms of reactive attachment disorder and pervasive developmental disorder

As there were only four children with a definite positive screen for PDD, this group was added to the children with a possible PDD screen, the (borderline) PDD group (*n* = 24). No significant association (X^2 (1, *n* = 102) = 0.97, *p* = 0.32, Phi = 0.10) was found between RAD (inhibited or disinhibited) and PDD [no PDD, PDD and (borderline) PDD, *n* = 28]), nor between RAD Inhibited (versus no RAD or RAD Disinhibited) and PDD classifications (X^2 (1, *n* = 102) = 0.14, *p* = 0.71, Phi = 0.04) and between RAD Disinhibited (versus no RAD or RAD Inhibited) and PDD classifications (X^2 (1, *n* = 102) = 0.07, *p* = 0.79, Phi = 0.03). Of the 43 children with a positive RAD-screen, 14 children (33%) showed symptoms in the possible or definite range for a PDD diagnosis. Of the 59 children with a negative RAD-screen, 14 children (24%) showed symptoms in the possible or definite range for a PDD diagnosis (Table 4). Positive screening for (probable or possible) PDD was not associated with positive screening for (probable or possible) RAD. Scoring with Oosterman & Schuengel (2007) criteria [at least one clear symptom (score 2) of RAD] had similar results.

Table 4 Frequency of RAD Classifications (RAD, No RAD, RAD Inhibited and RAD Disinhibited) and PDD Classifications (No PDD and (Borderline) PDD) (*n* = 102)

	PDD Classification			
	No PDD (*n* = 74)		(Borderline) PDD (*n* = 28)	
RAD Classification	***n***	**%**	***n***	**%**
RAD inhibited and/or disinhibited (*n* = 43)	29	67	14	33
No RAD (*n* = 59)	45	76	14	24
RAD Inhibited (*n* = 16)	11	69	5	31
No RAD Inhibited (*n* = 86)	63	73	23	27
RAD Disinhibited (*n* = 38)	27	71	11	29
No RAD Disinhibited (*n* = 64)	47	73	17	27

RAD, reactive attachment disorder; PDD, pervasive developmental disorder.

A set of t-tests for independent samples compared the incidence of the six AUTI-R cluster areas of PDD symptoms between children who screened positive on the classification of RAD Inhibited and children not screening positive on RAD Inhibited. The same set of t-tests was done between children who screened positive on the classification of RAD Disinhibited and children not screening positive on RAD Disinhibited. No significant associations were found (Table 5).

Table 5 Comparison Means of AUTI-R clusters areas between participants with RAD (inhibited or disinhibited) and no RAD (inhibited or disinhibited) behaviour (n = 102)

	RAD Classification									
	RAD Inhibited		No RAD Inhibited			RAD Disinhibited		No RAD Disinhibited		
AUTI-R clusters	**M**	**SD**	**M**	**SD**	**Cohen's *d***	**M**	**SD**	**M**	**SD**	**Cohen's *d***
Relationship disturbance	2.8	.57	2.8	.78	0.04	2.7	.73	2.9	.76	0.18
Language disturbance	2.2	.53	2.2	.77	0.09	2.3	.65	2.3	.79	0.03
Striking Motor phenomena	2.2	1.19	2.2	.95	0.10	2.3	1.11	2.2	.90	0.11
Striking Sensory phenomena	1.5	.73	1.5	.71	0.0	1.6	.78	1.5	.67	0.0
Resistance to change	2.0	.74	2.0	.91	0.0	2.0	.83	2.0	.93	0.01
Acute illogical fears	1.5	.85	1.6	1.14	0.16	1.4	.96	1.7	1.17	0.24

M = mean, DS = standard deviation

Discussion

Children with a mild or borderline ID, when referred for psychiatric assessment, showed high prevalence of disturbed social behaviour. Clear symptoms of disturbed attachment behaviour (RAD) were determined in four out of 10 children, while almost three out of 10 children showed enough symptoms of PDD that further diagnostic testing for PDD would be warranted. Positive screening of RAD symptoms could not be explained by the prevalence of PDD in this sample of children with borderline or mild IDs. Children with a classification of PDD were not more, but also not less, likely than others to display symptoms of RAD (Table 3). Therefore, RAD and PDD may together describe a significant portion of the social dysfunctioning of children with mild and borderline ID, and in 14% of cases, children appear to suffer from both types of problems. Important for the ongoing discussion of diagnostic criteria, the findings do not support using PDD as an exclusion criterion for the diagnosis of RAD.

In their review study Schuengel & Janssen (2006) described the underlying factors that might explain the high prevalence of problematic attachment, challenging behaviour and psychopathology in people with ID. Impaired affect regulation, vulnerability for stress, and deficient problem-solving skills (coping) are likely linked in people with ID. Support from attachment figures is therefore an important resilience factor. However,

caregivers may encounter considerable difficulties because of lack of adapted support, problematic interaction with the child, and needing time and resources to adapt psychologically to having a child with a disability. Current findings underscore the importance of pursuing these and other possible determinants of problems in attachment. Further studies are also needed to investigate whether children with ID who are not referred for psychiatric consultation may be at elevated risk for RAD symptoms as well.

The RAD is highly unlikely in the general population. Minnis *et al.* (2009) asked child mental health clinical teams and social workers in the Glasgow area (population of 28.000 children age 5-8) to refer children with symptoms of RAD. After clinical assessment 38 children were diagnosed as RAD. High prevalence rates of 56% RAD were found, however, in a study with children reared in profoundly deprived institutions in Romania (Smyke *et al.* 2002). Another study on toddlers in therapeutic foster care with a history of maltreatment found a 38% rate (Zeanah *et al.* 2004), whereas an 18% rate was reported in a study with toddlers and young children in regular foster care (Oosterman & Schuengel 2007). The current findings indicate that children with borderline or mild ID, referred for psychiatric assessment, are a high risk group as well.

As found in other studies (Gleason *et al.* 2011), symptoms of disinhibited RAD behaviour were more prevalent than symptoms of inhibited RAD. In addition, a substantial group of children showed symptoms of both types of RAD (Inhibited and Disinhibited). Although the DSM-IV prescribes assignment to either subtype but not both, Smyke *et al.* (2002) and Zeanah *et al.* (2004) also reported co-occurrence of inhibited and disinhibited symptoms. Zeanah & Gleason (2010) now propose to view inhibited and disinhibited symptoms as indicative for two distinct disorders rather than as indicative of either one or both subtypes of a single disorder. Both disorders still require the criterion of exposure to pathogenic care linked to symptom development. However, disinhibited, indiscriminately social behaviour symptoms are indicative of a disorder that could be more appropriately called Disinhibited Social Engagement Disorder, rather than attachment disorder, given evidence that children showing disinhibited symptoms were also found to develop selective attachment relationships to their foster caregivers (Gleason *et al.* 2011). In theory, these changes in the labelling of disorders open the possibility of having comorbid disorders of lack of attachment behaviour in combination with disinhibited social engagement. However, work should also focus on

the discriminant validity of these symptoms with respect to other disorders or personality characteristics.

The findings of this study support the statement of Zeanah & Gleason (2010) that overlap between RAD with PDD is possible. Although the criteria in the DSM-IV for Reactive Attachment Disorder require that the maladaptive social behaviour 'does not meet criteria for a pervasive developmental disorder' (p. 118), current findings show that PDD and RAD are unlikely to be confounded. Comorbidity between PDD and RAD could become, therefore, a diagnostic possibility. Furthermore, these results indicate that there is no lower or higher risk for RAD in children with PDD, at least in the population of children with ID.

Because the AUTI-R required teachers of the children as informants, while the DAI required parents or daily caregivers as respondents, shared informant bias was minimised. However, the lack of association between the two screening instruments can only be interpreted as evidence for discriminant validity insofar as both instruments show good criterion validity and inter-rater reliability. In previous studies DAI-results were compared with the outcome of concurrent diagnostic interviews and observational measurements (Gleason *et al.* 2011), the AUTI-R-results were compared with the psychiatric diagnosis according to the DSM-classification (Van Berckelaer-Onnes & Hoekman 1991). Inter-rater reliability of both instruments has proven good as was described in the Method section. The absence of any association between RAD and PDD symptoms on the level of total sums of symptoms as well as clusters and subtypes may therefore be substantively interpreted.

This study contributes to emerging insight in the different ways in which social functioning may become disordered. Findings suggest that not only neurodevelopmental processes but also environmentally mediated processes must be considered. For the field of practice, it is important that screening instruments, in this case AUTI-R and DAI, can help to differentiate maladaptive social behaviour patterns and provide guidance to further diagnostic investigation. Also the findings support the ongoing concern with potential difficulties in primary relationships of children with mild or borderline ID.

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Chapter 3

Convergence between observations and interviews in clinical diagnosis of reactive attachment disorder and disinhibited social engagement disorder

Giltaij, H.P., Sterkenburg, P.S., & Schuengel, C. (In press). Convergence between observations and interviews in clinical diagnosis of reactive attachment disorder and disinhibited social engagement disorder. *Clinical Child Psychology and Psychiatry.* DOI: 10.1177/1359104517709049

Abstract

Objective A comprehensive approach is needed for diagnosing disordered attachment behavior due to the multifaceted nature of attachment. Differences between various indicators can pose a challenge for deciding on the proper diagnosis. This study assessed the convergence between clinical interview assessment and observation-based clinical diagnosis, and their linkages with inadequate care.

Method Participating children ($N = 55$) had intelligence quotients (IQs) between 50 and 85 and were referred for psychiatric consultation. Data were obtained by structured review of medical records, the Disturbances of Attachment Interview (DAI), and the Clinical Observation of Attachment (COA) procedure.

Results Of the 18 children identified using the DAI with *Diagnostic and Statistical Manual of Mental Disorders-Fifth Edition* (DSM-5) diagnosis of Reactive Attachment Disorder (RAD) and/or Disinhibited Social Engagement Disorder (DSED), only 7 received a clinical DSM-5 diagnosis of RAD and/or DSED. Observed maladaptive attachment behavior in the COA was strongly associated with DAI scores and with clinical diagnosis of DSM-5 RAD and/or DSED. There was a significantly higher prevalence of extremes of insufficient care in children who were classified with RAD by DAI or DSM-5 and/or with DSED by DSM-5 compared to those with no attachment disorder.

Conclusions Using structured observation and record assessment leads to more conservative identification of RAD or DSED than using the DAI.

Keywords Attachment, Reactive Attachment Disorder (RAD), Disinhibited Social Engagement Disorder (DSED), assessment, low average intellectual functioning

Introduction

While research on attachment based on theoretical and empirical work by John Bowlby (e.g. Bowlby, 1984) and Mary Salter Ainsworth (e.g. Ainsworth, Blehar, Waters, & Wall, 1978) has transformed our overall understanding of socioemotional development and human relationships, progress has been more modest with respect to the etiology, diagnosis, and treatment of attachment-related disorders. The latest revision to a well-known definition of attachment-related disorders was the advent of the Diagnostic and Statistical Manual of Mental Disorders-5 (DSM-5; American Psychiatric Association, APA, 2013), 33 years after reactive attachment disorder (RAD) appeared in the DSM-III (APA, 1980). In the DSM-5, RAD is described as a pattern of disturbed social behavior similar to the emotionally withdrawn-inhibited subtype of RAD as defined in the previous edition of the DSM (DSM-IV) (APA, 1994). In contrast, the pattern previously subclassified as indiscriminate social-disinhibited RAD now qualifies for the diagnosis of Disinhibited Social Engagement Disorder (DSED). These changes were the result of theoretical and empirical work on the psychosocial functioning of children with backgrounds that were deemed to be pathogenic for attachment, such as children growing up in orphanages, foster care, and children exposed to maltreatment (Zeanah & Gleason, 2010, 2015).

The indicators of disordered attachment that diagnosticians in clinical practice should take into account have received relatively little research attention. While practice guidelines state that a variety of information sources should be accessed, all of which should yield converging data (Boris, Zeanah, & The Work Group on Quality Issues, 2005; Chaffin et al., 2006; Zeanah, Chesher, Boris, & The American Academy of Child and Adolescent Psychiatry (AACAP) Committee on Quality Issues (CQI), 2016; Zilberstein & Popper, 2014), the extent to which such convergence can be found in practice is largely unknown across clinical populations. As a result, there is little insight into the reliability and validity of diagnoses made using different approaches across populations.

The 2016 Practice Parameter of the American Academy of Child and Adolescent Psychiatry (Zeanah et al., 2016) for the assessment and treatment of children and adolescents with RAD and DSED (DSM-5) recommended taking a comprehensive history of the child's caregiving environment (foster care, adoption, institutional care or severe deprivation, and maltreatment), a history of the child's patterns of attachment behavior

plus direct in vivo clinical observation of children with familiar caregivers as well as with a stranger. A Clinical Observation of Attachment (COA) procedure, such as that developed and tested by Boris and colleagues (Boris et al., 2004), was recommended for this purpose. The American Professional Society on the Abuse of Children (APSAC) Task Force issued similar recommendations, adding that the child's behavior should be assessed across situations, contexts, and caregivers (Chaffin et al., 2006).

Structured clinical interviewing may also be used to collect data across contexts and caregivers, such as with the Disturbances of Attachment Interview (DAI; Smyke & Zeanah, 1999). The DAI has been used as a parent-report measure of disturbed attachment behavior in several studies in young at-risk children (Oosterman & Schuengel, 2007a; Smyke, Dumitrescu, & Zeanah, 2002; Zeanah et al., 2004; Zeanah, Smyke, & Dumitrescu, 2002; Zeanah, Smyke, Koga, & Carlson, 2005,) and has been examined on validity and reliability (Gleason et al., 2011; Oosterman & Schuengel, 2007a; Vervoort, De Schipper, Bosmans, & Verschueren, 2013).

Research on clinical observational methods used to assess symptoms of disordered attachment is more limited. Riley, Atlas-Corbett, and Lyons-Ruth (2005) developed an observational measure, the Rating of Infant and Stranger Engagement (RISE) based on the Strange Situation Procedure (SSP; Ainsworth, Blehar, Waters, & Wall, 1978). The SSP was originally developed to assess security of attachment by assessing variations in the way the child related to the parent after separation. In relation to the assessment of indiscriminate behavior, O'Connor and Zeanah (2003) argued that the underlying assumptions and traditional coding methods for assessing security of attachment are not adequate to capture disorders of attachment. Zeanah, Smyke, Koga, & Carlson (2005) and the BEIPCore Group found that 21% of children who were rated as secure on the SSP did not show fully developed attachment behaviors. This illustrates the likely inappropriateness of the traditional SSP coding scheme for children with attachment disorders, since those coding schemes assume the existence of a selective relationship and focus mostly on behavior toward the person assumed to be an attachment figure. To assess disinhibited social engagement, the focus should be not only on the behavior toward the parent but also on behavior toward a stranger. The RISE evaluates both the extent of the infant's affective engagement with the stranger compared to the caregiver and the extent to which the infant displays nonnormative acceptance of physical contact or response to soothing by the stranger. The RISE has been validated in studies

with at-risk home-reared toddlers (Lyons-Ruth, Bureau, Riley, & Atlas-Corbett, 2009) and institutionalized toddlers (Oliviera et al., 2012) by examining the convergence of observed disinhibited behavior of the young child with the (by the parent) reported indiscriminate behavior (in the DAI; Smyke & Zeanah, 1999).

One particular form of disinhibited behavior, namely leaving with a stranger without checking with the parent or caregiver, has been operationalized with the "stranger at the door procedure" (Gleason et al., 2011). Leaving with a stranger was associated with a caregiver report of disinhibited attachment behavior (Gleason et al., 2014). While this behavior can validate the caregiver's report of disinhibited attachment behavior, the stranger at the door procedure has limited clinical use. Studies using multi-informant diagnoses of disordered attachment are rare. McLaughlin, Epsie, and Minnis (2010) examined the reliability of a clinical observation schedule in a brief waiting room procedure. This observation procedure has recently been used with other parent report measures of disordered attachment and psychopathology as part in a large multi-informant study (N = 1654) to estimate the prevalence of RAD/DSED in children (age 6-8 years) in a deprived population (Minnis et al., 2013). Minnis et al. found a prevalence of RAD (including disinhibited attachment) in this population of 1.4%. Kay, Green, & Sharma (2016) investigated the prevalence of DSED in adopted children (age 6-11 years) using multi-informant cross-context measures including a researcher observation. Forty-nine percent of the children adopted from out-of-home care were classified with DSED, while 6% of the adopted children with no history of special care or child protection before adoption were classified with DSED.

According to Zeanah, Berlin and Boris (2011), it is important for clinicians to observe and analyze how children interact with their primary caregiver when they are distressed and their attachment behavioral system is activated. Features that are of interest in the interactions include the presence or absence of proximity-seeking, avoidance, resistance, or disorganization in their responses of distress, displays of affection toward the caregiver, controlling behavior toward the caregiver, and the ways in which a child interacts with an unfamiliar adult. Given the complexity of such observational diagnostic assessments, the convergence of the results of such assessments with those of more straightforward assessment procedures, such as use of the DAI, cannot be assumed. Indeed, the two types of assessments could potentially generate conflicting data.

A distinctive feature of RAD and DSED is that etiological factors are implied in the diagnosis itself. Thus, for these diagnoses, it must be established that the aberrant social behavior is reactive to exposure to extremes of insufficient care, such as social neglect and deprivation, frequent changes in primary caregivers, and growing up in caregiving arrangements that limit the formation of selective attachment relationships in early life (APA, 2013). Whether exposure to such environmental conditions is a necessary diagnostic criterion that improves diagnostic validity is disputed (Minnis et al., 2009). However, the AACAP Practice parameter (2016) has decided that "given that DSM-5 criteria require a history of severely 'insufficient care', the diagnosis should be questioned in any case in which a history of social neglect cannot be documented." In the large study of Minnis et al. (2013), information about possible pathogenic care was explored in a computerized parent-report interview with questions about physical abuse and witnessing domestic violence in the context of possible posttraumatic stress disorder (PTSD); other possible information sources about possible extreme insufficient care were not included in the study. Kay et al. (2016) used the Maltreatment Classification System (MCS; Barnett et al., 1993) for rating reported (by the adoptive parents) descriptions of maltreatment and neglect and information from the adoption files. In clinical practice, exposure to different caregiving environments can only be assessed retrospectively. Aside from the specific situations of children who grow up in foster or adoptive care or who are taken into foster or adoptive care from institutional settings, caregiver reporting is often problematic, because current caregivers (such as foster parents) may not be fully informed by previous caregivers and because social and legal implications may discourage caregivers from reporting any maltreatment they committed themselves. Case records can be examined as an alternative to interviewing current caregivers, but the records may also reflect selective or biased reporting by professionals, who often base their information on caregiver accounts. It can be argued that positive evidence for a reactive environment is not a necessary criterion if detailed behavioral study of the phenotype of attachment-related disorders (e.g. Lawler, Hostinar, Mliner, & Gunnar, 2014) reveals clear distinctions with the phenotypes of nonreactive disorders of social behavior, as was found when comparing symptoms of RAD, DSED, and autism spectrum disorder (Giltaij, Sterkenburg, & Schuengel, 2015). Nevertheless, in the absence of standardized and psychometrically sound behavioral tests, the inclusion of converging anamnestic evidence might increase the confidence in RAD diagnosis and protect against false-positive diagnoses, especially in clinical

populations that also have a high prevalence of neurodevelopmental disorders that impact social functioning (e.g. Riby, Kirk, Hanley, & Riby, 2014).

To support the development of valid diagnostic assessment procedures that identify children with disordered attachment, the current study explored whether the following methods for determining diagnostic information about RAD and DSED according to the DSM-5 yielded consistent results: semi-structured interviews with caregivers, structured clinical observations of attachment, and independent assessment of extremes of insufficient care.

The study was conducted with a clinical sample of children who were referred for psychiatric consultation to centers that specialize in treating children with low intellectual functioning. Low intellectual functioning is associated with caregiving risk and vulnerability to disorganized, atypical, or disordered attachment (Janssen, Schuengel, & Stolk, 2002). A previous study of this sample that used the DAI revealed that 42% of the children had caregiver-reported symptoms of inhibited and disinhibited behavior (Giltaij et al., 2015); accordingly, this population was expected to show a heightened base rate of disordered attachment, making it feasible to include an observational attachment assessment procedure.

Methods

Participants

The parents of 102 children who had been screened for disturbances of attachment and symptoms of autism spectrum disorder (Giltaij et al., 2015) were asked to participate in a follow-up, which was conducted 2 years after the first one. The participants were recruited through nine Dutch mental healthcare centers that specialize in the psychiatric assessment and treatment of children with developmental disabilities and low intellectual functioning. These centers are scattered throughout 8 of the 11 provinces in The Netherlands. The children were referred for diagnostic consultation mainly not only because of behavioral problems but also because of educational and emotional problems. The inclusion criteria were as follows: age between 5 and 11 years on the date of the assessment; low intellectual functioning with a measured intelligence quotient (IQ) of 50–85 or functional diagnosis of low average, borderline or mild

intellectual disability, and the ability to communicate in Dutch. The centers' psychiatrists and psychologists were instructed not to preselect cases. The parents of 55 of the 102 children gave permission for the children to participate in the study and returned the informed consent letter. The reasons that were given for not participating in the follow-up study were that the child had been discharged from the mental healthcare center system, there were time constraints, or the parents considered the procedure to be too stressful for the child.

The mean chronological age of the children at follow-up was 10.7 years (*SD* = 1.8), and 41 (75%) were boys. The mean IQ according to the case files was 72.1 (*SD* = 9.1), with 20 children scoring in the range of mild intellectual disability (IQ 50–70) and 35 children scoring in the range of borderline to low average intellectual functioning (IQ 71–85). The majority of the children, 43 (78%), had parents who were born in The Netherlands. The demographic characteristics of the participants in the follow-up study correspond with the characteristics from the first study, except for age (Table 1).

Table 1 Demographic characteristics of participants of the original sample (N = 102) and the follow-up sample (N = 55)

Categories	Characteristics	*Original Sample*	*Follow-up Sample*
Age, M (SD)		8.8 (1.7)	10.7 (1.8)
Gender, %	Boy	71%	75%
	Girl	29%	25%
Cognitive, M (SD)	IQ	71.7 (9.7)	72.1 (9.1)
Ethnicity, %	Dutch	81%	78%
	Non-Dutch, western	3%	4%
	Non-Dutch, non-western	16%	18%

Note: M = Mean, *SD* = standard deviation

Ethical approval was obtained from the Vrije Universiteit Medical Center Medical-Ethical Review Board (protocol number 2006/213). This board is licensed to approve research by the Central Committee on Research Involving Human Subjects, which monitors compliance with Dutch legislation on medical research. The participants received a small present (a voucher and a comic book).

Instruments

Symptoms for RAD and DSED.

The DAI (Smyke et al., 2002; Smyke & Zeanah, 1999) is a 12-item semi-structured interview conducted with a primary caregiver or another person who knows the child well, which is designed to assess behavioral symptoms of clinically disturbed or disordered attachment in children. Five items operationalize behaviors that are relevant to DSM-5 RAD: (1) absence of a discriminated, preferred adult; (2) lack of comfort seeking for distress; (3) failure to respond to comfort when offered; (4) lack of social and emotional reciprocity; and (5) emotion regulation difficulties. Three items operationalize the behavioral signs of DSM-5 DSED: (6) not checking back after venturing away from the caregiver, (7) lack of reticence with unfamiliar adults, and (8) a willingness to go off with relative strangers. Interviewers probed the respondent to acquire enough information on the child's behavior so that during the coding phase they could rate each item as "0"="no sign of RAD or DSED," "1"="somewhat or sometimes a sign of RAD or DSED," and "2"="considerable or frequently a sign of RAD or DSED'."

Previous studies have shown strong internal consistency and inter-rater reliability for scales encompassing inhibited (RAD) and disinhibited (DSED) behavior symptoms (Oosterman & Schuengel, 2007a; Smyke et al., 2002; Zeanah et al., 2004; Zeanah et al., 2002; Zeanah et al., 2005). The DAI scales converged with similar measures used in other studies of signs of RAD (Chisholm, 1998; O'Connor, Rutter, & The English and Romanian Adoptees Study Team, 2000) and diverged from measures of aggression, stereotypes, and language development (Zeanah et al., 2002), and autism (Giltaij et al., 2015). Although the DAI was not developed specifically for children with intellectual disabilities, there was good reliability in studies with Romanian orphanage children, among whom the IQs ranged from moderate intellectual disability to normal intelligence (Smyke et al., 2002; Zeanah et al., 2005). Independent psychiatric assessment of attachment disorder was found strongly associated with symptoms on the DAI (Gleason et al., 2011; Zilberstein & Popper, 2014).

Based on the work of Gleason and colleagues (2011), the presence of at least three signs of RAD and at least two signs of DSED was considered a positive score on the categorical diagnoses RAD and DSED. Given the validity evidence in their study, this scoring rule was also adopted in the current study. The interviewers were trained by M.

Oosterman, who conducted psychometric research on the Dutch version of the DAI (Oosterman & Schuengel, 2007b). N. Boris trained interviewers to score normal and aberrant attachment behavior. Interviews were conducted over the phone and were recorded. The interviewers subsequently determined the scores based on a review of the recording. Inter-rater reliability of the scale scores was good (Cohen's kappa = .88 for RAD and .98 for DSED).

Structured observation of adaptive and maladaptive attachment behavior.
The Clinical Observation of Attachment (COA; Boris et al., 2004; Boris et al., 2005) is a structured observational procedure conducted in an unfamiliar room (50 minutes) with the child, a caregiver, and an unfamiliar adult, who may be the diagnostician. The observational procedure for this study was based on the COA procedure developed by Boris and colleagues (2004). Elements of the COA that may elicit responses through the attachment behavioral system include an unfamiliar room and person, being approached by the unfamiliar person who joins in the child's play, separation from the caregiver followed by reunion, a sudden and loud alarm sound was then switched on by the unfamiliar person using a hidden remote control of a cd-music system, solicitation of physical contact by the unfamiliar person, and a co-operation task. A behavior transcript was made on the basis of video recordings of the sessions. Video recording and transcript were scored for adaptive and maladaptive attachment behavior using the List of Behavioral Signs of Disturbed Attachment in Young Children (BSDA; Boris, Fueyo, & Zeanah, 1997; Zeanah, Mammen, & Lieberman, 1993) as recommended in the AACAP Practice parameter (Boris et al., 2005; Zeanah et al., 2016). The BSDA list describes eight types of attachment behavior: (1) showing affection to the caregiver, (2) seeking comfort from the caregiver, (3) reliance on the caregiver for help, (4) cooperation with the caregiver, (5) exploratory behavior, (6) controlling behavior, (7) reunion responses, and (8) response to strangers. A five-point scale that ranged from adaptive (score = 1) to maladaptive (score = 5) was used to rate children's behavior toward their caregiver and toward the unfamiliar person. A sum score of maladaptive attachment behavior was calculated based on the eight behavior types. In the manual of the BSDA the behavioral expression of the extreme scores (scores 1 and 5) are described. The intermediate scores are related to these extreme scores (mostly adaptive, sometimes adaptive as well as maladaptive, mostly maladaptive). In this study, five master-students Special Education were trained for coding the attachment behavior using the BSDA. The inter-rater reliability was .73. The video recording and transcripts were randomly

divided among two coders who coded the video's independently the observed behavior of the child in the COA. If there was a difference of one point the coding was discussed until the coders reached consensus. If there was a difference of more than one point the video recording and transcript were coded a third time by an independent coder with no foreknowledge of the other coders. The coding was then discussed until consensus was met.

Extremes of insufficient care.

A research assistant who was not involved with other measurement activities in the study abstracted information from each child's case files at the mental health centers where the participants were recruited. These medical files contained all the reports (pediatric, psychological, child psychiatric, educational, juridical, and social work) that were used by the clinicians of the mental health center as input for assessment, classification, and treatment of the child. Coding of the files focused on the developmental history of the child, especially information about the first years of the child's life, as well as descriptions and experiences related to child-caregiver interactions. Extremes of insufficient care were coded positive in cases that showed (1) neglect of the child's emotional and social needs (references of an environment characterized as highly unstable, reported physical or sexual abuse, as confirmed by interference of childcare reports); (2) repeated changes in primary caregivers (multiple separations from primary caregivers in the first 3 years of age, such as hospital care, adversarial divorce of parents, and frequently changing caregivers); or (3) rearing in institutions with high child-to-caregiver ratios. The inter-rater reliability for extremes of insufficient care was high (intra-class correlation of .93). Given the recent shift from a broader definition of pathogenic care (DSM-IV) to the more narrowly defined extremes of insufficient care in the DSM-5, case records were also coded for exposure to harsh punishment and inept parenting and for exposure to domestic violence. Pakalniskiene (2008) defined harsh parenting as physical punishment and verbal or nonverbal aggression, such as anger outbursts, threats, stony silences, and rejection, thus combining both aspects – the physical and the nonphysical. Harsh punishment is considered as a form of "inept parenting" that includes coercive parent-child communication, dysfunctional disciplining practices, inconsistent control, harsh or violent physical punishment, negative attitudes and reasoning, limited use of praise, support, or warmth, and poor supervision and monitoring (Maccoby & Martin, 1983;

Patterson, Reid, & Dishion, 1992; Reid, Patterson, & Snyder, 2002; Robins & Rutter, 1990; Stoff, Breiling, & Maser, 1997).

Clinical diagnosis of RAD or DSED.

Clinical diagnoses were made independently by two highly experienced clinical psychologists using the DSM-5 criteria. The psychologists considered the assessments of behavior in the COA and the assessments of the attachment history without the DAI results. First, the (COA) observed (maladaptive) attachment behavior on the BSDA was matched with the DSM-5 criteria. Positive evidence for RAD is when the child demonstrates a consistent pattern of inhibited, emotionally withdrawn behavior toward adult caregivers, manifested by both of the following: (1) the child rarely or minimally seeks comfort when distressed, and (2) the child rarely or minimally responds to comfort when distressed, and at least two out of the following: (3) minimal social and emotional responsiveness to others; (4) limited positive affect; and (5) episodes of unexplained irritability, sadness, or fearfulness that are evident even during nonthreatening interactions with adult caregivers. Positive evidence for DSED is when the child shows a pattern of behavior in which the child actively approaches and interacts with unfamiliar adults and exhibits at least two of the next criteria: (1) reduced or absent reticence in approaching and interacting with unfamiliar adults; (2) overly familiar verbal or physical behavior (that is not consistent with culturally sanctioned and with age-appropriate social boundaries); (3) diminished or absent checking back with adult caregiver after venturing away, even in unfamiliar settings; and (4) willingness to go off with an unfamiliar adult with little or no hesitation. It is important to note that the criteria for the two disorders are not mutually exclusive, leaving open the possibility that both the RAD and DSED criteria may apply. However, given that children are prevented from venturing away on their own during the COA, it is unlikely that children may be diagnosed with DSED or DSED/RAD without displaying at least of the active disinhibited forms of social behavior (reduced reticence and/or overly familiar behavior).

Second, all information gained from the attachment history was matched to extremes of insufficient care according to the DSM-5 criteria (C). Third, since insufficient care is a necessary condition for the diagnosis, only children who met both conditions as meeting the DSM-5 criteria received the DSM-5 classification. Fourth, using the DSM-5 criteria A and B for RAD and criteria A for DSED both psychologists independently coded the

observed attachment behavior in the video-recorded COA as diagnosis RAD, DSED or RAD as well as DSED. The inter-rater reliability was $\kappa = 1.00$. Discrepancies in diagnosis were discussed until the consensus was met.

Data analysis

T-tests were conducted to investigate the association between positive diagnoses based on the DAI and the maladaptive attachment behavior scores based on the COA as well as between extremes of insufficient care and maladaptive attachment behavior scores on the COA. Chi-square tests were performed to investigate the convergence between positive diagnoses based on the DAI and extremes of insufficient care. Statistical analyses were performed using the software package SPSS version 21.

Results

Preliminary and descriptive analyses

To test for possible confounding factors for the convergence of the disordered attachment indicators, we tested for associations between indications for disordered attachment on the DAI (inhibited, disinhibited, or none), the scores for maladaptive attachment behavior on the COA, and insufficient care as reported in the records (extremes of insufficient care or no extremes of insufficient care), and the background variables, gender, ethnicity, age, and cognitive functioning. T-tests showed no significant associations (all $p > .05$).

Based on the DAI with the caregivers, inhibited and disinhibited behavior was below the threshold for a positive categorical diagnosis for RAD or DSED in 37 (67%) of the 55 children (Table 2). Positive DAI diagnoses for RAD were found for 9 children, and positive diagnoses for DSED were found for 15 children. Six children scored positive for RAD and DSED using the DAI.

Table 2 Frequency of DAI diagnoses of the original sample (*N* = 102), the non-participants in follow-up (*N* = 47) and the follow-up sample (*N* = 55).

Categories	Characteristics	*Original sample (N = 102, %)*	*Non-participants in follow-up (N = 47, %)*	*Follow-up sample (N = 55, %)*
DAI	No RAD and no DSED	58%	47%	67%
	RAD	5%	4%	6%
	DSED	27%	38%	16%
	RAD and DSED	11%	11%	11%

DAI: Disturbances of Attachment Interview; DSED: disinhibited social engagement disorder; RAD: reactive attachment disorder.

In the structured observation setting (COA), children's behavior toward their primary caregiver and the stranger was scored according to the eight BSDA signs of adaptive and maladaptive attachment behavior. With this five-point scale, the minimum possible total score is 8 (fully adaptive attachment behavior), and the maximum possible total score is 40 (fully maladaptive attachment behavior). The average score on the BSDA was 17.98 (SD = 6.39), ranging from 9 to 33.

Exposure to extremes of insufficient care was found in the medical records of 12 children (22%). This included repeated changes in caregivers in 10 (18%) cases, living in institutions in 7 (13%) cases, and neglect of their social and emotional needs in 9 (16%) cases. In eight cases, harsh punishment or inept parenting was coded in addition to extremes of insufficient care, and in two cases, harsh punishment or inept parenting was mentioned without extremes of insufficient care. Domestic violence was coded in seven cases in addition to extremes of insufficient care, and domestic violence was coded without exposure to extremes of insufficient care in two cases. The two cases that were positive for harsh punishment or inept parenting did not overlap with the two cases that were positive for domestic violence.

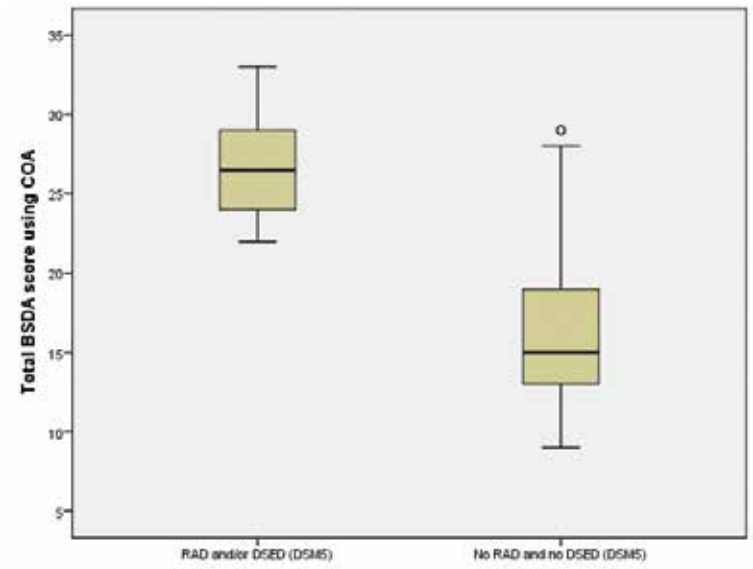
Based on the diagnostic information that was collected, 10 children received a DSM-5 RAD and/or DSED diagnosis: 7 children were diagnosed with RAD and with DSED, 2

children were diagnosed with DSM-5 RAD only, and 1 child was diagnosed with DSM-5 DSED only.

Convergence between DAI diagnoses and COA outcomes

An independent samples *t*-test found that children who scored positive for RAD or DSED on the DAI (n = 18, M = 21.61, SD = 7.2) showed more maladaptive attachment behavior according to the BSDA than those who scored negative for RAD or DSED (n = 37, M = 16.22, SD = 5.2), $t(53) = 3.17$, $p = .003$, $d = 0.91$. Children who scored positive for RAD only (n = 9, M = 25.78, SD = 5.8) showed more maladaptive attachment behavior than those who scored negative for RAD (n = 46, M = 16.46, SD = 5.3), $t(53) = -4.72$, $p < .001$, $d = -1.72$. Likewise, children who scored positive DSED only (n = 15, M = 22.2, SD = 7.7) showed more maladaptive attachment behavior than those who scored negative for DSED (n = 40, M = 16.4, SD = 5.1), $t(53) = -2.70$, $p = .014$, $d = 0.82$. Figure 1 shows the distribution of the BSDA scores for the DAI scoring RAD and/or DSED (RAD Inhibited and/or RAD disinhibited) and no RAD and no DSED.

Figure 1 Total BSDA score using COA and DAI screening for RAD and/or DSED and no RAD / no DSED (N = 55)



Convergence between DAI diagnoses and extremes of insufficient care

Extremes of insufficient care were significantly less prevalent in the records of children with a DAI diagnosis of no RAD and no DSED (Fisher's exact test, $p = .043$, $\kappa = .19$). Significantly more children who scored positive for RAD had extremes of insufficient care compared to children who scored negative for RAD (Fisher's exact test, $p = .017$, $\kappa = .36$); similarly, significantly more children who scored positive for DSED had extremes of insufficient care compared to children who scored negative for DSED (Fisher's exact test, $p = .011$, $\kappa = .36$) (Table 3).

Table 3 Frequency of extremes of insufficient care and no RAD and no DSED, RAD and DSED according to the DAI ($N = 55$)

Extremes of insufficient care	Extremes of insufficient care Total	Classification according to DAI					
		No RAD and no DSED		RAD		DSED	
		Yes	No	Yes	No	Yes	No
Yes	12	5	7	5	7	7	5
No	43	32	11	4	39	8	35

Note: DAI = Disturbance of Attachment Interview, RAD = Reactive Attachment Disorder.

Convergence between COA outcomes and extremes of insufficient care

The mean total score for the observed BSDA behavior of children who were not found to have experienced extremes of insufficient care was 16.07 ($SD = 5.14$, $n = 43$); children with records reporting extremes of insufficient care had a mean total BSDA score of 24.83 ($SD = 5.86$, $n = 12$). Children who had experienced extremes of insufficient care had a significantly higher total score on the BSDA than those who had not experienced extremes of insufficient care or those with insufficient information in their medical file to determine whether they had experienced extremes of insufficient care: $t(53) = 5.07$, $p < .001$, $d = 1.66$.

Convergence between DAI diagnoses and a DSM-5 classification of attachment disorder

A total of 34 children who were negative for a RAD and/or DSED using the DAI were also independently diagnosed negative according to DSM-5 for RAD and/or DSED. Seven children had positive DAI and an independently positive DSM-5 diagnosis of RAD

and/or DSED. Thus, in 41 children (75%) the DAI diagnosis and the DSM-5 diagnosis were consistent. Three children who scored negative according to the DAI had a positive classification for attachment disorder according to the combination of observed behavior (COA) plus a history of exposure to extremes of insufficient care (DSM-5 RAD or DSED). Eleven children who scored positive according to the DAI had a negative DSM-5 diagnosis (Table 4). The predictive validity of the DAI for a DSM-5 diagnosis of RAD or DSED therefore showed only fair strength (Fisher's exact test, $p = .01$ $\kappa = .35$). For 25% of the children, the DAI was inconsistent with a DSM-5 RAD or DSED diagnosis.

Table 4 Frequency of the classification according to the DAI and frequency of clinical diagnosis according to the DSM-5 ($N = 55$)

Classification according to DAI	**Clinical diagnosis according to the DSM-5 (Including the criterion of evidence of extremes insufficient care)**		**Total**
	No RAD (DSM-5) or DSED (DSM-5) (according to COA and IC)	**RAD (DSM-5) and/or DSED (DSM-5) (according to COA and IC)**	
No RAD	34	3	37
RAD Inhibited and/or RAD Disinhibited	11	7	18
Total	45	10	55

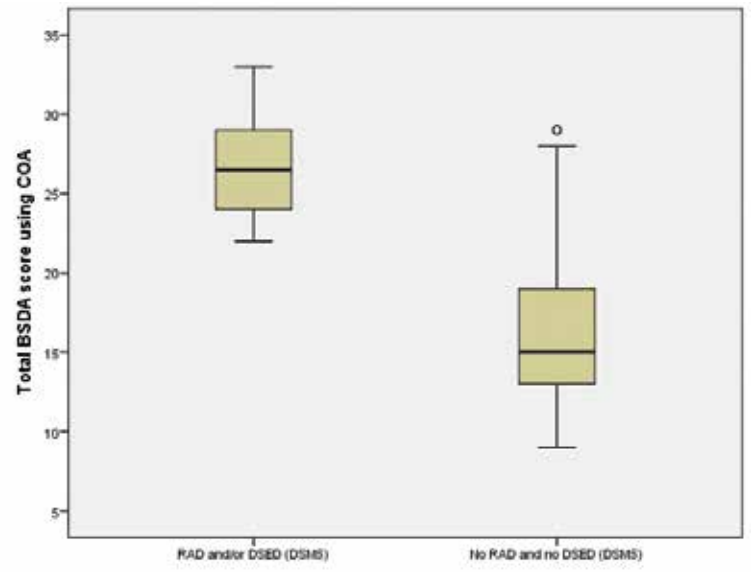
Note: COA = Clinical Observation of Attachment, DAI = Disturbance of Attachment Interview, DSED = Disinhibited Social Engagement Disorder, DSM = Diagnostic and Statistical manual of Mental disorders, IC = Extremes of insufficient care, RAD = Reactive Attachment Disorder.

Convergence between COA outcomes and a clinical (DSM-5) diagnosis of RAD and DSED

The mean total score for the observed BSDA behavior of children who were not diagnosed with a DSM-5 RAD or DSED was 16.0 ($SD = 5.03$, $n = 45$); children who only had a DSM-5 RAD diagnosis had a mean total score (BSDA) of 23.5 ($SD = .71$, $n = 2$), and the child who only had a DSM-5 DSED diagnosis had a total score (BSDA) of 25. The children with both DSM-5 RAD and DSED diagnoses had a mean total score (BSDA) of 28.2 ($SD = 3.72$, $n = 7$). Children with a DSM-5 RAD and/or DSED diagnosis had a significantly higher total score on the BSDA than those without DSM-5 RAD and/or

DSED diagnosis: $t(53) = -5.07$, $p < .001$, $d = -1.77$. Figure 2 shows the BSDA scoring distribution for RAD and/or DSED and no RAD and no DSED.

Figure 2 Total BSDA score using COA and DSM-5 diagnose RAD and/or DSED and no RAD / no DSED ($N = 55$)



Discussion

The diagnostic steps recommended by Boris et al. (2005) and Chaffin et al. (2006) led to high convergence between indicators of disordered attachment, supporting the validity of diagnoses made of RAD or DSED for children with low intellectual functioning. Using these recommended diagnostic procedures (the association between observation of the attachment behavior in a structured setting and a history of extreme insufficient care, gathered from a comprehensive history of the child's early caregiving environment) led to a positive clinical diagnosis of RAD and/or DSED according to the criteria of the DSM-5 in 18% of the cases in this sample of children with low intellectual functioning referred for mental health assessment, despite having been under treatment for 2 years. With a rate of 33% positive RAD and/or DSED diagnosis based on the DAI as a structural clinical interview, the DAI identified a considerable higher number of cases than based on more comprehensive diagnosis using observations.

Observations of maladaptive attachment behavior during the COA procedure using the BSDA proved feasible, reliable, and valid according to associations with the other measures. The mean BSDA score was higher for children who screened positive for RAD and/or DSED using the DAI and for the DSM-5 diagnosed children than for those with negative screens or diagnoses. Still, some children who displayed high rates of maladaptive attachment behavior screened negative on the DAI and did not fulfil the criteria for exposure to extremes of insufficient care, illustrating the importance of using multiple sources of information, even if observations are made. A lack of evidence for insufficient care does not mean that the child was not exposed to it. Caregivers may be unaware of what insufficient care is, or may have reasons not to disclose this information during the assessment (shame, guilt, or fear for consequences). Although some studies (e.g. Minnis et al., 2009) did not include the criteria for extremes of insufficient care because the information may be unreliable, in this study the recommendations of the AACAP were followed using the classification criteria of the DSM-5. However, the use of the AACAP increased the risk that false-negative scores on insufficient care may be reported, as the participants' files may be incomplete.

A surprisingly high number of children were comorbid on RAD and DSED, given the decision that was made for the DSM 5 to define two disorders (RAD and DSED), rather than define two distinct types of inhibited and disinhibited RAD. Past research with the DAI has suggested that so-called passive forms of indiscriminate behavior, such as failing to check with caregiver after venturing and willingness to go off with stranger, may confound comorbid classification if not combined with more active disinhibited social engagement (Zeanah & Gleason, 2010). The DSM-5 criteria for DSED also do not exclude the possibility that children are diagnosed with DSED based on observed "passive" indiscriminate behavior alone, which is on the basis of the two criteria of failing to check back after venturing away and willingness to go off with stranger. However, it is unlikely that comorbidity is accounted for by these passive indiscriminate behaviors only, because children are prevented from venturing away during the COA procedure, requiring that at least one of the other, more active, criteria needs to be fulfilled.

This study had some limitations. The sample of 55 children was modest and originated from a study on inhibited and disinhibited attachment behavior and symptoms of autism (Giltaij et al., 2015). Dropout was considerable (47 out of 102); this may have

introduced bias, although analyses of the families who declined participation in the follow-up did not reveal significant differences in gender, ethnicity and cognitive functioning.

Another limitation was that the DAI used in this study was originally developed for administrating disturbed attachment behavior in children until 5 years of age. Although the DSM-5 criteria for RAD and DSED do not distinguish between young and school-aged children, the phenomenology of the behavior may differ. In 2010 (Zeanah & Smyke, 2010), a version of the DAI was presented for school-aged children, adding some adaptations. Relative to the original child version (e.g. Gleason et al., 2011), little is known about the psychometric attributes of the school-age version. Also, the studies using the school-age version are very limited (Vervoort et al., 2013), while other studies on school-aged children used the original version (Oosterman & Schuengel, 2007a, 2007b).

Furthermore, children were only observed with their primary caregiver. Although it is presumed that attachment-related disorders are a characteristic of the child, attachment itself is a relational concept. Nevertheless, the maladaptive attachment behavior in the children scoring positive on the clinical diagnosis RAD and/or DSED is, in this study, only observed in relation with one primary caregiver, and it is therefore insufficient to define it as a within-the-child disorder. Obtaining diagnostic information on disordered attachment within other relationships as well is likely due to increased diagnostic confidence. The AACAP Practice parameter on assessments of RAD and DSED (Boris et al., 2005; Zeanah et al., 2016) recommends serial observations of the child interacting with at least both primary caregivers in a minimum of two or three visits.

Finally, it should be kept in mind that the DSM-5 criteria and diagnostic guidelines refer to disorders that begin in early childhood, while the chronological ages of the children in the current sample ranged from age 7 to 13 years. Previous work has shown that disinhibited attachment behavior is highly stable from age 6 to 11 years (Rutter et al., 2007), but the validity of observed and reported inhibited attachment behavior in school aged children is less clear. Resolution of this issue is hampered by the low frequency of inhibited attachment and RAD in the current sample as well as in other samples.

The classification criteria for the DSM-5 attachment disorders (i.e. RAD and DSED) include observable and distinct social behaviors of children that can be evaluated by clinicians who are trained in attachment theory and in the observation of attachment. The current study supports a best practice for assessing disordered attachment that consists of interviewing the primary caregivers or other adults who are closely familiar with the child, using a checklist of extremes of insufficient care, reviewing a developmental history of the child's care experiences, and performing a structured observation procedure (COA) with the child and primary caregiver plus a stranger (Boris et al., 2005; Zeanah et al., 2016). Combining these instruments bolsters the validity of the diagnosis and, thus, improves the subsequent treatment plan. These findings suggest that the DAI should be used as a screening instrument with caution, as it cannot replace full clinical assessment for diagnosing RAD and/or DSED. Thus, future research may consider the DAI as a screening instrument. Furthermore, clinicians should always obtain careful histories from as many sources as are relevant about insufficient care. Developing and testing intervention programmes that reduce maladaptive attachment behaviors in the context of promoting secure attachment remain a priority for further research. Although studies have found encouraging results for interventions that focus on secure attachment in high-risk families (Bernard et al., 2012; Moss et al., 2011), with the exception of placement from institutional care to foster care (Smyke et al., 2012), little is known about the potential of treatment to reduce disordered attachment.

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Chapter 4

Adaptive behaviour, comorbid psychiatric symptoms, and attachment disorders

Giltaij, H.P., Sterkenburg, P.S., & Schuengel, C. (2016). Adaptive behaviour, comorbid psychiatric symptoms, and attachment disorders. *Advances in Mental Health and Intellectual Disabilities, 10 (1)*, 82-91.

Abstract

Purpose – The purpose of this paper is to describe the mental and intellectual developmental status of children with combined intellectual disabilities, reactive attachment disorder (RAD), and/or disinhibited social engagement disorder (DSED), and to describe the presence of comorbid diagnoses.

Design/methodology/approach – The study included 55 children that were referred for psychiatric consultation due to low intellectual functioning (borderline or mild; IQ 50-84). Attachment diagnoses were based on the Clinical Observation of Attachment (COA) procedure. Development was measured with the Dutch version of the Vineland Adaptive Behaviour Scales. Psychopathology was measured with the DISC-IV and AUTI-R. Emotional and behavioural problems were measured with the Dutch version of the Developmental Behaviour Checklist.

Findings – Children with and without attachment diagnoses had similar IQs. However, children with disturbed attachment, RAD, and/or DSED had lower levels of adaptive behaviour than those without attachment diagnoses. No comorbidity was associated with autism or ADHD. However, 80 per cent of children with RAD and/or DSED were also diagnosed with ADHD. Parents of children with DSED often reported disruptive anti-social behaviour.

Practical implications – Children with RAD and/or DSED may have unused developmental potential. Disturbed attachment should be considered in some cases of ADHD.

Originality/value – Findings suggested that diagnostic expertise provided added value for distinguishing disordered attachment in young clients with intellectual disability.

Keywords Comorbidity, Adaptive behaviour, DSED, Low intellectual functioning, Psychiatric symptoms, RAD

Introduction

Several studies have documented that children with intellectual disability are at high risk for developing psychopathology (Dekker *et al.*, 2006; Einfeld *et al*., 2011). Janssen *et al.* (2002) stated that people with intellectual disability are more vulnerable to stress, and they linked this susceptibility to the quality of the attachment relationship with their caregivers. Their stress-attachment model provided an alternative explanation for the high prevalence of challenging behaviour among people with intellectual disability compared to other explanations based on neurocognitive or behavioural learning models. The stress-attachment model also proposed that children with intellectual disability may be particularly vulnerable to environmental factors that may lead to both low quality attachment relationships and disorders in attachment at a more basal, individual level. These types of disturbances may develop concurrent with other mental health disorders, although the extent to which this occurs remains unknown.

Disruptions in attachment relationships, such as separations, neglect, or abuse, are thought to have wide-ranging ramifications that may lead to emotional and behavioural dysfunctioning (MacLean, 2003). Bos *et al.* (2011) reported that early conditions of severe deprivation and neglect were linked to the later health and development of a child. They found higher levels of internalizing symptoms and disorders (anxiety and depressive disorders) and externalizing symptoms and disorders (ADHD, oppositional defiant disorder, and conduct disorder) in a group of children that had been severely deprived and neglected, compared to a control group. In a Romanian study, psychiatric disorders were reported in 53 per cent of young children with any history of institutional rearing, compared to 22 per cent in children without that history (Zeanah *et al.*, 2009). Zeanah *et al.* (2005) also reported that children that lived in institutions where they experienced early social deprivation and neglect demonstrated heightened levels of inhibited and disinhibited attachment behaviours, compared to children in a community comparison sample.

DSM-5 (American Psychiatric Association (APA), 2013) describes inhibited and disinhibited attachment behaviours within the diagnoses of Reactive Attachment Disorder (RAD) and Disinhibited Social Engagement Disorder (DSED). RAD is defined as "a consistent pattern of inhibited, emotionally withdrawn behaviour towards adult caregivers" (pp.141-142). DSED is defined as "a pattern of behaviour in which a child

actively approaches and interacts with unfamiliar adults" (pp. 142-143). For both attachment-related disorders, the presumption is made that disturbances in attachment behaviour are a reaction to pathogenic care. Pathogenic care is defined as conditions where a child grows up without a limited, stable number of caregivers that provide individualized attention to children (e.g. in an orphanage); where a child is exposed to serious social neglect or deprivation in the form of persistent lack of having basic emotional needs for comfort, stimulation, and affection met by caring adults.

Because the environmental factors associated with RAD and DSED may also increase the chances for other mental health problems, RAD and DSED may be associated with other common disorders. For children with intellectual disability, this potential association gives rise to the question of whether a specialized diagnosis of attachment-related disorders would provide added value to their mental healthcare, given the scarcity of resources and the fact that additional diagnostic procedures represents a burden to children and families. Therefore, we investigated the prevalence of psychiatric psychopathic comorbidities in children diagnosed with RAD and/or DSED. We also tested whether children with RAD or DSED possessed lower adaptive functioning than children with other diagnoses; this potential association may be an additional reason for improving our awareness of these diagnostic categories.

Method

Participants

This study was part of a larger research project which included 102 children with a mean age of 8.8 years (SD = 1.7). That project focused on the prevalence of disordered attachment symptoms in children with an intellectual disability and the diagnostic overlap between symptoms of disordered attachment and symptoms of pervasive developmental disorder (Giltaij *et al.,* 2015). All the participants were referred for psychiatric assessment to nine mental health centres scattered over eight of the 11 provinces of The Netherlands. All the centres provided specialized services in psychiatric assessments and treatment for children with intellectual disabilities. There was no pre-selection of cases. That large study included children between five and 11 years old, that had an IQ between 50 and 85 and could speak Dutch. One year after that first study, the parents of the 102 children were invited to participate in the present (second)

study. In total, 55 parents returned informed consent letters. Part of the lack of response was due to the facts that some parents no longer attended the same mental healthcare centre; some parents felt that it was too time consuming; and/or some parents assumed that it would be too stressful for their child.

The participants in this study (*n* = 55) had a mean age of 10.7 years (*SD* = 1.8). The cohort included 41 (75 per cent) boys and 14 (25 per cent) girls. The mean level of cognitive functioning (IQ) was 72.1 (*SD* = 9.1). The range of cognitive functioning was between mild intellectual disability (IQ 50-70) (*n* = 20) and borderline intellectual functioning (IQ 71-85) (*n* = 35). Most participating children (*n* = 43; 78 per cent) had parents born in the Netherlands; the other children had at least one non-Dutch parent (*n* = 12; 22 per cent).

The VU University Medical Centre Medical-Ethics Review Board approved the ethics of the study (No. 2006/213). All participants received a small voucher and a comic book at completion of the assessment.

Measurements

Disordered attachment behaviour.

The Clinical Observation of Attachment (COA) (Boris *et al*., 2004, 2005) is a structured observational procedure (50 min). The procedure takes place in an unfamiliar observation room, with the child, the primary caregiver, and an experienced and professional clinician that is unknown to the child. The caregiver is instructed about the procedure, but the child is minimally prepared with the explanation that both are going to the assessment centre for an appointment. The observational procedure performed in this study was an adapted version of the procedure described by Boris *et al.* (2004). Briefly, the procedure included segments that gradually introduced cues to the child that indicated danger or challenges. The stranger participated in the child's play, the caregiver left the room, a frightening alarm sounded, the stranger initiated close physical contact for a short time, and the child was involved in a cooperation task. The observation session was videotaped, and the child's behaviour was written down. Two experienced clinicians (the first and second authors) independently encoded all the child's behaviours, based on the videos and written descriptions. Discrepancies were resolved by discussions that led to a consensus code for each item. The raters encoded signs of disturbed attachment behaviour, based on the List of Behavioural Signs of

Disturbed Attachment in Young Children (BSDA; Zeanah *et al.,* 1993; Boris *et al.*, 1997), which was recommended by the American Academy of Child and Adolescent Psychiatry (AACAP) Practice Parameter on assessments of RAD (Boris *et al.*, 2005). This list distinguished eight signs of attachment behaviour: showing affection to the caregiver, seeking comfort from the caregiver, reliance on the caregiver for help, cooperation with the caregiver, exploratory behaviour, controlling behaviour, reunion responses, and response to strangers. Based on the interactive behaviours of the child towards the primary caregiver and stranger, each item on the list was scored on a five-point scale, which ranged from adaptive (score 1) to maladaptive (score 5) attachment behaviour. Theoretically, the summed scores ranged from 8 to 40. The inter-rater reliability of the two coders, before achieving consensus, was adequate (Cronbach's $\alpha = 0.73$).

Adaptive functioning.

The Vineland Screener 0-12 (NL) questionnaire (Van Duijn *et al.*, 2009) was derived for research purposes from the Vineland Adaptive Behaviour Scales (Sparrow *et al.*, 1993). This questionnaire had to be completed by a primary caregiver. The 90 items in this instrument covered four domains of adaptive behaviour, including Communication (26 items), Daily Living Skills (23 items), Socialization (23 items), and Motor Skills (18 items). Items were encoded with scores as follows: "yes, usually" = 2, "sometimes or partial" = 1, "no, never" = 0, or "don't know" = DK. The scores reflected a developmental age group (in months) associated with the individual's adaptive (behavioural) level. The internal consistency of the domains and the test-retest reliability of the screener instrument were high: 0.90 or higher.

Psychiatric symptomatology.

The Dutch version of the Diagnostic Interview Schedule for Children, version IV (DISC-IV; Ferdinand *et al.*, 1998) is a translation of the National Institute of Mental Health Diagnostic Interview Schedule for Children, Version IV (NIMH-DISC-IV; Shaffer *et al.*, 2000). The DISC-IV is a highly structured diagnostic interview designed to assess more than 30 psychiatric disorders, based on DSM-IV criteria, that occur in children and adolescents. In this study, we used the DISC-P for parents or knowledgeable caregivers of children aged 6-17 years. The DISC-P comprises an interview organized into six modules of related diagnoses. In this study, we administered three modules that comprised a total of 16 psychiatric disorders, including anxiety disorders ($n = 9$), mood disorders ($n = 3$), and disruptive disorders ($n = 4$). The DISC-IV is commonly used in

research to obtain diagnostic information about psychiatric disorders. The most common internalizing and externalizing disorders covered in the NIMH-DISC-IV generally showed test-retest reliability estimates in the acceptable range (Shaffer *et al.*, 2000).

The AUTI-Revised scale (AUTI-R; Van Berckelaer-Onnes and Hoekman, 1991) is a 51-item questionnaire designed to be completed by a professional, such as a teacher, that knows the child well. This instrument assesses the classification of autistic disorders in children. In this study, we used the version designed for children 6-12 years old. The AUTI-R can also be used for children with intellectual disability. The 51 items of the AUTI-R cover seven areas, including relationship disturbance (12 items), language disturbance (15 items), striking motor phenomena (five items), striking sensory phenomena (nine items), resistance to change (six items), acute, illogical fears (two items), and two other items. Each item is scored based on a 6-point scale that ranges from "no" to "very frequently". The total score leads to the classification of "non-autistic", "borderline autistic" or "autistic". The inter-rater reliability of the AUTI-R was reported as good (r = 0.97; n = 73). The validity of the AUTI-R criterion was rated as excellent (of 220 individuals in the survey group, 92 per cent was correctly classified; Van Berckelaer-Onnes and Hoekman, 1991; Evers *et al.*, 2000). The AUTI-R was used in a follow-up study that focused on Romanian adopted children with a background of severe neglect (Hoksbergen *et al.*, 2005). In that study, 16 per cent of adopted children were classified in the autistic range (autistic or borderline autistic) and exhibited symptoms of autistic-like behaviour.

Emotional and behavioural problems.

The Developmental Behaviour Checklist (DBC; Einfeld and Tonge, 1991, 1994, 1995; Einfeld *et al.,* 1999) was especially developed to determine emotional and behavioural problems in children and adolescents, aged 6-18 years, with intellectual disabilities. This study employed the Dutch version of the DBC, known as the Vragenlijst over Ontwikkeling en Gedrag (VOG; Koot and Dekker, 2001). There are two parallel versions of the VOG: one is to be completed by the parents of the child or by a familiar caregiver (VOG-O/DBC-P), and the other is to be completed by a professional that regularly has contact with the child, such as the teacher or daycare staff (VOG-L/DBC-T). The checklist contains 93 items organized into five problem categories, including disruptive and anti-social, self-absorbed, disturbance in communication, anxiety, and relating in social

situations. Each item is given a score of 0 ("not applicable"), 1 ("somewhat or sometimes applicable"), or 2 ("clearly or often applicable"). The total scores can be transformed into percentile scores, which can be compared with scores from other children with a similar cognitive level of functioning, age, and sex. The test-retest reliability of the VOG in the five categories was between 0.76 and 0.89 (VOG-O/DBC-P) and between 0.69 and 0.91 (VOG-L/DBC-T).

Data analysis

SPSS software package, version 21, was used for statistical analyses. The BSDA scores reflected observed maladaptive behaviour of the child towards the attachment figure and stranger. The *t*-test for independent samples was used to compare BSDA scores between children with and without a clearly reported history of pathogenic care. Child development was evaluated by setting a threshold for the Vineland screener score equal to the mean developmental age, minus 2 standard deviations. Scores above the threshold were classified as normal adaptive behaviour, and scores below the threshold were classified as maladaptive behaviour. X^2 tests were performed to evaluate differences in adaptive behaviour development (Vineland Adaptive Behaviour Scale) between children with and without a DSM-5 diagnosis of RAD and/or DSED.

Comorbid psychiatric disorders (DSM-IV) were diagnosed with the DISC-IV and the AUTI-R. The *t*-test for independent samples was used to determine the statistical significance of differences in comorbid psychiatric disorders between children with and without attachment-related DSM-5 disorders (RAD and/or DSED).

To examine the links between emotional and behavioural problems and attachment diagnoses, the Mann-Witney *u*-test was first used to compare the scores for emotional and behavioural problems determined by the parents (VOG-O/DBC-P) to those determined by the teachers (VOG-L/DBC-T). Next, Pearson's X^2 test was used to determine whether the difference between the parent and teacher scores was similar or different between the groups of children with or without attachment disorders (DSM-5). For this analysis, the highest percentile scores (score > 0.90) were recorded as severe psychopathology and the group with percentile scores lower than 0.90 were recorded as having mild psychopathology.

Results

Attachment-related diagnoses

Among a total of 55 children, ten (18 per cent) had scores on the DSM-5 criteria that indicated an attachment-related disorder diagnosis of RAD and/or DSED (APA, 2013). These children showed maladaptive attachment behaviour during the COA. Pathogenic care was convincingly reported in the medical files of these children, which is required for a DSM-5 RAD and DSED diagnosis (APA, 2013) (Table I).

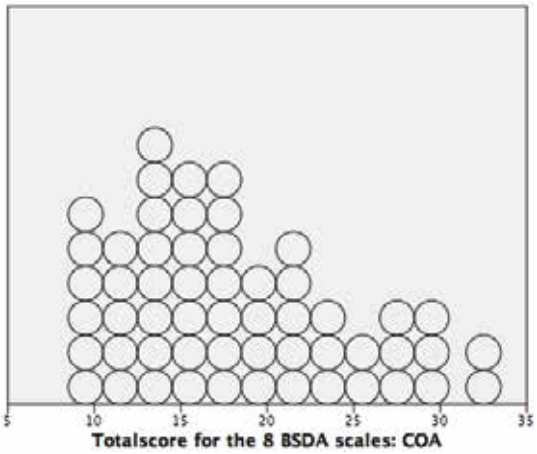
Table I Frequency of DSM-5 diagnoses for the total sample of children (*n* = 55)

DSM-5 diagnoses	*n*	%
DSM-5: No RAD and no DSED	45	82
DSM-5: RAD	3	5
DSM-5: DSED	1	2
DSM-5: RAD and DSED	6	11

RAD, reactive attachment disorder; DSED, disinhibited social engagement disorder

The clinically observed maladaptive attachment behaviour (COA) was scored based on the BSDA (Zeanah *et al.*, 1993; Boris *et al.*, 1997). The total scores varied from nine to 33 (mean = 17.98, *SD* = 6.39). Figure 1 shows the distribution of the clinically observed (COA) total BSDA total scores.

Figure 1 Distribution of Clinical Observation of Attachment (COA) scores (*n* = 55)



The medical files of 15 out of 55 children reported a history of pathogenic care. The *t*-test showed that children that did not receive pathogenic care scored significantly lower on the BSDA (mean = 15.98, *SD* = 5.24) compared to children that received pathogenic care (mean = 23.33, *SD* = 6.24): ($t(53) = -4.40$, $p < 0.001$). No significant differences were found for age or intellectual functioning between children that received and those that did not receive pathogenic care (Table II).

Table II Total BSDA scores, age, and IQ for children without (no stated) and those with (stated) a history of pathogenic care (*n* = 55)

	Mean (SD)	
Group characteristics	**No stated Pathogenic care (*n* = 40)**	**Stated Pathogenic care (*n* = 15)**
Total BSDA score	15.98 (5.24)	23.33 (6.24)
Age (years)	10.53 (1.89)	11.04 (1.75)
Intellectual functioning (IQ)	71.80 (8.84)	73.00 (10.07)

BSDA, List Behavioural Signs of Disturbed Attachment in Young Children

Adaptive functioning

For skills, as reported with the Vineland Adaptive Behaviour Scales, significant differences were found between the children with and without diagnosed RAD and/or DSED (DSM-5) in the socialization domain (Fisher's exact test, $p = 0.035$) and in the motor skills domain (Fisher's exact test, $p = 0.012$). Also, the children diagnosed with and without RAD (DSM-5) showed significant differences in the motor skills domain (Fisher's exact test, $p = 0.047$). Children diagnosed with and without DSED (DSM-5) showed significant differences in three domains, including daily living skills (Fisher's exact test, $p = 0.017$), socialization (Fisher's exact test, $p = 0.044$), and motor skills (Fisher's exact test, $p = 0.016$). No significant associations were found between the level of intellectual functioning and the scores on the Vineland Adaptive Behaviour Scales. No significant differences in intellectual functioning were found between children with and without diagnosed RAD and/or DSED (DSM-5).

Psychiatric symptomatology

One participant was excluded from this analysis, because the scores on the DISC-IV indicated the presence of 12 out of the 16 psychiatric disorders, which gave rise to doubt about the validity of that evaluation. According to the DISC-IV and AUTI-R, 44 of the 54 children exhibited one or more diagnosed psychiatric disorder; of these, 25 children were diagnosed with two or more disorders. The children with RAD and/or DSED had comorbid disorders (n = 10, mean = 2.30, SD = 1.77) more frequently than children with no RAD and no DSED (n = 44, mean = 1.48, SD = 1.13), but the difference was not significant: $t(52) = -1.86$, $p = 0.069$. Children with and without attachment diagnoses were not significantly different in individual diagnoses of psychiatric disorders (e.g. anxiety, mood, disruptive behaviour), or autistic disorders (DISC-IV and AUTI-R; Table III). Also, the groups were not significantly different in ADHD prevalence; ADHD was found in eight out of ten children (80 per cent) diagnosed with RAD and/or DSED and in 24 out of 44 children (55 per cent) without a diagnosis of RAD and/or DSED. ADHD was the most frequently mentioned diagnosis.

Table III DISC (DSM-IV)-classifications for children that displayed disturbed (RAD, DSED) and non-disturbed (no RAD or DSED) attachments

Instrument and classification (n=54 children)	*No RAD or DSED (n=44)*	*RAD (n=9)*	*DSED (n=7)*	*RAD and/or DSED (n=10)*
DISC (DSM-IV): Anxiety - Social phobia (*n*=8)	6	1	1	2
DISC (DSM-IV): Anxiety - Separation anxiety (*n*=3)	2	1	1	1
DISC (DSM-IV): Anxiety - Specific phobia (*n*=2)	2	0	0	0
DISC (DSM-IV): Anxiety - Panic disorder (*n*=1)	1	0	0	0
DISC (DSM-IV): Anxiety - Agoraphobia (*n*=0)	0	0	0	0
DISC (DSM-IV): Anxiety - Generalized anxiety (*n*=7)	6	1	0	1
DISC (DSM-IV): Anxiety - Selective mutism (*n*=0)	0	0	0	0
DISC (DSM-IV): Anxiety - Obsessive compulsive disorder (*n*=1)	1	0	0	0
DISC (DSM-IV): Anxiety - Posttraumatic stress disorder (*n*=1)	1	0	0	0
DISC (DSM-IV): Mood - Major depressive episode/ Dysthymia (*n*=3)	2	1	1	1
DISC (DSM-IV): Mood - (Hypo)manic episode (*n*=1)	0	1	1	1
DISC (DSM-IV): Disruptive - ADHD (*n*=32)	24	7	5	8
DISC (DSM-IV): Disruptive - ODD (*n*=13)	9	3	4	4
DISC (DSM-IV): Disruptive - Conduct disorder (*n*=2)	1	1	1	1
DISC (DSM-IV): Other - Pervasive developmental disorder (*n*=15)	10	3	3	4
AUTI-R: Any Anxiety disorder (*n*=16)	13	2	2	3
AUTI-R: Any Mood disorder (*n*=3)	2	1	1	1
AUTI-R: Any Disruptive disorder (*n*=36)	27	8	6	9

Emotional and behavioural problems

Teachers ratings on the DBC indicated that significantly more disruptive and anti-social behaviour was observed in children with mild mental retardation (IQ 50-70) than in children with borderline intellectual functioning (IQ 71-85) (X^2 (1, n = 55) = 4.26, p = 0.039). No other significant differences were found between the level of intellectual functioning and the severity of emotional and behavioural problems.

With the Mann-Witney U-test a significant positive association was found between disruptive anti-social behaviour, based on parent reports (VOG-O/DBC-P), and

attachment-related disorders (DSM-5) (U = 137.5, Z = -1.987, p = 0.047). That is, children with an attachment diagnosis showed significantly more disruptive anti-social behaviour than children without attachment diagnoses. Children diagnosed with DSED showed more disruptive anti-social behaviour (VOG-O/DBC-P) (U = 94.5, Z = 2.323, p = 0.020*)* than children not diagnosed with DSED. There was also a significant positive association (U = 76, Z = -2.784, p = 0.005*)* between self-absorbed behaviour (VOG-O/DBC-P) and the DSED diagnosis. No significant associations were found between emotional and behavioural problems, based on teacher reports (VOG-L/DBC-T), and attachment-related disorders.

Self-absorbed behaviour, based on parent reports (VOG-O/DBC-P) was significantly different among children with and without a DSM-5 attachment related disorder (X^2 (1, n = 55) = 7.69, p = 0.006), among children with and without RAD (X^2 (1, n = 55) = 5.14, p = 0.023), and among children with and without DSED (X^2 (1, n = 55) = 8.93, p = 0.003). Communicative disturbances, based on teacher reports (VOG-L/DBC-T), were significantly different among children diagnosed with and without RAD X^2 (1, n = 55) = 4.34, p = 0.037), and the total emotional problems scores were significantly different among children diagnosed with and without DSED (X^2 (1, n = 55) = 5.17, p = 0.023).

Discussion

This study evaluated children (aged 7-14 years) with a mild or borderline intellectual disability (IQ 50-85) that had been referred for psychiatric assessment and followed up one year later. We found that nearly one in five (18 per cent) of these children was diagnosed with an attachment disorder RAD or DSED, based on the DSM-5 criteria (APA, 2013) and the assessment procedure recommended by the AACAP (2005) Practice Parameter. We also found that the BDSA, a clinical scoring instrument for eight behavioural signs of disturbed attachment behaviour, was helpful in distinguishing disorders in attachment behaviour.

Among children with an attachment-related disorder diagnosed as RAD and/or DSED, those with a history of pathogenic care and observed maladaptive behaviour also showed a significant disadvantage in adaptive behaviour compared to children referred for psychiatric assessment but not diagnosed with an attachment-related disorder. This

suggested that attachment-related disorders may hamper a child's developmental potential more dramatically than other disorders. According to the stress-attachment model (Janssen *et al.*, 2002), a lack of perceived secure attachment relationships in times of stress may lead to escalating distress and undermine opportunities for learning and exploring. Integrative therapy for attachment and behaviour was recently developed for treating children and adolescents with severe challenging behaviour that were diagnosed with RAD. The participants showed more exploratory behaviour during sessions with the integrative therapist than during sessions with a control therapist. During integrative therapy, at first, the child showed more exploratory behaviour towards the therapist; then, later in therapy, the participants explored their environment; e.g., objects (Sterkenburg and Schuengel, 2011).

In the present study, parents of children with attachment related disorders indicated that the children showed severe pathological self-absorbed behaviour (e.g. eats non-food items, preoccupied with trivial items, hums, grunts). This finding was consistent with the results from a previous study by Ashworth *et al.* (2009), who studied adults with intellectual disability that resided in institutions. Pica behaviour (a form of pathological self-absorbing behaviour) was associated with a lack of social and family relationships and with an absence of participation in (recreational) activities.

What are the practical implications?

The results showed that RAD and/or DSED could be diagnosed with DSM-5 criteria (APA, 2013) and the assessment procedure described by the AACAP (2005) Practice Parameter. Therefore, the DSM-5 can be implemented according to the recommendations for the Practice Parameter.

No IQ differences were found between children without and those with RAD and/or DSED. However, lower adaptive functioning was found for children with RAD and/or DSED. Therefore, in addition to ameliorating emotional and behavioural problems, a multi-disciplinary and multi-treatment approach may contribute to unlocking a child's developmental potential. However, treatment may not automatically resolve all challenging behaviour. In an intervention that starts with an attachment-based intervention, followed by behaviour therapy, positive results will require a combination of building a therapeutic attachment and behaviour modification (Sterkenburg *et al.*, 2008, Schuengel *et al.*, 2009).

Furthermore, clinicians should be alert to comorbid diagnoses. In this study, a majority of children diagnosed with ADHD was also diagnosed with RAD or DSED.

What new information does this paper have?

This study provides new information about the functioning of children with attachment disorders, in the domains of adaptive behaviour and mental state.

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Chapter 5

General Discussion and Summary

This chapter is partly published as

Giltaij, H., Sterkenburg, P., & Schuengel, C. (2016). Gehechtheidsstoornissen [attachment disorders]. In: Didden, R., Troost, P., Moonen, X., & Groen, W. (Eds.). *Handboek Psychiatrie en lichte verstandelijke beperking [Handbook psychiatry and mild intellectual disability] (pp. 127-144).* Utrecht, De Tijdstroom.

Introduction

The main aim of this thesis was to contribute to the description of attachment related disorders and to improvements in the clinical mental health practice regarding diagnostic assessment of children presenting with attachment-related problems, as described in DSM-IV (RAD inhibited type and RAD disinhibited type) and the DSM-5 (RAD and DSED). This study included children aged 5-11 years at baseline, cognitively subnormal or with a mild intellectual disability, who had been referred for child psychiatric consultation. The second aim was to contribute to the dissemination of insights gained, and thereby adding to the knowledge and clinical expertise of professionals encountering attachment and attachment related disorders in daily practice, in particular concerning caregivers and clinicians of children with intellectual disability. In this chapter I reflect on the main findings and conclusions of this study project. These findings will also be discussed with respect to the limitations of the study and the implications for further research and clinical practice.

This chapter starts with a description of current opinions about the concept of attachment related disorders. On the one hand publications describing a continued differentiation and specification of variants of disordered attachment (Zeanah & Lieberman, 2016) are becoming more numerous, and the guidelines for assessment and treatment on the basis of research from the past 10 years have been expanded (Zeanah, Chesher, Boris, & the American Academy of Child and Adolescent Psychiatry [AACAP] Committee on Quality Issues [CQI], 2016). On the other hand, the entire concept of disordered attachment is being questioned and calls are being made to remove the disorder, assessment, and treatment of disordered attachment from clinical practice (Allen, 2016).

Developments in the concept of disordered attachment resulted in the concept being revised through an amendment of DSM-IV to DSM-5 (American Psychiatric Association [APA], 2013) (see Chapter 1, Introduction), in which the two subtypes of Reactive Attachment Disorder (RAD) are defined as two distinct disorders, RAD and Disinhibited Social Engagement Disorder (DSED). The aetiology of both disorders is rooted in severe social and emotional neglect or deprivation during infancy. RAD is described explicitly as a disorder in attachment development with a characteristic pattern of disordered attachment behaviour. This is a child-specific disorder. In the case of DSED, it is less clear

whether it can be understood as a disorder in attachment development, given that the characteristic behavioural pattern of culturally inappropriate, overly familiar relations with relative strangers is not only observed among children who presumably lack a selective attachment relationship, but also among those who do appear to have developed an attachment relationships with their a primary caregiver. That is why the extent to which DSED can be considered as an attachment disorder is currently being hotly debated (Zeanah et al., 2016; Zeanah & Gleason, 2010, 2015). For this reason we have chosen to use in this chapter the term "attachment related disorders" for the combination of RAD and DSED.

The discussion of the extent to which, in addition to the RAD and DSED disorders referred to here, certain psychopathological behavioural patterns in young children in a specific relationship with an adult caregiver may also be defined as a diagnosable disordered relationship is also important. To that end, Zeanah and Lieberman (2016) proposed incorporating a Relationship-Specific Disorder of Early Childhood in the Diagnostic Classification of Mental Health and Developmental Disorders of Infancy and Early Childhood DC: 0-5 (Zero to Three, 2016). In this document they describe children who have had serious negative experiences in their attachment development with a specific primary caregiver, which, within the relationship between the child and the specific caregiver (and not in the relationship with other primary caregivers) have led to a persistent pattern of emotional or behavioural disorders, such as strongly oppositional behaviour, hypervigilance, putting themselves in a self-endangering situation, refusal to eat or sleep, or overly controlling behaviour.

Broadening the focus of disordered attachment as a child-specific disorder into a spectrum of child-specific to relationship-specific disorders is based in part on the assumed serious consequences of disordered attachment development in the early years of life for children's functioning and developmental outcomes. In spite of the lack of research into the linkages between relationship-specific disordered attachment and developmental outcomes, investigations into individual differences in the quality of attachment relationships have shown that this quality of relationships has predictive value for a broad spectrum of development outcomes, albeit with modest effect sizes (Groh, Fearon, van IJzendoorn, Bakermans-Kranenburg & Roisman, 2017). The clinical relevance of problematic attachment relationships on the one hand and the hitherto lack of diagnostic instruments on the other hand added to the confusion about the

concept of disordered attachment in clinical practice and the broad variation in its application. Zeanah and Lieberman's (2016) proposal is an attempt to resolve this confusion and to provide a coherent clinical conceptual framework for attachment and mental health.

Allen (2016) refers to the lack of consensus within clinical practice regarding the use and meaning of the terminology of disordered attachment and attachment therapy, which frequently causes confusion about the usage and meaning of these terms. Consequently, attachment related disorders (RAD and DSED) tend to be both overdiagnosed and underdiagnosed in clinical practice (Woolgar & Baldock, 2015). Allen (2016) claims that disordered attachment as a concept is debatable in current practice, is often misunderstood and misused, is exceptionally rare, and that clinical intervention would appear unnecessary, save for placing the child with a supportive primary caregiver. Allen does not dispute that some children, on account of a background of serious neglect or maltreatment (in their early years), may exhibit patterns of severely disordered behaviour in their relations with others and in self-regulation. However, a majority of children with a comparable background do not exhibit these atypical behaviours. Allen (2016) poses the question whether it might also be possible to describe these deviant behaviours as (a combination of) other forms of psychopathology, such as externalizing behavioural disorder, and points out the danger that designating the behaviour as attachment related disorder may overshadow other disorders that might exist. Allen is correct in asserting that the establishment of the final diagnosis is very important, as this provides the basis for the therapeutic treatment or intervention. Therefore, incorrect, incomplete, or invalid diagnosis increase the chances of incorrect or inadequate treatment.

While Allen's critical account of disordered attachment, as a concept within clinical practice, is interesting and invites discussion, his proposal to abandon the diagnosis of RAD and/or DSED altogether may indeed be too radical. This proposal would affect a group of children, however small that group may be, that do exhibit observable patterns of severe maladaptive social behaviours that are not captured within other diagnostic categories. Moreover, these behaviours are found among children growing up in institutions or in living conditions in which serious neglect and absence of stable attachment opportunities occur, and thereby highlight the toxicity of these environments for children. By repudiating these distinct patterns of disordered and maladaptive

behaviour, a group of children may be denied a diagnosis and consequently appropriate treatment.

The ongoing confusion and the inconsistent use of terms such as disordered attachment, attachment assessment and attachment treatment may be understood as the result of ongoing investigation and debate by a small number of research groups. The lack of diagnostic measurement instruments accepted as reliable and valid, and treatment methods shown to be effective offers a great deal of room to clinical professionals for an individual interpretation of the above-mentioned terms. This calls for more coordination and integration between the scientific research and the clinical practice on development and –implementation of attachment knowledge.

An important step was the development and approval of the Practice Parameter for the assessment and treatment of children with Reactive Attachment Disorder (according to the DSM-IV) by the American Academy of Child and Adolescent Psychiatry (AACAP) in 2005 with guidelines for clinical practice as well as scientific research, based on the then prevailing state-of-the-art. While the Practice Parameter revealed many issues of clinical consensus, other issues were left unresolved, awaiting more clinical research. These issues regarded specific diagnostic instruments and interventions, as well as the application of the concepts in different populations. Recently Fletcher, Flood, and Hare (2016) explored the clinical difficulties associated with attachment relations in people with intellectual and developmental disabilities, and described a Clinician's Guide to practice and research. In the Netherlands a series of single case experiments with six children with visual and intellectual disability and a history of severe insufficient care was conducted, demonstrating the viability of protocol-based treatment (Sterkenburg & Schuengel, 2014). The encouraging findings of this small-sample effectstudy led to the publication of the Best Practice for diagnosing problematic attachment in children with visual and mild intellectual disability (Dekker-van der Sande & Janssen, 2010). This Best Practice was the basis for the Dutch Guidelines for problematic attachment in childcare and child protection, approved by the professional associations of psychologists and special educators (De Wolff et al., 2014).

The central objective of the studies described in this document was to describe and investigate a diagnostic research process for Reactive Attachment Disorder (RAD) and Disinhibited Social Engagement Disorder (DSED) as described in DSM-5, among children

in the school-aged stage, functioning at a low average, borderline or mildly intellectual disabled (IQ 50-85) level and referred for child psychiatric consultation. This research process also generated information about the prevalence of RAD and DSED and the diagnosis of disordered attachment behaviour in this specific vulnerable group of children. Comorbidity with other psychiatric disorders and adaptive functioning was also reviewed.

The outline of the diagnostic process was based on the guidelines and recommendations as set out in the AACAP Practice Parameter for the diagnosis and treatment of children and adolescents with reactive attachment disorder (2005, revised 2016). Distinction is made here between the following elements: information about the early developmental and educational background of the child, in particular about the attachment behaviour of the child with its primary caregivers, and direct observation of the interaction of the child with its primary caregiver and an unknown stranger in a relatively structured procedure with stress-enhancing moments. Links to other forms of psychopathology and the level of development of the child's performance were studied by means of the completion of specific diagnostic questionnaires.

This chapter will summarize and discuss the findings of the studies done for dissertation, as presented in Chapters 2, 3, and 4. The studies' limitations as well as implications for future investigation have also been added. And finally, a description of the clinical implications will be included, as will a description of the clinical research process for determining RAD and DSED.

Summary and Conclusions

Attachment related disorders and ASD

While DSM-5 no longer defines the disorders RAD and DSED as 'markedly disturbed and developmentally inappropriate social relatedness', but as 'a behavioural pattern of inhibited (RAD) or disinhibited (DSED) behaviour towards adults', with associated behavioural characteristics, a DSM-5 RAD diagnosis is still ruled out if Autism Spectrum Disorder (ASD) is also diagnosed. Research on the distinctions between both disorders is therefore important to determine whether this criterion should be preserved in future editions of classification and diagnostic systems. Chapter 2 reports on the

investigation of the overlap and differences between the behavioural characteristics of ASD versus RAD (Inhibited) and DSED (Disinhibited). The behavioural characteristics of ASD were measured using the AUTI-R screening instrument (Van Berckelaer-Onnes & Hoekman, 1991); the behavioural characteristics of RAD and DSED were measured using the Disturbances of Attachment Interview screening instrument (DAI, Smyke & Zeanah, 1999). The participants were children (from 5 to 11 years of age) with an intellectual disability (IQ 50-85), who had been referred for psychiatric assessment to a specialized centre for child and adolescent psychiatry on account of psychological, psychiatric or behavioural problems. On the basis of the AUTI-R scores, it was shown that 27% of the children had either possible or definite ASD, with a clear majority of boys over girls. No association was found between the ASD classification and intellectual functioning level, ethnic background or age. Analyses at the level of classification and clusters of behavioural characteristics did not reveal any significant association between ASD (measured using AUTI-R) and the RAD and/or DSED disorders (measured using DAI). It may therefore be concluded that, as intended, these instruments measure different phenomena, making them suitable for structured assessment and differential diagnosis of ASD, RAD and DSED. These findings also show that foregoing the exclusion criterion of ASD will not necessarily lead to overidentification of RAD and DSED.

Attachment related disorders and multi-measurement assessment

The Disturbances of Attachment Interview (DAI) is a semi-structured interview for determining behavioural signals of disordered attachment as perceived by adults who regularly interact with the child. If the clinician is well trained and experienced, this instrument can be used to obtain a score in a relatively short time (20 to 40 minutes). Interviewers are trained to pursue their line of questioning to clarify whether the informant has a sufficiently adequate overview of the attachment behaviour. Still, it is important to remain aware that the information elicited from the interviewed parent or caregiver is influenced by their interpretation of children's behaviour as abnormal and worrisome. This level is partly a function of presuppositions and selective attention on the part of the informant, but also of the natural variation in situations in which the disordered behaviour may manifest itself.

It is therefore imperative that positive findings on the DAI be followed up by direct observations of children's interactions with parents or primary caregivers, in standardized situations in which the child is exposed to a stress factor (Zeanah et al., 2016). Moreover,

the child's attachment behaviour vis-à-vis the parent or primary caregiver must be compared with the attachment behaviour towards an unfamiliar adult in comparable situations. To that end, as recommended in the Practice Parameter we have used the observation protocol of the Clinical Observation of Attachment (COA) for this study, for which the protocol's stressors have been adapted to the age range of the study population. Children's observed behaviours are then assessed on the eight scales of (disturbed) attachment behaviour of the List of Behavioural Signs of Disturbed Attachment in Young Children (BSDA) (Zeanah, Mammen, & Lieberman, 1993), resulting in an overall score between 8 and 40. Given that the RAD and DSED disorders are presumed to be a response to a developmental background in which the child is exposed to a pattern of serious neglect and insufficient care in order to form a stable attachment relationship, it is essential to pursue questioning and study the child's life history, in particular its early years.

Follow-up investigations were conducted in order to determine the DSM-5 RAD and DSED diagnosis for 55 of the children who participated in the study described in Chapter 2, during which the above-mentioned instruments (DAI, COA, BSDA) were used and children's development history was taken into account. Chapter 3 describes the results obtained with these instruments. Associations were tested between the child's (disordered) attachment behaviour (DAI) as described by the parent/primary caregiver, the child's observed (disordered) attachment behaviour (COA and BSDA,) and the described background of extreme insufficient care for the child. Experienced clinical researchers determined the clinical diagnosis of RAD and/or DSED on the basis of factors including the observed behaviour and the child's life history.

The scores for both described and observed attachment behaviour, and also those for the occurrence of extreme insufficient care were found not to be associated with the child's gender, ethnic background, age or level of cognitive functioning. No associations were found between the characteristics and occurrence of RAD and/or DSED and these child characteristics. One out of three children in this group of 55 was described by the parent/primary caregiver as having behavioural characteristics that might suggest RAD and/or DSED. On the basis of the described development history of their early years one out of five children (22%) were assessed as having a background of extreme insufficient care, as a result of which the child would have experienced serious difficulty entering

into a stable attachment relationship with the parent/primary caregiver. A clinical diagnosis of RAD and/or DSED was made for 10 of the 55 children (18%).

The BSDA was used for scoring the observed disordered attachment behaviour by independent and trained researchers. The scores for the 55 children varied from 9 to 33. The BSDA scores of children who were described (on the DAI) by the parent/primary caregiver as having RAD and DSED, reflected a BSDA score of 26 to 33, with an average of 29.3 (SD = 2.7), which meant there was a significant difference with the children whose parents described them as not having RAD and/or DSED, who had scores varying from 9 to 29, with an average of 16.2 (SD = 5.2). There were significant differences in average scores and ranges for RAD as an individual disorder (with scores from 17 to 21 (M = 18.7, SD = 2.1) and DSED (with a BSDA score of 10 to 28 (M = 17.4, SD = 6.0) between children who had been described by the parent/primary caregiver as having RAD and/or DSED, and those who had not been categorized as such. Similarly, there was significantly more evidence of extreme insufficient care, determined on the basis of the described developmental anamnesis, for children whose parent/primary caregiver described them as having RAD and/or DSED, than for children who had not been categorized as such.

Children whose development history revealed a background of extreme insufficient care had significantly higher scores for observed disordered attachment behaviour (with an average BSDA score of 25) than children for whom no extreme insufficient care could be determined (an average BSDA score of 16). This confirms that: 1) there is an association between pathogenic care and disordered attachment behaviour observed during the COA and scored as such by means of BSDA, 2) BSDA contributes to the adequate assessment of disordered attachment behaviour and therefore 3) confirms the importance of including BSDA in the diagnostic protocol.

Attachment related disorders and comorbidity

Children with an intellectual disability experience a three to four times higher risk of developing psychopathological disorders than children without this disability (Dekker, Douma, de Ruiter, & Koot, 2006; Einfeld, Ellis, & Emerson, 2011). Various studies have demonstrated an association between problematic attachment and vulnerability to developing psychopathological disorders (Mikulincer & Shaver, 2012). Children who have grown up in orphanages or institutions are at a high risk of delayed development

and disorders, including psychiatric disorders (MacLean, 2003). Chapter 4 compares the children (*n* = 55) diagnosed for Chapter 3 with RAD and/or DSED with children who had not been given this diagnosis. The Vineland Adaptive Behaviour Scales (Sparrow, Carter, & Cicchetti, 1993) revealed that children with RAD and/or DSED were significantly more delayed in the development of their adaptive functioning. Psychiatric comorbidity was investigated with the National Institute of Mental Health Diagnostic Interview Schedule for Children, Version IV (NIMH-DISC-IV; Shaffer, Fisher, Lucas, Dulcan, & Schwab-Stone, 2000) of the parent/primary caregiver. A psychiatric disorder (DSM-IV anxiety, mood, behaviour or pervasive development disorder) was found in more than 81% of the children. Two or more psychiatric disorders were found in 46% of the children. Children for whom RAD and/or DSED were found had an average of 2.3 comorbid disorders, while the group of children without RAD and/or DSED exhibited positive scores for an average of 1.5 psychiatric disorders; this difference was not statistically significant. ADHD was by far the most frequent disorder in this sample.

Teachers filled out the Developmental Behaviour Checklist (DBC) (Einfeld & Tonge, 1991, 1994, 1995; Einfeld, Tonge, & Parmenter, 1999). Results showed that transgressive and antisocial behaviours, included within diagnoses such as ADHD, occurred significantly more frequently in this study group among children functioning at a lower cognitive level. Conversely, the responses of parents on this checklist indicated an association between the occurrence of these behavioural problems and a diagnosed RAD and/or DSED disorder. Parents' responses also indicated a positive association between introverted behaviour and the RAD or DSED diagnosis. Children with an RAD and/or DSED disorder had significantly more emotional and behavioural problems than children from this study group without such a disorder. These results show that RAD and DSED are more prevalent among children showing also other forms of psychopathology. Nevertheless, not all children with RAD and/or DSED received other comorbid diagnoses.

Limitations of the study and implications for future research

The prevalence of attachment related disorders among children in the general population is estimated to be very low (Richters & Volkmar, 1994), although others have found a prevalence of over 1% and called this considerable (Minnis et al., 2013), as it is comparable to the prevalence of for example autistic disorders. The prevalence of disordered attachment behaviour appears to be substantial in clinical samples, although hardly any studies are conducted into the prevalence of the RAD and DSED disorders as established according to the AACAP guidelines. Previous studies into the behavioural characteristics of disordered attachment report that such characteristics are observed among 40% of young institutionalized children (Smyke, Dumitrescu, & Zeanah, 2002; Zeanah, Smyke, & Dumitrescu, 2002) and among 18% (Oosterman & Schuengel, 2008) to 38% (Zeanah, Scheeringa, Boris, Heller, Smyke, & Trapani, 2004) of children in therapeutic foster care. Gleason et al. (2011) published their large-scale study focusing on the prevalence of the RAD and DSED diagnosis in a specifically high-risk group (young children who have spent a significant part of their lives growing up in neglected conditions in Romanian orphanages, some of whom were later placed in foster care). Of these children, 4% received a RAD diagnosis, while 17 to 18% received a DSED diagnosis. Minnis et al. (2013) found in their large multi-informant study a prevalence of 1.4% of attachment related disorders in children (age 6-8 years) in a deprived urban area. Vervoort, de Schipper, Bosmans and Verschueren (2013) examined the psychometric properties of screening questionnaires for symptoms of RAD and DSED in school-aged children with severe emotional or behavioural disorders, and the convergence of these screening instruments with observed behaviour in a structured observational schedule for attachment related disorders. Scheper and colleagues (2016) reported RAD behaviour (9% and 27%) and DSED behaviour (42% and 51%) in two groups of young children referred for treatment of emotional and/or behavioural problems: the subgroup of home-reared children and the subgroup of children in therapeutic foster care after out-of-home placement.

Table 1 Overview multi-informant/-instruments studies of children with disordered attachment behaviour or attachment related disorders.

Study	*Study group*	*Focus*	*Sample*	*Used instruments*	*Key findings*
Smyke, Dumitrescu, & Zeanah (2002)	Severely deprived, institutionalized children (BEIP project) (age 1-7 years)	Prevalence of signs of RAD or DSED	*N* = 94 children: Standard Unit (*n* = 32), Pilot Unit (*n* = 29), Never-institutionalized children (*n* = 33)	*Care history*: **No Measure** *Interview:* Disturbances of Attachment Interview (DAI) *Observation:* **No Measure** *Psychopathology*: **No Measure**	Standard Unit: RAD signs: 37% DSED signs: 43% Pilot Unit: RAD signs: 14% DSED signs:17 %
Zeanah, Scheeringa, Boris, Heller, Smyke, & Trapani (2004)	Maltreated children (age 10-48 months) in foster care	Prevalence of signs of RAD or DSED	*N* = 94 children	*Care history:* maltreatment legally established *Interview:* Disturbances of Attachment Interview (DAI) *Observation*: **No Measure** *Psychopathology*: **No Measure**	RAD signs: 45% DSED signs: 37%
Zeanah, Smyke, Koga, Carlson, and the BEIP Core Group (2005)	Severely deprived, institutionalized children (BEIP project) (age 12-31 months)	Differences of signs of RAD and DSED in Institutio-nalized and never-institutio-nalized children	*N* = 145 children: Institutionalized children (*n* = 95) and never-institutionalized children (*n* = 50)	*Care history:* Observational Record of the Caregiving Environment (ORCE) *Interview:* Disturbances of Attachment Interview (DAI) *Observation:* Strange Situation Procedure (SSP) *Psychopathology:* Infant Toddler Social Emotional Assessment (ITSEA)	Significant differences between institutionalized and never-institutionalized children for RAD ($p<.001$) and for DSED ($p<.01$)

Study	*Study group*	*Focus*	*Sample*	*Used instruments*	*Key findings*
Oosterman & Schuengel (2008)	Children (age 2-7 years) in foster care	Prevalence of signs of RAD or DSED	*N* = 69 children	*Care history*: **No Measure** *Interview:* Disturbances of Attachment Interview (DAI) *Observation:* Attachment Q-set (AQS) on videotaped home-observation *Psychopathology:* Child Behaviour Checklist (CBCL)	RAD/DSED signs: 18%
Gleason, Fox, Drury, Smyke, Egger, Nelson & Zeanah (2011)	Previously institutionalized children (BEIP project) (age 22 to 54 months)	Prevalence and stability over time of RAD and DSED	*N* = 122-135 children	*Care history:* Observational Record of the Caregiving Environment (ORCE) *Interview:* Disturbances of Attachment Interview (DAI) *Observation:* Strange Situation Procedure (SSP) *Psychopathology:* Preschool Age Psychiatric Assessment (PAPA), Infant Toddler Social Emotional Assessment (ITSEA)	RAD: from 4.6% to 4.1% DSED: from 31.8% to 17.6%

Study	*Study group*	*Focus*	*Sample*	*Used instruments*	*Key findings*
Minnis, Macmillan, Pritchett, Young, Wallace, Butcher, Sim, Baynham, Davidson & Gillberg (2013)	Children (age 6-8 years) in a deprived UK population	Prevalence of RAD and/or DSED	Main group: *N*=1646 children (SDQ, RPQ, SIMD), out which 49 likely RAD cases (DAWBA, CAPA-RAD, Observation)	*Care history:* Scottish Index of Multiple Deprivation (SIMD) *Interview:* Child and Adolescent Psychiatric Assessment, Reactive Attachment Disorder module (CAPA-RAD), Relationship Problems Questionnaire (RPQ) *Observation:* Observational Checklist for Reactive Attachment Disorder *Psychopathology:* Strengths and Difficulties Questionnaire (SDQ), Development and Wellbeing Assessment (DAWBA),	RAD and/or DSED: 1.4% (2.3% borderline/ suspected RAD/DSED)
Kay & Green (2013)	Maltreated, out-of-home Looked After Care adolescents (age 10-16 years) without history of institutional care	Characteri-zation of RAD/DSED behaviours	*N* = 153 adolescents, with control group of low risk group (*n*=42).	*Care history:* Maltreatment Classification System (MCS) *Interview:* Child and Adolescent Psychiatric Assessment, Reactive Attachment Disorder module (CAPA-RAD) *Observation:* **No Measure** *Psychopathology:* Child Behaviour Checklist (CBCL)	RAD/DSED signs: 63% DSED signs: 56%

Study	*Study group*	*Focus*	*Sample*	*Used instruments*	*Key findings*
Vervoort, de Schipper, Bosmans, & Verschueren (2013)	Children (age 6-10 years)from schools for special education	Examination psychometric properties of the Relationship Problems Question-naire (RPQ)	*N* = 152 children	*Care history*: Indicated by school psychologist *Interview:* Disturbances of Attachment Interview (DAI), Relationship Problems Questionnaire (RPQ) *Observation:* Observational Checklist for Reactive Attachment Disorder *Psychopathology:* Strengths and Difficulties Questionnaire (SDQ)	The RAD and DSED subscales of RPQ and DAI showed moderate, positive association. The DSED subscale of the RPQ and DAI showed moderate to strong positive correlation with the observed subscale Approach to Stranger
Scheper, Abrahamse, Jonkman, Schuengel, Lindauer, de Vries, Doreleijers & Jansen (2016)	Children (age 2-8 years), home reared and foster care, referred for treatment of emotional and behavioural problems	Prevalence of RAD and DSED behaviour	*N* = 200 children: Referred home reared children (*n* = 141) and referred foster children (*n* = 59)	*Care history:* Maltreatment Classification System (MCS) *Interview:* Disturbances of Attachment Interview (DAI) *Observation:* **No Measure** *Psychopathology:* Child Behaviour Checklist (CBCL)	Referred home reared children: RAD behaviour: 9% DSED behaviour: 42% Referred foster children: RAD behaviour: 27% DSED behaviour: 51%

Study	*Study group*	*Focus*	*Sample*	*Used instruments*	*Key findings*
Kay, Green & Sharma (2016)	Adopted children (age 6-11 years) without history of institutional care	Prevalence of DSED	*N* = 60 children, with control group of clinic-referred, non-maltreated children with externalizing disorder (ED, *n*=26) and low risk group (LR, *n*=55).	*Care history:* Maltreatment Classification System (MCS) *Interview:* Child and Adolescent Psychiatric Assessment, Reactive Attachment Disorder module (CAPA-RAD), Relationship Problems Questionnaire (RPQ) *Observation:* registration of DSED behaviour during 2,5 hour home visit *Psychopathology:* Development and Wellbeing Assessment (DAWBA)	Matching criteria DSED: 49% of Adopted children; 4% of ED-children; 6% of LR-children
Giltaij, Sterkenburg & Schuengel (2017)	Children with mild intellectual disability (age 5-11 years), referred for assessment of emotional and behavioural problems	Prevalence of RAD and DSED	*N* = 55 children	*Care history:* Structured review of medical records on DSM-5 defined extremes of insufficient care *Interview:* Disturbances of Attachment Interview (DAI) *Observation:* Clinical Observation of Attachment 6-15 (COA 6-15), List of Behavioural Signs of Disturbed Attachment in Young Children (BSDA) *Psychopathology:* Diagnostic Interview Schedule for Children, version IV (DISC-IV)	According to DAI: Signs RAD or DSED: 35% RAD signs: 16% DSED signs: 27% According to DSM-5: RAD and/or DSED: 18% RAD: 16% DSED: 15%

Children with an intellectual disability who exhibit psychological or behavioural problems also constitute a group that is at risk of developing disordered attachment. The level of elevated risk of behavioural characteristics and a RAD or DSED diagnosis was as yet unknown among this group. This is why considerable efforts were made to recruit children into this study group. Recruitment took place at all the large centres for child and adolescent psychiatry specializing in children with mild or borderline intellectual functioning, across the Netherlands.

Figure 1 Overview participating centres for child and adolescent mental health care, specialized in children with mild or borderline intellectual functioning.

This resulted in a study group of 102 children who met the criteria of age, level of cognitive functioning and language competence (Chapter 2). The parents of the children in this study group were again approached for a follow-up study between one and two years later. However, it turned out that a considerable proportion of the study group no longer had any care relationship with the psychiatric center, and for some of the parents, participation in the follow-up study was considered to be an additional pressure for the child or the parents, on top of the on-going care being provided by the

center. The parents of 55 children gave their informed consent for participation in the follow-up study (Chapters 3 and 4). On the basis of the scores for the Disturbances of Attachment Interview, this study showed that, according to the information from the primary caregiver, 42% of the children exhibit sufficient behavioural characteristics for an RAD Inhibited (RAD) and/or RAD Disinhibited (DSED) classification. The finding that 11% of the children meet the criteria for both RAD Inhibited and RAD Disinhibited classifications confirms the description of the two separate RAD and DSED disorders in DSM-5 instead of the incompatible classification types of Inhibited RAD or Disinhibited RAD as stated in DSM-IV. No correlation was found between the RAD and DSED classifications and gender, level of intellectual performance, ethnic background or age. The clinical diagnosis of RAD and/or DSED was made for 18% of these children, where the AACAP guidelines were used for making the diagnosis.

Notwithstanding the efforts to collect as large a sample as possible, the study group was too small to make any reliable statements at a subgroup level (RAD, DSED and RAD/DSED subgroups). The prevalence figures could also be biased on account of selective non-response. A limitation of this study is the lack of a clear understanding of the factors associated with this non-response. That makes it impossible to correct the results in order to account for this. Due to these limitations, no conclusions can be drawn in relation to the prevalence figures for the diagnoses in the target population.

Both DAI and COA were originally developed for the assessment and observation of disordered attachment behaviour among children up to the age of five. The use of these instruments was originally limited to the population of children up to the age of five, given that the phenomenology of disordered attachment behaviour among older children remains unclear. Longitudinal research is needed to develop better understanding and description of symptoms of RAD and DSED among older children, such as the Bucharest Early Intervention Project (Smyke et al., 2012), in which the development of children diagnosed with disordered attachment is monitored over a prolonged period and compared in various conditions of growing up. Just like attachment behaviour develops as the child matures, it might be assumed that disordered attachment behaviour develops with age as well.

Reasons for expecting developmental changes are that children over the age of five resolve problems on their own more frequently, that they will make contact with

unknown individuals more frequently, and that children internalize safety more readily. Similarly, in the case of disinhibited behaviour among adolescents with DSED, this may be demonstrated by the way in which the young person shapes their friendships. Kay and Green (2013) found in their study with non-institutionalized Looked-After-Care adolescents (10-16 years) with a history of early maltreatment or neglect, high prevalence of disinhibited indiscriminate behaviour (overfriendly behaviour, asking strangers personal questions, seeking comfort from strangers and wandering away from caregivers), attention seeking (demanding and possessive behaviour, wary and watchful behaviour), superficial relationships and unpredictability. The social impairment phenotype of the disorder seems persistent through the early development.

Adolescents show their disinhibited behaviour in superficial and frequently changing friendships, and in rapid reference to a contact as being their 'best friend' (Zeanah et al., 2016). There is no reason why the behavioural characteristics surveyed in DAI would not be applicable to children in the school-aged stage (Humphreys, Nelson, Fox, & Zeanah, 2017). This instrument has been used to determine disordered attachment behaviour in several recent studies among children older than five years of age (Jonkman et al., 2014; Smyke et al., 2012). However, when questioning and assessing the behaviour described by the caregiver, account must be taken of the phenomenology of this behaviour at this time of life. For example, a child in the school-aged stage distancing itself from its caregivers in an unfamiliar environment would not be considered as maladaptive as in the case of a pre-schooler. Pursuing a clear line of questioning is important in that case. When surveying behavioural characteristics relating to DAI, account must also be taken of the child's intellectual disability. For example, pursuing a line of questioning will be important for the item relating to exhibiting dangerous behaviour. Children with an intellectual disability will be less aware of the hazards in many situations. The original COA protocol (AACAP, 2005; Boris et al., 2004) was adjusted for the age range for the school-aged stage to include other factors that increase levels of stress. The other age range requires other stressors for triggering (disordered) attachment behaviour. There were no significant associations between the scores for maladaptive attachment behaviour on the COA and age for children who had been designated as having disordered attachment behaviour (Chapter 3). This means it is unlikely that increasing the age band for which the instruments were used has resulted in any overreporting of disordered attachment behaviour.

The assumed factor for the development of the attachment related disorders (RAD and DSED) is extreme insufficient care (pathogenic care). DSM-IV and DSM-5 describe extremes of insufficient care in relatively general terms, leading to a risk of over- or underassessment of the care situation for individual children. The Maltreatment Classification System (MCS) (Barnett, Manly, & Cicchetti, 1993) is a proven reliable and valid instrument for measuring forms of neglect and maltreatment (Cicchetti, Rogosch, & Thibodeau, 2012; English et al., 2005), used for determining pathogenic care in various studies of RAD according to the DSM-IV criteria. However, DSM-5 shifted this criterion more in the direction of (patterns of extremes of insufficient) care situations that are a serious limitation to form selective and stable attachment relationships. We have yet to find a reliable and valid instrument with which to determine this. In the current study, we had access to the complete clinical case files of the participants and clear criteria were defined for assessing 'extremes of insufficient care': the child's caregivers were frequently changed during the first three years of its life, or the child spent long periods in hospital or was admitted to hospital on frequent occasions, or the child was removed from its home at a young age, or the child was subject to an order of the Child Care and Protection Board or a juvenile court. While this wording defines the care situation during early childhood more concretely, making it easier to assess, it still leaves room for subjective judgment. It is also difficult to assess the level of insufficient care in the child's development history on the basis of subjective descriptions by the caregivers. It is difficult to determine whether there is (or has been) any neglect or maltreatment among very young children who are incapable of describing their experiences (verbally) (Zeanah & Gleason, 2010). This may have led to false negative assessments about extremes of insufficient care. Interesting is the approach of Cyr, Euser, Bakermans-Kranenburg, and Van IJzendoorn (2010), who conducted a meta-analysis of the correlation between key socio-economic risk indicators for the attachment development of children. They concluded that children exposed to at least five of the six risk indicators exhibited the same prevalence of disorganized attachment as children with a background of established child abuse. Such a study into the correlation of risk indicators and disordered attachment behaviour would contribute to a more objective determination of possible extreme insufficient care for the child.

In order to compare the various research measurement instruments (interviews, questionnaires, observation situation), a decision was made to include one of the two caregivers in the study. However, it is possible that the other primary caregiver, if

present, might have responded differently to, or provided a different description of the relation with the child. That is why the results may only be interpreted as being part of the relationship with this specific parent. However, for diagnosis in clinical practice it is important to observe the AACAP guidelines and to interview several caregivers and observe them in their relationship with the child. The current study focused on the child-related RAD and DSED disorders, but the method applied means we cannot discount the fact that for some children, the observed disordered attachment behaviour was relationship-specific. Follow-up studies are needed into the diagnostic classification of disordered attachment behaviour as a relationship-specific disorder (Zeanah & Lieberman, 2016).

The lack of unequivocal behavioural descriptions (thresholds) in line with the coding variation for the eight behavioural signals of adaptive and maladaptive attachment behaviour in BSDA may have left considerable room for coder bias. This issue was addressed by deploying and training five independent researchers. They received random video recordings from COAs, and each COA was scored by two researchers, independently of each other and without any prior knowledge about the child. If the scores given for a particular item differed by one point, the researchers would discuss this in order to reach a consensus. If the scores given for the behavioural signal by the two researchers differed by more than one point, the item in question was scored again by a third independent researcher, who was unaware of the scores originally given. This third score was included in the discussion in order to reach a consensus. In this way agreement arose about the thresholds to handle BSDA encodings, necessary for good interrater reliability. In this study the interrater reliability was good (.73).

Children who had received a clinical diagnosis of RAD and/or DSED on the basis of an established background of extreme insufficient care in combination with observed maladaptive attachment behaviour (in COA) achieved a score between 22 and 33 for the BSDA, with an average of 26.9. Children for whom no background of extreme insufficient care could be established achieved a BSDA score of 9 to 29, with an average score of 16.0. Four of these children were observed as having maladaptive attachment behaviour (scores of 23 to 29), although no RAD or DSED could be determined, given the lack of a background of extreme insufficient care from the available development history. These figures show that BSDA is an useful observation list for diagnosing attachment related disorders. However, further investigation is necessary into both

standardization, norms, as well as potential profiles within BSDA. There is also need to develop a semi-structured interview about the development history and circumstances of the child focused on possible pathogenic care and inadequate opportunities to form stable, selective attachment relationships. Such follow-up investigation into BSDA profiles and development background must also include the relationship-specific form of disordered attachment (Groh, Fearon, van IJzendoorn, Bakermans-Kranenburg, & Roisman, 2016; Zeanah & Lieberman, 2016).

Clinical implications

This study contributes to the development of clinical protocols for the assessment of the RAD and DSED disorders among children with a mild intellectual disability. Three guidelines are currently in place in the clinical practice of youth care in the Netherlands and mental health care for young people. First there is the internationally accepted Practice Parameter for the assessment and treatment of RAD and DSED, as published by AACAP (2005, revised 2016), which provides guidelines based on the results of current state-of-the-art scientific studies into RAD and DSED for conducting diagnostic procedures and treatment. There is also the Best Practice for identifying disordered attachment behaviour as described by Dekker-van der Sande and Janssen (2010), comprising three phases: screening, diagnosis and internal working model. These guidelines were subsequently developed into the Problematic Attachment Guideline (Richtlijn Problematische Gehechtheid), which is incorporated into the database of guidelines of the Youth Care and Protection body (Jeugdhulp en Jeugdbescherming) of the Dutch Youth Institute (Nederlands Jeugd Instituut) (De Wolff et al., 2014).

As far as we know, this dissertation is the first study to report on assessment using multiple assessment instruments, the combination of which complies with the guidelines of the Practice Parameter for diagnosing and treating children and adolescents with attachment related disorders (Zeanah et al., 2016) for the diagnoses of RAD and DSED in children with mild intellectual disability referred for psychiatric assessment because of severe emotional and/or behavioural problems. This study sets out the correlation and the limitations of these assessment instruments for the screening and diagnosis of RAD and DSED, and with that, the consequences for the clinical assessment protocol of these attachment related disorders. While the entire process of conducting diagnostic

investigation in accordance with AACAP guidelines is a time-consuming process in clinical practice, the findings of this study show that it is very important to use all the guidelines and to work through the entire protocol in order to arrive at a well-founded diagnosis of RAD or DSED. The study showed that restricting the work-up to the use of a structured clinical interview such as DAI may lead to an incorrect diagnosis for some 25% of the participants. The consequence of this finding is that the DAI in clinical practice should be regarded only as a screening instrument in the diagnostic assessment for attachment related disorders. The DAI shows to be a good instrument for obtaining an inventory of possible disordered attachment behaviour, as reported by the primary caregiver, in a relatively short space of time. It is recommended that this interview be conducted separately for both caregivers. The clinical diagnosis of RAD and/or DSED can then be determined by examining the child's (attachment) development history, focusing on any possible extreme insufficient care, in particular during the child's very early years, and by means of observing (disordered) attachment behaviour in the structured COA. BDSA would appear to be a suitable instrument for scoring the observed behaviour.

On the basis of the study results described in this document, it is recommended to apply a 'stepped-care model' in order to diagnose the RAD and DSED disorders, in which a more thorough diagnosis is made in accordance with the above-mentioned guidelines when the results of previous phases in the diagnostic process point towards a potential classification of disordered attachment.

Phase 1 comprises the completion of the *Child Behaviour Checklist* (CBCL) (Achenbach & Rescorla, 2001) or the *Developmental Behaviour Checklist* (DBC) (Einfeld & Tonge, 1991, 1994, 1995; Einfeld, Tonge, & Parmenter, 1999) for the primary caregivers and the teacher, and the completion of the development history, including the development of the attachment behaviour with the primary caregivers. If the checklist scores are high and the information from the development history (also) leads to the assumption of attachment problems and/or extreme insufficient care, it is necessary to proceed to the next phase of the assessment.

Phase 2 comprises the completion of the *Disturbances of Attachment Interview* (DAI) (Smyke et al., 2002) for at least one primary caregiver. Additionally and as needed, a screening or diagnostic investigation into potential differential diagnosis or comorbidity

of Autism Spectrum Disorder (ASD) may be conducted using the *AUTI-R* (Van Berckelaer-Onnes & Hoekman, 1991) or the *Autism Diagnostic Observation Scale* (ADOS-2) (De Bildt, Greaves-Lord, & De Jonge, 2013). If the scores from these instruments suggest disordered attachment behaviour, it is necessary to proceed to the final phase of the diagnosis.

Phase 3 comprises the completion of the *Clinical Observation of Attachment* (COA) (Boris et al., 2004, amended by Giltaij & Sterkenburg, 2017, appendix of this manuscript), with the child, the primary caregiver and an unfamiliar adult. Wherever possible, this procedure is conducted for both primary caregivers in order to determine whether the disordered attachment behaviour manifests itself in more than one caregiver relationship (child-specific disorder). A video recording is made of the observation session, which is then analysed and scored on the basis of the *List Behavioural Signs of Disturbed Attachment in Young Children* (BSDA) (Zeanah et al., 1993). High BSDA scores contribute to the diagnosis of an attachment disorder. The behavioural characteristics set out in DSM-5 are decisive factors for diagnosing RAD or DSED or both disorders. For the determination of a differential diagnosis and comorbidity of other disorders, the *Diagnostic Interview Schedule for Children, version IV* (DISC-IV) (Shaffer et al., 2000) is recommended.

It should be noted that Phases 1 and 2 are primarily intended for triaging, and are therefore liable to return incomplete information and subjective descriptions. Phase 3 relates to the child's actual observed behaviour in a standardised situation that invokes (disordered) attachment behaviour, which is then analysed and scored by a trained clinical diagnostician. Based on these results it should be possible for an experienced and trained clinician to establish the diagnosis.

This is an initial study into the prevalence and comorbidity of RAD and DSED among children with a low average, borderline or mild intellectual disability, who have been referred for further child psychiatric assessment on account of severe psychological or behavioural problems. The results show that they fall within a high-risk group for the psychiatric disorders RAD and DSED (Chapter 3). The limited number of participants (55 children) and the fact that the observation and the interview were conducted for one primary caregiver are worth mentioning in this regard. Giving more attention to the attachment development and the assessment of these disorders in clinical practice, as

well as incorporating a triage for further diagnosis of disordered attachment behaviour in the standard assessment procedure will benefit the accuracy and completeness of the diagnostic landscape of the problems. Once the assessment is better implemented, the evidence base for treatment can also be further developed.

Children with RAD or DSED were also diagnosed with elevated numbers of comorbid psychiatric disorders (anxiety, mood and/or behavioural disorders) (Chapter 4), compounding the complexity of both diagnosis and treatment. This increases the risk of focusing on comorbid disorders in the diagnostic work-up, thereby missing the underlying RAD or DSED disorder and not including it in the treatment. The DSM-5 explicitly requires in order to the diagnosis of RAD that it must be possible to differentiate maladaptive behaviour from behaviours associated with the Autism Spectrum Disorder (ASD). To this end it is very important that the development history be thoroughly examined (background of severe social neglect, selective attachment behaviour) and the various behavioural characteristics that are typical of the maladaptive behaviour associated with ASD and/or RAD be accurately described. This study shows that it is quite possible to use focused research to distinguish between disordered attachment behaviour and autistic behaviour (Chapter 2). This finding has implications for the Practice Parameter (Zeanah et al,, 2016) and the substantiation of the RAD and DSED classifications in the Diagnostic Manual - Intellectual Disabilities - 2 (DM-ID-2) (Fletcher, Barnhill, & Cooper, 2016; Fletcher, Barnhill, McCarthy & Strydom, 2016). While extremes of insufficient care during the first years of the child's life are presumed to account for the diagnosis of RAD and DSED, the origin of ASD are explained from disordered neurobiological development. The possibility to distinguish between these attachment related disorders and ASD makes it possible to propose a suitable course of treatment, which increases the likelihood of treating the disorder(s) successfully.

For suitable, high-quality treatment and care, it is also important that clinicians understand that children with RAD or DSED may suffer from relatively more severe delays in adaptive ability (Chapter 4).

General conclusions

Children with intellectual disability are a high-risk group for developing severe mental or behavioural disorders (psychopathology), that is 3 to 4 times higher risk as compared to children without intellectual disability (Dekker & Koot, 2003). The vulnerability of this group indicates the need for special attention in Mental Health research and clinical practice. The last decade the Mental Health Care for people with intellectual disability has improved, both in research as well as in clinical practice, but this improvement is progressing very slow and there is still a large backlog compared with the Mental Health Care for the general population (Flynn, 2012). Attachment and disordered attachment were chosen as a topic on the assumption that children with intellectual disability are also a high-risk group for development of disordered attachment (Green & Goldwyn, 2002; Schuengel & Janssen, 2006). In this study this assumption held. Almost one out of the five school-aged children with intellectual disability, referred for assessment and treatment because of mental health problems, were diagnosed positive on an attachment related disorder RAD and/or DSED.

The aim of this study was to contribute to the description of the clinical diagnosis of the DSM-5 attachment related disorders RAD and DSED, and to the improvement of the diagnostic assessment of these disorders in the clinical mental health practice. We found that it is well possible to discriminate the clinical diagnosis of Reactive Attachment Disorder (RAD) from the clinical diagnosis of Autism Spectrum Disorder (ASD), which is one of the conditions in the (DSM-IV and) DSM-5 classification of RAD. With the guidelines of the Practice Parameter, recommended by the American Academy of Child and Adolescent Psychiatry (2005, 2016) we constructed a multi-informant diagnostic composite for testing school-aged children on attachment related disorders. We used questionnaires, interviews, structured observations and the information from the medical files of the children. Informants were the primary caregivers, teachers, the Medical Health Centre, and the child. Convergence between the elements of the composite was examined and resulted in the construction of a three phased stepped-care model for diagnosing attachment related disorders in the clinical practice of Child Mental Health Care. Using this model the results confirmed the need to invest in the assessment of attachment related disorders, for we found more comorbid psychopathology and developmental (functional) delay in children with classification of attachment related disorders than children without. These findings underscore the

need for developing and testing adequate treatment and support programs for children and their caregivers.

The second aim of this study was to disseminate the found knowledge into the field of research and clinical practice. The findings of this study have been transferred in the update of the Diagnostic Manual Intellectual Disability (DM-ID) to accompany the DSM-5. This Textbook of Diagnosis of Mental Disorders in Persons with Intellectual Disability, edited by the National Association for the Dually Diagnose (NADD) of the USA (Fletcher, Barnhill, & Cooper, 2016) is published as DM-ID2. In the Netherlands the results of the study and the diagnostic model have been described as a chapter in the first Manual Psychiatry and Mild Intellectual Disability (edited by Didden, Troost, Moonen, & Groen, 2016). The findings from this study are incorporated into a multi-days training course for clinical and research professional practitioners in using the Disturbances of Attachment Interview (DAI) as instrument in the diagnostic process for attachment related disorders. A similar training course in using the Clinical Observation of Attachment 6-12 (COA 6-12) is in preparation. The continued demand for this training shows the sustained interest in the unique behaviours within RAD and DSED, and therefore the relevance of continued research efforts in this area.

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Appendix

Protocol Clinical Observation of Attachment 6-12

(COA 6-12)

(English and Dutch version)

(English version)

Clinical Observation of Attachment 6-12 (COA 6-12)

The Clinical Observation of Attachment 6-12 (COA 6-12, Giltaij & Sterkenburg) is an adaptation of the COA, developed by Boris, Fueyo and Zeanah (1997).

The child (C) is unaware of being observed and videotaped. The parent/caregiver (P) is instructed to tell C that they have an appointment for visiting the center. P is informed that in a videotaped observation the interactions of C with P and with the diagnostician (S) will be examined. The day of the assessment P is instructed in detail by the research assistant about the procedure of Episode I of the COA 6-12. P and C are than guided into the observation room (OR) and the assistant tells C that it is allowed to play with the toys or games. The assistant closes the door, leaving P and C in the OR. After leaving the OR P will receive instruction from the assistant about Episode II and III.

C = Child P = Parent/caregiver S = Stranger (diagnostician)

Episode	time	time cum.	present	description
I	5	5	C P	* Entering OR * P demonstrates the toys and starts reading a magazine, leaving C with the toys; P is reactive on the initiatives of C.
	5	10	C P S	* S enters OR and sits down on distance of C; starts reading magazine
	2	12	C P S	* S starts talking to P.
	3	15	C P S	* S initiate participation in play C.
	3	18	C S	* P leaves OR (first separation); S continues play with C.
II	2	20	C P S	* P enters OR (first reunion) and sits down close to C; P reacts on initiatives of C.
	3	23	C P	* S leaves OR; P stays close to C and reacts on initiatives of C.
	2	25	C	* P leaves OR (second separation).
	2	27	C S	* S enters OR and sits down close to C; S reacts on initiatives of C.
	3	30	C	* S leaves OR

Episode	time	time cum.	present	description
III	2	32	C P	* P enters OR (second reunion) and sits down near to C; P reacts on initiatives of C.
	3	35	C P S	* S enters OR and sits down close to C with same distance from C as P; S reacts on initiatives of C.
	1	36	C P S	* Short (20-30 seconds) loud noise (alarm); P and S do not react on the noise, but react on C.
	10	46	C P S	* S gives C the choice out of three activities: a. Reading aloud by S; b. Playing a party game with S (like Ladders and snakes, playing cards, memory); c. Puzzle game with S. * S initiates physical close contact with C during play. After 5 minutes P takes over from S (who withdraws) and then P initiates physical close contact with C during play.
	2	48	C P S	* S asks P to clean up the toys with C.; S leaves OR.
	2	50	C P	* P and C leave OR.

Boris, N., Fueyo, M., & Zeanah, C.H. (1997). The clinical assessment of attachment in children under five. *Journal American Academy of Child and Adolescent Psychiatry, 36.* pp: 291–293

(Dutch version)

Clinical Observation of Attachment 6-12 (COA 6-12)

The Clinical Observation of Attachment 6-12 (COA 6-12, Giltaij & Sterkenburg) is een aanpassing van de COA, ontwikkeld door Boris, Fueyo and Zeanah (1997).

Het kind (K) is onwetend dat het geobserveerd en gefilmd wordt. De ouder/verzorger (O) is geïnstrueerd K te vertellen dat zij voor een afspraak de instelling zullen gaan bezoeken. O heeft informatie ontvangen dat de observatiesessie op video zal worden opgenomen en dat het interactiegedrag van K naar O en naar de diagnosticus zal worden onderzocht. Op de onderzoeksdag krijgt O van de onderzoeksassistent gedetailleerde instructie over de procedure van Episode I van de COA 6-12. O en K worden vervolgens naar de observatieruimte (OR) geleid en de onderzoeksassistent vertelt K dat deze mag spelen met het aanwezige speelgoed en spelmateriaal. De assistent sluit vervolgens de deur en laat O en K achter in de OR. Op de momenten dat O volgens de procedure de OR verlaat ontvangt O instructies over Episode II en III.

K = Kind O = Ouder/verzorger V = Vreemde (onderzoeker)

Episode	tijd	tijd cum.	betrok-kenen	beschrijving
I	5	5	K + O	* Binnenkomst Observatieruimte * O laat speelgoed zien en gaat vervolgens een tijdschrift lezen; reageert op en gaat in op initiatieven van K
	5	10	K + O + V	* V komt binnen en gaat zitten, tijdschrift lezen
	2	12	K + O + V	* V praat met O
	3	15	K + O + V	* V probeert in te voegen in spel K
	3	18	K + V	* O verlaat ruimte (1ste scheiding); V zet spel met K voort
II	2	20	K + O + V	* O komt binnen (1ste hereniging) en neemt plaats in nabijheid van K en gaat in op initiatieven van K

Episode	tijd	tijd cum.	betrok-kenen	beschrijving
	3	23	K + O	* V verlaat ruimte; O blijft in nabijheid van K, zet activiteit voort
	2	25	K	* O verlaat ruimte (2de scheiding)
	2	27	K + V	*V komt binnen en neemt plaats in nabijheid van K en gaat in op initiatieven van K
	3	30	K	* V verlaat ruimte
III	2	32	K + O	* O komt binnen (2de hereniging) en neemt plaats in nabijheid van K en gaat in op initiatieven van K
	3	35	K + O + V	* V komt binnen en neemt plaats in nabijheid van K op gelijke afstand als O; en gaat in op initiatieven van K
	1	36	K + O + V	* Kortdurend (20-30 seconden) hard geluid van brandalarm; O en V reageren niet uit zichzelf, maar op actie van K
	10	46	K + O + V	V stelt kind voor keuze uit drie activiteiten: a. Voorgelezen worden uit een voorleesboek (eerst door V, daarna overgenomen door O); b. gezelschapsspel (bijvoorbeeld ladders en slangen, memory, kwartet), waarbij K eerst speelt tegen V, daarna tegen O c. Puzzelen, waarbij eerst geassisteerd wordt door V, daarna door O * V initieert tijdens de activiteit lichamelijk contact bij K (op schoot nemen, arm om heen slaan, tegen aan leunen); O initieert tijdens diens activiteit een zelfde lichamelijk contact bij K
	2	48	K + O + V	* V neemt afscheid en geeft O en K opdracht om samen spelmateriaal op te ruimen
	2	50	K + O	* K en O verlaten ruimte

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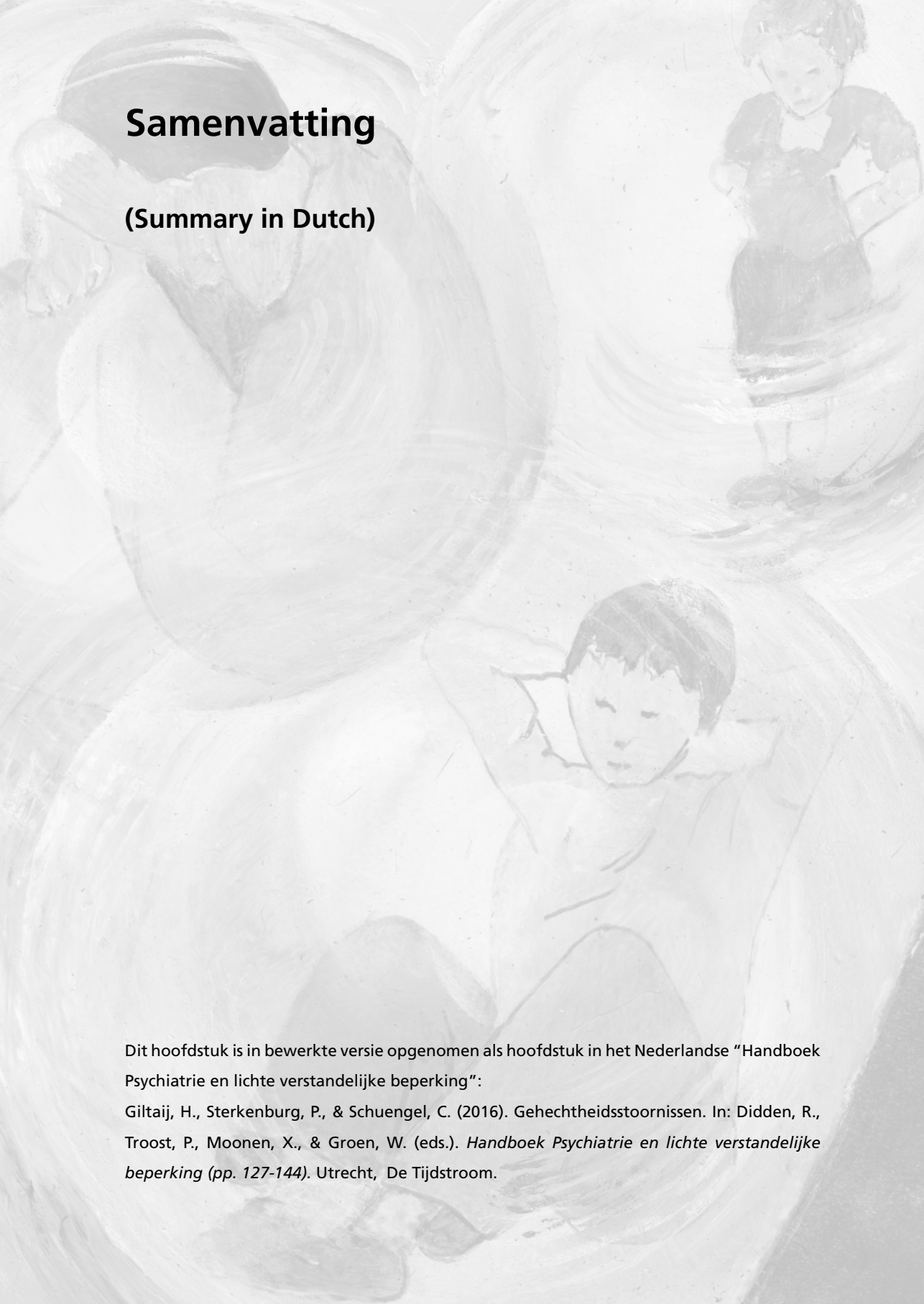
Samenvatting

(Summary in Dutch)



Dit hoofdstuk is in bewerkte versie opgenomen als hoofdstuk in het Nederlandse "Handboek Psychiatrie en lichte verstandelijke beperking":
Giltaij, H., Sterkenburg, P., & Schuengel, C. (2016). Gehechtheidsstoornissen. In: Didden, R., Troost, P., Moonen, X., & Groen, W. (eds.). *Handboek Psychiatrie en lichte verstandelijke beperking (pp. 127-144).* Utrecht, De Tijdstroom.

Inleiding

De hoofddoelstelling van deze dissertatie was het leveren van een bijdrage aan de verheldering van het concept van gehechtheid gerelateerde stoornissen en verbetering van de diagnostische praktijk in de geestelijke gezondheidszorg voor kinderen, die gehechtheid gerelateerde gedragsproblemen vertonen, zoals deze gedefinieerd zijn in de DSM-5 (Reactieve Hechtingsstoornis, RHS, en Ontremd-sociaalcontactstoornis, OSCS). Deze studie is uitgevoerd met kinderen, in de leeftijd van 5 tot 11 jaar op moment van eerste participatie in het onderzoek, functionerend op zwakbegaafd of licht verstandelijk beperkt intelligentieniveau, die vanwege emotionele of gedragsproblemen verwezen zijn voor een jeugdpsychiatrische beoordeling. De tweede doelstelling van dit studie project was het bijdragen aan de verspreiding van verworven kennis en daarmee het verhogen van de kennis en klinische expertise van professionals aangaande gehechtheid en gehechtheid gerelateerde stoornissen in hun dagelijkse praktijk, in het bijzonder betreffende verzorgers en professionals betrokken bij kinderen met een verstandelijke beperking. In dit hoofdstuk vat ik samen en reflecteer ik op de belangrijkste bevindingen en conclusies van dit studieproject. Deze bevindingen zullen ook worden bediscussieerd met betrekking tot de beperkingen van de studie en de implicaties voor voortgaand wetenschappelijk onderzoek en klinische praktijk.

Dit hoofdstuk begint met een beschrijving van huidige visies over het concept van gehechtheid gerelateerde stoornissen. Enerzijds verschijnen steeds meer publicaties, die een verdere differentiatie en specificering van varianten van verstoorde gehechtheid beschrijven (Zeanah & Lieberman, 2016), en richtlijnen voor het uitvoeren van diagnostiek en behandeling naar aanleiding van onderzoeksresultaten uit de laatste 10 jaar worden aangevuld (Zeanah, Chesher, Boris, & the American Academy of Child and Adolescent Psychiatry [AACAP] Committee on Quality Issues [CQI], 2016). Anderzijds wordt het hele concept van verstoorde gehechtheid ter discussie gesteld en gepleit voor verwijdering van de stoornis, diagnostiek en behandeling van verstoorde gehechtheid uit de klinische praktijk (Allen, 2016).

De ontwikkeling aangaande het concept van verstoorde gehechtheid heeft geleid tot een revisie van het concept, welke is opgenomen in de wijzigingen van de DSM-IV naar de DSM-5 (American Psychiatric Association [APA], 2013) (zie Hoofdstuk 1) en waarin de twee subtypen van de Reactieve Hechtingsstoornis (RHS) zijn gedefinieerd als twee

afzonderlijke stoornissen, RHS en OSCS. Beide stoornissen hebben gemeen dat zij een etiologie hebben met ernstige sociale en emotionele verwaarlozing of deprivatie in de vroege jaren. De stoornis RHS wordt expliciet beschreven als een stoornis in de gehechtheidsontwikkeling met een kenmerkend patroon van verstoord gehechtheidsgedrag. Het betreft hier een kind-specifieke stoornis. Bij de stoornis OSCS is minder duidelijk of dit begrepen kan worden als een stoornis in de gehechtheidsontwikkeling, aangezien het kenmerkende gedragspatroon van cultureel ongepast, te familiaire omgang met relatieve vreemden niet alleen kan worden waargenomen bij kinderen met een vermoedelijk ontbreken van een selectieve gehechtheidsrelatie, maar ook bij kinderen die wel lijken een gehechtheidsrelatie met een primaire opvoeder te hebben ontwikkeld. Het is daarom volop in discussie in hoeverre OSCS beschouwd kan worden als een stoornis in de gehechtheid (Zeanah et al., 2016; Zeanah & Gleason, 2010, 2015). Om deze reden hebben wij gekozen voor het gebruik van de term "gehechtheid gerelateerde stoornissen" als we het hebben over de combinatie van RHS en OSCS.

Belangrijk is ook de discussie in hoeverre naast de hier genoemde stoornissen RHS en OSCS ook bepaalde psychopathologische gedragspatronen van het jonge kind in een specifieke relatie met een volwassen verzorger gedefinieerd kunnen worden als een diagnosticeerbare verstoorde relatie. Zeanah en Lieberman (2016) doen hiervoor een voorstel tot het opnemen van een Relatie-Specifieke Stoornis van de Vroege Kindertijd (Relationship Specific Disorder of Early Childhood) in de Diagnostic Classification of Mental Health and Developmental Disorders of Infancy and Early Childhood DC: 0-5 (Zero to Three, 2016). Zij beschrijven hierin kinderen die in de gehechtheidsontwikkeling met een specifieke primaire verzorger ernstig negatieve ervaringen hebben opgedaan, die binnen deze relatie tussen kind en opvoeder (en niet in de relatie met andere primaire verzorgers) geleid hebben tot een persistent patroon van emotionele of gedragsstoornissen, zoals sterk oppositioneel gedrag, hyperwaakzaamheid, zichzelf in gevaar brengend gedrag, weigering tot eten of slapen, of overmatig controlerend gedrag.

Het verbreden van de focus van verstoorde gehechtheid als kind-specifieke stoornis naar een spectrum van kind-specifiek naar relatie-specifieke stoornissen is mede gebaseerd op de veronderstelde ernstige gevolgen van verstoorde gehechtheidsontwikkeling in de vroege levensjaren voor het uiteindelijk functioneren en ontwikkeling van het kind. Ondanks het ontbreken van onderzoek naar de verbanden

tussen relatie-gebonden verstoorde gehechtheid en ontwikkelingsuitkomsten, hebben onderzoek naar individuele verschillen in de kwaliteit van gehechtheidsrelaties aangetoond dat die kwaliteit van relaties voorspellende waarde heeft over een breed spectrum van ontwikkelingsuitkomsten, zij het met bescheiden effectmaten (Groh, Fearon, van IJzendoorn, Bakermans-Kranenburg & Roisman, 2017). De klinische relevantie van problematische gehechtheidsrelaties enerzijds, en het tot nu toe ontbreken van diagnostische instrumenten anderzijds, hebben bijgedragen aan verwarring over het begrip gehechtheid in de klinische praktijk en grote variatie in de toepassing ervan. Zeanah en Lieberman's (2016) voorstel is een poging deze verwarring op te lossen en te voorzien in een samenhangend klinisch conceptueel kader voor gehechtheid en geestelijke gezondheid.

Allen (2016) verwijst naar het ontbreken van consensus binnen de klinische praktijk over het gebruik en de betekenis van de terminologie verstoorde gehechtheid en gehechtheidstherapie, waardoor vaak sprake is van verwarring over het gebruik en de betekenis van deze termen. Dientengevolge vindt in de klinische praktijk zowel over- als onder-diagnostisering van verstoorde gehechtheid (RHS en OSCS) plaats (Woolgar & Baldock, 2015). Allen (2016) stelt dat in de huidige praktijk de gehechtheidsstoornis als concept discutabel is, vaak verkeerd wordt begrepen en gebruikt, uitzonderlijk zeldzaam is, en dat klinische interventie, anders dan het kind plaatsen bij een ondersteunende verzorger of opvoeder, onnodig lijkt. Allen (2016) bestrijdt niet dat er kinderen zijn die vanwege een achtergrond van (in hun jonge jaren) ernstige verwaarlozing of mishandeling patronen van ernstig verstoord gedrag kunnen gaan vertonen in de relatie met anderen en in de zelfregulatie. Niettemin, een meerderheid van kinderen met vergelijkbare achtergrond vertoont deze atypische gedragingen niet. Allen stelt de vraag of deze afwijkende gedragingen ook niet beschreven zouden kunnen worden als een (combinatie van) andere vormen van psychopathologie, zoals externaliserende gedragsstoornis, en wijst op het gevaar dat de toeschrijving van het gedrag als gehechtheidsstoornis overschaduwen van andere stoornissen tot gevolg kan hebben. Allen wijst er terecht op dat de vaststelling van de uiteindelijke diagnose van groot belang is omdat deze de basis vormt voor de therapeutische behandeling of interventie. En een onjuiste, onvolledige of invalide diagnose verhoogt de kans op een onjuiste of onvoldoende behandeling.

Hoewel Allen's kritische weergave van de gehechtheidsstoornis als concept binnen de klinische praktijk interessant is en uitnodigt tot discussie, is zijn voorstel tot het geheel afschaffen van de diagnose RHS en/of OSCS toch wellicht te radicaal. Dit voorstel zou gevolgen hebben voor een groep kinderen, hoe klein die groep ook wellicht is, die wel waarneembare patronen van ernstig maladaptief sociale gedragingen vertonen, die niet ondergebracht kunnen worden binnen andere diagnostische categorieën. Bovendien worden deze gedragingen waargenomen onder kinderen die opgroeien in instituten of leefomstandigheden, waarin sprake is van ernstige verwaarlozing en ontbreken van stabiele hechtingsmogelijkheden, en daarmee benadrukken de giftigheid van deze omgevingen voor kinderen. Met het ontkennen van deze duidelijke patronen van verstoord en maladaptieve gedrag, kan een groep van kinderen de diagnose en daarmee ook de gepaste behandeling worden ontnomen.

De lopende verwarring en het niet eenduidig gebruiken van termen als verstoorde gehechtheid, gehechtheidsdiagnostiek en gehechtheidsbehandeling kan worden begrepen als het resultaat van de lopende onderzoeken en het debat door een klein aantal van onderzoeksgroepen. Het ontbreken van diagnostische meetinstrumenten, geaccepteerd als valide en betrouwbaar, en behandelmethodieken, die gebleken effectief zijn, bieden veel ruimte aan klinische professionals voor individuele invulling van de genoemde termen. Dit noodzaakt tot meer afstemming en integratie tussen de wetenschappelijk onderzoekspraktijk en de klinische praktijk op het gebied van kennisontwikkeling en –implementatie betreffende gehechtheid.

Een belangrijke stap was de ontwikkeling en aanvaarding van de Practice Parameter for the assessment and treatment of children with Reactive Attachment Disorder (volgens de DSM-IV), door de American Academy of Child and Adolescent Psychiatry (AACAP) in 2005, met richtlijnen voor zowel de klinische praktijk als wetenschappelijk onderzoek, gebaseerd op de dan heersende state-of-the-art. Hoewel de Practice Parameter veel onderwerpen van klinische overeenstemming publiceerde, bleven sommige vraagstukken liggen in afwachting van meer klinisch onderzoek. Deze vraagstukken betreffen zowel specifieke diagnostische instrumenten en interventies, als de toepassing van de modellen in verschillende doelgroepen. Recent hebben Fletcher, Flood en Hare (2016) onderzocht de klinische moeilijkheden verband houdend met gehechtheidsrelaties bij mensen met verstandelijke en ontwikkelingsbeperkingen, en hebben op grond daarvan een Richtlijn voor de praktijk en wetenschappelijk

onderzoek beschreven. In Nederland is een serie van single-case experimenten uitgevoerd met zes kinderen met een visueel- en-verstandelijke beperking en een achtergrond van ernstige verwaarlozing, en daarmee aantonend de toepasbaarheid van geprotocolleerde diagnostiek (Sterkenburg & Schuengel, 2014). De bemoedigende resultaten van deze kleine steekproef effectstudie hebben geleid tot de publicatie van de best-practice voor diagnosticeren van problematische gehechtheid bij kinderen met visuele-en-verstandelijke beperking (Dekker-van der Sande & Janssen, 2010). Deze best-practice vormde de basis voor de Nederlandse Richtlijn voor problematische gehechtheid in de Jeugdzorg en Kinderbescherming, aanvaard door de beroepsverenigingen van psychologen en pedagogen (De Wolff et al., 2014).

De centrale doelstelling van de in dit proefschrift opgenomen studies was het beschrijven en onderzoeken van een diagnostisch onderzoekstraject voor het vaststellen van de Reactieve Hechtingsstoornis, RHS, en de Ontremd-sociaalcontactstoornis, OSCS, zoals beschreven in de DSM-5, bij kinderen in de latentiefase, functionerend op zwakbegaafd of een licht verstandelijk beperkt (IQ 50-85) intelligentieniveau en verwezen voor kinderpsychiatrische consultatie. Tevens leverde de uitvoering van dit onderzoekstraject resultaten op, die een bijdrage leveren over de prevalentie van RHS en OSCS en de vaststelling van verstoord gehechtheidsgedrag bij deze specifieke kwetsbare doelgroep. Daarnaast werd de comorbiditeit met andere psychiatrische stoornissen en het niveau van functioneren besproken.

Het ontwerp van het diagnostisch traject was uitgegaan van de richtlijnen en aanbevelingen zoals deze in de American Academy of Child and Adolescent Psychiatry (AACAP) Practice Parameter voor de diagnostiek en behandeling van kinderen en adolescenten met reactieve hechtingsstoornis (2005, met revisie in 2016) zijn beschreven. Hierin worden de volgende elementen onderscheiden: informatie over de vroege ontwikkelings- en opvoedingsgeschiedenis van het kind, in het bijzonder van het gehechtheidsgedrag van het kind met de primaire opvoeders, en directe observatie van het interactiegedrag van het kind met de primaire opvoeder/verzorger en een onbekende vreemde in een relatief gestructureerde procedure met stress verhogende momenten. Verbanden met andere vormen van psychopathologie en het ontwikkelingsniveau van functioneren van het kind zijn bestudeerd door afname van specifieke diagnostische vragenlijsten.

In dit hoofdstuk zullen de bevindingen van deze studies die onderdeel uitmaken van deze dissertatie, zoals gepresenteerd in Hoofdstuk 2, 3, en 4, worden samengevat en bediscussieerd. Beperkingen van deze studie evenals implicaties voor toekomstig onderzoek worden toegevoegd. Tenslotte zullen de klinische implicaties en de beschrijving van het klinisch onderzoekstraject voor vaststelling van RHS en OSCS worden beschreven.

Samenvatting en Conclusies

Gehechtheid gerelateerde stoornissen en ASS

Alhoewel de DSM-5 de stoornissen RHS en OSCS niet langer definieert als 'gestoorde sociale bindingen', maar als 'een gedragspatroon van geïnhibeerd (RHS) of gedisinhibeerd (OSCS) gedrag naar volwassenen toe', met daarbij verbonden gedragskenmerken, is bij de DSM-5 stoornis RHS nog steeds opgenomen dat deze stoornis wordt uitgesloten indien tevens sprake is van de diagnose autisme-spectrum-stoornis (ASS). Onderzoek naar de onderscheidende kenmerken tussen beide stoornissen is daarom van belang om vast te stellen of dit criterium moet worden behouden in toekomstige edities van classificatie en diagnostische systemen. Hoofdstuk 2 doet verslag van het onderzoek naar de overlap en verschillen tussen de gedragskenmerken van ASS versus RHS (Inhibited) en OSCS (Disinhibited). ASS-gedragskenmerken werden gemeten met het screeningsinstrument AUTI-R (Van Berckelaer-Onnes & Hoekman, 1991), gedragskenmerken van RHS en OSCS werden gemeten met het screenings-instrument Disturbances of Attachment Interview (DAI, Smyke & Zeanah, 1999). De participanten waren kinderen (leeftijd 5-11 jaar) met een verstandelijke beperking (IQ 50-85), die vanwege psychische, psychiatrische of gedragsproblemen voor psychiatrisch onderzoek waren verwezen naar een gespecialiseerd centrum voor Kinder- en Jeugdpsychiatrie. Op grond van de AUTI-R scores bleek bij 27% van de kinderen sprake van mogelijke of duidelijke ASS, met een duidelijke meerderheid van jongens boven meisjes. Er werd geen verband gevonden tussen de ASS classificatie en het niveau van intellectueel functioneren, etnische achtergrond of leeftijd. Analyses op het niveau van classificatie en clusters van gedragskenmerken lieten geen significante verbanden zien tussen ASS (gemeten met de AUTI-R) en de stoornissen RHS en/of OSCS (gemeten met de DAI). Daarmee kan de conclusie worden getrokken dat beide instrumenten, zoals waar zij voor bedoeld zijn, een ander fenomeen van gedrag meten en daarmee goed

bruikbaar zijn voor gestructureerd beoordeling en differentiëren tussen de ASS, RHS en OSCS. Deze bevindingen laten ook zien dat het weglaten van het exclusie criterium van ASS niet noodzakelijk zal leiden tot over-identificatie van RHS en OSCS.

Gehechtheid gerelateerde stoornissen en multi-instrumentele assessment

Het Disturbances of Attachment Interview (DAI) is een semigestructureerd interview voor vaststelling van gedragssignalen van verstoorde gehechtheid zoals waargenomen door volwassenen die regelmatig omgaan met het kind. Indien de onderzoeker goed getraind en ervaren is, is dit een instrument dat in relatief korte tijd (20-40 minuten) afgenomen en gescoord kan worden. Interviewers zijn getraind om goed door te vragen om duidelijk te krijgen of de informant een voldoende adequaat beeld van het gehechtheidsgedrag heeft. Echter, het is belangrijk waakzaam te blijven met het gegeven dat de bij de geïnterviewde ouder of hulpverlener uitgevraagde informatie beïnvloed wordt door diens interpretatie van het gedrag van het kind als abnormaal en zorgelijk. Die gradatie is deels een functie van vooronderstellingen en selectieve aandacht van de kant van de informant, maar ook van de natuurlijke variatie in gelegenheden waarop het verstoorde gedrag zich kan voordoen.

Het is dan ook van groot belang dat positieve bevindingen op de DAI gevolgd worden door live observaties van de interacties van de kinderen met de ouders of primaire verzorgers in gestandaardiseerde situaties waarin het kind wordt blootgesteld aan een stressfactor (Zeanah et al., 2016). Tevens dient het gehechtheidsgedrag van het kind ten opzichte van de ouder of primaire verzorger vergeleken te worden met het gehechtheidsgedrag ten opzichte van een onbekende volwassene in vergelijkbare situaties. In deze studie hebben wij, zoals aanbevolen in de Practice Parameter, daarvoor het observatieprotocol van de Clinical Observation of Attachment (COA) gebruikt, waarbij de stressoren in het protocol aangepast zijn voor de leeftijdsgroep van de onderzoekspopulatie. Het waargenomen gedrag van de kinderen werd vervolgens gescoord op de acht schalen van (verstoord) gehechtheidsgedrag van de Lijst Gedragssignalen van Verstoorde Gehechtheid (List of Behavioural Signs of Disturbed Attachment in Young Children, BSDA, Zeanah et al., 1993), resulterend in een totaalscore tussen de 8 en 40. Omdat verondersteld wordt dat beide stoornissen RHS en OSCS een reactie zijn op een ontwikkelingsachtergrond waarin het kind is blootgesteld aan een patroon van ernstige verwaarlozing en ontoereikende zorg om een stabiele

gehechtheidsrelatie te kunnen vormen, is uitvragen en bestudering van de levensgeschiedenis, met name van de eerste jaren, essentieel.

Voor het stellen van de DSM-5 diagnose RHS en OSCS is bij 55 van de kinderen die participeerden in de studie van Hoofdstuk 2 vervolgonderzoek gedaan, en werden bovengenoemde instrumenten (DAI, COA, BSDA) en de ontwikkelingsgeschiedenis van de kinderen afgenomen. In Hoofdstuk 3 worden de resultaten van deze instrumenten beschreven. Verbanden werden onderzocht tussen het door de ouder/primaire verzorger beschreven (verstoorde) gehechtheidsgedrag van het kind (DAI), het geobserveerde (verstoorde) gehechtheidsgedrag van het kind (COA en BSDA) en de beschreven achtergrond van ernstige ontoereikende zorg voor het kind. Door ervaren klinische onderzoekers is, mede op grond van het geobserveerde gedrag en de levensgeschiedenis van het kind, de klinische diagnose voor RHS en/of OSCS vastgesteld.

De scores op zowel beschreven als geobserveerd gehechtheidsgedrag, evenals het voorkomen van extreme ontoereikende zorg bleken niet verbonden aan het geslacht, de etnische achtergrond, de leeftijd of het niveau van cognitief functioneren van het kind. Er is geen verband gevonden tussen kenmerken en voorkomen van RHS en/of OSCS en deze kindkenmerken. In deze groep van 55 kinderen werd één op de drie kinderen door de ouder/primaire opvoeder beschreven met gedragskenmerken die zouden wijzen op RHS en/of OSCS. Bij één op de vijf kinderen (22%) kon op grond van de beschreven ontwikkelingsgeschiedenis van de eerste levensjaren beoordeeld worden als een achtergrond van ernstige ontoereikende zorg, waardoor het kind ernstige belemmeringen zal hebben ervaren om een stabiele gehechtheidsrelatie aan te gaan met de ouder/primaire opvoeder. De klinische diagnose van RHS en/of OSCS werd bij 10 van de 55 (18%) kinderen vastgesteld.

Voor het scoren van het geobserveerde (verstoorde) gehechtheidsgedrag door onafhankelijke en getrainde codeurs werd gebruik gemaakt van de BSDA. De scores van de 55 kinderen lieten een variantie zien van 9 tot 33. De scores op deze lijst door kinderen, die door de ouder/primaire verzorger beschreven waren (op de DAI) als RHS en OSCS, hadden een BSDA score van 26 tot 33 met een gemiddelde van 29.3 (SD = 2.7), en verschilden daarmee significant van de kinderen die door de ouders beschreven waren als niet RHS en/of OSCS met scores variërend van 9 tot 29, met een gemiddelde van 16.2 (SD = 5.2). Ook voor de afzonderlijke stoornis RHS met een score

van 17 tot 21 (*M* = 18.7, SD = 2.1) en OSCS met een BSDA score van 10 tot 28 (*M* = 17.4, SD = 6.0) geldt dat sprake is van een significant verschil in scoregemiddelde en -bereik tussen de kinderen die wel danwel niet door de ouder/primaire verzorger als RHS en/of OSCS worden beschreven. Evenzo was significant meer bewijs van ernstig ontoereikende zorg, vastgesteld vanuit de beschreven ontwikkelingsanamnese, bij kinderen die door ouder/primaire verzorger als RHS en/of OSCS waren beschreven, dan bij de kinderen die niet als zodanig beschreven waren.

De kinderen bij wie de ontwikkelingsgeschiedenis een achtergrond van ernstig ontoereikende zorg liet zien, scoorden significant hoger op het geobserveerde verstoorde gehechtheidsgedrag (met een gemiddelde score op de BSDA van 25), dan de kinderen bij wie geen ernstig ontoereikende zorg kon worden vastgesteld (gemiddelde score op de BSDA van 16). Hiermee wordt bevestigd dat 1) er een relatie is tussen pathogene zorg en het gedurende de COA geobserveerde en met de BSDA gescoorde verstoorde gehechtheidsgedrag, 2) de BSDA bijdraagt tot het adequaat beoordelen van verstoord gehechtheidsgedrag en daarmee 3) bevestigt het belang van het includeren van de BSDA in het diagnostisch protocol.

Gehechtheid gerelateerde stoornissen en comorbiditeit

Kinderen met een verstandelijke beperking hebben een drie tot vier keer hoger risico op het ontwikkelen van psychopathologie als kinderen zonder deze beperking (Dekker, Douma, de Ruiter, & Koot, 2006; Einfeld, Ellis, & Emerson, 2011). Diverse studies hebben aangetoond dat sprake is van een verband tussen problematische gehechtheid en kwetsbaarheid voor ontwikkeling van psychopathologie (Mikulincer & Shaver, 2012). Kinderen die opgegroeid zijn in kindertehuizen of instellingen hebben een hoog risico op een ontwikkelingsachterstand en stoornissen, inclusief psychiatrische stoornissen (MacLean, 2003). Voor Hoofdstuk 4 zijn de kinderen (*n* = 55) die voor Hoofdstuk 3 gediagnostiseerd waren met RHS en/of OSCS vergeleken met de kinderen die niet deze diagnose hebben gekregen. De Vineland Adaptive Behaviour Scales (Sparrow, Carter, & Cicchetti, 1993) liet zien dat kinderen met RHS en/of OSCS een significant grotere ontwikkelingsachterstand in hun adaptief functioneren hadden. Psychiatrische comorbiditeit werd onderzocht door afname met de National Institute of Mental Health Diagnostic Interview Schedule for Children, Version IV (NIMH-DISC-IV; Shaffer, Fisher, Lucas, Dulcan, & Schwab-Stone, 2000) bij de ouder/primaire opvoeder. Bij ruim 81% van de kinderen werd een psychiatrische stoornis (DSM-IV angst-, stemmings-,

gedrags- of pervasieve ontwikkelingsstoornis) vastgesteld. Bij 46% van de kinderen werden twee of meer psychiatrische stoornissen vastgesteld. Kinderen bij wie sprake was van RHS en/of OSCS hadden, naast deze gehechtheid gerelateerde stoornis, gemiddeld 2.3 comorbide stoornissen, terwijl de groep kinderen zonder vastgestelde RHS en/of OSCS positief scoorden op gemiddeld 1.5 psychiatrische stoornissen; dit verschil was niet statistisch significant. ADHD was veruit de meest voorkomende stoornis in deze onderzoeksgroep.

Leerkrachten vulden de Developmental Behaviour Checklist (DBC; Einfeld & Tonge, 1991, 1994, 1995; Einfeld, Tonge, & Parmenter, 1999) in, en lieten zien dat normoverschrijdend en antisociaal gedrag, waaronder ADHD, in deze onderzoeksgroep significant vaker gezien werd bij kinderen die op een lager cognitief niveau functioneerden. Daarentegen gaven de reacties van de ouders op deze checklist aan, dat een relatie bestond tussen het voorkomen van deze gedragsproblematiek en een vastgestelde stoornis RHS en/of OSCS. De reacties van de ouders gaven ook aan een positief verband te zien tussen in-zichzelf gekeerd gedrag en de diagnose van RHS of OSCS. Kinderen uit deze onderzoeksgroep met een stoornis RHS en/of OSCS hadden significant meer emotionele en gedragsproblemen dan kinderen uit deze onderzoeksgroep zonder deze stoornis. Deze resultaten tonen aan, dat RHS en OSCS vaker voorkomen bij kinderen die ook andere vormen van psychopathologie vertonen. Niettemin, niet alle kinderen met RHS en/of OSCS hebben andere comorbide stoornissen.

Beperkingen van de studie en implicaties voor toekomstig onderzoek

De prevalentie van verstoorde gehechtheid bij kinderen in de algemene bevolking wordt als zeer laag ingeschat (Richters & Volkmar, 1994), hoewel anderen een prevalentie van meer dan 1% hebben gevonden en dit aanzienlijk noemen, vergelijkbaar met de prevalentie van bijvoorbeeld autistische stoornissen. Bij zogenaamde hogerisicogroepen lijkt deze prevalentie van verstoord gehechtheidsgedrag aanzienlijk te zijn in klinische onderzoeksgroepen, hoewel er nog nauwelijks studies zijn uitgevoerd naar de prevalentie van de stoornissen RHS en OSCS, vastgesteld volgens de richtlijnen van de AACAP. Eerdere studies naar gedragskenmerken van verstoorde gehechtheid beschrijven dat bij 40% van jonge geïnstitutionaliseerde kinderen (Smyke, Dumitrescu,

& Zeanah, 2002; Zeanah, Smyke, & Dumitrescu, 2002) en bij 18% (Oosterman & Schuengel, 2008) tot 38% (Zeanah et al., 2004) van kinderen in therapeutische pleegzorg dergelijke gedragskenmerken waargenomen zijn. Er is slechts één grote studie gepubliceerd gericht op de prevalentie van de diagnose RHS en OSCS in een specifieke hoog-risicogroep (jonge kinderen die belangrijk deel van hun leven zijn opgegroeid in verwaarloosde toestand in Roemeense kindertehuizen en deels later in pleegzorg zijn geplaatst) (Gleason et al., 2011). Bij 4% van deze kinderen werd de diagnose RHS en bij 17-18% de diagnose OSCS vastgesteld. Minnis et al. (2013) vonden in hun uitgebreide multi-informant studie een prevalentie van 1,4% gehechtheid gerelateerde stoornissen bij kinderen (leeftijd 6-8 jaar) in een achtergesteld stedelijk gebied. Vervoort, de Schipper, Bosmans en Verschueren (2013) onderzochten de psychometrische eigenschappen van screening vragenlijsten voor symptomen van RHS en OSCS onder schoolkinderen met ernstige emotionele of gedragsstoornissen, en overeenkomst van deze screening instrumenten met geobserveerd gedrag in een gestructureerde observatie situatie voor gehechtheid gerelateerde stoornissen. Scheper en collega's (2016) rapporteren RHS gedrag (9% en 27%) en OSCS gedrag (42% en 51%) in twee subgroepen van jonge kinderen, die verwezen zijn voor behandeling van emotionele en/of gedragsproblemen: de subgroep van thuis opgegroeide kinderen en de subgroep van uithuisgeplaatste kinderen in therapeutische pleegzorg.

Tabel 1 Overzicht van multi-informant/-instrumenten studies van kinderen met verstoord gehechtheidsgedrag of gehechtheid gerelateerde stoornissen.

Studie	*Onderzoeksgroep*	*Focus*	*Steekproef*	*Gebruikte instrumenten*	*Belangrijkste bevindingen*
Smyke, Dumitrescu, & Zeanah (2002)	Ernstig verwaarloosde, geïnstitutio-naliseerde kinderen (BEIP project) (leeftijd 1-7 jaar)	Prevalentie van signalen van RHS of OSCS	*N*=94 kinderen: Standard Unit (*n*=32), Pilot Unit (*n*=29), Nooit-geïnstitutio-naliseerde kinderen (n=33)	*Zorg geschiedenis*: **Geen meting** *Interview:* Disturbances of Attachment Interview (DAI) *Observatie:* **Geen meting** *Psychopathologie:* **Geen meting**	Standard Unit: RHS signalen: 37% OSCS signalen: 43% Pilot Unit: RHS signalen: 14% OSCS signalen:17 %
Zeanah, Scheeringa, Boris, Heller, Smyke, & Trapani (2004)	Mishandelde kinderen (leeftijd 10-48 maand) in pleegzorg	Prevalentie van signalen van RHS of OSCS	*N*=94 kinderen	*Zorg geschiedenis:* verwaarlozing juridisch vastgesteld *Interview:* Disturbances of Attachment Interview (DAI) *Observatie:* **Geen meting** *Psychopathologie:* **Geen meting**	RHS signalen: 45% OSCS signalen: 37%

Studie	*Onderzoeksgroep*	*Focus*	*Steekproef*	*Gebruikte instrumenten*	*Belangrijkste bevindingen*
Zeanah, Smyke, Koga, Carlson, and the BEIP Core Group (2005)	Ernstig verwaarloosde, geïnstitutionaliseerde kinderen (BEIP project) (leeftijd 12-31 maand)	Verschillen tussen signalen van RHS en OSCS in geïnstitutionaliseerde en nooit-geïnstitutionaliseerde kinderen	*N*=145 kinderen: geïnstitutionaliseerde kinderen (*n* = 95) en nooit-geïnstitutionaliseerde kinderen (*n* = 50)	*Zorg geschiedenis:* Observational Record of the Caregiving Environment (ORCE) *Interview:* Disturbances of Attachment Interview (DAI) *Observatie:* Strange Situation Procedure (SSP) *Psychopathologie:* Infant Toddler Social Emotional Assessment (ITSEA)	Significante verschillen tussen geïnstitutionaliseerde en nooit-geïnstitutionaliseerde kinderen voor RHS ($p<.001$) en voor OSCS ($p<.01$)
Oosterman & Schuengel (2008)	Kinderen (leeftijd 2-7 jaar) in pleegzorg	Prevalentie van signalen van RHS of OSCS	*N*=69 kinderen	*Zorg geschiedenis:* **Geen meting** *Interview:* Disturbances of Attachment Interview (DAI) *Observatie:* Attachment Q-set (AQS) tijdens video vastgelegde thuisobservatie *Psychopathologie:* Child Behaviour Checklist (CBCL)	RHS/OSCS signalen: 18%

Studie	*Onderzoeksgroep*	*Focus*	*Steekproef*	*Gebruikte instrumenten*	*Belangrijkste bevindingen*
Gleason, Fox, Drury, Smyke, Egger, Nelson & Zeanah (2011)	Voorheen geïnstitutio-naliseerde kinderen (BEIP project) (leeftijd 22-54 maand)	Prevalentie en stabiliteit over tijd van RHS en OSCS	*N*=122-135 kinderen	*Zorg geschiedenis:* Observational Record of the Caregiving Environment (ORCE) *Interview:* Disturbances of Attachment Interview (DAI) *Observatie:* Strange Situation Procedure (SSP) *Psychopathologie:* Preschool Age Psychiatric Assessment (PAPA), Infant Toddler Social Emotional Assessment (ITSEA)	RHS: van 4,6% tot 4,1% OSCS: van 31,8% tot 17,6%
Minnis, Macmillan, Pritchett, Young, Wallace, Butcher, Sim, Baynham, Davidson & Gillberg (2013)	Kinderen (leeftijd 6-8 jaar) in een achtergesteld stedelijk gebied (UK)	Prevalentie van RHS en/of OSCS	Hoofdgroep: *N*=1646 kinderen (SDQ, RPQ, SIMD), waaruit 49 waarschijnlijke RHS gevallen (DAWBA, CAPA-RAD, Observatie)	*Zorg geschiedenis:* Scottish Index of Multiple Deprivation (SIMD) *Interview:* Child and Adolescent Psychiatric Assessment, Reactive Attachment Disorder module (CAPA-RAD), Relationship Problems Questionnaire (RPQ) *Observatie:* Observational Checklist for Reactive Attachment Disorder *Psychopathologie:* Strengths and Difficulties Questionnaire (SDQ), Development and Wellbeing Assessment (DAWBA),	RHS en/of OSCS: 1,4% (2,3% aangrenzend/ verdacht RHS/ OSCS)

Studie	*Onderzoeksgroep*	*Focus*	*Steekproef*	*Gebruikte instrumenten*	*Belangrijkste bevindingen*
Kay & Green (2013)	Mishandelde thuiswonende adolescenten in Jeugdzorg begeleiding (leeftijd 10-16 jaar) zonder achtergrond van institutionalisering	Karakterisering van RHS/OSCS gedragingen	*N*=153 adolescenten, met als controlegroep een laag-risico groep (*n*=42).	*Zorg geschiedenis:* Maltreatment Classification System (MCS) *Interview:* Child and Adolescent Psychiatric Assessment, Reactive Attachment Disorder module (CAPA-RAD) *Observatie:* **Geen meting** *Psychopathologie:* Child Behaviour Checklist (CBCL)	RHS/OSCS signalen: 63% OSCS signalen: 56%
Vervoort, de Schipper, Bosmans, & Verschueren (2013)	Kinderen (leeftijd 6-10 jaar) van scholen voor Speciaal Onderwijs	Toetsing van psychometrische eigenschappen van de Relationship Problems Questionnaire (RPQ)	*N*=152 kinderen	*Zorg geschiedenis:* Beoordeeld door school psycholoog *Interview:* Disturbances of Attachment Interview (DAI), Relationship Problems Questionnaire (RPQ) *Observatie:* Observational Checklist for Reactive Attachment Disorder *Psychopathologie:* Strengths and Difficulties Questionnaire (SDQ)	De RHS en OSCS subschalen van RPQ en DAI laten matig positieve verbanden zien. De OSCS subschaal van de RPQ en DAI laten matig tot sterk positieve samenhang zien met de geobserveerde subschaal Benadering van Vreemde

Studie	*Onderzoeksgroep*	*Focus*	*Steekproef*	*Gebruikte instrumenten*	*Belangrijkste bevindingen*
Scheper, Abrahamse, Jonkman, Schuengel, Lindauer, de Vries, Doreleijers & Jansen (2016)	Kinderen (leeftijd 2-8 jaar) verwezen voor behandeling van emotionele en gedrags-problemen; subgroep thuiswonend versus subgroep pleegzorg.	Prevalentie van RHS en OSCS gedrag	*N* = 200 kinderen: Verwezen thuis-wonende kinderen (*n*=141) en verwezen pleegzorg kinderen (*n*=59)	*Zorg geschiedenis:* Maltreatment Classification System (MCS) *Interview:* Disturbances of Attachment Interview (DAI) *Observatie:* **Geen meting** *Psychopathologie:* Child Behaviour Checklist (CBCL)	Verwezen thuiswonende kinderen: RHS gedrag: 9% OSCS gedrag: 42% Verwezen pleegzorg kinderen: RHS gedrag: 27% OSCS gedrag: 51%
Kay, Green & Sharma (2016)	Adoptie kinderen (leeftijd 6-11 jaar) zonder achter-grond van institutionalisering	Prevalentie van OSCS	*N* = 60 kinderen, met een controle-groep van verwezen, niet-verwaarloosde kinderen met externaliserende gedragsstoornis (ED, *n*=26) en een laag-risico groep (LR, *n*=55).	*Zorg geschiedenis:* Maltreatment Classification System (MCS) *Interview:* Child and Adolescent Psychiatric Assessment, Reactive Attachment Disorder module (CAPA-RAD), Relationship Problems Questionnaire (RPQ) *Observatie:* registratie van OSCS gedrag tijdens 2,5 uur durend huisbezoek *Psychopathologie:* Development and Wellbeing Assessment (DAWBA)	Overeenkomstig criteria OSCS: 49% van Adoptie kinderen; 4% van ED-kinderen; 6% van LR-kinderen

Studie	*Onderzoeksgroep*	*Focus*	*Steekproef*	*Gebruikte instrumenten*	*Belangrijkste bevindingen*
Giltaij, Sterkenburg & Schuengel (2017)	Kinderen met licht verstandelijke beperking (leeftijd 5-11 jaar) verwezen voor psychiatrische beoordeling vanwege emotionele en gedrags-problemen.	Prevalentie van RHS en OSCS	*N* = 55 kinderen	*Zorg geschiedenis:* Gestructureerde inspectie van medische rapportages op DSM-5 gedefinieerde vormen van extreem ontoereikende zorg *Interview:* Disturbances of Attachment Interview (DAI) *Observatie:* Clinical Observation of Attachment 6-15 (COA 6-15), List of Behavioural Signs of Disturbed Attachment in Young Children (BSDA) *Psychopathologie:* Diagnostic Interview Schedule for Children, version IV (DISC-IV)	Volgens de DAI: Signalen RHS of OSCS: 35% RHS signalen: 16% OSCS signalen: 27% Volgens de DSM-5: RHS en/of OSCS: 18% RHS: 16% OSCS: 15%

Kinderen met een verstandelijke beperking, die psychische of gedragsproblemen vertonen, vormen eveneens een risico groep voor ontwikkeling van verstoorde gehechtheid. De mate van verhoogd risico op gedragskenmerken en diagnose van RHS en OSCS bij deze groep, was nog onbekend. Er is daarom breed ingezet voor de werving van kinderen voor deze studiegroep. Bij alle grote centra voor kinder- en jeugdpsychiatrie, gespecialiseerd voor kinderen met LVG, verdeeld over Nederland heeft de werving plaatsgevonden (Figuur 1).

Figuur 1 Overzicht van in studie participerende centra voor Kinder- en Jeugdpsychiatrie, gespecialiseerd voor kinderen met LVG

Dit resulteerde in een studiegroep van 102 kinderen, die voldeden aan de criteria van leeftijd, niveau van cognitief functioneren en taalkennis (Hoofdstuk 2). Na één tot twee jaar werden de ouders van deze studiegroep opnieuw benaderd voor vervolgonderzoek. Een aanzienlijk deel van de studiegroep bleek echter geen zorgrelatie meer te hebben met het psychiatrisch centrum, en voor een deel van de ouders werd de deelname aan het vervolgonderzoek naast de lopende behandeling vanuit het centrum als een belasting voor het kind of de ouders ingeschat. De ouders

van 55 kinderen gaven hun informed consent voor deelname aan de vervolgstudie (Hoofdstuk 3 en 4). Op grond van de scores op de Disturbances of Attachment Interview bleek in dit onderzoek volgens de informatie van de primaire opvoeder 42% van de kinderen voldoende gedragskenmerken te vertonen voor de classificatie RHS Inhibited (RHS) en/of RHS Disinhibited (OSCS). De vaststelling dat 11% van de kinderen zowel voldoet aan de criteria van RHS Inhibited als aan RHS Disinhibited onderschrijft de beschrijving van de twee afzonderlijke stoornissen RHS en OSCS in de DSM-5 in plaats van de in de DSM-IV gestelde onverenigbare typen indeling Inhibited RHS of Disinhibited RHS. Er werd geen verband gevonden tussen de RHS- en OSCS-classificatie en geslacht, niveau van intellectueel functioneren, etnische achtergrond of leeftijd. De klinische diagnose van RHS en/of OSCS werd bij 18% van deze kinderen vastgesteld, waarbij de richtlijnen van de AACAP voor de vaststelling van de diagnose werden gevolgd.

Niettegenstaande de inspanningen voor het verzamelen van een zo groot mogelijke steekproef, was de onderzoeksgroep te klein om uitspraken te doen op subgroep niveau (subgroep RHS, OSCS en RHS/OSCS). Daarnaast kunnen de prevalentiecijfers mogelijk ook vertekend zijn door selectieve non-respons. Een beperking aan de studie is het ontbreken van zicht op de factoren die met deze non-respons samenhingen. Hierdoor is het niet mogelijk de resultaten daarop te corrigeren. Op grond van deze beperkingen kunnen geen conclusies verbonden worden aan de prevalentiecijfers van de diagnoses in de doelpopulatie.

Zowel de DAI als de COA is oorspronkelijk ontwikkeld voor de screening en observatie van verstoord gehechtheidsgedrag bij kinderen in de leeftijdscategorie tot vijf jaar. Aanvankelijk was het gebruik van deze instrumenten beperkt tot de doelpopulatie van kinderen tot vijf jaar, aangezien het nog onduidelijk is hoe de fenomenologie van verstoord gehechtheidsgedrag bij oudere kinderen eruit ziet. Longitudinaal onderzoek is nodig om te komen tot een beter begrip en beschrijving van symptomen van RHS en OSCS bij oudere kinderen, zoals het Bucharest Early Intervention Project (Smyke et al., 2012), waarbij de ontwikkeling van kinderen met vastgestelde verstoorde gehechtheid voor langere tijd wordt gevolgd en vergeleken in verschillende condities van opgroeien. Zoals gehechtheidsgedrag zich met de leeftijd ontwikkelt, kan ook worden verondersteld dat verstoord gehechtheidsgedrag zich met de leeftijd ontwikkelt. Verondersteld kan worden dat kinderen ouder dan vijf jaar problemen vaker zelf oplossen, vaker contact zullen leggen met onbekende personen en dat veiligheid door

kinderen meer geïnternaliseerd wordt. Op dezelfde wijze zou het voor OSCS kenmerkende disinhibited gedrag zich in de adolescentie wellicht laten zien in de wijze waarop de jeugdige vriendschappen vormgeeft: oppervlakkige en veelvuldig wisselende vriendschappen, bij nieuwe contacten snel gebruik maken van term als zijnde hun 'best friend' (Zeanah et al., 2016). Kay en Green (2013) vonden in hun onderzoek bij niet-geïnstitutionaliseerde, jeugdzorg-begeleide adolescenten (10-16 jaar) met een achtergrond van vroege verwaarlozing en mishandeling, een hoge prevalentie van disinhibited indiscriminate gedrag (overmatig familiair gedrag, vreemden persoonlijke vragen stellen, troost zoeken bij vreemden, en weglopen van verzorgers), aandacht trekkend gedrag (veeleisend en bezittelijk gedrag, voorzichtig en waakzaam gedrag), oppervlakkige relaties en onvoorspelbaarheid. De sociale beperking als uitingskenmerk van deze stoornis lijkt voortgezet te worden gedurende de ontwikkeling. Er is geen aanleiding om te veronderstellen dat de in de DAI uitgevraagde gedragskenmerken van verstoorde gehechtheid bij kinderen in de leeftijd van de latentiefase niet van toepassing zou zijn (Humphreys, Nelson, Fox & Zeanah, 2017). Dit instrument wordt in enkele recente studies bij kinderen ouder dan vijf jaar, gebruikt voor de vaststelling van verstoord gehechtheidsgedrag (Jonkman et al., 2014; Kay et al., 2016; Smyke et al., 2012). Bij het uitvragen en scoren van het door de opvoeder beschreven gedrag dient echter wel rekening gehouden te worden met de fenomenologie van dit gedrag in deze leeftijdsfase. Zo zal van een kind in de latentiefase het nemen van afstand van de opvoeders in een onbekende omgeving minder snel als maladaptief worden beschouwd dan bij een peuter. Goed doorvragen is dan belangrijk. In het uitvragen van de DAI gedragskenmerken zal ook rekening moeten worden gehouden met de verstandelijke beperking van het kind. Zo zal bijvoorbeeld doorvragen van belang zijn bij het item aangaande het vertonen van gevaarlijk gedrag. Kinderen met een verstandelijke beperking zullen zich minder bewust zijn van het gevaar in veel situaties. Het oorspronkelijke protocol van de COA (AACAP, 2005; Boris et al., 2004) is voor de leeftijdsgroep van de latentiefase aangepast met andere stress verhogende onderdelen. De andere leeftijdsgroep verlangt andere stressoren om (verstoord) gehechtheidsgedrag op te roepen. Er was geen significant verband tussen de scores voor maladaptief gehechtheidsgedrag op de COA en de leeftijd voor kinderen die werden aangemerkt met verstoord gehechtheidsgedrag (Hoofdstuk 3). Het is daarmee onwaarschijnlijk dat het oprekken van de leeftijdsperiode waarin de instrumenten werden gebruikt heeft geleid tot een overrapportage van verstoord gehechtheidsgedrag.

De veronderstelde factor voor het ontstaan van de stoornissen RHS en OSCS is extreem ontoereikende zorg (pathogene zorg). Extreem ontoereikende zorg is in de DSM-IV en DSM-5 in betrekkelijk algemene termen beschreven, waardoor het gevaar bestaat van over- en onderbeoordeling van de zorgsituatie van individuele kinderen. Voor het meten van vormen van verwaarlozing en mishandeling vormt de Maltreatment Classification System (MCS, Barnett, Manly, & Cicchetti, 1993) een bewezen betrouwbaar en valide instrument (Cicchetti, Rogosch, & Thibodeau, 2012; English et al., 2005), dat in diverse studies naar RHS volgens de criteria van de DSM-IV is gebruikt om pathogene zorg vast te stellen. In de DSM-5 is echter dit criterium meer verschoven naar (patronen van extreem ontoereikende) verzorgingsomstandigheden die het aangaan van selectieve en stabiele gehechtheidsrelaties ernstig beperken. Voor de vaststelling hiervan hebben wij nog geen betrouwbaar en valide instrument gevonden. In de huidige studie hadden we toegang tot het volledige klinisch-medisch dossier van de participanten en bij het scoren van 'extreem ontoereikende zorg' zijn heldere criteria beschreven, namelijk: het kind heeft in de eerste drie jaar veelvuldig met wisselende opvoeders te maken gehad; of het kind was langdurig of frequent in het ziekenhuis opgenomen; of het kind is op jonge leeftijd uit huis geplaatst; of er is sprake van een maatregel vanuit de Raad voor de Kinderbescherming c.q. de kinderrechter. Hoewel deze formulering de vroegkinderlijke zorgomstandigheden meer concreet definieert en daarmee duidelijker te scoren valt, geeft deze formulering nog ruimte voor kwalitatieve verschillen. Het is daarnaast ook moeilijk de mate van ontoereikende zorg in de ontwikkelingsgeschiedenis van het kind te beoordelen op grond van subjectieve beschrijvingen door de opvoeders. Het is moeilijk vast te stellen of sprake is (geweest) van verwaarlozing of mishandeling bij zeer jonge kinderen die niet in staat zijn om (verbaal) hun ervaringen te beschrijven (Zeanah & Gleason, 2010). Dit kan mogelijk geleid hebben tot vals negatieve beoordelingen over extreem ontoereikende zorg. Interessant is de benadering van Cyr, Euser, Bakermans-Kranenburg en van IJzendoorn (2010), die een meta-analyse uitgevoerd hebben naar de samenhang van belangrijke socio-economische risico indicatoren voor de gehechtheidsontwikkeling van kinderen. Er werd geconcludeerd dat kinderen die waren blootgesteld aan tenminste vijf van de zes risico indicatoren een zelfde prevalentie van gedesorganiseerde gehechtheid lieten zien als bij kinderen met een achtergrond van vastgestelde kindermishandeling. Een dergelijke studie naar samenhang van risico indicatoren en verstoord gehechtheidsgedrag zou bijdragen aan een meer objectieve vaststelling van mogelijke extreem ontoereikende zorg voor het kind.

Om de verschillende onderzoeksmeetinstrumenten (interviews, vragenlijsten, observatiesituatie) met elkaar te kunnen vergelijken is gekozen voor het betrekken van één van beide opvoeders in het onderzoek. Echter, de mogelijkheid bestaat dat de andere primaire opvoeder, indien aanwezig, een andere reactie of beschrijving zou hebben gegeven op en in de relatie met het kind. De resultaten kunnen daarom alleen worden geïnterpreteerd als in de relatie met deze specifieke ouder. Voor diagnostisering in de klinische praktijk is het echter belangrijk om de AACAP richtlijn te volgen en meerdere opvoeders te interviewen en in de relatie met het kind te observeren. De huidige studie richtte zich op de kind-gebonden stoornissen RHS en OSCS, maar door de gehanteerde methode kunnen wij niet uitsluiten dat het gesignaleerde verstoorde gehechtheidsgedrag bij sommige kinderen relatie-gebonden was. Vervolgonderzoek is nodig naar de diagnostische classificatie van verstoord gehechtheidsgedrag als een relatie-gebonden stoornis (Zeanah & Lieberman, 2016).

Het ontbreken van eenduidige gedragsbeschrijvingen (drempels) passend bij de coderingsvariatie bij de acht gedragssignalen van adaptief en maladaptief gehechtheidsgedrag op de BSDA is opgelost door het inzetten en trainen van vijf onafhankelijke codeerders. Deze kregen ad random video opnames van de COA's toebedeeld, waarbij elke COA door twee codeerders werd gescoord, onafhankelijk van elkaar en zonder voorkennis over het kind. Indien de toegekende scores op een item één punt verschilde, vond door beide codeerders overleg plaats om tot consensus te komen. Indien het gedragssignaal door de twee codeerders met meer dan één punt verschil werd beoordeeld, werd dit item nogmaals gescoord door een derde onafhankelijke codeerder, die geen kennis had van de oorspronkelijk toegekende scores. Deze derde score werd meegenomen in het overleg om tot consensus te komen. Op deze wijze werd overeenstemming bereikt over de drempels van de gehanteerde BSDA codering, hetgeen nodig is voor een goede interbeoordelaars-betrouwbaarheid. In deze studie was de interbeoordelaars-betrouwbaarheid goed (.73).

Kinderen bij wie op grond van een vastgestelde achtergrond van ernstig ontoereikende zorg in combinatie met geobserveerd maladaptief gehechtheidsgedrag (in COA) de klinische diagnose van RHS en/of OSCS kregen toebedeeld lieten een score zien tussen 22 en 33 op de BSDA, met een gemiddelde van 26,9. Kinderen bij wie geen achtergrond van ernstig ontoereikende zorg kon worden vastgesteld, lieten een score op de BSDA zien van 9 tot 29, met een gemiddelde score van 16,0. Bij vier van deze kinderen werd

wel maladaptief gehechtheidsgedrag in de observatie waargenomen (scores 23 tot 29), maar kon op grond van het ontbreken van achtergrond van ernstig ontoereikende zorg vanuit de beschikbare ontwikkelingsanamnese geen RHS of OSCS vastgesteld worden. Deze cijfers geven aan dat de BSDA een geschikte observatielijst is voor gebruik in het diagnostisch proces naar gehechtheid gerelateerde stoornissen, maar dat nog verder onderzoek vraagt zowel naar standaardisatie, normstelling en mogelijke profielen binnen de BSDA. Er is ook behoefte tot het ontwikkelen van een semi-gestructureerd interview over de ontwikkelingsgeschiedenis en de omstandigheden van het kind gericht op eventuele pathogene zorg en onvoldoende mogelijkheden tot het aangaan van een stabiele, selectieve gehechtheidsrelatie. In een dergelijk vervolgonderzoek naar BSDA-profielen en ontwikkelingsachtergrond dient dan tevens meegenomen te worden de relatie-specifieke vorm van verstoorde gehechtheid (Groh, Fearon, van IJzendoorn, Bakermans-Kranenburg, & Roisman, 2016; Zeanah & Lieberman, 2016).

Klinische implicaties

Deze studie draagt bij aan de ontwikkeling van klinische protocollen voor de diagnostiek van de stoornissen RHS en OSCS bij kinderen met een licht verstandelijke beperking. In de klinische praktijk van de Nederlandse Jeugdzorg en Jeugd Geestelijke Gezondheidszorg zijn momenteel drie richtlijnen beschikbaar. Allereerst is er de internationaal geaccepteerde Practice Parameter voor de diagnostiek en behandeling van RHS en OSCS, vastgesteld door de AACAP (2005, met revisie in 2016), die op grond van de huidige state-of-the-art aan wetenschappelijke onderzoeksbevindingen inzake RHS en OSCS onderbouwde richtlijnen geeft voor het uitvoeren van diagnostiek en behandeling. Daarnaast is er de door Dekker-van der Sande en Janssen (2010) beschreven Best Practice voor het signaleren van verstoord gehechtheidsgedrag, bestaande uit drie fasen: screening, diagnose en intern werkmodel. Deze richtlijn is vervolgens verder uitgewerkt tot de Richtlijn Problematische Gehechtheid, welke is opgenomen in de richtlijnen databank Jeugdhulp en Jeugdbescherming van het Nederlands Jeugd Instituut (De Wolff et al., 2014).

Zover bekend is dit de eerste studie die rapporteert over diagnostiek waarin meerdere meetinstrumenten gebruikt zijn, die in combinatie voldoen aan de richtlijnen van de

Practice Parameter voor de diagnostiek en behandeling van kinderen en adolescenten met hechtingsstoornissen (Zeanah et al., 2016) voor de diagnostiek van RHS en OSCS bij kinderen met licht verstandelijke beperking, die verwezen zijn voor psychiatrische beoordeling vanwege ernstige emotionele en/of gedragsproblemen. Deze studie formuleert de samenhang en beperkingen van deze onderzoeksinstrumenten voor de screening en diagnosestelling van RHS en OSCS, en daarmee, de gevolgen voor het klinisch onderzoeksprotocol van deze gehechtheid gerelateerde stoornissen. Hoewel het volledig uitvoeren van het diagnostisch onderzoek volgens de AACAP richtlijn in de klinische praktijk een tijdrovend proces is, laten de bevindingen in deze studie zien dat het uitermate belangrijk is om voor het stellen van een verantwoorde RHS of OSCS diagnose de volledige richtlijn te gebruiken en het volledige protocol te doorlopen. Zo bleek uit deze studie, dat het zich beperken tot het afnemen van een semigestructureerd interview als de DAI nog bij 25% van de deelnemers tot een foutieve diagnose zou leiden. De consequentie van deze bevinding is dat de DAI in de klinische praktijk gebruikt zou moeten worden als een screeningsinstrument in de diagnostische beoordeling van gehechtheid gerelateerde stoornissen. De DAI blijkt een goed bruikbaar instrument om in relatief korte tijd een beeld te krijgen van mogelijk verstoord gehechtheidsgedrag, zoals dat door de primaire opvoeder wordt weergegeven. Afzonderlijke afname van dit interview bij beide opvoeders is aan te raden. De klinische diagnose van RHS en/of OSCS kan vervolgens vastgesteld worden aan de hand van afname van de (gehechtheids)ontwikkelingsgeschiedenis van het kind, met het focus op eventuele extreem ontoereikende zorg met name in de eerste levensjaren, en aan de hand van observatie van (verstoorde) gehechtheidsgedrag in de gestructureerde COA. Voor het scoren van het geobserveerde gedrag lijkt de BSDA een passend instrument te zijn.

Voor de diagnostiek van de stoornissen RHS en OSCS wordt op grond van de hier beschreven studieresultaten geadviseerd een 'stepped-care-model' te hanteren, waarbij uitvoeriger diagnostiek volgens de hierboven beschreven richtlijnen wordt verricht naarmate de resultaten van eerdere fasen in het diagnostisch traject wijzen in de richting van mogelijke classificatie van verstoorde gehechtheid.

Fase 1 bestaat uit het standaard afnemen van de vragenlijst *Child Behavior Checklist* (CBCL, Achenbach & Rescorla , 2001) of *Developmental Behaviour Checklist* (DBC; Einfeld and Tonge, 1991, 1994, 1995; Einfeld, Tonge, & Parmenter, 1999) bij de primaire

opvoeders en leerkracht, en afname van de ontwikkelingsanamnese, inclusief ontwikkeling van het gehechtheidsgedrag bij de primaire opvoeders. Indien de scores op de vragenlijst hoog zijn en de informatie van de ontwikkelingsanamnese aanleiding geeft om (ook) gehechtheidsproblematiek en/of ernstige ontoereikende zorg te veronderstellen, dan wordt overgegaan naar de volgende diagnostiekfase.

Fase 2 bestaat uit het afnemen van het *Disturbances of Attachment Interview* (DAI, , Smyke et al., 2002) bij minimaal één primaire verzorger. Zo nodig kan dit uitgebreid worden met afname van een screenings- of diagnostisch onderzoek naar mogelijke differentiaaldiagnose of comorbiditeit van ASD met de *AUTI-R* (Van Berckelaer-Onnes & Hoekman, 1991) of *Autism Diagnostic Observation Scale* (ADOS-2, De Bildt, Greaves-Lord, & De Jonge, 2013). Indien de scores op deze instrumenten wijzen op verstoord gehechtheidsgedrag wordt overgegaan naar de laatste diagnostiekfase.

Fase 3 bestaat uit het afnemen van de observatieprocedure *Clinical Observation of Attachment* (COA, Boris et al., 2004, aangepast door Giltaij & Sterkenburg., 2017, bijlage bij dit manuscript) met het kind, de primaire verzorger en een onbekende volwassene. Deze procedure wordt zo mogelijk afgenomen bij beide primaire verzorgers om vast te stellen of het verstoord gehechtheidsgedrag zich laat zien in meerdere opvoedrelaties (kind-gebonden stoornis). De observatiesessie wordt opgenomen op video en vervolgens geanalyseerd en gescoord met de *List Behavioral Signs of Disturbed Attachment in Young Children* (BSDA, Zeanah et al., 1993). Hoge scores op de BSDA dragen bij aan diagnose van een gehechtheid gerelateerde stoornis. De in de DSM-5 beschreven gedragskenmerken geven uitsluitsel of sprake is van RHS, OSCS dan wel beide stoornissen. Voor de bepaling van differentiaaldiagnose en comorbiditeit van andere stoornissen is de afname van de *Diagnostic Interview Schedule for Children, version IV* (DISC-IV; Shaffer et al., 2000) aan te bevelen.

Opgemerkt dient te worden dat de fasen 1 en 2 vooral bestemd zijn voor triage beoordeling en derhalve onderhevig kunnen zijn aan onvolledige informatie en subjectieve beschrijvingen. Fase 3 betreft het werkelijke geobserveerde gedrag van het kind in een gestandaardiseerde situatie die (verstoord) gehechtheidsgedrag oproept, en dat vervolgens door een getrainde diagnosticus geanalyseerd en gescoord wordt. Op grond van deze resultaten dient de ervaren clinicus de diagnose vast te kunnen stellen.

Dit is een eerste studie naar de prevalentie en comorbiditeit van RHS en OSCS bij kinderen met een licht verstandelijke beperking, die vanwege ernstige psychische of gedragsproblemen verwezen werden voor nader kinderpsychiatrisch onderzoek. De resultaten tonen aan dat het een risicogroep is voor de psychiatrische stoornissen RHS en OSCS (Hoofdstuk 3). Kanttekening hierbij is het beperkte aantal deelnemers (55 kinderen) en het gegeven dat observatie en interview is uitgevoerd bij één primaire verzorger. Door in de klinische praktijk de gehechtheidsontwikkeling en diagnostiek van deze stoornissen een prominente plaats te geven en de triage beoordeling voor verdere diagnose van verstoord gehechtheidsgedrag op te nemen in het standaard diagnostisch traject zal dit ten goede komen aan de juistheid en volledigheid van het diagnostisch beeld van de problematiek. Als de diagnostiek beter is ingevoerd, dan kan de empirische onderbouwing van de behandeling ook verder ontwikkeld worden.

Kinderen met RHS of OSCS werden ook gediagnostiseerd met verhoogde aantallen van comorbide psychiatrische stoornissen (angst-, stemmings- en/of gedragsstoornissen) (Hoofdstuk 4), hetgeen de complexiteit van zowel diagnostiek als behandeling vergroot. Dit vergroot het risico dat binnen de diagnostiek het focus op comorbide stoornissen komt te liggen en de onderliggende stoornis RHS of OSCS niet wordt vastgesteld en in de behandeling wordt betrokken. De DSM-5 vereist uitdrukkelijk met het oog op de diagnose van RHS dat het mogelijk moet zijn het maladaptieve gedrag van de RHS te onderscheiden van gedrag dat verband houdt met de autisme spectrumstoornis (ASS). Hiervoor is een gedegen uitvragen van de ontwikkelingsgeschiedenis van groot belang (achtergrond van ernstige sociale verwaarlozing, selectief gehechtheidsgedrag) en nauwkeurige beschrijving van de verschillende gedragskenmerken die typerend zijn voor het maladaptieve gedrag passend bij ASS en/of RHS. In deze studie is vastgesteld dat het goed mogelijk is om met gericht onderzoek onderscheid te maken tussen verstoord gehechtheidsgedrag en autistisch gedrag (Hoofdstuk 2). Deze bevinding heeft implicaties gehad voor de Practice Parameter (Zeanah et al., 2016) en de onderbouwing van de classificaties RHS en OSCS in de Diagnostic Manual-Intellectual Disabilities-2 (DM-ID-2, Fletcher, Barnhill, & Cooper, 2016; Fletcher, Barnhill, McCarthy & Strydom, 2016). Terwijl extreem ontoereikende zorg gedurende de eerste levensjaren van het kind wordt verondersteld de oorsprong te zijn voor de ontwikkeling van de gehechtheid gerelateerde stoornissen RHS en OSCS, wordt de oorsprong van de stoornis ASS verklaard vanuit een verstoorde neurobiologische ontwikkeling. De mogelijkheid tot het onderscheiden tussen gehecht-

heid gerelateerde stoornissen en ASS maakt het mogelijk een passende behandeling aan te bieden, hetgeen de kans op een succesvolle behandeling van de stoornis(sen) vergroot.

Voor geschikte, kwalitatief goede behandeling en zorg is het ook belangrijk dat clinici zich realiseren dat kinderen met een gehechtheid gerelateerde stoornis kunnen leiden aan relatief meer ernstige achterstanden in adaptief vermogen (Hoofdstuk 4).

Algemene conclusies

Kinderen met een verstandelijke beperking zijn een hoog-risico groep voor het ontwikkelen van ernstige psychische en gedragsstoornissen (psychopathologie), die drie tot vier keer zo hoog is als bij kinderen zonder verstandelijke beperking (Dekker & Koot, 2003). De kwetsbaarheid van deze groep toont de behoefte aan voor speciale aandacht van het wetenschappelijk onderzoek en de klinische praktijk van de geestelijke gezondheidszorg. Het afgelopen decennium is de geestelijke gezondheidszorg voor de mensen met een verstandelijke beperking verbeterd, zowel in wetenschappelijk onderzoek als in de klinische praktijk, maar deze verbetering verloopt erg langzaam en er is nog steeds sprake van een grote achterstand vergeleken met de geestelijke gezondheidszorg voor de algemene populatie (Flynn, 2012). Gehechtheid en verstoorde gehechtheid werden gekozen als studie onderwerp vanuit de aanname dat kinderen met een verstandelijke beperking een hoog-risico groep voor de ontwikkeling van verstoorde gehechtheid zijn (Green & Goldwyn, 2002; Schuengel & Janssen, 2006). In deze studie wordt deze aanname bevestigd. Bijna één op de vijf latentiekinderen met een verstandelijke beperking, verwezen voor psychiatrische beoordeling en behandeling vanwege psychische gezondheidsproblemen, werden positief gediagnostiseerd op een gehechtheid gerelateerde stoornis RHS en/of OSCS.

De doelstelling van deze studie was om een bijdrage te leveren aan de beschrijving van de klinische diagnose van de DSM-5 gehechtheid gerelateerde stoornissen RHS en OSCS, en tot verbetering van de diagnostische beoordeling van deze stoornissen in de klinische geestelijke gezondheidspraktijk. Wij vonden dat het goed mogelijk is om onderscheid te maken tussen de klinische diagnose van Reactieve Hechtingsstoornis (RHS) en de klinische diagnose van Autismespectrumstoornis (ASS), hetgeen één van de

voorwaarden van de DSM-IV en DSM-5 is voor de classificatie van RHS. Met de richtlijnen van de Practice Parameter, aanbevolen door de American Academy of Child and Adolescent Psychiatry (2005, 2016) hebben wij een multi-informant diagnostisch model samengesteld voor het testen van latentiekinderen op gehechtheid gerelateerde stoornissen. Wij gebruikten vragenlijsten, interviews, gestructureerde observaties en de informatie van de klinisch medische dossiers van het kind. Informanten waren de primaire verzorgers, onderwijzers, het jeugdpsychiatrisch centrum, en het kind. De samenhang tussen de elementen van het model werd onderzocht en resulteerde in de constructie van een drie-fase stepped-care model voor het diagnosticeren van gehechtheid gerelateerde stoornissen in de klinische praktijk. Gebruik makend van dit model bevestigden de resultaten de noodzaak om te investeren in de beoordeling van gehechtheid gerelateerde stoornissen, want wij vonden meer comorbide psychopathologie en functionele ontwikkelingsachterstand bij kinderen met classificatie van gehechtheid gerelateerde stoornissen dan bij kinderen zonder deze stoornis. Deze bevindingen onderschrijven de behoefte naar ontwikkeling en testen van passende behandeling en begeleidingsprogramma's voor kinderen en hun verzorgers.

De tweede doelstelling van deze studie was het verspreiden van de verworven kennis naar het veld van het wetenschappelijk onderzoek en de klinische praktijk. De bevindingen van deze studie zijn opgenomen in de herziende uitgave van het Diagnostic Manual Intellectual Disability (DM-ID), in navolging van de samenstelling van de DSM-5. Dit Textbook of Diagnosis of Mental Disorders in Persons with Intellectual Disability, opgesteld door the National Association for the Dually Diagnose (NADD) of the USA (Fletcher, Barnhill, & Cooper) is gepubliceerd in 2016 als de DM-ID2. In Nederland zijn de resultaten van de studie en het diagnostisch model beschreven als een hoofdstuk in het eerste Handboek Psychiatrie en Licht Verstandelijke Beperking (opgesteld door Didden, Troost, Moonen, & Groen, 2016). De in deze studie verzamelde kennis heeft geleid tot een meerdaagse trainingscursus voor klinisch-werkende professionals en wetenschappelijk onderzoekers in het gebruik van het Disturbances of Attachment Interview (DAI) als instrument in het diagnostische proces voor gehechtheid gerelateerde stoornissen. Een vergelijkbaar trainingscursus in het gebruik van de Clinical Observation of Attachment 6-12 (COA 6-12) is in voorbereiding. De aanhoudende vraag naar deze training toont de voortdurende belangstelling in de unieke gedragingen van RHS en OSCS aan, en daarmee de relevantie van vervolgonderzoek op dit gebied.

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Dankwoord

(Acknowledgements)

Op een maandagochtend in april 2004 reed ik op de snelweg A1 richting mijn werkadres in Almelo. Ik reed deze route van 115 kilometer heen, 115 kilometer terug, drie dagen in de week. Ik deed dat al bijna 8 jaar, dus de weg was zeer vertrouwd. Wel leek de afstand de laatste 2-3 jaren steeds langer te worden, en werd als steeds vermoeiender ervaren. Ter hoogte van Apeldoorn, tijdens het inhalen van een vrachtwagen (130 km/uur), gebeurde het: ik zag een baksteen recht op mij afkomen! In een reflex wist ik het stuur een ruk te geven, waarna de baksteen via de motorkap afketste en rechts de zijspiegel afsneed …. Op de vluchtstrook heb ik vervolgens 20 minuten doodstil achter het stuur gezeten, emoties reguleren (zouden we nu zeggen …). Het is op dat moment dat ik mijzelf de vraag stelde: "Waar ben ik mee bezig?". Daarbij werd alleen maar de intense binding met mijn jonge kinderen Willemijn (3 jaar) en Maarten (half jaar) gevoeld, en die met Yvonne, mijn levenspartner en maatje. Al het andere verdween naar de achtergrond en leek niet belangrijk. Het is op die plaats, dat ik het besluit nam om de risico's van die lange autoritten niet langer te tarten, maar naar een andere werkplek te zoeken, die dichter bij huis en gezin zou liggen. Ik gaf mijzelf hiervoor vijf maanden ….

Op 26 augustus 2004 was op mijn andere werkplek (Bartiméus) een bijeenkomst van de klankbordgroep, verbonden aan het promotieonderzoek van mijn collega Paula Sterkenburg. Als voorzitter van deze klankbordgroep nodigde ik de leden na afloop van de bespreking uit voor lunch in het restaurant. Tijdens deze lunch sprak ik met de promotor van Paula, Carlo Schuengel. We spraken over ideeën die bij de afdeling psychotherapie bestonden voor onderzoeksthema's die in de samenwerking met de Vrije Universiteit wellicht uitgewerkt zouden kunnen worden. Er volgde een gesprek waarbij we beide steeds enthousiaster werden, waarna Carlo plots de woorden uitsprak: "Waarom doe je het niet zelf?". Tja, die had ik niet zien aankomen, en lag op dat moment ver buiten mijn voorstellingsvermogen. Maar, het was wel de laatste week van die vijf maanden …. Toeval? Een teken? In ieder geval een heel bijzondere kans om naast mijn klinische carrière ook kennis te maken met de wereld van het wetenschappelijk onderzoek ……..!

Het zou uiteindelijk nog ruim een jaar duren van voorbereidend werk alvorens duidelijk werd dat dit ambitieuze plan daadwerkelijk kon gaan starten. Ik nam afscheid van mijn collegae en de adolescenten van De Bleskolk in Almelo, en stapte als parttime wetenschappelijk onderzoeker in de trein naar de Vrije Universiteit in Amsterdam.

Het eind van een zeer uitdagend traject is nu in zicht: HEERLIJK !. Dit traject heeft beduidend langer geduurd (ruim 12 jaar!) dan bij aanvang was bedoeld en waarop was ingestoken. Het is een traject geweest van veel aanpassen en opnieuw piketpaaltjes slaan: niet alleen van mij en van mijn begeleiders, maar zeker ook van mijn meest dierbaren, mijn gezin. De eerlijkheid gebiedt om hier ook aan te geven dat soms de moedeloosheid de overhand dreigde te krijgen en de aandrang opstak om te doen wat slecht bij mij past: opgeven. Toch waren er dan steeds personen die de juiste steunende woorden zeiden om voort te gaan. Het is een traject geweest dat mij heel veel heeft geleerd over mensen, over relaties, en zeker ook over mijzelf. Ik kan hierover alleen maar dankbaar zijn.

Op deze plek wil ik ieder die een bijdrage geleverd heeft, direct of indirect, aan de uitvoering van dit onderzoek en de totstandkoming van dit proefschrift, bedanken.

In de eerste plaats wil ik dank zeggen tegen alle kinderen en ouders die hebben meegewerkt. Terwijl zij zelf vaak zaten in een moeilijke periode in het leven, waarbij hulp ingeroepen moest worden, en waarbij zij zich naar hulpverleners kwetsbaar moesten opstellen, was er toch nog de bereidheid om aan dit onderzoek mee te werken: vragenlijsten in te vullen, op interviews in te gaan en naar instellingen te gaan om zich te laten filmen in observatiesituaties. En dat alles zonder eigen belang, maar omdat zij weten hoe het is om kinderen in psychische nood te hebben/zijn. Diep respect is hier op zijn plaats ….

Ook een woord van dank aan de leerkrachten en begeleiders van de kinderen, die de tijd namen om tussen de drukke werkzaamheden heen, informatie te verstrekken en vragenlijsten in te vullen over de kinderen. Ontroerend was de manier waarop door hen zorg en medeleven met het kind en ouders werd gedeeld.

Dank ook aan de medewerkers (coördinatoren, psychiaters, psychologen, administratie) van de participerende instellingen. Zonder de toestemming van de directie van deze instellingen en de inzet en enthousiasme van deze medewerkers zouden we nooit de ouders en kinderen hebben kunnen bereiken en zouden we nooit dit onderzoek hebben kunnen uitvoeren. De steun en het enthousiasme van deze medewerkers werd veelvuldig door hen kenbaar gemaakt door het benadrukken hoe sterk de behoefte in

de klinische praktijk bestaat naar meer kennis en expertise betreffende verstoorde gehechtheid.

Zonder financiële steun en facilitering is onderzoek niet mogelijk. Zowel Bartiméus (en de Vereniging Bartiméus Sonneheerdt) als de Vrije Universiteit hebben dit onderzoeksproject omarmt en besloten samen dit te ondersteunen met een budget dat uiteindelijk zes jaar van het project (tot en met de fase van dataverzameling) heeft kunnen financieren. Ik ben hen zeer dankbaar in het gestelde vertrouwen in mij.
En dan wil ik mensen bij naam bedanken, die voor mij een bepalende rol hebben gespeeld gedurende dit traject.

Carlo, mijn promotor en degene die mij de kans heeft voorgehouden om deze bijzondere ervaring van dit onderzoeksproject aan te gaan. Carlo, ik dank je voor het vertrouwen dat je destijds in mij hebt gesteld. Ik heb je geduld zeer op de proef gesteld, maar hoop dat je tevreden bent met het bereikte resultaat. Ik zal nooit vergeten die eerste bijeenkomst met Cees Janssen, Paula en jou: jij en Cees waren in enthousiast overleg over een mogelijke onderzoeksopzet. Ik begreep helemaal niets van alle wetenschappelijke termen die jullie hanteerden, maar als een soort tovenaar verscheen uit jouw krijtje op jouw (ouderwetse) krijtbord een schets van het onderzoek. Ik voelde mij vereerd dat ik getuige mocht zijn van dit moment.

Paula, mijn copromotor, ….. ja, copromotor, maar jij was en bent zoveel meer!! Via jouw promotieonderzoek werd het contact gelegd met Carlo en de Vrije Universiteit. Jij was niet alleen de aangever van het onderzoek, maar in de uitvoering was jij op alle fronten betrokken: scoring van de DAI, 'vreemde' in de COA, scoring van de BSDA, maken van de syntax en analyses met SPSS, schrijven van de publicaties, meedenker en discussiant, aanjager en motivator, …. Ja, zelfs financieel ondersteuner. Zonder jou, jouw vertrouwen en jouw aanmoedigingen zou ik het niet hebben kunnen volhouden tot het huidige resultaat. Eigenlijk zou het passend zijn geweest als je naast mij zou staan en je tweede doctoraat zou ontvangen! Ik ben dan ook trots en blij dat je naast mij staat als copromotor en daarmee formeel en onlosmakelijk verbonden bent aan dit proefschrift. Maar bovenal ben ik vereerd en gelukkig met jou als maatje in het werk met mensen met een beperking. "Dank" zeggen is te weinig. Daarom in jouw taal: "Baie, baie dankie vir alles wat jij vir mij gedoen en beteken het !".

Neil Boris, my lecturer on disordered attachment. During the training in Amsterdam in 2005 you shared your knowledge on the, at that time, state-of-the-art on disordered attachment. During the training you created the common frame of mind between the researchers and clinicians who participated in this study. Your inspiring presentations and your enthusiastic personality has been an echo for me during this 12-years project. Thanks and hope to meet you again soon.

Cees, Clasien, Mirjam en Marielle, mijn ankerpunten op de VU. Cees, de eerste jaren ben je mijn werkbegeleider geweest. Dank dat je altijd je deur open had staan voor het mogen stellen van vragen, zelfs als je aan het klussen was in je boerderij in Frankrijk. Voor mij, omdat ik slechts twee dagen in de week aanwezig was op de VU, was die voortdurende beschikbaarheid erg belangrijk. Dank ook voor de financiële steun toen jijzelf met pensioen ging en er nog een budget onderzoeksgeld achterbleef (en mijn budget op was). Clasien, jij was (als overbuurvrouw) voor mij altijd de baken van rust en warm welkom als ik weer op de VU aankwam. Een dag zonder jouw aanwezigheid was altijd een koudere dag. Ik ben heel blij dat wij (samen met Mirjam) onze samenwerking voortzetten in het geven van landelijke trainingen in het gebruik van het Disturbances of Attachment Interview. Mirjam en Marielle, jullie waren jarenlang mijn kamergenoten in een periode waarin we alle drie in de fase van jonge ouder zaten. Apetrots deelden we bijna dagelijks de laatste foto's en belevenissen van onze kinderen, die we toch echt de mooiste en slimste van alle kinderen vonden. Dank voor al die momenten.

Mathilde, Janna en Carlijn, mijn onderzoeksassistenten. Dank voor het coördinerende werk tijdens de projectfase van de dataverzameling. Doordat jullie het onderhouden van de contacten en afstemmen van afspraken met de instellingen, coördinatoren, administraties, professionals, ouders en onderzoekers van mij hebben overgenomen, en dit zo zorgvuldig deden, kon ik mij tenminste richten op het uitvoeren van de verschillende onderzoeks- en observatie instrumenten op de verschillende locaties in het land. Jullie zijn een enorme steun voor mij geweest!

Lia en Marjoke, orthoptisten bij Bartiméus. Dank voor de samenwerking bij de vele observatie onderzoeken op locaties in het land. Het was fijn om met jullie die vele kilometers op te rijden. Jammer dat het visuele functie onderzoek bij de kinderen te weinig informatie heeft opgeleverd om in het onderzoek mee te nemen.

Ook dank aan de vele studenten, die in het kader van hun masterthese gekozen hebben om bijdrage te leveren aan het onderzoek, zoals het coderen en verwerken van gegevens.

Dank aan prof. dr. D. de Ruyter, prof. dr. H. Grietens, prof. dr. X. Moonen, prof. dr. H. van Bakel, dr. M. de Wolff en dr. M. Oosterman voor hun deelname in de leescommissie van mijn proefschrift. Ik ervaar het als een eer mijn werk aan u ter beoordeling te hebben mogen voorleggen.

Mijn team van de afdeling Psychotherapie van Bartiméus wil ik hier bij naam noemen: Paula, Francien, Hannelies, Floor, Marieke, Kobie, Marise, Maaike, Judith, Leonien, Eline, Willemijn, maar ook de collegae die de afgelopen jaren zijn vertrokken vanwege afronden van opleiding of aangaan van een andere betrekking: Eva, Esther, Tessa, Bernardien, Eva, Lisette, Lotte, Tineke en Thera. Dank voor de samenwerking en jullie steun en vertrouwen. Wat een geweldige groep mensen en professionals zijn jullie! Gedurende de afgelopen jaren heb ik regelmatig momenten van wanhoop ervaren en dan vormden jullie de 'veilige haven', waar ik ontvangen werd met een kop koffie, een luisterend oor en weer in mijn krachten werd gezet. Sinds ik met jullie werk, ben ik in verwarring waar de relatie als collegae overgaat in relatie als vrienden.

Ik ben zeer dankbaar voor hetgeen dit onderzoeksproject heeft opgeleverd voor de wetenschap, de klinische praktijk, en voor mijn persoonlijke ontwikkeling. Dat deze 12 jaar onderzoek, in combinatie met een intensieve en verantwoordelijke klinische functie, en in combinatie met een jong gezin, ook een prijs kende, heb ik ook ervaren. Ik heb gedurende dit traject mijn persoonlijke grenzen op alle gebied ervaren: lichamelijk, psychisch, sociaal en relationeel. Ik heb gedurende dit traject ook de andere kant van die grenzen ervaren. En dat waren moeilijke periodes, niet alleen voor mij persoonlijk, maar ook voor mijn meest dierbaren met wie ik het leven deel. In dit verband wil ik mijn onmetelijke dank uitspreken aan Anky en Jos, die mij, ieder op hun eigen terrein, hebben geholpen de weg te vinden om weer aan de juiste kant van de grens te komen.

Ik wil mijn vrienden bedanken voor hun geduld, begrip en belangstelling. De tijd breekt aan om samen weer afspraken te maken en leuke dingen te ondernemen! In het

bijzonder wil ik hier Millie noemen: Millie, jij kan mij laten voelen wat vriendschap is op een manier die ik niet onder woorden kan brengen.

Ik wil ook hier Jan Bijl bedanken, in het bijzonder voor de jarenlange vriendschap (evenzo geldt dat voor Wil), maar ook voor jouw prachtige omslag voor dit proefschrift. Op basis van mijn beschrijving van het onderzoeksthema kwam jij tot de verbeelding zoals opgenomen als omslag. Jan, je had het niet mooier kunnen weergeven!

Marieke en Thijs Jan, ik wil jullie danken dat jullie zo enthousiast hebben gereageerd op mijn vraag om mijn paranimf te willen zijn tijdens de verdediging van mijn proefschrift. Ik kan mij geen betere rustbakens wensen.

Nu kom ik steeds dichter bij mijn eigen 'veilige basis'. Dank aan mijn broers, schoonzussen, zwagers, en hun kinderen. Het is een geruststellend gevoel te weten dat jullie er zijn als dat echt nodig is. Dank voor jullie steun en belangstelling voor hetgeen ik voor dit onderzoek deed.

En ma ……, wat ben ik blij dat ik en wij allen dit moment met jou mogen delen. Jij hebt zo ontzettend meegeleefd met alle ups en downs deze afgelopen jaren, zoals je dat gedurende mijn hele leven hebt gedaan. Dank voor je onvoorwaardelijke liefde en alle zorg en aandacht die je had (en hebt) voor wat werkelijk belangrijk is in ons leven. En pa ……, wat zou jij trots zijn geweest en hier graag bij zijn geweest. Door hier ook mijn dankbaarheid naar jou te noemen ben jij er toch ook bij.

Lieve Yvonne, wat een reis hebben wij samen doorgemaakt. Dit project heeft ons niet alleen naar alle uithoeken van de wereld gebracht, maar ook naar die van ons samen. Jaren zat ik avonden, nachten en weekenden boven 'in mijn toren' te werken en was ik te weinig beschikbaar voor jou en ons gezin. Ik ben je heel dankbaar dat jij er altijd was en bleef voor ons gezin, ook in de periode dat ik moeilijk bereikbaar was. Dank voor je zorg, steun, liefde en vertrouwen in mij en ons samen. Dit heeft ons uiteindelijk juist gebracht naar een nabijheid en verbondenheid, die diep geworteld voelt. We gaan samen het Noorderlicht opzoeken ….

Lieve Willemijn en Maarten, wat een bijzondere kinderen zijn jullie en wat houd ik zielsveel van jullie! Daar op die snelweg bij Apeldoorn was het mijn liefde voor jullie

dat de aanzet vormde, die uiteindelijk geleid heeft tot dit onderzoekstraject. Ik kon toen niet overzien wat dit allemaal met zich mee ging brengen. De onvoorwaardelijke liefde voor en van jullie vormde de inspiratiebron voor dit onderzoek en de motor om dit door te zetten om tot een geslaagd eind te brengen. Het is eigenlijk best wel wrang dat we daardoor ook juist nogal wat momenten met elkaar gemist hebben, omdat ik zo nodig weer hiervoor aan het werk moest. Het is wrang om enerzijds zo bezig te zijn met het onderwerp van gehechtheid, en anderzijds mijn eigen kinderen te horen zeggen: "Papa, ik mis je". Ik heb dat als de meest pijnlijke kant van het onderzoek ervaren: ik had graag meer momenten met jullie willen delen. Ondanks die gemiste momenten hebben we gelukkig ook onze momenten van samenzijn genomen, zoals elke avond voor het slapen gaan het voorlezen uit de zevendelige serie van Harry Potter. De wederzijdse liefde en verbondenheid laat zich ook zien in de edelsteen die jullie samen voor mij van jullie vakantiegeld kochten, en die ik sindsdien elke dag bij mij draag.

Lieve, lieve Willemijn en Maarten, Jullie zijn zo verschillend, en toch samen zo één. Wat is het toch mooi om getuige te mogen zijn (en bij tijd en wijle iets te sturen) van jullie opgroeien van die mooiste, maar ook kwetsbare baby tot eigen persoonlijkheden. En wat prijs ik mijzelf gelukkig dat jullie mij deelgenoot maken van jullie observatievermogen, jullie sensitiviteit, jullie wijze woorden, jullie zorgzaamheid, gevoel voor sociale rechtvaardigheid en liefde. Wat ben ik trots op jullie! Willemijn, wat is het heerlijk om met mijn dochter jouw belevingen, visie en gevoelens over het leven te kunnen en mogen bespreken. Dank hiervoor. Maarten, wat is het heerlijk om met mijn zoon de belangstelling voor natuur, scouting en 'weetjes' te mogen delen. Dank hiervoor.

En ja, Maarten, …. een dergelijk project zal ik niet meer aannemen; hand erop!

Curriculum Vitae

Hans Giltaij was born on July 25th, 1958 in Arnhem (The Netherlands). He finished his pre-university education at Christelijk Lyceum in Arnhem and Revius Lyceum in Doorn (1976). In the years 1976 - 1979 he went to the Pedagogische Academie Jan van Nassau in Utrecht to become teacher for primary education. After graduation Hans did his military service, where he was trained to become an operation-room assistant and did his clinical training in Leyenburg Hospital in The Hague (The Netherlands). In 1981 Hans started the study Special Education (Orthopedagogiek) at the University Utrecht and graduated in 1988.

During this study Hans worked first as teacher at the school of Beukenrode in Doorn, an observation-institute for adolescent boys with severe behavioral problems, than as teacher at the primary school Lage Engh in Bunnik. From 1989 till 1993 Hans worked as sociotherapist in the Triangel in Amsterdam. The Triangel was an institute where multi-problem families (parents, children, even house pets) lived 24 hours a day in therapeutic family groups for a period of 9-12 months. He worked with highly complex family systems, with child maltreatment and family violence, and with attachment problems. The special interest for the development and position of the child in this complex context motivated Hans to study for child and adolescent psychotherapist in Utrecht (1991-1998). He worked as a junior child and adolescent psychotherapist in Laanzicht (The Hague), a mental health clinic for adolescents with severe psychiatric disorders (1993) and RIAGG Westelijk Utrecht (Utrecht), a mental health care institute for assessment and treatment of children and young adults with psychological, psychiatric and behavior disorders (1994-1998).

The interest and affinity for more complexity in psychopathology and the dual individual as well as system-orientated approach brought Hans to the Bleskolk (SKJPON) in Almelo, a mental health clinic for children and adolescents, where he worked as child and adolescent psychotherapist, especially for the division adolescents (1997-2005). Leading psychiatrists and psychologists in this clinic had special interest in attachment as an important keystone for the developmental pathway of the child. The complexity of the pathology urged sometimes for more (at that time) experimental treatments of these mostly multi-traumatized children with disturbed attachment relations. There was support for treatments as EMDR, Parent-child treatment, attachment-focused phase therapy, Mentalization Based Therapy (MBT). The first experiences with writing publications enhanced case studies of these treatments (2004-2006).

Since 1999 Hans combined this work with a new challenging function and a new challenging special interest population: he became senior psychotherapist and head of the department Psychotherapy (mental health care) in Bartiméus, a large organization for care, work, and education for people with visual or multiple (visual-and-cognitive) disability. Diagnosing psychiatric pathology as comorbid to the visual and/or intellectual disability is very challenging and has been an important focus prior to treatment. Together with his colleagues he developed this experimental department into the first, and until now only, officially acknowledged and qualified mental health care center for people with visual and/or intellectual disability. An important focus of treatment of this mental health care center has been attachment disorders. The importance of assessment of attachment related disorders and the lack of knowledge was the start of his PhD study in 2005.

Apart from the clinical and research work Hans is since 2005 freelance lecturer and trainer at the post-academic educational center RINO Group and supervisor (Vereniging Kinder- en Jeugdpsychotherapie, Nederlandse Vereniging voor Psychotherapie) for psychologists and psychotherapists.

Hans is registered in the Dutch Government Register (BIG) for professionals as Mental Health Care Psychologist, as Mental Health Care Psychologist-Specialist, and as Psychotherapist. In the Dutch Institute of Psychologists (NIP) he is registered as Child- and Youth Psychologist-Specialist. He is also a registered EMDR-therapist.

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