Chapter 5

General Discussion and Summary

This chapter is partly published as
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 effect study 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; Einfield, 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.

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<thead>
<tr>
<th>Study</th>
<th>Study group</th>
<th>Focus</th>
<th>Sample</th>
<th>Used instruments</th>
<th>Key findings</th>
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<tbody>
<tr>
<td>Smyke, Dumitrescu, &amp; Zeanah (2002)</td>
<td>Severely deprived, institutionalized children (BEIP project) (age 1-7 years)</td>
<td>Prevalence of signs of RAD or DSED</td>
<td>N = 94 children: Standard Unit (n = 32), Pilot Unit (n = 29), Never-institutionalized children (n = 33)</td>
<td>Care history: <strong>No Measure</strong>&lt;br&gt;Interview: Disturbances of Attachment Interview (DAI)&lt;br&gt;Observation: <strong>No Measure</strong>&lt;br&gt;Psychopathology: <strong>No Measure</strong></td>
<td>Standard Unit: RAD signs: 37%&lt;br&gt;DSED signs: 43%&lt;br&gt;Pilot Unit: RAD signs: 14%&lt;br&gt;DSED signs: 17%</td>
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<td>Zeanah, Scheeringa, Boris, Heller, Smyke, &amp; Trapani (2004)</td>
<td>Maltreated children (age 10-48 months) in foster care</td>
<td>Prevalence of signs of RAD or DSED</td>
<td>N = 94 children</td>
<td>Care history: maltreatment legally established&lt;br&gt;Interview: Disturbances of Attachment Interview (DAI)&lt;br&gt;Observation: <strong>No Measure</strong>&lt;br&gt;Psychopathology: <strong>No Measure</strong></td>
<td>RAD signs: 45%&lt;br&gt;DSED signs: 37%</td>
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<td>Zeanah, Smyke, Koga, Carlson, and the BEIP Core Group (2005)</td>
<td>Severely deprived, institutionalized children (BEIP project) (age 12-31 months)</td>
<td>Differences of signs of RAD and DSED in Institutionalized and never-institutionalized children</td>
<td>N = 145 children: Institutionalized children (n = 95) and never-institutionalized children (n = 50)</td>
<td>Care history: Observational Record of the Caregiving Environment (ORCE)&lt;br&gt;Interview: Disturbances of Attachment Interview (DAI)&lt;br&gt;Observation: Strange Situation Procedure (SSP)&lt;br&gt;Psychopathology: Infant Toddler Social Emotional Assessment (ITSEA)</td>
<td>Significant differences between institutionalized and never-institutionalized children for RAD (p&lt;.001) and for DSED (p&lt;.01)</td>
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| 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% |
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% |
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| 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 | Characterization 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% |
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| Vervoort, de Schipper, Bosmans, & Verschueren (2013) | Children (age 6-10 years) from schools for special education                  | Examination psychometric properties of the Relationship Problems Questionnaire (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% |
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| 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 |
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
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 theAUTI-R(Van Berckelaer-Onnes & Hoekman, 1991) or theAutism Diagnostic Observation Scale(ADOS-2)(DeBildt, 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 theClinical 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 theList 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, theDiagnostic 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.
References


Derde Proef