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 IJzendoorn, & Van Berkelaeer-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)

<table>
<thead>
<tr>
<th>Code</th>
<th>Description</th>
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<tr>
<td>313.89</td>
<td>Reactive Attachment Disorder</td>
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<tr>
<td>A)</td>
<td>A consistent pattern of inhibited, emotionally withdrawn behaviour toward</td>
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<td>adult caregivers, manifested by both of the following:</td>
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<td>1) The child rarely or minimally seeks comfort when distressed.</td>
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<td>2) The child rarely or minimally responds to comfort when distressed.</td>
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<td>B)</td>
<td>A persistent social and emotional disturbance characterized by at least two of</td>
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<td>the following:</td>
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<td></td>
<td>1) Minimal social and emotional responsiveness to others.</td>
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<td>2) Limited positive affect.</td>
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<td>3) Episodes of unexplained irritability, sadness, or fearfulness that are</td>
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<td></td>
<td>evident even during nonthreatening interactions with adult caregivers.</td>
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<td>C)</td>
<td>The child has experienced a pattern of extremes of insufficient care as</td>
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<td>evidenced by at least one of the following:</td>
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<td></td>
<td>1) Social neglect or deprivation in the form of persistent lack of having</td>
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<td>basic emotional needs for comfort, stimulation, and affection met by</td>
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<td>caregiving adults.</td>
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<td>2) Repeated changes of primary caregivers that limit opportunities to form</td>
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<td>stable attachments (e.g., frequent changes in foster care).</td>
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<td>3) Rearing in unusual settings that severely limit opportunities to form</td>
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<td>selective attachments (e.g., institutions with high child-to-caregiver</td>
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<td>ratios).</td>
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<td>D)</td>
<td>The care in Criterion C is presumed to be responsible for the disturbed</td>
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<td>behaviour in Criterion A (e.g., the disturbances in Criterion A began</td>
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<td>following the lack of adequate care in Criterion C).</td>
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<td>E)</td>
<td>The criteria are not met for autism spectrum disorder.</td>
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<td>F)</td>
<td>The disturbance is evident before the age 5 years.</td>
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<td>G)</td>
<td>The child has a developmental age of at least 9 months.</td>
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</table>
Table 2  DSM-5 Criteria for Disinhibited Social Engagement Disorder (APA, 2013)

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 intrerrater 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.
References


