MENTAL HEALTH IN YOUNG CHILDREN REFERRED FOR TREATMENT

Frederike Scheper

TEMPERAMENT, DISTURBED ATTACHMENT BEHAVIOR AND COURSE

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The studies described in this thesis were performed at the VU University Medical Center, Department of Child and Adolescent Psychiatry, Amsterdam, the Netherlands.

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Mental health in young children referred for treatment

Temperament, disturbed attachment behavior and course

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SUMMARY

This thesis responded to the growing interest in early childhood mental health care (Dougherty, Leppert, et al., 2015; Shonkoff, 2003) by contributing to closing the gap in knowledge about assessment and treatment of mental health problems between young children and older children (Angold & Egger, 2007).

Research in the field of developmental psychopathology has provided evidence for specific temperament traits and child maltreatment being associated with mental health problems in children (Barnett et al., 1993; Caspi et al., 1995; Frick, 2004; Nigg, 2006). Temperament traits have been defined as heritable characteristics, expressed as individually based differences in emotional reactivity and self-regulation (Rothbart & Derryberry, 1981). In young children, three broad temperament dimensions have been identified by Rothbart and colleagues to be relatively stable from age three: negative affectivity, extraversion/surgency, and effortful control (Rothbart et al., 2001). Negative affectivity and extraversion/surgency represent the tendency of children to react with either negative or positive emotions to daily situations. Children with high negative affectivity tend to react to situations with fear, sadness, anger/frustration, and distress and are not easily soothed (low soothability). Children with high extraversion/surgency are inclined to react with smiling/laughter, pleasure, as well as impulsivity, activity, and approaching. Effortful control, also referred to as self-regulation, represents the ability to regulate attention and activate or inhibit the emotional and behavioral responses. The studies that addressed the relation between child temperament and psychopathology have mostly been done in general population samples. Studying the relation between specific temperament traits and mental health problems in young children referred for treatment, as well as follow-up after treatment, likely contributes to the fine-tuning of diagnostic and treatment trajectories in early childhood.

Serious aberrant attachment and social behaviors in children have been found in children who were exposed to social deprivation, maltreatment and neglect (Gleason, Fox, et al., 2011; Tizard & Rees, 1975; Zeanah et al., 2016). Inhibited attachment behavior is characterized by a lack of preference for a specific caregiver, unresponsiveness to primary caregivers, and negative emotionality. Disinhibited social engagement behavior is characterized by

indiscriminate social behavior, failing to maintain proximity to the caregiver in new situations, and willingness to go off with strangers. Children typically show inappropriate approach by verbally and physically interacting with unfamiliar adults. Evidence supporting clinical relevance of the symptoms of Reactive Attachment Disorder (RAD) and Disinhibited Social Engagement Disorder (DSED) pertains mostly children reared in institutional care. However, child maltreatment and neglect also occur in children reared with biological parents. Moreover, child maltreatment occurs more often in children referred for mental health care than in community populations (Lau & Weisz, 2003; Walrath et al., 2006). While there is emerging attention for the effect of trauma, including child maltreatment, and attachment problems in young children, there is little knowledge about inhibited social engagement behavior and disinhibited social engagement behavior in home-reared children.

This thesis aimed to extend our knowledge about mental health problems in referred young children and its course during and after treatment, with a focus on child temperament and disturbed attachment behavior. The three main research questions were: 1. What is the relation between child temperament traits and mental health problems in young children referred to mental health care? 2. Does inhibited attachment behavior and disinhibited social engagement behavior occur in referred home-reared young children, and what is the clinical relevance of these specific behaviors? 3. What is the course of mental health problems after treatment in early childhood, and are child temperament traits and parental psychopathology related to the course?

Study design and population

This dissertation is the result of a research project, which assessed referred young children and their parents and evaluated the outpatient treatment offered at MOC 't Kabouterhuis, a specialized psycho-medical health center for children (0 through 7 years) with developmental and behavioral problems in the region of Amsterdam, the Netherlands. Children were included in the studies after referral for social-emotional- and/or behavioral problems. Referral to the center was conducted by general practitioners, other medical specialists and child workers of (public) health care centers or child protection services. Parent training was offered for all children, including improved access to care via home visitations.

Video feedback could be used by trained specialists addressing sensitivity in parenting and attunement to the needs of the child. At least one home visit during the treatment trajectory was planned, but more home visits were possible when parents could not easily attend the clinic. Depending on the developmental needs, children were offered treatment in an individual or group setting. Individual treatment consisted of weekly sessions with the parent(s) at the clinic or at home. Day treatment in a group setting for children could be one to four days a week. Multimodal treatment addressed the developmental needs through multiple modalities, such as behavioral training, social-skills training, medication, (parent-infant or child) psychotherapy, physiotherapy, and speech therapy. Interventions in day treatment included social skills training to improve functioning within a peer group and activities to stimulate development of speech/language, motor and cognitive skills.

For the study addressing aim 1 (described in Chapter 2), young children referred for treatment at MOC 't Kabouterhuis were compared to a general population sample derived from a study on child temperament (Majdandžić & van den Boom, 2007b). For the study addressing aim 2 (described in Chapter 3), data from young children referred to MOC 't Kabouterhuis were combined and compared with data from children who were referred for treatment at de Bascule, an academic center for child- and adolescent psychiatry. For the study addressing aim 2 and 3 (described in Chapters 4 and 5), longitudinal data of young children referred for treatment at MOC 't Kabouterhuis were included to answer questions about the course of mental health problems. The children from MOC 't Kabouterhuis were examined three times: at the start of treatment, at the end of treatment, and on average three years after treatment (see Appendix 1 for the study outline with instruments).

Summary of findings

In **Chapter 2**, associations between child temperament traits and mental health problems were investigated in 216 clinically referred children, aged 3.0 - 7.3 years. A subsample of 115 children was compared with an age and gender matched general population sample of 115 children (see Table 1 for the main findings). Higher levels of negative affectivity and lower levels of effortful control (self-regulation) in child temperament each were associated with more internalizing and externalizing problems, with associations equal in magnitude for referred and general population children. Results showed a difference between referred children and general population children in levels of emotional reactivity and self-regulation. The referred children showed less smiling in response to daily situations and more difficulty to recover from distress, demonstrated by lower levels of soothability, than the general population children. Also, referred children had more problems in their ability to regulate their emotions and behavior, represented by lower levels of effortful control, than the general population children. Furthermore, there was a difference between referred and non-referred children in the relation between extraversion/surgency and externalizing problem behavior. In referred children, impulsivity was more strongly related to externalizing problems compared to general population children.

In referred children, we found specific patterns concerning the relations between fine-grained traits of surgency/extraversion with internalizing and externalizing problem behavior. More shyness and less smiling/laughter were related to internalizing problem behavior and more impulsivity was specifically related to externalizing problem behavior. Low levels of soothability, low levels of inhibitory control and high levels of frustration were associated with clinically relevant levels of co-occurring internalizing and externalizing problem behavior. Soothability was the only trait that was negatively associated with more internalizing and externalizing problems when viewed separately, as well as with (sub)clinical levels of comorbid internalizing and externalizing problem behavior.

We concluded that more negative affectivity and less effortful control might well be temperament traits that vary across a continuum and in extreme levels represent psychopathology in young children. At the same time, children with a tendency to react more impulsively to daily situations might be vulnerable to develop externalizing problems. Furthermore, it was concluded that low

soothability in children could be an important temperament trait in relation to internalizing and externalizing problems. It was proposed that assessment of temperament in clinically referred children could be of help when customizing diagnostic procedures and tailoring treatment interventions in early childhood.

Chapter 3 addressed symptoms of attachment and social engagement disorders in 200 children, aged 2.0 - 7.9 years, referred for treatment of emotional or behavioral problems (see Table 1 for the main findings). The symptoms were examined in relation to child maltreatment, parenting stress, and child internalizing and externalizing behaviors. Home-reared children (*n* = 141) were compared to foster children (*n* = 59). Emotional or physical maltreatment or neglect was found in 54% of the clinical records of the home-reared children. While this shows high prevalence of exposure to pathogenic caregiving, foster children had been exposed even more often to such pathogenic environments (90%). Also, inhibited attachment behavior was significantly less prevalent in home-reared children (9%) than in foster children (27%). A combination of inhibited attachment behavior and maltreatment/neglect, suggesting a possible RAD diagnosis, was found in 6% (*n* = 9) of the home-reared children. Disinhibited social engagement behavior was found to be similarly prevalent in clinically referred young home-reared children (42%) compared to children in treatment foster care (51%). However, only in half of the home-reared children with disinhibited social engagement behavior we found exposure to child maltreatment or neglect reported in clinical records. Disinhibited social engagement behavior in combination with maltreatment/neglect, suggesting a possible diagnosis of DSED, was found in 21% (*n* = 29). For home-reared and foster children, no unique covariance was found between inhibited attachment nor disinhibited social engagement behavior and child maltreatment/neglect, meaning that parent reported symptoms of RAD and DSED were not unique to children with documented exposure to patterns of inadequate caregiving.

More inhibited attachment behavior was associated with clinical levels of internalizing and externalizing problem behavior, but the associations were only significant for the home-reared children. More disinhibited social engagement behavior was associated with clinical levels of externalizing problem behavior in referred home-reared children as well as in foster children, regardless of exposure to maltreatment. Also, when accounting for the influence of child internalizing and externalizing problem behavior on parenting stress, disinhibited social

engagement behavior was associated with higher levels of parenting stress.

The conclusion was that inhibited attachment and disinhibited social engagement behaviors in referred home-reared children have clinical significance, despite the unclear etiology of such behaviors. It was proposed to address the specific social aberrant behaviors in the diagnostic and treatment process in relation to child internalizing and externalizing problem behavior and parenting stress.

In **Chapter 4**, the course of disinhibited social engagement behavior was studied by interviewing parents of 124 preschool children, aged 1.9 – 5.9, from referral in early childhood until on average four years later (see Table 1 for the main findings). Disinhibited social engagement behavior at follow-up was assessed not only with a semi-structured interview with parents but also by observation during home assessment. In the sample participating in this follow-up study, 29% had maltreatment/neglect (including physical abuse). Neglect or emotional maltreatment was found in 25% of the children (Chapter 4). Results showed relative persistence of parent reported disinhibited social engagement behavior after treatment (57%), and convergence between measurement methods including observational rating in middle childhood. The course of disinhibited social engagement behavior was neither associated with neglect/emotional maltreatment nor with children's level of self-regulation (effortful control). Results did not show an association between disinhibited social engagement behavior and Autism Spectrum Disorder (ASD), based on a semi-structured interview. Results showed that disinhibited social engagement behavior at preschool age was associated with a psychiatric disorder with externalizing behavior (ADHD and/or ODD) four years later. Also, children with persistent disinhibited social engagement behavior had a higher prevalence of ADHD (with or without ODD) than children without disinhibited social engagement behavior.

We concluded that the longitudinal findings suggest clinical relevance of disinhibited social engagement behavior in referred home-reared children by showing relative persistence and linkages with externalizing behaviors over a four-year-period. Assessment and treatment of home-reared children should address these specific aberrant social behaviors in children in relation to their caregivers.

In **Chapter 5**, the course of mental health problems was examined during and after early childhood treatment (see Table 1 for the main findings). We assessed internalizing and externalizing problem behavior in referred preschool children aged 1.8 – 6.1 from start of treatment (M = 4.0 years, SD = .93) to the end (n = 246, M = 4.9 years, SD = .92), and on average three years after treatment (n = 164, M = 8.0 years, SD = 1.4). All children had been referred with social-emotional- or behavioral problems. Also, 59% had reports of language/speech problems in their referral files. All parents participating in our follow-up study received parent guidance/training and most children (82%) participated in day treatment. Parents reported clinically relevant levels of internalizing and/or externalizing problem behavior in 70% of the children, with similar levels of internalizing and externalizing behaviors. At the end of treatment, parents reported internalizing and/or externalizing problem behavior in 48% of the children and at follow-up in 52%. Co-occurring internalizing and externalizing problem behavior at the start of treatment was found in 34%, at the end of treatment in 25% and at follow-up in 23% of the children. Results showed that child problem behavior significantly improved after treatment in early childhood and retained the same level at follow-up on average three years later. At the same time, 72% fulfilled criteria for one or more psychiatric disorders (without including serious distress as criterion). More improvement of child problem behavior during treatment was associated with a better quality of life at follow-up. High levels of child problem behavior at the start of treatment was associated with more child problem behavior over time, after treatment and at follow-up. Furthermore, results showed that low soothability in children at the start of treatment was associated with more internalizing problem behavior over time, also when accounting for parental depression and hostility. Also, difficulties in regulating emotions and behaviors (low effortful control) and a tendency to respond with frustration to daily situations in children was associated with more externalizing problem behavior over time. More symptoms of parental depression were significantly associated with more internalizing problem behavior in children over time after treatment. Because specific temperament traits and parental depression were associated with child internalizing and externalizing behavior over time, it was proposed to further fine-tune interventions attending child temperament as well as parental psychopathology.

Table 1. Summary of main findings

Aim 1: What is the relation between child temperament traits and mental health problems in referred young children?

Chapter 2:

- Referred children had lower levels of effortful control, soothability and smiling/laughter than general population children.
- Impulsivity was more strongly associated with externalizing problems in referred children than in general population children.
- Lower levels of soothability, inhibitory control and higher levels of frustration were associated with clinical levels of co-occurring internalizing and externalizing problems in referred children.

Aim 2: Does disturbed attachment behavior occur in referred home-reared young children, and what is the potential clinical relevance?

Chapter 3:

- Symptoms of disorders of attachment (IAB) and social engagement (DSEB) were found in clinically referred home-reared children; DSEB as prevalent as in children in treatment foster care.
- There was no association between IAB/DSEB and a history of child maltreatment or neglect.
- IAB and DSEB were both associated with clinical levels of child problem behavior in home-reared children and DSEB was also associated with more parenting stress.

Chapter 4:

- Persistence of DSEB was found in 57% of referred home-reared children examined on average four years later.
- Course of DSEB was not related to neglect/emotional maltreatment, effortful control or ASD, but was associated with ADHD/ODD.

Aim 3: What is the course of mental health problems after treatment in early childhood, and are child temperament traits and parental psychopathology related to the course?

Chapter 5:

- There was a reduction in mental health problems directly after multimodal treatment in early childhood, which remained at the same level on average three years later.
- More improvement in child problem behavior during treatment was associated with a better quality of life at follow-up.
- On average three years after treatment, 34% had clinically relevant emotional- and behavioral problems and 72% psychiatric disorders.
- More problem behavior at the start of treatment, difficulties in emotional reactivity and effortful control, and parental depression were associated with more child problem behavior over time.

IAB: inhibited attachment behavior

DSEB: disinhibited social engagement behavior

ASD: autism spectrum disorder

ADHD: attention-deficit/hyperactivity disorder

ODD: oppositional defiant disorder

GENERAL DISCUSSION

Child temperament traits and mental health problems (aim 1)

Temperament traits, defined as constitutionally based individual differences in emotional reactivity and self-regulation, have been proposed to vary across a continuum with extreme levels representing psychopathology, also described as the *spectrum model* (Krueger & Tackett, 2003). Consistent with the spectrum model, negative affectivity and less effortful control each were associated with more internalizing and externalizing problems with associations equal in magnitude for referred and general population children (Chapter 2). Former research also found evidence for a *vulnerability model,* in which temperament traits either predispose for or protect against psychopathology in certain circumstances (Belsky & Pluess, 2009). No evidence for a vulnerability of low effortful control was found in our study addressing the course of disinhibited social engagement behavior. Effortful control did not moderate the association between neglect/emotional maltreatment and disinhibited social engagement behavior (Chapter 4). Our finding that impulsivity in referred children was more strongly related to externalizing problem behavior, compared to general population children, implies that referred children who are impulsive in their reactions are vulnerable for externalizing behaviors. When children tend to react impulsively in daily situations, parents of referred children might respond more negatively than parents of general population children, adding to the risk of developing externalizing problem behavior. It is also possible that parents perceive temperament traits of referred children differently than parents of non-referred children. Furthermore, psychopathology could change the expression of temperament in children (Zentner & Shiner, 2012). Trait-by-trait moderation was found in other studies, with evidence that the relation between negative affectivity and externalizing behavior is dependent on the level of effortful control (Gartstein et al., 2012; Murray & Kochanska, 2002). However, we did not find evidence for this trait-by-trait moderation in our study (Chapter 2).

Regarding fine-grained temperament traits in relation to mental health problems in clinically referred young children, shyness and low levels of smiling/laughter were associated with internalizing problem behavior and impulsivity was associated with externalizing problem behavior. These findings were in line with expectations because these traits represent internally focused behavior (i.e.,

shyness and smiling/laughter) and externally focused behavior (impulsivity).

High frustration was associated with clinical levels of concurrent internalizing and externalizing problem behavior as well as with poor outcome on externalizing behavior over time after treatment, after controlling for parental depression and hostility (Chapters 2 and 5). This finding is in line with research addressing the importance of preschool irritability in children referred for mental health problems (Dougherty, Smith, et al., 2015). Preschool irritability, defined as high levels of frustration in children and characterized by anger and temper outbursts, has been found related to severe mental health problems and poor outcome later in life (Birmaher, 2016; Dougherty, Smith, et al., 2015; Vidal-Ribas et al., 2016). Chronic irritability during early childhood in general population children was associated with internalizing as well as externalizing disorders in childhood (Dougherty, Smith, et al., 2015).

In our studies, we also found evidence for the importance of 'soothability' in clinically referred young children. Soothability is a feature depending on the child's ability to regulate emotions after distress and how the child reacts to soothing by its parent/caregiver. Less soothability was associated with more internalizing and externalizing problems when viewed separately, as well as with (sub)clinical levels of concurrent internalizing and externalizing problem behavior at referral (Chapter 2). Furthermore, children who were more difficult to sooth were more prone to retain high levels of internalizing behavior over time (Chapter 5). Rothbart (2011) has described how important soothability is for the development of self-regulation.

The findings regarding low levels of effortful control (self-control) in referred young children add to research suggesting that mental health problems in young children are related to a constitutionally based impaired ability to voluntarily regulate behavior and emotions (Caspi et al., 2014; Gartstein & Fagot, 2003; Olino et al., 2014; Zentner & Shiner, 2012). Results showed that children with difficulties in regulating their emotions and behavior (low effortful control) had severe internalizing and externalizing problems at the start of treatment, and were prone to retain high levels of externalizing problems over time (Chapter 2 and 5). These findings are in line with other studies showing that poor self-regulation is related to various types of psychopathology, and it has been proposed that poor self-regulation is an early developmental feature of general

psychopathology (Caspi et al., 2014; Olino et al., 2014). There is growing evidence for a 'general psychopathology factor' or 'common factor' that is associated with a wide range of mental health problems and dysfunction in life (Caspi et al., 2014; Olino et al., 2014). Our findings described above also suggest that high frustration and low soothability are temperament traits that could be important in relation to general psychopathology and dysfunction, because they were both associated with internalizing as well as externalizing problems and the course of mental health problems over time. The general psychopathology factor is suggested to explain the co-occurrence of different types of mental health problems and change in type of mental health problems over time (heterotypical continuity) (Basten et al., 2016; Caspi et al., 2014). Furthermore, the existence of such a factor may explain why finding specific risk factors or etiological variables for specific mental disorders is often difficult (Caspi et al., 2014). For instance, child maltreatment is not related to specific mental disorders but is related to general psychopathology with various types of mental health problems (Caspi et al., 2014).

Symptoms of disorders of attachment and social engagement (aim 2)

In young children, examined after referral for treatment of emotional and behavioral problems, we found a high prevalence of maltreatment and/or neglect (54%, Chapter 3) when compared to the 3% found in a general population study in the Netherlands (Euser, van IJzendoorn, Prinzie, & Bakermans-Kranenburg, 2010). This is in line with research showing that child maltreatment/neglect is more prevalent in children referred to mental health care than in community populations and associated with psychopathology and dysfunction in life (Lau & Weisz, 2003; Walrath et al., 2006). As clinical records are also vulnerable to underreporting, the prevalence of maltreatment/neglect underlines the high risk of inadequate caregiving in these young referred children. The studies in this thesis (Chapter 3 and 4) have added knowledge to the literature about inhibited attachment behavior and disinhibited social engagement behavior in relation to maltreatment/neglect, parenting stress, and mental health problems in referred young home-reared children.

In Chapter 3, we reported that inhibited attachment behavior was found to be

less prevalent in referred home-reared children (9%) than in foster children (27%). Disinhibited social engagement behavior was found in 42% of the referred home-reared children, which was similar when compared to children in treatment foster care (Chapter 3). No full diagnostic clinical assessment of RAD and DSED was undertaken, which would have required information from multiple informants and observation sessions at the two measurement occasions (Zeanah et al., 2016). However, results from our cross-sectional and follow-up study (Chapter 3 and 4) showed acceptable reliability and construct validity of the parent report (a semi-structured interview with parents), as well as significant associations between the parent report and observation measures of disinhibited social engagement behavior at follow-up. Therefore, the results do suggest that both inhibited attachment behavior and disinhibited social engagement behavior occur in referred home-reared children.

The finding that inhibited attachment behavior was less prevalent compared with foster children was expected, because children in foster care had been separated from their parents and were therefore more prone to develop attachment problems. Although we found no direct association with maltreatment/neglect, 75% of the home-reared children whose parents reported inhibited attachment behavior in a semi-structured interview had maltreatment or neglect reported in clinical records. Therefore, a diagnosis of RAD could be considered in 6% (n = 9) of the clinically referred home-reared children, which is higher than the 1.4% RAD/DSED found in a large general population sample of 1646 children (Minnis et al., 2013). The finding that inhibited attachment behavior was associated with clinical levels of internalizing problem behavior in referred home-reared children, is in line with earlier studies in institutionalized children (Gleason, Fox, et al., 2011). In addition, an association with clinical levels of externalizing problem behavior was found in the home-reared children. Although the sample size of home-reared children with inhibited attachment behavior was small (n = 12), and findings should be replicated in other samples with observation measures, results do indicate occurrence and clinical relevance of inhibited attachment behavior in referred children.

Regarding disinhibited social engagement behavior, half of the home-reared children with this type of aberrant social behavior had reports of maltreatment or neglect in their patient records. Therefore, a diagnosis of DSED could be considered in 21%, which is less than in our foster care sample but more than

the 1-2% found in the few studies examining community samples (Gleason, Zamfirescu, et al., 2011; Minnis et al., 2013). The relative persistence of disinhibited social engagement behavior (57%, Chapter 4) is in line with the rate of persistence found in (post) institutionalized children (54%) (Rutter et al., 2007). However, it is much higher than in a group of adopted children (6.5%) who did not experience severe deprivation (Rutter et al., 2007). Also, we found no association between (the course of) disinhibited social engagement behavior and neglect/emotional maltreatment of the children. An important question is therefore whether parent reported disinhibited social engagement behavior represents something different in home-reared children than in (post) institutionalized children, especially when records reveal no exposure to neglect or emotional maltreatment.

A direct association between maltreatment/neglect and disinhibited social engagement behavior was not found in our sample. Still, the high percentages of maltreatment/neglect reported in the records of the children (Chapters 3 and 4) suggest that these children are at risk for adverse caregiving. Also, clinical records are vulnerable to underreporting of emotional maltreatment and neglect. Another possibility is that other adverse childrearing factors are related to disinhibited social engagement behavior in referred home-reared children. There is some evidence showing that disruptions in caregiving and disruptive emotional interactions between parent and child are associated with disinhibited social engagement behavior in children. In an earlier study, disinhibited social engagement behavior in non-institutionalized adopted children was not associated with maltreatment, but was associated with out-of-home placement between the age of 7 and 24 months (Kay et al., 2016). In yet another study, disinhibited social engagement behavior was associated with a history of psychiatric disturbance in mothers of home-reared children (Lyons-Ruth et al., 2009; Zeanah et al., 2004). Disruptive emotional interactions between a primary caregiver and the infant are known to influence the development of the child (Lyons-Ruth et al., 2009), and these disruptive interactions are not always captured within the concept of child maltreatment. We found that disinhibited social engagement behavior in children was associated with more parenting stress (Chapter 3). It might well be possible that high parenting stress concurs with atypical parenting and parent-relation problems that could underlie disinhibited social engagement behavior found in these young children.

There is evidence that the interplay between biological and environmental factors

influences the onset and course of disinhibited social engagement behavior in children (Kay et al., 2016; Minnis et al., 2007). Possibly, children with a certain biological make-up are more prone to develop symptoms of disorders of attachment and social engagement in reaction to inadequate caregiving (Zeanah & Fox, 2004). An important point to address is the role of child temperament. Low effortful control may be an indicator of early exposure to insufficient care due to biological programming but effortful control may also influence children's response to caregiving (Bakermans-Kranenburg et al., 2011; Zeanah & Fox, 2004). Lower levels of self-control in child temperament were found to be associated with more disinhibited social engagement behavior in non-maltreated home-reared children (Pears, Bruce, et al., 2010). We also found that referred home-reared children with disinhibited social engagement behavior had lower effortful control than children without symptoms of disorders of attachment or social engagement (Scheper, Jansen, de Vries, Doreleijers, & Schuengel, 2016), but we did not replicate this finding in our follow-up sample (Chapter 4). As mentioned earlier, low effortful control as well as child maltreatment has been related to general psychopathology. It is therefore not surprising that disinhibited social engagement behavior has also been associated with other mental health problems and disorders.

Our study showed no association between disinhibited social engagement behavior and ASD. This finding is in line with other research, suggesting that the social aberrant behaviors of autism and DSED can be discriminated with standardized assessment procedures (Giltaij et al., 2015). Results of our study did show that children with persistent disinhibited social engagement behavior over four years' time had a higher prevalence of ADHD than children without disinhibited social engagement behavior. Also, parent reported disinhibited social engagement behavior in early childhood was associated with ADHD/ODD at follow-up, but not with ADHD alone. This suggests that disinhibited social engagement behavior in young children is mainly associated with mixed externalizing behavior over time. These results are in line with earlier research in home-reared infants, showing that disinhibited social engagement behavior predicted hostile and hyperactive behavior at the age of five (Lyons-Ruth et al., 2009). Although there is a possibility that parent-reported disinhibited social engagement behavior was miss-classified in some of the children with ADHD, other studies have shown that symptoms of DSED and ADHD can concur

(Gleason, Fox, et al., 2011; Pritchett et al., 2013). Also, symptoms of DSED were found to be qualitatively distinct from ADHD and conduct problems, supporting the clinical relevance of disinhibited social engagement behavior (Gleason, Fox, et al., 2011; Minnis et al., 2007).

In conclusion, the specific pathogenic pathway(s) to disinhibited social engagement behavior in home-reared children are still unclear. Although findings should be replicated in home-reared children, including observation of children's behavior as well as atypical parenting and specific information about the caregiving history, results do indicate occurrence and clinical relevance of both inhibited attachment behavior and disinhibited social engagement behavior in treatment referred young children.

Course of mental health problems after treatment at preschool age (aim 3)

Despite referral for treatment of behavioral and social-emotional problems, not all children were reported to have clinically relevant levels of child problem behavior at the start of treatment. This is in line with other multimodal preschool evaluation studies, in which clinically relevant levels were reported in 64 - 86% of the treatment-referred children (Martin et al., 2013; Muller et al., 2015). It is possible that some children were referred in an early stage, when mental health problems not yet reached clinically relevant levels. It is also possible that other caregivers (other family members, day care or teachers) recognized more problems than the primary caregiver who reported at the start of treatment. Furthermore, there could also be other problems that were more relevant for the parents than the internalizing and externalizing problem behaviors that were measured at the start of treatment.

After referral for early childhood treatment of social-emotional and behavioral problems, we found an improvement of internalizing and externalizing problem behavior (Chapter 5). Moreover, the longitudinal findings suggest stability of mental health improvement and no deterioration until three years after treatment. Because no control group was included, we cannot report on effectiveness of early childhood multimodal treatment. There is growing evidence for the effectiveness of parent training and multimodal day treatment from studies

showing that early childhood treatment is beneficial for preschool children with externalizing problem behavior (Piquero et al., 2016; Posthumus et al., 2012; Tse, 2006; van der Veen-Mulders, Hoekstra, Nauta, & van den Hoofdakker, 2017). Also, there are other studies which found a reduction of internalizing problem behavior after multimodal early childhood treatment (Martin et al., 2013; Muller et al., 2015).

Our findings showed that improvement of child problem behavior during treatment was associated with a higher quality of life for children on average three years after treatment. This could be an important finding, suggesting that when parents perceive improvement of child problem behavior after early childhood treatment, they also report better mental health functioning later in childhood. Other studies already suggested the importance of early childhood intervention to improve functioning in later life (Campbell et al., 2014; Tremblay et al., 2004).

However, the findings do not imply that there is always remission of psychopathology after treatment. For some children, there might be remission of psychopathology, but the results also revealed that, in spite of an overall decrease in problem behavior, 48% of the children still showed clinically relevant internalizing or externalizing problem behavior after treatment. Also, on average three years after treatment still 72% fulfilled criteria for one or more psychiatric disorders, although serious distress was not included in the prevalence at follow-up. Preschool children referred with emotional- and behavioral problems are a vulnerable group of which many children likely need further mental health care (Visser et al., 2003). Still, the findings are promising and suggest that improvement of child problem behavior during early childhood supports better functioning in later childhood (Campbell et al., 2014; Shonkoff, 2003).

An important issue is to fine-tune the assessment and multimodal treatment in early childhood to further improve functioning of children. Because we did not examine the effect of treatment, it is unclear whether treatment would have been more effective when addressing specific child or parent related factors. However, we did find that specific temperament traits and symptoms of parental depression were associated with problem behavior over time.

There is much evidence for the relation between parental psychopathology

(mostly mothers' depression) and poor mental health outcome in children (Bufferd et al., 2014; Gartstein & Fagot, 2003; Mesman et al., 2009). We also found that parental depression was associated with more internalizing problem behavior in children over time (Chapter 5). The relation between parental depression and child mental health problems can be reciprocal. Parents with symptoms of depression could have more difficulty in parenting their child, which could enhance mental health problem in children, but mental health problems in children could also enhance the depressive feeling in parents (Gross, Shaw, & Moilanen, 2008).

We also found that high levels of frustration in children's temperament as well as low levels of effortful control at the start of early childhood treatment were associated with more externalizing behavior after treatment. As mentioned above on the relation between temperament traits and mental health problems, these findings are in line with previous research (Dougherty, Smith, et al., 2015; Eisenberg et al., 2009; Vidal-Ribas et al., 2016). The findings imply that not only effortful control, but also the level of frustration with regards to more irritability in children is important to address in early childhood treatment. Furthermore, we found that low soothability in preschool children was associated with more internalizing problem behavior after treatment. Possibly, soothability can be seen as a linking pin between the fields of child development, infant and early childhood mental health, and child psychiatry. Young children's ability to regulate emotions after distress and the response to soothing by parents/caregivers could be important in the development and course of various mental health problems and disorders.

Limitations and strengths

The findings in this thesis should be interpreted in light of study limitations. Important limitations to discuss are participant attrition, the heterogeneity of the study sample and of treatment modalities, and the lack of a control group in the longitudinal studies. In all studies, parents had to actively participate by providing information and giving consent that the results would be used for research purposes, which could have led to participation bias. The studies tried to address this by comparing basic descriptive characteristics of participants with characteristics of non-participants. Ethnicity seemed to be the most important factor resulting in bias; children from non-western ethnicity were less represented

in our studies then in the non-participants. Regarding heterogeneity, we examined children with various mental health problems. Also, parents and children received various modalities of early childhood treatment. Regarding study design, we could not include a non-referred control group in the studies on symptoms of disorders of attachment and social engagement and the course of mental health problems (Chapters 3, 4 and 5). The study described in Chapter 5 could therefore be limited by spontaneous improvement of mental health problems as well as regression to the mean. Also, the lack of a control group limited the possibility to control for other factors that could be related to the course of child mental health problems. On the other hand, a control group of non-referred children could be included in the study on associations between temperament traits and mental health problems (Chapter 2). An important strength of the current study is that clinical practice was evaluated concerning a broad range of mental health problems in children, including longitudinal outcome after early childhood treatment (Chapter 5).

Future research implications

Future research should focus on advancing clinical understanding and treatment of inhibited attachment and disinhibited social engagement behavior in young children. Because of the clinical relevance, including the relative persistence, more research is needed on understanding and treatment of disinhibited social engagement behavior in home-reared children. Evidence of earlier research suggested that a physical, rather than a nonphysical, type of disinhibited social engagement behavior should be understood as atypical behavior (Lawler, Hostinar, Mliner, & Gunnar, 2014). Still, it is unknown whether differentiating physical from nonphysical disinhibited behavior could be clinically relevant in referred home-reared children. Disinhibited social engagement behavior should be assessed through parent interviews as well as observational measures, which could fill the gap on validity evidence for observational measurement of disinhibited social engagement behavior in home-reared young children as well as children in middle childhood and beyond (Vervoort et al., 2013). Also, studies addressing detailed information about early caregiving disruptions, the quality of the parent-child relation, as well as biological and temperamental factors in home-reared referred and non-referred populations are needed for better understanding of disinhibited social engagement behavior. Furthermore, disinhibited behavior of parents and atypical parenting associated with trauma

and disorganized attachment (Lyons-Ruth et al., 2009) should be addressed in research for diagnostic as well as treatment perspectives. There are a few studies suggesting that interventions addressing social boundaries and self-regulation as well as training parents to manage their child's behavior could be beneficial (Dickmann & Allen, 2017; Zeanah & Gleason, 2015). However, research on treatment of disinhibited social engagement behavior particularly in home-reared children is still scarce.

Future research addressing outcome of early childhood treatment should take temperament traits into account, as well as parental depression. Developmental functions and characteristics as underlying processes in various types of mental health problems, across diagnostic boundaries, should be addressed (Basten et al., 2016; Olino et al., 2014). Furthermore, evidence for differential susceptibility indicates that research should take interaction effect into account, such as by examining the influence of child temperament traits for better and for worse (Belsky & Pluess, 2009). Regarding parental depression, it could be clinically relevant to examine indications for when to treat parental depression first, at the same time during early childhood treatment or after childhood treatment. When ethically feasible, outcome research of multimodal early childhood treatment should include a control group (such as a waiting list condition) and specifically register the modalities used in treatment. Multiple case studies with multiple assessments could also be helpful in answering specific questions about outcome of early childhood treatment of mental health problems.

Clinical implications

Regarding the findings of improvement after early childhood treatment (Chapter 5) and results of other studies suggesting the importance of intervention in early childhood to improve functioning in later life (Campbell et al., 2014; Tremblay et al., 2004), investing in this period of life is extremely important. Results from our studies on mental health problems in young referred children suggest fine-tuning of assessment and intervention in early childhood mental health care, with knowledge about children's temperament traits, disturbed attachment and social engagement behavior as well as parental psychopathology.

Assessment

Assessment of temperament in clinically referred children could be of help when customizing diagnostic procedures and tailoring treatment interventions in early childhood. Findings of our studies (Chapter 2 and 5) suggest that specific characteristics in children are related to emotional and behavioral problems. Extreme shyness and little smiling in reaction to daily situations should be addressed in early assessment of emotional problems by observations and talking to parents about these features. Impulsivity in reaction to daily situations as well as inhibitory control should be addressed in early assessment of behavioral problems. At the same time, negative emotionality should be addressed in prevention and early childhood treatment of various mental health problems (Olino et al., 2014; Vidal-Ribas et al., 2016). When children tend to respond with irritability, including high levels of frustration in reaction to situations, and children are not easily soothed, interaction with caregivers becomes more challenging. Low soothability refers to the child's ability to regulate emotions but also the response to soothing by parents in situations of distress. A circle of reciprocal dysregulation can occur when a child suffers from emotion regulation problems, parents perceive the child as difficult to sooth, parenting stress increases, there is dysregulating caregiver behavior (such as withdrawal, ignoring the child, or intrusive negative behavior), while regulation problems of the child worsens.

Also, training mental health workers in recognizing inhibited attachment behavior and disinhibited social engagement behavior could be useful in further addressing mental health problems in children and parenting stress. It should be considered in assessment procedures using information from parents about the behaviors as well as parent-child observation and interaction with relative strangers. Important features of inhibited attachment behavior are when children show a lack of preference for a caregiver and do not seek comfort in situations of distress. These children have been described as emotionally withdrawn. Children with disinhibited social engagement behavior are typically indiscriminately social in interaction with strangers, which can be perceived as awkward and unpleasant. These children tend to leave easily with strangers, and some children will even protest upon the departure of an unfamiliar mental health worker. However, recognizing disinhibited social engagement behavior should not lead to the assumption of child neglect or maltreatment. It should not be used as

an indicator for maltreatment, as was also noted for disorganized attachment (Granqvist et al., 2017). A lack of understanding of aetiology of disinhibited social engagement behavior in clinically referred home-reared children requires cautious interpretation in home-reared children.

It is important to note that some temperament traits as well as aberrant social behaviors could be highly functional in certain circumstances; for instance impulsivity and approach when childen are being neglected. Some temperament traits or behaviors of children could be adaptive to environmental factors. Therefore, the clinical relevance of temperamental features and behaviors in children must always be placed into perspective of other child and contextual factors related to mental health problems. Regarding parent psychopathology, symptoms of depression should be assessed when young children are referred for mental health care. Symptoms of depression, but also anxiety and stress, in parents could limit the ability to reflect upon the child's mental state and attune to the needs of the child. Regarding pathogenic caregiving conditions, prevention and early detection of child maltreatment and neglect should be target in all mental health care programs, because young children with developmental and behavioral problems are a high-risk group for maltreatment especially when parents perceive parenting stress and symptoms of depression (Lau & Weisz, 2003; Rodriguez & Richardson, 2007; Walrath et al., 2006). In a recent report and national program in the Netherlands (Rijksoverheid, 2018), a multidisciplinary approach with system focus is advised with the first aim to create a safe environment and secondly to address the risk factors and treat the possible negative outcomes of the maltreatment.

Intervention

Early intervention could possibly alter dysfunctional extremes of temperament traits, as they unfold through maturation and experience. Temperament-based intervention has been suggested and used in general prevention programs, selective prevention programs (when children show features but not yet mental disorders), and treatment programs to reduce serious mental health problems in children (McClowry & Collins, 2015). Caregiver education and training parenting skills could be used to learn how to recognize specific temperament traits of children and how to deal with the traits in order to reduce potential negative consequences. Effortful control, including inhibitory and attention

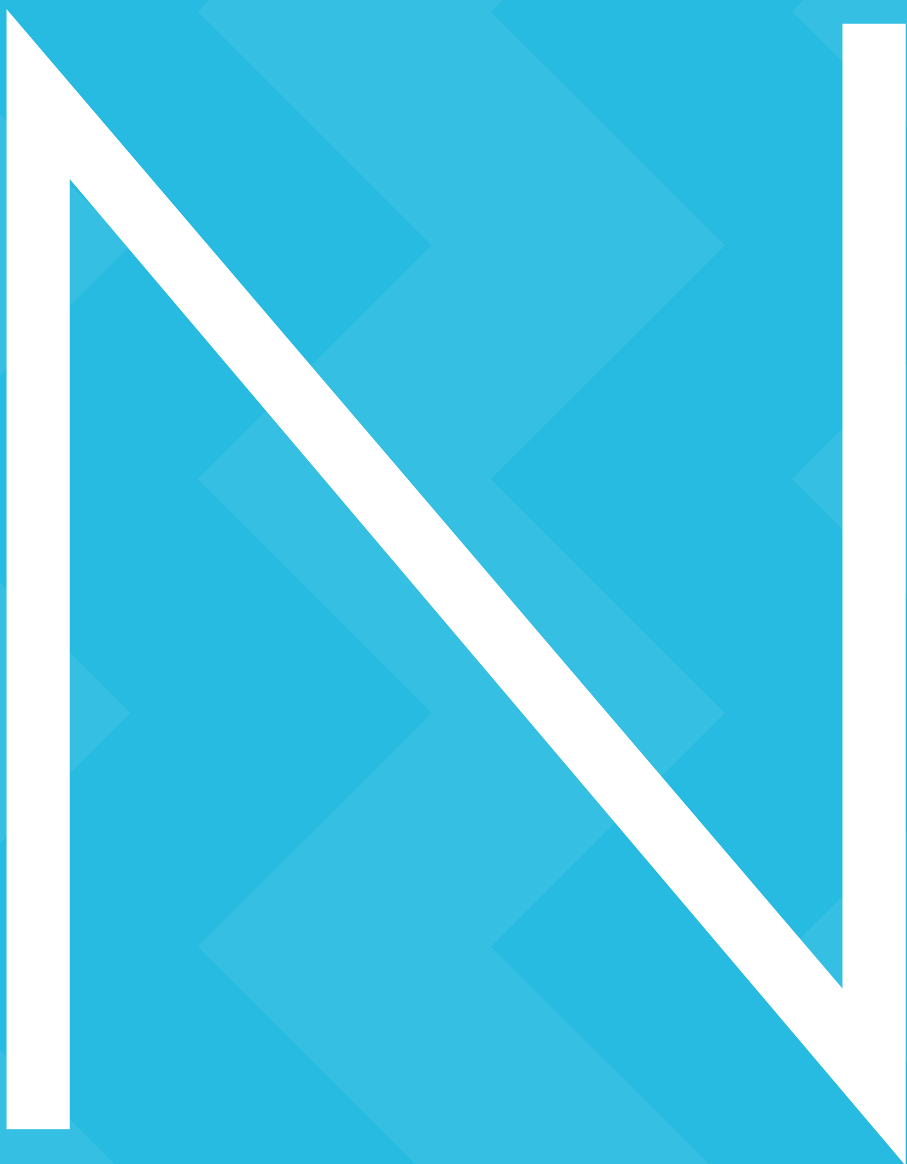
regulation, and emotion regulation in children are already addressed in treatment programs for children with externalizing disorders (De Mey & Braet, 2011; Posthumus et al., 2012). However, temperament-based intervention could be incorporated in treatment of various mental health problems, including disorders. It should be mentioned that some temperament traits could be dysfunctional in low as well as in high levels. While most research addresses low levels of effortful control in relation to mental health problems, high levels of effortful control have also been associated with emotional problems (Murray & Kochanska, 2002).

Very young children are still dependent on their primary caregivers to help them learn to regulate their emotions and behavior. Addressing soothability in intervention should therefore always include caregivers and children together. Video feedback could be used in order to improve attunement between parents and child. When parents experience stress or symptoms of depression that are hampering the parent-child relation, treatment should specifically focus on these parental problems in order to improve functioning outcome in children. Previous studies have found some evidence that a focus on child temperament and parental depression could be effective in improving outcome (Cuijpers et al., 2015; McClowry & Collins, 2015; McClowry et al., 2005; Posthumus et al., 2012). Also, the association between parental depression and child mental health problems is likely to be partially bidirectional suggesting that treatment of mental health problems in children has a positive effect on parental mood and vice versa (Gross et al., 2008; Neece, Green, & Baker, 2012).

When inhibited attachment and disinhibited social engagement behavior are recognized, treatment can be considered aimed at reducing social aberrant behavior and improving the parent-child relation. Disinhibited social engagement behavior should be addressed in treatment, because of the relative persistence over time and associations with more parenting stress and externalizing problems (Chapter 3 and 4). Children are also prone for victimization due to their inappropriate approach of strangers. Disinhibited social engagement behavior can be overshadowed by comorbid ADHD symptoms and lead to selective treatment of ADHD symptoms in children without focus on parent-child relation and social interaction, as has been discussed by Pritchett et al. (2013). Treatment should focus on children, parents and the parent-child relation, addressing behavior regarding social boundaries, self-control and emotion regulation (Dickmann & Allen, 2017; Zeanah & Gleason, 2015).

Conclusion

The results of this thesis should contribute to further improvement of clinical approaches of mental health problems during early childhood, aiming at proper functioning in later life. Since we could not assess all possible factors that might influence the development and course of mental health problems, our focus was on child temperament traits and disturbed attachment behavior. Results indicated clinical relevance of inhibited attachment behavior and specifically of disinhibited social engagement behavior in young children referred for mental health care. We proposed that these behaviors should be addressed in clinical assessment and treatment in early childhood as well as in future research. Of note, the behaviors were not associated to a history of child maltreatment/neglect and therefore cannot be used as an indicator of maltreatment. Still, maltreatment/neglect was found to be prevalent in referred home-reared children. Prevention and early detection of child maltreatment and neglect should be a major target in all early childhood mental health care programs. Furthermore, the present findings show that, next to the influence of parental depression, specific temperament traits were associated with more severe mental health problems after treatment. Higher levels of irritability in children and difficulties in regulating emotions and behaviors were associated to mental health problems at referral and poor outcome after treatment. Possibly, soothability in children can be seen as a linking pin between the fields of child development, infant and early childhood mental health, and child psychiatry. Young children's ability to regulate emotions after distress and the response to soothing by parents/caregivers could be important in the development and course of various mental health problems and disorders. Mental health care in early childhood may benefit from more specifically addressing their emotion regulation temperament traits, disinhibited social engagement behavior and symptoms of depression in their parents.



Nederlandse samenvatting

PSYCHISCHE PROBLEMATIEK BIJ JONGE KINDEREN VERWEZEN VOOR BEHANDELING

TEMPERAMENT, VERSTOORD GEHECHTHEIDSGEDRAG EN BELOOP

Frederike Scheper

Achtergrond

Net als oudere kinderen kunnen ook peuters en kleuters sociaal-emotionele problemen en gedragsproblemen hebben die hen belemmeren in hun functioneren (Angold & Egger, 2007). Er is steeds meer aandacht voor preventie en behandeling op de jonge leeftijd om alle kinderen een kans te geven op een gezonde ontwikkeling (Dougherty, Leppert, et al., 2015; Shonkoff, 2003). Desondanks staan diagnostiek en behandeling van psychische problematiek bij jonge kinderen nog in de kinderschoenen vergeleken met oudere kinderen. De grote stappen in ontwikkeling die kinderen in hun eerste levensjaren doormaken en het belang van gehechtheid en veiligheid van opgroeien maken het belangrijk om breed te kijken naar jonge kinderen die worden aangemeld vanwege sociaal-emotionele en gedragsproblemen. Onderzoek naar de relatie tussen psychische problematiek, temperament en verstoord gehechtheidsgedrag kan helpen om diagnostiek en behandeling beter af te stemmen op de specifieke problematiek van jonge kinderen en hun ouders binnen de context waarin zij leven. Dit proefschrift beoogt onze kennis te vergroten over temperament, verstoord gehechtheidsgedrag en het beloop van internaliserende en externaliserende gedragsproblemen bij jonge kinderen na behandeling.

Temperament wordt gedefinieerd als aangeboren persoonlijkheidsstijl met verschillende kenmerken die in de loop van het leven worden beïnvloed door een wisselwerking tussen rijping van de hersenen en de omgeving (Rothbart & Derryberry, 1981). Vanaf jonge leeftijd worden er al herkenbare individuele verschillen gezien in reactie op dagelijkse situaties. Drie dimensies in temperament zijn relatief stabiel vanaf het derde levensjaar: positieve emotionaliteit, negatieve emotionaliteit en zelfcontrole (Rothbart et al., 2001). Positieve emotionaliteit kenmerkt zich door een neiging om positief en actief te reageren op dagelijkse situaties, zoals met blijheid, plezier en enthousiasme. Kinderen die vaak reageren met angst, verdriet, boosheid/frustratie en die moeilijk te kalmeren zijn, hebben juist een hoge mate van negatieve emotionaliteit. Zelfcontrole vertegenwoordigt de mogelijkheid om emoties en gedrag te reguleren, zoals het vermogen om gedrag te stoppen (inhibitie). De verschillen tussen mensen in temperament zijn gradueel. Dat wil zeggen dat er in meer of mindere mate sprake van kan zijn. In algemeen bevolkingsonderzoek is gevonden dat de combinatie van een hoge mate van negatieve emotionaliteit

en een lage mate van zelfcontrole bij jonge kinderen voorspellend is voor internaliserende problematiek (zoals angst en depressie) en externaliserende problematiek (zoals agressie) (Gartstein et al., 2012). Er is echter nog weinig onderzoek gedaan naar de relatie tussen temperament en internaliserende en externaliserende problemen in klinische populaties jonge kinderen.

Verstoord gehechtheidsgedrag is met name onderzocht bij kinderen die opgroeien in instellingen, adoptie- of pleeggezinnen. Dit gedrag kan zich op verschillende manieren uiten. Zo zijn er kinderen die emotioneel teruggetrokken gedrag laten zien en weinig onderscheid maken tussen bekende en onbekende volwassenen. Dit wordt geïnhibeerd gehechtheidsgedrag genoemd. Andere kinderen laten juist vrijpostig gedrag zien, waarbij ze bijvoorbeeld zomaar met vreemden kunnen meegaan of op schoot gaan zitten. Dit wordt ook wel sociaal ontremd of gedisinhibeerd gehechtheidsgedrag genoemd. Geïnhibeerd gehechtheidsgedrag is kenmerkend voor een reactieve hechtingsstoornis (reactive attachment disorder, RAD) en gedisinhibeerd gehechtheidsgedrag is kenmerkend voor de ontremd-sociaal contactstoornis (disinhibited social engagement disorder, DSED); beide beschreven in de DSM-5 (American Psychiatric Association, 2013). Symptomen van RAD verminderen vaak na interventie, maar symptomen van DSED blijven vaak bestaan (Zeanah & Gleason, 2015). Opgroeien in ongunstige omstandigheden, waar bijvoorbeeld sprake is van emotionele mishandeling en/of verwaarlozing en continue wisselende verzorgers, kan leiden tot verstoord gehechtheidsgedrag (Zeanah & Gleason, 2015). Verstoord gehechtheidsgedrag is nauwelijks onderzocht bij kinderen die opgroeien bij hun eigen ouders, zonder uit huis geplaatst te zijn, terwijl ook thuiswonende kinderen emotioneel mishandeld en/of verwaarloosd kunnen worden. We weten dat kinderen die verwaarloosd of mishandeld zijn ook ernstige gedragsproblemen kunnen hebben (Lau & Weisz, 2003), maar niet of jonge thuiswonende kinderen die worden verwezen vanwege sociaal-emotionele problemen en gedragsproblemen ook verstoord gehechtheidsgedrag vertonen.

Het beloop van psychische problematiek vanaf jonge leeftijd is belangrijk om te onderzoeken als we beter willen weten welke kinderen problemen behouden. Het kan inzicht geven in de noodzaak tot het aanscherpen van hulpverlening en verdere hulp na behandeling. Eerder onderzoek heeft laten zien dat jonge kinderen met emotionele- en gedragsproblemen vaak ook later nog psychische problemen hebben (Bufferd et al., 2012). Met name jonge kinderen met

een combinatie van internaliserende problemen (zoals angst en depressieve symptomen) en externaliserende problemen (zoals hyperactiviteit en agressie) lopen risico op psychische problematiek later in het leven (Basten et al., 2016). Daarom is er steeds meer aandacht voor preventie en vroege behandeling van psychische problematiek bij jonge kinderen. Ten aanzien van het stimuleren van een positieve ontwikkeling bij kinderen, is bekend dat behandeling van gedragsproblemen bij jonge kinderen effectief kan zijn (Muller et al., 2015). Desondanks zijn er ook veel kinderen die jaren na behandeling problemen houden (Visser et al., 1999). Er is daarom meer kennis nodig over welke kinderen na behandeling op de vroege leeftijd toch problemen behouden, zodat behandeling daar mogelijk op aangepast dient te worden.

Opzet van dit onderzoek

Allereerst hebben we inzicht willen krijgen in de relatie tussen temperamentskenmerken en psychische problematiek bij jonge kinderen die vanwege sociaal-emotionele problemen en/of gedragsproblemen werden verwezen naar het Medisch Orthopedagogisch Centrum (MOC) 't Kabouterhuis in Amsterdam. MOC 't Kabouterhuis helpt gezinnen met jonge kinderen met een ontwikkelingsachterstand, met opvoedings- en/of psychiatrische problemen. De kinderen werden onderzocht door middel van door de ouders in te vullen vragenlijsten. Deze kinderen werden vergeleken met kinderen uit de algemene bevolking. Ook werden ze vergeleken met niet-verwezen kinderen uit de algemene bevolking die mee hadden gedaan aan een longitudinaal onderzoek naar temperament in Amsterdam (Majdandžić & van den Boom, 2007b).

Het tweede doel was om de klinische waarde te onderzoeken van verstoord gehechtheidsgedrag, symptomen van RAD en DSED, bij thuiswonende kinderen die waren verwezen vanwege emotionele- en/of gedragsproblemen. Hiervoor werden kinderen onderzocht die verwezen waren naar MOC 't Kabouterhuis en kinderen die verwezen waren naar de Bascule, een instelling in Amsterdam voor kinder- en jeugdpsychiatrie. Voor dit onderzoek werd gebruikt gemaakt van een groep kinderen waarbij informatie beschikbaar was over verstoord gehechtheidsgedrag, internaliserende en externaliserende problemen en ouderlijke stress. Deze informatie was verzameld door middel van interviews met de ouders en vragenlijsten. Met behulp van een checklist werden dossiers

gescoord op aanwijzingen voor het voorkomen van mishandeling/verwaarlozing. Gemiddeld drie jaar na behandeling werden ouders van de kinderen die in behandeling waren bij 't Kabouterhuis opnieuw benaderd voor een follow-up onderzoek. Tijdens het follow-up onderzoek onderzochten we het beloop van gedisinhibeerd gehechtheidsgedrag door middel van interviews en observaties. Het onderzoek behelsde ook het verband tussen het beloop van gedisinhibeerd gehechtheidsgedrag en emotionele mishandeling/verwaarlozing, de mate van zelfcontrole bij kinderen en psychiatrische stoornissen.

Het derde doel was om de kennis te vergroten over het beloop van emotionele- en gedragsproblemen na behandeling op de vroege kinderleeftijd en te achterhalen of specifieke temperamentskenmerken naast psychische problemen bij ouders van invloed zijn op dit beloop. Hiervoor werden kinderen die verwezen waren naar 't Kabouterhuis onderzocht door middel van vragenlijsten en interviews: bij de start van de behandeling, aan het einde van de behandeling en bij follow-up. (Zie ook Appendix 1 voor een studie-opzet met instrumenten). De behandeling was afgestemd op de hulpvraag, rekening houdend met de mogelijkheden en beperkingen van de kinderen/gezinnen. Gezinsbegeleiders maakten samen met psychologen en orthopedagogen gebruik van gedragstherapeutische adviezen, oplossingsgericht werken, competentie vergrotend werken en video feedback. Kinderen kregen zo nodig logopedie, fysiotherapie, psychotherapie, medicatie en/of één tot vier dagen per week groepsbehandeling gericht op het vergroten van ontwikkelingsvaardigheden.

Bevindingen

In **hoofdstuk 2** wordt de relatie beschreven tussen temperament en psychische problematiek bij 216 jonge kinderen, verwezen voor behandeling van emotionele- of gedragsproblemen. Van deze groep werden 115 verwezen kinderen vergeleken met 115 niet-verwezen kinderen uit de algemene bevolking (gematcht op leeftijd en geslacht). Het bleek dat verwezen kinderen meer moeite hadden om hun emoties en gedrag te reguleren (minder zelfcontrole) dan kinderen uit de algemene bevolking. Ook bleek dat verwezen kinderen minder gemakkelijk te kalmeren waren. Verder waren hogere scores op negatieve emotionaliteit en lagere scores op zelfcontrole bij kinderen gerelateerd aan meer internaliserende en externaliserende problemen. Deze relatie was net zo sterk bij verwezen

kinderen als bij kinderen in de algemene bevolking. De extremen van deze temperamentskenmerken kunnen mogelijk tot disfunctioneren leiden waarvoor behandeling wordt geïndiceerd. Opvallend was dat impulsiviteit wel gerelateerd was aan externaliserend probleemgedrag bij verwezen kinderen, maar niet bij de kinderen uit de algemene bevolking. Mogelijk zijn er andere factoren in het temperament van kinderen of factoren in ouderschap die deze relatie beïnvloeden.

Verwezen kinderen met zowel ernstige internaliserende als externaliserende problematiek hadden meer problemen om hun gedrag af te remmen (onderdeel van zelfcontrole), meer moeite om zichzelf te kalmeren tijdens stress en een hogere mate van frustratie/irritatie dan kinderen zonder een combinatie van ernstige internaliserende en externaliserende problematiek.

Hoofdstuk 3 beschrijft onderzoek naar geïnhibeerd en gedisinhibeerd gehechtheidsgedrag, symptomen van RAD en DSED, bij 200 jonge kinderen die waren verwezen voor behandeling vanwege emotionele- of gedragsproblemen. Kinderen die opgroeiden bij hun biologische ouders (n = 141) werden vergeleken met kinderen die in een pleeggezin waren geplaatst (n = 59). Van de thuiswonende kinderen was 54% (ooit) mishandeld/verwaarloosd, terwijl dit bij de pleegkinderen voorkwam bij 90%. Geïnhibeerd gehechtheidsgedrag werd minder vaak gevonden bij kinderen die opgroeiden bij hun biologische ouders (9%) dan bij pleegkinderen (27%). Er werd geen verband gevonden met mishandeling/verwaarlozing. Alleen bij thuiswonende kinderen was geïhibeerd gehechtheidsgedrag gerelateerd aan zowel internaliserende als externaliserende problematiek bij kinderen. Gedisinhibeerd gehechtheidsgedrag kwam in vergelijkbare mate voor bij thuiswonende kinderen (42%) als bij pleegkinderen (51%). Er werd geen verband gevonden met verwaarlozing/mishandeling, maar gedisinhibeerd gehechtheidsgedrag was wel gerelateerd aan een hogere mate van ouderlijke stress (opvoedbelasting). Ook was het gerelateerd aan een ernstige mate van externaliserende gedragsproblemen bij zowel thuiswonende kinderen als pleegkinderen. De bevindingen laten zien dat geïnhibeerd en gedisinhibeerd gehechtheidsgedrag voorkomen bij thuiswonende kinderen die zijn verwezen voor behandeling van emotionele- en gedragsproblemen. Deze gedragingen kunnen klinisch van belang zijn, gezien het verband met emotionele- en gedragsproblemen bij kinderen als ook de opvoedbelasting bij ouders. De bevindingen laten de vraag onbeantwoord waar geinhibeerd en gedisinhibeerd gehechtheidsgedrag vandaan komt.

Vervolgonderzoek naar het beloop van gedisinhibeerd gehechtheidsgedrag wordt beschreven in **hoofdstuk 4**. Voor dit onderzoek zijn 124 kinderen (82% jongens, gemiddeld 4 jaar oud bij de start van de behandeling en gemiddeld 8 jaar oud bij follow-up) onderzocht. Onder de 47 kinderen met vroeg gedisinhibeerd gehechtheidsgedrag werd bij 57% ditzelfde gedrag nog steeds bij follow-up gevonden. Emotionele mishandeling of verwaarlozing in de vroege kinderjaren vertoonde geen verband met gedisinhibeerd gehechtheidsgedrag. Er werd evenmin een relatie gevonden met de mate van zelfcontrole bij de kinderen op jonge leeftijd. Daarmee is onduidelijk wat het beloop van gedisinhibeerd gehechtheidsgedrag bij jonge thuiswonende kinderen verklaart. Wel blijkt een aanzienlijk deel van de kinderen te persisteren in dit gedrag.

Vanwege gerezen vragen naar een mogelijke overlap met symptomen van een autisme spectrum stoornis (ASS), waarbij kinderen ook sociaal onaangepast gedrag kunnen laten zien, werd gekeken naar de samenhang van ASS met gedisinhibeerd gehechtheidsgedrag. Gedisinhibeerd gehechtheidsgedrag hing niet samen met een autisme spectrum stoornis. Deze resultaten sluiten aan bij ander onderzoek dat laat zien dat er onderscheid bestaat tussen symptomen van ASS en DSED. Kinderen die het gedisinhibeerd gedrag bleven vertonen, hadden bij follow-up wel vaker dan de kinderen die dit gedrag niet vertoonden een aandachtsdeficiëntie-/hyperactiviteitsstoornis (ADHD), met of zonder oppositionele gedragsstoornis (ODD). Gedisinhibeerd gehechtheidsgedrag op jonge leeftijd voorspelde de aanwezigheid van ADHD in combinatie met ODD bij follow-up, maar niet ADHD alleen. Dit suggereert dat gedisinhibeerd gehechtheidsgedrag bij jonge thuiswonende kinderen met name geassocieerd is met ADHD wanneer er ook grensoverschrijdend agressief gedrag is.

In **hoofdstuk 5** wordt onderzoek beschreven waarbij het beloop van internaliserende en externaliserende problemen na behandeling op vroege kinderleeftijd in kaart werd gebracht in relatie tot temperament en ouderlijke psychopathologie. Voor dit onderzoek werden 360 peuters en kleuters (81% jongens, gemiddelde leeftijd 4 jaar) samen met hun ouders onderzocht. De kinderen waren zowel voor ambulante als voor dagbehandeling verwezen naar MOC 't Kabouterhuis vanwege emotionele- en/of gedragsproblemen. Ze werden driemaal onderzocht: bij de start van de behandeling, bij einde van de behandeling (n = 246) en gemiddeld 3 jaar later (n = 164). We hebben een significante daling van de psychische problemen gevonden: de kinderen

vertoonden minder internaliserende en externaliserende problemen zowel direct na de behandeling als 3 jaar later ten opzichte van de start. Verbetering van probleemgedrag tijdens de behandeling was gerelateerd aan een betere kwaliteit van leven bij follow-up. Tegelijkertijd werden na behandeling nog wel bij 48% van de kinderen emotionele- of gedragsproblemen gerapporteerd. Een lagere score op zelfcontrole en een hogere score op frustratie bij jonge kinderen was gerelateerd aan externaliserende problematiek tot minstens drie jaar na behandeling. Problemen om zichzelf te kalmeren (lagere scores op kalmeerbaarheid bij kinderen), naast meer depressieve symptomen bij de ouder, waren juist gerelateerd aan meer internaliserende problemen bij kinderen na behandeling. Eerder is door Caspi et al. (2014) gesuggereerd dat een lage zelfcontrole een risicofactor kan zijn die gerelateerd is aan verschillende psychiatrische stoornissen later in het leven. Ook prikkelbaarheid op jonge leeftijd, waar een hoge mate van frustratie deel van uitmaakt, is eerder geassocieerd met psychiatrische problematiek en disfunctioneren later in het leven (Dougherty et al., 2015). Dit onderzoek sluit aan bij deze bevindingen. Daarnaast voegt het eraan toe dat problemen met kalmeerbaarheid, dus wanneer kinderen zichzelf moeilijk kunnen kalmeren, een risicofactor vormen.

Beperkingen

De generaliseerbaarheid van de bevindingen zijn beperkt doordat niet alle gezinnen meededen. Er waren ouders die bijvoorbeeld vanwege een taalbarrière niet mee konden doen. Daardoor hebben we een zekere selectie in onze groep van meer autochtone kinderen. Ook is de groep kinderen die is onderzocht een heterogene groep wat betreft het type problematiek en wat betreft het type behandelingen dat is uitgevoerd, waardoor niet gezegd kan worden wat er precies voor wie in welke context heeft gewerkt. Ook wijkt de populatie van het Kabouterhuis mogelijk af van die van andere kinderpsychiatrische instellingen. Een ander beperking is dat niet alle mogelijke relevante factoren (zoals o.a. taalproblemen, medische aandoeningen) konden worden meegenomen in dit onderzoek. Een beperking met betrekking tot de gehechtheid is dat we de kwaliteit van de ouder-kind relatie niet hebben onderzocht. Dit had te maken met de keuze om de ouders en kinderen niet te veel te belasten.

Implicaties voor onderzoek

Het blijft belangrijk om te onderzoeken welke kinderen problemen houden en of behandeling kan worden aangepast met betere resultaten tot gevolg. Verder is het, net als voor oudere kinderen, ook voor jonge kinderen belangrijk om te onderzoeken of er behandelingen zijn die voor specifieke groepen kinderen onder bepaalde omstandigheden effectief zijn of juist niet. Daarbij kan het relevant zijn om rekening te houden met depressieve symptomen bij de ouder(s) en met temperamentskenmerken van kinderen. Vervolgonderzoek is verder nodig om het verstoord gehechtheidsgedrag dat we vonden bij thuiswonende kinderen nog beter te begrijpen. Daarbij kan gedacht worden aan het specifieker beschrijven van het soort gedrag van de kinderen, bijvoorbeeld of de kinderen alleen verbale sociale disinhibitie laten zien of ook fysieke disinhibitie. Ook is het belangrijk om ouderschap en de kwaliteit van de relatie met de ouders te onderzoeken, met name in stressvolle situaties. Daarbij dient ook aandacht te zijn voor sociaal maatschappelijk factoren, armoede en sociaal isolement.

Implicaties voor de klinische praktijk

Aandacht in het veld neemt toe voor kenmerken die, stoornis overstijgend, een rol kunnen spelen in de behandeling. Dit worden ook wel transdiagnostische kenmerken genoemd. Emotieregulatie en gehechtheid zijn daar voorbeelden van. Waar een diagnostische benadering zich richt op symptomen van stoornissen, richt een transdiagnostische benadering zich op factoren of processen die van belang zijn bij meerdere stoornissen (van Amelsvoort et al., 2018).

Een hoge mate van frustratie, moeite om zichzelf te kalmeren in situaties van stress en een lage zelfcontrole zijn uit ons onderzoek naar voren gekomen als temperamentskenmerken die van belang kunnen zijn voor het latere functioneren. Onderdeel van veel behandelingen voor jonge kinderen is het trainen van zelfcontrole. Behandeling zou echter ook gericht moeten zijn op andere temperamentskenmerken. Wanneer ouders en kind beter leren omgaan met de specifieke eigen temperamentskenmerken, naast of bij eventuele psychiatrische

stoornissen, en ouders en kind beter op elkaar kunnen afstemmen, kan dit een positieve invloed hebben op de ontwikkeling van een kind (McClowry & Collins, 2015). Er zijn aanwijzingen dat kinderen verschillend op omstandigheden kunnen reageren (Belsky & Pluess, 2009). In eerder onderzoek is gevonden dat kinderen die vanuit hun temperament geneigd zijn om negatief te reageren op situaties meer dan kinderen zonder deze manier van reageren ontvankelijk zijn voor een negatieve manier van opvoeden door hun ouders om externaliserende problemen te ontwikkelen (van Zeijl, et al. 2007). Uit dit onderzoek bleek dat deze kinderen ook extra ontvankelijk zijn voor verbetering van probleemgedrag als ouders positief gaan opvoeden, wat kansen biedt voor behandeling.

Het onderzoek beschreven in dit proefschrift laat ook zien dat het belangrijk is om depressieve symptomen bij ouders te herkennen en mee te nemen in de behandeling, omdat dit geassocieerd is met een slechter beloop van psychische problemen na behandeling. Aangezien de manier waarop jonge kinderen zichzelf kunnen kalmeren in stressvolle situaties ook afhankelijk is van de ouders en ouder-kind relatie, kan het extra belangrijk zijn om te investeren in verbetering van de ouder-kind relatie wanneer er problemen worden gezien.

Tegelijkertijd is het voor diagnostiek en behandeling ook van belang dat er beter zicht komt op eventueel verstoord gehechtheidsgedrag. Daarvoor is het nodig dat er met de ouders worden gesproken over de ouder-kind relatie en het gedrag van het kind, alsook dat de ouder-kind relatie wordt geobserveerd en het gedrag van het kind ten opzichte van een relatieve onbekende wordt beoordeeld. Wanneer geïnhibeerd (emotioneel teruggetrokken, nauwelijks voorkeur voor een volwassene) en gedisinhibeerd (vrijpostig, sociaal ontremd en ongepast) gehechtheidsgedrag beter wordt herkend, kan diagnostiek en behandeling daarop worden aangepast. Het is echter belangrijk dat men zich realiseert dat wanneer een kind geïnhibeerd of gedisinhibeerd gehechtheidsgedrag vertoont, er niet automatisch vanuit moet worden gegaan dat er sprake is van mishandeling of verwaarlozing. Uit eerder onderzoek en ook uit dit onderzoek blijkt wel dat jonge kinderen met emotionele- en gedragsproblemen risico lopen op kindermishandeling (Lau & Weisz, 2003; Walrath et al, 2006). Maar er is altijd zorgvuldig diagnostisch onderzoek nodig naar de omstandigheden van opgroeien. Ook is zorgvuldig onderzoek nodig om onderscheid te maken tussen gedisinhibeerd gehechtheidsgedrag passend bij DSED en andere psychiatrische stoornissen zoals ODD en ADHD. Er is eerder al gesuggereerd dat symptomen

van DSED verward kunnen worden voor ADHD (Pritchett et al., 2013). Goede diagnostiek is nodig om onderscheid te maken tussen de stoornissen, maar de stoornissen kunnen ook samen voorkomen. Het is belangrijk om gedisinhibeerd gehechtheidsgedrag te onderkennen, aangezien dit gedrag vaak blijft bestaan en een risico kan zijn voor het optreden van slachtofferschap van mishandeling/misbruik.

Slotwoord

Voor diagnostiek en behandeling kan het in beeld brengen van verstoord gehechtheidsgedrag en manieren van emotie-regulatie als kenmerken van temperament bij kinderen, naast eventuele depressieve symptomen bij de ouder(s), zinvol zijn om de zorg beter af te stemmen. Middels gesprekken, observaties, video-feedback en ouder-kind behandeling kan gewerkt worden aan een verbetering van de afstemming tussen ouders/verzorgers en het jonge kind. Naast een stoornis-gerichte benadering kan een transdiagnostische benadering helpen om diagnostiek en behandeling persoonlijk af te stemmen. Een interdisciplinaire werkwijze, waarbij er met meerdere disciplines wordt gewerkt aan integratieve beeldvorming en behandeling, is daarvoor van groot belang.