PARENTING A CHILD WITH AUTISM

Support for early parent-child interaction

Irina Elizabeth Poslawsky

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Het opvoeden van kinderen met autisme
Ondersteuning van de vroege ouder-kind interactie
(met een samenvatting in het Nederlands)

Proefschrift

ter verkrijging van de graad van doctor aan de Universiteit Utrecht
op gezag van de rector magnificus, prof. dr. G.J. van der Zwaan,
ingevolge het besluit van het college voor promoties
in het openbaar te verdedigen op
donderdag 9 januari 2014 des ochtends te 10.30 uur

door

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geboren op 27 januari 1954
te Utrecht

Voor Marti

Argeloos samen opgegroeid

Contents

Chapter 1

Introduction

Introduction

"Because I am getting tired of people making remarks like "it'll be OK"; "Just be stricter with him" and "It's just fashionable to put a label on a child".

It'll be OK*: Well, no. Neil really does have autism, even though he is nothing like a drooling, contact-disturbed Rain Man. He has most trouble dealing with busy and noisy activities, people he doesn't know, unexpected situations, loud noises, food; and regarding his language skills, he is "slow in understanding things". He takes everything so literally. For example, when the doorbell rings and I ask him to open the door, he will do that, but then he will walk straight back, leaving the person standing by the door. He was asked to open the door, right? He also puts things together wrongly. A few months ago, he hit his head during gym class and he is now scared to hit his head again during gym class because gym class equals hitting his head. If something doesn't goes as planned, or the furniture is rearranged, or we are in a busy or noisy place, it confuses him and a tantrum results. And there are about 83 other things that Neil has difficulties with. So, no, it's not OK.*

Just be stricter with him*: That won't help. If Neil doesn't want something, most of the time it's because he doesn't understand or because (he thinks) he won't be able to manage. Getting angry will make it worse and then there will be another huge tantrum (again).*

It's just fashionable to put a label on a child*: It might look like that because we are learning more and more about autism. I am overjoyed with his label. Now I (and school and others) can help him and understand him better. If there had been no such diagnosis, he would probably be 'that difficult and annoying child'. Now we can do something about it, or at least we can understand his situation better. Didn't we all know a kid in school who didn't fit in with the rest when we were little? Well, that kid is my kid….*

By the way: my child is the sweetest child in the world!

This citation is from the mother of a four-year-old boy, recently diagnosed with Autism Spectrum Disorder (ASD). It illustrates the impact of ASD on parenting. Parents have to explain to others what a diagnosis within the autism spectrum actually means: that the child struggles with the demands of daily situations and that effective parental strategies differ from those applicable to a typically-developing child. Therefore, the relevance of an ASD diagnosis is that it creates the opportunity to provide adequate support for children and their families.

Diagnosis and prevalence of ASD

ASD comprises a broad psychiatric diagnostic category of pervasive developmental disorders, ranging from mild to severe symptomatology (American Psychiatric Association [APA], 2013). Previously, ASD was classified in distinctive disorders: the Autistic Disorder (AD), Pervasive Developmental Disorder - Not Otherwise Specified (PDD-NOS), Asperger Syndrome, and the less common Rett's Disorder and Childhood Disintegrative Disorder (APA, 2000). The prevalence of ASD is continually being revised because of an increase in the rate of identification of the characteristics, changes over time in case definition, and heterogeneity of study methods and study populations (Wing & Potter, 2002; Fombonne, 2003a; Williams, et al., 2006; Posserud, et al., 2010). Currently, the prevalence of ASD is estimated at 1% (Baird, et al., 2006; Fombonne, 2009; Rice, 2009), though rates up to 2% have been discussed recently (Charman, 2011; Kim et al., 2011; Lord, 2011). More boys than girls are affected, with a sex ratio of about four to one. Comorbidity with mental retardation ranges from 12% (PDD-NOS) to 68% (AD) (Chakrabarti & Fombonne, 2001; Chakrabati & Fombonne, 2005). Other common comorbid conditions are language deficits, sensory and motor problems, anxiety, disruptive (e.g. Attention Deficit Hyperactivity Disorder) or self-injurious behavior, epilepsy, eating and sleep problems (Levy et al., 2009).

Characteristics of ASD

ASD is a neurodevelopmental disorder characterized by impairments of social interaction and communication, combined with repetitive or restricted behaviors and interests. The onset is before 36

months of age (APA, 2013; Volkmar et al., 2004). Structural brain abnormalities, including atypical organization within the cortex and neuropathological alterations, are sometimes found in patients suffering from ASD (Rumsey & Ernst, 2000; Palmen et al., 2004). Twin and family studies demonstrate genetic influences with an estimated heritability of more than 90% (Lauritsen & Ewald, 2001; Nicolson & Szatmari, 2003). Several genome-wide association studies have identified risk genes in patients with ASD, although genetic differences only explain a small part of the variance in autistic symptomatology (Weiss & Arking, 2009; Van der Zwaag et al., 2009, Vorstman et al., 2011). New insights into human genome function are now emerging through ongoing research by the ENCODE project consortium (Encyclopedia of DNA Elements) (see Birney et al., 2007).

The course of ASD is chronic with a disadvantageous prognosis. The majority of adults diagnosed with ASD in childhood remain dependent on the support of their families or special services (Nordin &Gillberg, 1998; Howlin et al., 2004). Frequently, social and communication skills remain impaired and stereotypical behaviors or interests persist into adulthood. However, long-term outcomes are heterogeneous and not straightforwardly predictable. A prospective follow-up study of preschool children with ASD ($N = 219$) revealed developmental and symptomatic changes over time (Baghdadli et al., 2007). The severity of autistic symptoms and the level of intellectual, linguistic and adaptive functioning at age five were found to predict future developmental outcome.

The population of children diagnosed with ASD under the age of five is growing. In the United States, an increase of more than 40% between 2000 and 2050 is anticipated (Fombonne, 2003b). In addition, the focus on early symptom detection has shifted to the first year of life. In a prospective longitudinal study of 6- and 12-month-old infants, early behavioral risk markers were found (Zwaigenbaum et al., 2005). Infants later diagnosed with ASD demonstrated atypical behaviors and delayed language development, i.e. atypical visual engagement, limited social responsiveness, relatively few vocalizations, lack of joint attention, sensory-oriented behaviors including stereotypical, self-stimulating use of play materials, fixation on particular objects, and

an unusual temperamental pattern varying from passivity to extreme distress reactions. The stability of the ASD diagnosis at a very young age was demonstrated to be 79-88% (Barbaro & Dissanayake, 2009). Although most diagnoses continue within the autism spectrum, Van Daalen et al. (2009) reported reduced symptom severity between 23 and 42 months of age in groups of children both with and without a stable diagnosis, and improvement of cognitive functioning including language development.

Even with early diagnosis, there is no cure for the core deficits of ASD. Nowadays, consensus of opinion about treatment of ASD is that interventions should focus on: (1) stimulation of developmental progress, (2) prevention of secondary (behavioral) disorders and (3) support for the family in order to enhance parental efficacy, coping and stress reduction (e.g. Van Engeland & Buitelaar, 2009).

Early interventions for children with ASD

A world-wide survey on 552 parents revealed the use of 111 different therapies for children with ASD, with an average current usage per family of seven individual treatments (Green et al., 2006). Parental preferences for treatments were not associated with evidence of therapeutic effectiveness. Some parents used ten different treatments at the same time, but it is not clear how the interventions were related to the complexity of children's disorders. The number of treatments declined as the child became older. Speech-language therapy was used most frequently, followed by skill-based training, including applied behavior analysis. Half of the parents used pharmacological therapy for their children. Pharmacological treatments commonly address comorbid symptoms (Levy et al., 2009), e.g. the atypical antipsychotic risperidone for disruptive behavior or harmful stereotypical behavior and melatonin for sleep problems (Hollander et al., 2003). In addition to conventional therapy, complementary and alternative treatments are frequently used, e.g. special diets and vitamin supplements (Levy & Hyman, 2008). Comprehensive behavioral and educational intervention programs including language training are recommended for young children with ASD (New York State Department of Health Clinical Practice Guideline, 1999).

The most well-researched and effective early interventions are based on applied behavior analysis (ABA; see meta-analyses of Makrygianni & Reed, 2010; Reichow et al., 2012; Virués-Ortega, 2010; Peters-Scheffer et al., 2011). Children in ABA-based groups show significantly better outcomes in the domains of cognition, language and adaptation than control groups, although most sample sizes are small (Ospina et al. 2008; Rogers & Vismara, 2008). ABA interventions are based on operant conditioning approaches such as prompting, modeling, fading and reinforcement. The use of ABA techniques differs among programs in the degree of reinforcement control, the setting, and their focus on children's deficits or strengths, but common elements are parental involvement, individualized programming and intensity (15-40 weekly hours) (Erba, 2000). Parental involvement is crucial for the individualization of interventions, i.e. adapting the individual goals and needs of the child and family to the intervention and, conversely, supporting the child in incorporating new skills and behavior in daily life (New York State Department of Health Clinical Practice Guideline, 1999). Moreover, parental involvement enhances the therapeutic effectiveness of comprehensive ABA-based programs (Strauss et al., 2013).

For toddlers with ASD, the Early Start Denver Model (ESDM) demonstrates improvements of the child's cognitive and language skills, adaptive behavior and ASD diagnosis (Dawson et al., 2010). The ESDM is a comprehensive early intervention program, comprising an integration of ABA with developmental and relationship-based approaches in which improvements on child outcome are associated with normalizing patterns of brain activity (Dawson et al., 2012). However, these comprehensive and intensive intervention programs are expensive and not always available or accessible to parents. The intensity of ABA programs can increase family stress, especially when parents are heavily involved (Schwichtenberg & Poehlmann, 2007). Parents of young children with ASD are often in a phase of life during which they are building their careers and expanding their families. During this phase they are especially vulnerable to conflict between family and work demands (Demerouti et al., 2012). It can be extra stressful for parents to balance the time devoted to their child's intensive therapy regarding

ASD with their engagement in other family and work demands. For this reason, there is also a need for early, feasible and short-term intervention programs enhancing parents' competences to raise their children with ASD. Short-term, behavioral early interventions for children with and without developmental risks have proved effective in parental competence and the parent-child relationship, when they focus explicitly on parent-child interactions (Bakermans-Kranenburg et al., 2003; Juffer et al., 2009). Such treatment programs are tailored to individual parent-child dyads and can also benefit parents of children with ASD.

The impact of ASD on parenting and the parent-child relationship

Parenting is defined as the process of promoting and supporting the child's physical, social and environmental development from infancy to adulthood (Davies, 2001). Generally, parents of children with ASD report more parenting stress than parents of typically-developing children or children with mental retardation, resulting in higher levels of depression, anxiety, somatic complaints, family dysfunction and burnout (Benjak et al., 2009; Eisenhower et al., 2005; Higgins et al., 2005; Koegel et al., 1992; Weiss, 2002). The social deficits of young children newly diagnosed with ASD are associated with greater stress than delays in children's cognitive and verbal development (Davis & Carter, 2008). Furthermore, especially when the child starts to receive special services, parents of children with ASD are found to experience high levels of distress (Schieve et al. (2007). Parental coping style and social support are found to be moderators of stress in such parents (Drabowska & Pisula, 2010; Sivberg, 2002; Dunn et al., 2001).

The quality of the parent-child relationship during infancy is important for child development, e.g. for their level of cognitive functioning (Murray et al., 1996), brain maturation (Johnson, 2001) and the risk of anxiety or depression in adulthood (Murray &Greenberg, 2006). An important aspect of the parent-child relationship is parental sensitivity; that is the extent to which parents respond adequately to the needs of the child (Bakermans-Kranenburg et al., 2003; De Wolf & Van IJzendoorn, 1997). There is no difference in parental sensitivity

between parents of children with or without ASD (Van IJzendoorn et al., 2007). However, the unusual affective behavior of children with ASD (Dawson et al. 1990), including their deficits in processing social information (Noens & Van Berckelaer-Onnes, 2005; Ploog, 2010) and their insensitivity to social stimuli (Gervais et al., 2004), challenges parent-child interactions. For example, parental support can lose its effectiveness when a child reacts aversely to hugs and kisses. The child's unusual behaviors or interaction-perception need explicit parental stimuli, different from the stimuli commonly used for typically-developing children (Kasari et al., 1988; Doussard-Roosevelt et al., 2003). In other words, parents have to attune sensitively to the autistic traits of their children for adequate and mutual interaction. Therefore, we assume that parents can benefit from early support that enables them to learn how to relate ASD symptomatology successfully to their individual responsiveness during interaction with their child. Supportive services to parents that offer early parent-mediated interventions for young children with ASD are indeed effective at improving the parent-child interaction and some of the children's developmental outcomes (McConachie & Diggle, 2007; Oono et al., 2013). However, to our knowledge, no evidence-based intervention programs with the explicit aim of enhancing early parent-child relationships in the presence of ASD have been published.

Aims and outline of this thesis

The essence of this thesis is to study ways of supporting parents of children with ASD by improving early parent-child interactions with limited interference in families' daily routines. We focus on the following study objectives:

1. To explore correlations between the stability of parents' experienced feelings about their child's recent ASD diagnosis and their subsequent coping style.
2. To adapt a tailored, short-term intervention protocol with the aim of enhancing the quality of the parent-child relationship with respect to the individual competences of parents in rearing a child with ASD.
3. To test the effectiveness of the adapted, short-term intervention in a randomized controlled trial, assessing parental sensitive

responsiveness to the child with ASD before and after the intervention.

4. To test the effectiveness of the adapted, short-term intervention in a randomized controlled trial with regard to the children's joint attention and play behavior.

5. To improve parent-child interactions, using oxytocin as a pharmacological supplement.

Knowledge of a parent's expectations, hopes and beliefs about their child's functioning could provide an important foundation from which care providers can tailor interventions. Therefore, in **chapter 2** we present an exploratory study of parental reactions to their child's diagnosis in order to investigate the parents' coping strategies. We hypothesized a negative association between parents' adequate coping strategies and the severity of ASD-symptomatology as well as influences from prediagnostic circumstances. Furthermore, we explored the stability of parental reaction to ASD diagnosis over time.

In **chapter 3** we describe the development and content of a tailored intervention program: the attachment-based Video feedback Intervention to promote Positive Parenting adapted to Autism (VIPP-AUTI). VIPP-AUTI was designed in a collaborative project of the Center for Child and Family Studies, Leiden University and the Department Psychiatry of the University Medical Center, Utrecht. VIPP-AUTI is no cure for ASD, but it is meant to empower parental self-management in guiding a child with ASD. The program involves home training, short in duration in order to limit the costs and burden for families. We also report on intervener criteria and the evaluations of parents who received VIPP-AUTI. Moreover, a single-blind randomized controlled trial of the effectiveness of the VIPP-AUTI program was carried out. The results concerning parent outcome are presented in **chapter 4**. We hypothesized that VIPP-AUTI would improve parent-child interactions by enhancing parental sensitive responsiveness to the child's signals and parents' perceived feelings of competence as well as by reducing parental stress. In addition, we tested the effectiveness of VIPP-AUTI on some child developmental outcomes as reported in **chapter 5**. We hypothesized a therapeutic effect of VIPP-AUTI on children's joint attention and play skills.

Alongside the study of effects of VIPP-AUTI on parent-child interactions, we focused on neurobiological mechanisms related to early parent-child interactions. The neuropeptide oxytocin is associated with nurturing and parenting by both mothers and fathers (Carter & Porges, 2013, Feldman et al., 2012) and was found to enhance the father's sensitivity during play-interaction with his child (Naber et al., 2010). In **chapter 6** we describe the results of a double-blind, placebo-controlled, within-subject experiment on the effects of intranasally administered oxytocin to fathers of children with ASD. The objective was to investigate whether paternal sensitivity increases during play interaction with their children with ASD after intranasal administration of oxytocin, similar to the effect on fathers of typically-developing children. Finally, in **chapter 7**, we summarize and discuss the findings of the various studies described in this thesis, and we offer some recommendations for implementing the VIPP-AUTI program.

References

American Psychiatric Association (2013). *Diagnostic and Statistical Manual of Mental Disorders (5ᵗʰ ed.)*. Washington DC.

American Psychiatric Association. (2000). *Diagnostic and statistical manual of mental disorders* (4ᵗʰ ed., Tekst Revision).* Washington DC

Baghdadli, A., Picot, M., Michelon, C., Bodet, J., Pernon, E., Burstezjn, C., et al. (2007). What happens to children with PDD when they grow up? prospective follow-up of 219 children from preschool age to mid-childhood. *Acta Psychiatrica Scandinavica, 115*(5), 403-412.

Baird, G., Simonoff, E., Pickles, A., Chandler, S., Loucas, T., Meldrum, D., et al. (2006). Prevalence of disorders of the autism spectrum in a population cohort of children in south thames: The special needs and autism project (SNAP). *Lancet, 368*(1474-547; 9531), 210-215.

Bakermans-Kranenburg, M. J., van IJzendoorn, M. H., & Juffer, F. (2003). Less is more: Meta-analyses of sensitivity and attachment interventions in early childhood. *Psychological Bulletin, 129*(2), 195-215.

Barbaro, J., & Dissanayake, C. (2009). Autism spectrum disorders in infancy and toddlerhood: A review of the evidence on early signs, early identification tools, and early diagnosis. *Journal of Developmental and Behavioral Pediatrics : JDBP, 30*(5), 447-459.

Benjak, T., Vuletic Mavrinac, G., & Pavic Simetin, I. (2009). Comparative study on self-perceived health of parents of children with autism spectrum disorders and parents of non-disabled children in Croatia. *Croatian Medical Journal, 50*(4), 403-409.

Birney, E., Stamatoyannopoulos,J.A., Dutta, A., Guigó, R., Gingeras, T.R., Margulies,E.H., et al. (2007). Identification and analysis of functional elements in 1% of the human genome by the ENCODE pilot project. *Nature, 447.* doi: 10.1038/nature05874

Carter, C., & Porges, S.W. (2013). The biochemistry of love: An oxytocin hypothesis. *EMBO Reports, 14*(1), 12-16.

Chakrabarti, S., & Fombonne, E. (2001). Pervasive developmental disorders in preschool children. *JAMA : The Journal of the American Medical Association, 285*(24), 3093-3099.

Chakrabarti, S., & Fombonne, E. (2005). Pervasive developmental disorders in preschool children: Confirmation of high prevalence. *The American Journal of Psychiatry, 162*(6), 1133-1141.

Charman, T. (2011). *The highs and lows of counting autism. The American Journal of Psychiatry, 168(9)*, 873-875.

Dabrowska, A., & Pisula, E. (2010). Parenting stress and coping styles in mothers and fathers of pre-school children with autism and down syndrome. *Journal of Intellectual Disability Research, 54*, 266-280.

Davies M. (ed.) (2001). *The Blackwell encyclopaedia of social work (reprint).* Oxford: Wiley-Blackwell.

Davis, N. O., & Carter, A. S. (2008). Parenting stress in mothers and fathers of toddlers with autism spectrum disorders: Associations with child characteristics. *Journal of Autism and Developmental Disorders, 38*(7), 1278-1291.

Dawson, G. Hill, D. Spencer, A. Galpert, L., & Watson, L. (1990). Affective exchanges between young autistic children and their mothers. *Journal of Abnormal Child Psychology, 18*(3), 335-345.

Dawson, G., Rogers, S., Munson, J., Smith, M., Winter, J., Greenson, J., et al. (2010). Randomized, controlled trial of an intervention for toddlers with autism: The early start Denver model. *Pediatrics, 125*(1), e17-23.

Dawson, G. J., Merkle, E.J.M., Venema, K., Lowy, K., Faja, R., Kamara,S., et al. (2012). Early behavioral intervention is associated with normalized brain activity in young children with autism. *Journal of the American Academy of Child & Adolescent Psychiatry, 51*(11), 1150-1159.

De Wolff, M. S., & Van IJzendoorn, M. H. (1997). Sensitivity and attachment: A meta-analysis on parental antecedents of infant attachment. *Child Development, 68*(4), 571-591.

Demerouti, E., Peeters, M. C. W., & Van der Heijden,B. I. J. M. (2012). Work-family interface from a life and career stage perspective: The role of demands and resources. *International Journal of Psychology, 47*(4), 241-258.

Doussard-Roosevelt, J. A., Joe, C. M., Bazhenova, O. V., & Porges, S. W. (2003). Mother-child interaction in autistic and nonautistic children: Characteristics of maternal approach behaviors and child social responses. *Development and Psychopathology, 15*(2), 277-295.

Dunn, M. E., Burbine, T., Bowers, C. A., & Tantleff-Dunn, S. (2001). Moderators of stress in parents of children with autism. *Community Mental Health Journal, 37*(1), 39-52.

Eisenhower, A. S., Baker, B. L., & Blacher, J. (2005). Preschool children with intellectual disability: syndrome specificity, behavior problems, and maternal well-being. *Journal of Intellectual Disability Research : JIDR, 49*(Pt 9), 657-671.

Erba, H. W. (2000). Early intervention programs for children with autism: Conceptual frameworks for implementation. *American Journal of Orthopsychiatry, 70*(1), 82-94.

Feldman, R., Zagoory-Sharon, O., Weisman, O., Schneiderman, I., Gordon, I., Maoz, R., et al. (2012). Sensitive parenting is associated with plasma oxytocin and polymorphisms in the OXTR and CD38 genes. *Biological Psychiatry, 72*(3), 175-181.

Fombonne, E. (2003a). The prevalence of autism. *JAMA : The Journal of the American Medical Association, 289*(0098-7484; 1), 87-89.

Fombonne, E. (2003b). Epidemiological surveys of autism and other pervasive developmental disorders: An update. *Journal of Autism and Developmental Disorders, 33*(4), 365-382.

Fombonne, E. (2009). Epidemiology of pervasive developmental disorders. *Pediatric Research, 65*(6), 591-598.

Gervais, H., Belin, P., Boddaert, N., Leboyer, M., Coez, A., Sfaello, I., et al. (2004). Abnormal cortical voice processing in autism. *Nature Neuroscience, 7*(8), 801-802.

Green, V. A., Pituch, K. A., Itchon, J., Choi, A., O'Reilly, M., & Sigafoos, J. (2006). Internet survey of treatments used by parents of children with autism. *Research in Developmental Disabilities, 27*(1), 70-84.

Higgins, D. J., Bailey, S. R., & Pearce, J. C. (2005). Factors associated with functioning style and coping strategies of families with a child with an autism spectrum disorder. *Autism : The International Journal of Research and Practice, 9*(2), 125-137.

Hollander, E., Phillips, A., & Yeh, C. (2003). Targeted treatments for symptom domains in child and adolescent autism. *Lancet, 362*(9385), 732-734.

Howlin, P., Goode, S., Hutton, J., & Rutter, M. (2004). Adult outcome for children with autism. *Journal of Child Psychology and Psychiatry, 45*(2), 212-229.

Johnson, M. H. (2001). Functional brain development in humans. *Nature Reviews Neuroscience, 2*(7), 475-483.

Juffer, F., Bakermans-Kranenburg, M. J., & Van IJzendoorn, M. H. (2009). Attachment-based interventions: Heading for evidence-based ways to support families. *ACAMH Occasional Papers, 29*, 47-57.

Kasari, C., Sigman, M., Mundy, P., & Yirmiya, N. (1988). Caregiver interactions with autistic children. *Journal of Abnormal Child Psychology, 16*(1), 45-56.

Kim, Y. S., Leventhal, B. L., Koh, Y. J., Fombonne, E., Laska, E., Lim, E. C., et al. (2011). Prevalence of autism spectrum disorders in a total population sample. *The American Journal of Psychiatry, 168*(9), 904-912.

Koegel, R. L., Schreibman, L., Loos, L. M., Dirlich-Wilhelm, H., Dunlap, G., Robbins, F. R., et al. (1992). Consistent stress profiles in mothers of children with autism. *Journal of Autism and Developmental Disorders, 22*(2), 205-216.

Lauritsen, M., & Ewald, H. (2001). The genetics of autism. *Acta Psychiatrica Scandinavica, 103*(0001-690; 6), 411-427.

Levy, S. E., & Hyman, S. L. (2008). Complementary and alternative medicine treatments for children with autism spectrum disorders. *Child and Adolescent Psychiatric Clinics of North America, 17*(4), 803-20.

Levy, S. E., Mandell, D. S., & Schultz, R. T. (2009). Autism. *Lancet, 374*(9701), 1627-1638.

Lord, C. (2011). Epidemiology: How common is autism? *Nature, 474*(7350), 166-168.

Makrygianni, M. K., & Reed, P. (2010). A meta-analytic review of the effectiveness of behavioral early intervention programs for children with autistic spectrum disorders. *Research in Autism Spectrum Disorders, 4*(4), 577-593.

McConachie, H., & Diggle, T. (2007). Parent implemented early intervention for young children with autism spectrum disorder: A systematic review. *Journal of Evaluation in Clinical Practice, 13*(1), 120-129.

Murray, L., FioriCowley, A., Hooper, R., & Cooper, P. (1996). The impact of postnatal depression and associated adversity on early mother-infant interactions and later infant outcome. *Child Development, 67*(5), 2512-2526.

Murray, C., & Greenberg, M. (2006). Examining the importance of social relationships and social contexts in the lives of children with high-incidence disabilities. *Journal of Special Education, 39*(4), 220-233.

Naber, F., IJzendoorn van, M. H., Deschamps, P., Engeland van, H., & Bakermans-Kranenburg, M. J. (2010). Intranasal oxytocin increases fathers' observed responsiveness during play with their children: A double-blind within-subject experiment. *Psychoneuroendocrinology,35*, 1583-1586.

New York State Department of Health Clinical Practice Guideline (1999). *Quick Reference Guide for parents and professionals:* a*utism/pervasive developmental disorders, assessment and intervention for young children (age 0-3 years).* http://www.health.ny.gov/publications/4216.pdf

Nicolson, R., & Szatmari, P. (2003). Genetic and neurodevelopmental influences in autistic disorder. *Canadian Journal of Psychiatry.Revue Canadienne De Psychiatrie, 48*(8), 526-537.

Noens, I. L., & van Berckelaer-Onnes, I. A. (2005). Captured by details: Sense-making, language and communication in autism. *Journal of Communication Disorders, 38*(2), 123-141.

Nordin, V., & Gillberg, C. (1998). The long-term course of autistic disorders: Update on follow-up studies. *Acta Psychiatrica Scandinavica, 97*(2), 99-108.

Oono, I., Honey,E., & McConachie, H. (2013). Parent-mediated early intervention for young children with autism spectrum disorders (ASD). *Cochrane Database of Systematic Reviews, 4*, CD009774-CD009774. doi:10.1002/14651858.CD009774.pub2

Ospina, M. B., Krebs Seida, J., Clark, B., Karkhaneh, M., Hartling, L., Tjosvold, L., et al. (2008). Behavioral and developmental interventions for autism spectrum disorder: A clinical systematic review. *PloS One, 3*(11), e3755.

Palmen, S. J. M. C., Van Engeland, H., Hof, P. R., & Schmitz, C. (2004). Neuropathological findings in autism. *Brain, 127*, 2572-2583.

Peters-Scheffer, N., Didden, R., Korzilius, H., & Sturmey, P. (2011). A meta-analytic study on the effectiveness of comprehensive ABA-based early intervention programs for children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 5*(1), 60-69.

Ploog, B. O. (2010). Stimulus overselectivity four decades later: A review of the literature and its implications for current research in autism spectrum disorder. *Journal of Autism and Developmental Disorders, 40*(11), 1332-1349.

Posserud, M., Lundervold, A. J., Lie, S. A., & Gillberg, C. (2010). The prevalence of autism spectrum disorders: Impact of diagnostic instrument and non-response bias. *Social Psychiatry and Psychiatric Epidemiology, 45*(3), 319-327.

Rice ,C. (2009). Prevalence of autism spectrum disorders - Autism and Developmental Disabilities Monitoring Network, United States, 2006. *MMWR.Surveillance Summaries : Morbidity and Mortality Weekly Report.Surveillance Summaries / CDC, 58*(10), 1-24.

Reichow, B., Barton, E.E., Boyd, B. A., & Hume, K. (2012). Early intensive behavioral intervention (EIBI) for young children with autism spectrum disorders (ASD). *Cochrane Database of Systematic Reviews, 10*, CD009260-CD009260. doi:10.1002/14651858.CD009260.pub2

Rogers, S. J., & Vismara, L. A. (2008). Evidence-based comprehensive treatments for early autism. *Journal of Clinical Child and Adolescent Psychology : The Official Journal for the Society of Clinical Child and Adolescent Psychology, American Psychological Association, Division 53, 37*(1), 8-38.

Rumsey, J. M., & Ernst, M. (2000). Functional neuroimaging of autistic disorders. *Mental Retardation and Developmental Disabilities Research Reviews, 6*(3), 171-179.

Schieve, L. A., Blumberg, S. J., Rice, C., Visser, S. N., & Boyle, C. (2007). The relationship between autism and parenting stress. *Pediatrics, 119 Suppl 1*, S114-21.

Schwichtenberg, A., & Poehlmann, J. (2007). Applied behavior analysis: Does intervention intensity relate to family stressors and maternal well-being? *Journal of Intellectual Disability Research, 51*(8), 598-605.

Sivberg, B. (2002). Family system and coping behaviors: A comparison between parents of children with autistic spectrum disorders and parents with non-autistic children. *Autism : The International Journal of Research and Practice, 6*(4), 397-409.

Strauss, K., Mancini, F., & Fava, L. (2013). Parent inclusion in early intensive behavior interventions for young children with ASD: A synthesis of meta-analyses from 2009 to 2011. *Research in Developmental Disabilities, 34*(9), 2967-2985.

Van Daalen, E., Kemner, C., Dietz, C., Swinkels, S. H., Buitelaar, J. K., & Van Engeland, H. (2009). Inter-rater reliability and stability of diagnoses of autism spectrum disorder in children identified through screening at a very young age. *European Child & Adolescent Psychiatry, 18*(11), 663-674.

Van der Zwaag, B., Franke, L., Poot, M., Hochstenbach, R., Spierenburg, H. A., Vorstman, J. A., et al. (2009). Gene-network analysis identifies susceptibility genes related to glycobiology in autism. *PloS One, 4*(5), e5324.

Van Engeland, H., & Buitelaar, J. K. (2009). In *Rutter's Child and Adolescent Psychiatry (5th ed.),* Rutter, M., Bishop, D.V.M., Pine, D.S., Scott, S., Stevenson, J., Taylor E., & Thapar, A. (Eds). Blackwell Publishing Ltd. doi:10.1002/9781444300895.ch46

Van IJzendoorn, M. H., Rutgers, A. H., Bakermans-Kranenburg, M. J., Swinkels, S. H., van Daalen, E., Dietz, C., et al. (2007). Parental sensitivity and attachment in children with autism spectrum disorder: comparison with children with mental retardation, with language delays, and with typical development. *Child Development, 78*(2), 597-608.

Virues-Ortega, J. (2010). Applied behavior analytic intervention for autism in early childhood: Meta-analysis, meta-regression and dose-response meta-analysis of multiple outcomes. *Clinical Psychology Review, 30*(4), 387-399.

Volkmar, F. R., Lord, C., Bailey, A., Schultz, R. T., & Klin, A. (2004). Autism and pervasive developmental disorders. *Journal of Child Psychology and Psychiatry, and Allied Disciplines, 45*(0021-9630; 1), 135-170.

Vorstman, J. A., van Daalen, E., Jalali, G. R., Schmidt, E. R., Pasterkamp, R. J., de Jonge, M., et al. (2011). A double hit implicates DIAPH3 as an autism risk gene. *Molecular Psychiatry, 16*(4), 442-451.

Weiss, L. A., & Arking, D. E., Gene Discovery Project of Johns Hopkins & the Autism Consortium, Daly, M. J., & Chakravarti, A. (2009). A genome-wide linkage and association scan reveals novel loci for autism. *Nature, 461*(7265), 802-808.

Weiss, M. J. (2002). Hardiness and social support as predictors of stress in mothers of typical children, children with autism, and children with mental retardation. *Autism: The International Journal of Research and Practice, 6*(1), 115-130.

Williams, J. G., Higgins, J. P., & Brayne, C. E. (2006). Systematic review of prevalence studies of autism spectrum disorders. *Archives of Disease in Childhood, 91*(1), 8-15.

Wing, L., & Potter, D. (2002). The epidemiology of autistic spectrum disorders: Is the prevalence rising? *Mental Retardation and Developmental Disabilities Research Reviews, 8*(1080-4013; 3), 151-161.

Zwaigenbaum, L., Bryson, S., Rogers, T., Roberts, W., Brian, J., & Szatmari, P. (2005). Behavioral manifestations of autism in the first year of life. *International Journal of Developmental Neuroscience : The Official Journal of the International Society for Developmental Neuroscience, 23*(2-3), 143-152.

Chapter 2

Parental reaction to early diagnosis of their children's Autism Spectrum Disorder: an exploratory study

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Abstract

This study explores parental reactions subsequent to receiving their child's ASD-diagnosis. Seventy seven parents of recently diagnosed children participated in the Reaction to Diagnosis Interview. Within this group, associations between parental reaction to diagnosis, parental and child characteristics and prediagnostic circumstances were analyzed. In a sub-sample, the stability of reaction to diagnosis was examined.

The majority of parents were classified as 'resolved' regarding their child's diagnosis. Conversely, parents of children with more severe ASD symptoms or Non–Dutch parents were more likely to be classified as 'Unresolved'. Sub-sample analysis revealed stability of reaction to ASD-diagnosis.

The majority of parents adapted well to the circumstances and the care for their child. Autism severity and parental nationality were significant factors affecting parental reactions. Thus, early identification of parental reaction to children's ASD-diagnosis may aid in providing more tailored parental support programs.

Introduction

Autism or Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder characterized by atypical and delayed development of reciprocal social interaction and communication, and the presence of stereotyped or restricted behaviors and interests (American Psychiatric Association [APA], 2013). The onset of ASD is before the child's age of 36 months, followed by a chronic course.

Parenting a child with autism is found to be more stressful than parenting typically developing children or children with mental retardation (Benjak et al., 2009; Eisenhower et al., 2005; Higgins et al., 2005; Koegel et al., 1992; Weiss, 2002). This parental burden is especially associated with children's social deficits (Davis & Carter, 2008) and increases even further during times when children need treatment (Schieve et al., 2007). After diagnosis, parents use several interventions (Green et al., 2006). Although the number of interventions reduces over time, parents experience permanent stress (Osborne et al., 2008). Particularly, maternal stress may increase over time, associated with

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more (social) environmental demands when the child becomes older (Rutgers et al., 2007; Warfield et al., 1999). Parental coping style appears to be an important moderator of stress in parents of children with ASD. While lack of coping leads to negative parental outcome, such as depression, isolation and spousal relation problems, active problem solving was found to reduce stress (Dabrowska & Pisula, 2010; Dunn et al., 2001; Sivberg, 2002). Parental coping style is associated with parent's personality, which was found to be mediated by parental efficacy (De Haan et al., 2009). Parental efficacy refers to Bandura's theory of self-efficacy with respect to parenting, meaning parents' perceived confidence in their ability to raise their child successfully (Bandura, 1995). Greater parental efficacy was found to increase parents' adjustment of the challenging demands while raising their child with ASD (Bekhet et al., 2012; Pakenham et al., 2004). Adjustment, including rearranging daily life was found a continuous activity of parents in order to cope and to adapt to their child's deficits associated with ASD (Luong et al., 2009). Therefore, parent's experiences of rearing stress, their feelings of efficacy, the child's severity of symptomatology and its age are expected to be associated with parental coping strategies to their child's ASD.

Regardless various coping strategies, levels of stress and timing to adapt to the child's disabilities, a commonality is that parents' hopes and expectations for their child's development and future functioning are challenged by the diagnosis of ASD. Former expectations need to be changed into new, more realistic ones (Luther et al., 2005). Letting go of the old expectations can cause emotional pain and has been compared to a process of grieving. Marvin and Pianta (1996) describe the expressed feelings of such grieving as (non)resolution with respect to the diagnosis of an atypically developing child. The concept of 'resolution of diagnosis' is based on attachment theory and research (Bowlby, 1969; Cassidy & Shaver, 2008; Marvin & Pianta, 1996). Parents who come to terms with their child's diagnosis manage to control their emotions and deal with the new situation of their child's condition and their own parenthood. Parents with a lack of resolution find it difficult to change their hopes and believes, which results in permanent grieving (Marvin & Pianta, 1996). Resolution of diagnosis allows integration of the parental experience into an adequate way of caregiving (Marvin &

Pianta, 1996; Oppenheim et al., 2009). Adequate caregiving has been associated with parental sensitivity; that is the extent to which parents adequately respond to the needs of the child. Parental sensitivity in turn is associated with securely attached children (Bakermans-Kranenburg et al., 2003; De Wolff & Van IJzendoorn, 1997; Oppenheim et al, 2009). Both the deviant development of the child, as well as the grief of the parents related to the diagnosis itself can interfere with adequate caregiving.

Parental resolution of the child's diagnosis can be assessed with the 'Reaction to Diagnosis Interview' (RDI; Marvin & Pianta, 1996). RDI-outcomes are divided into two main classifications: 'Resolved' or 'Unresolved' (Barnett et al., 2006; Kearney et al., 2011; Pianta et al., 1996). A resolved classification indicates successful adaptation and coping, having moved on from the crisis of the diagnosis. On the contrary, parents classified as unresolved show ongoing negative emotions associated with the diagnosis. Resolved and unresolved classifications are labeled in separate categories (Milshtein et al., 2010; Pianta et al., 1996; Rentinck et al., 2010; Sheeran et al., 1997). Parents classified as resolved move on with their lives by so called action, thinking or feeling oriented coping strategies. An action oriented coping strategy is characterized by supporting the child's functioning through direct action, such as arranging resources or adapting routines of caregiving which emphasize the child's special needs. Thinking oriented parents focus on knowing and understanding the child's condition, while feeling oriented parents demonstrate balanced emotions with respect to the experience. In contrast, parents with an unresolved main classification on the RDI show little or no change in response to the child's diagnosis. They seem to be stuck in negative emotions which obstruct adaptation of the diagnosis and demonstrate inadequate coping strategies, e.g. blaming others, false believes of the child's condition, or simply ignoring what is going on.

To our knowledge, only a few studies used the RDI when a child was diagnosed with ASD (Milshtein et al., 2010; Oppenheim et al., 2009; Wachtel & Carter, 2008). Sample sizes of these studies range from 45 to 63 children. All three studies used a cross sectional design. Associations were found between resolution of diagnosis

and optimal maternal interaction style (Wachtel & Carter, 2008) or between resolution of diagnosis and children's security of attachment (Oppenheim et al., 2009). Similar findings were described in previous studies of children who were diagnosed with other developmental disabilities. However, no associations were found between parents' classifications as resolved and child characteristics (age, gender, autism severity, mental age, daily living skills), parental characteristics (age, gender, IQ, traits of broad autism phenotype, maternal depressive symptoms, social economic status), or the time interval between the child's diagnosis and the RDI-assessment (Milshtein et al., 2010; Wachtel & Carter, 2008). Nevertheless, the mean duration of time since parents received their child's diagnosis and RDI-assessment was rather long, ranging from nearly ten months to more than four years. Moreover, these studies lack information on stability of RDI-results over time. Two previous longitudinal studies looking at children with 1) neurological or disfigurement congenital disorders and 2) cerebral palsy only, demonstrated predominant stable outcomes at the main classifications of parental reaction to children's diagnoses (Barnett et al., 2006; Rentinck et al., 2010) and changes with respect to RDI-subclassifications (Rentinck et al., 2010). In these diagnostic populations, associations were found between RDI-outcomes and the type or severity of diagnosis. Unresolved classifications of the RDI were found overrepresented in parents of children with a neurological diagnosis and lower mental ability (Barnett et al., 2006) and in parents of children with more severe forms of cerebral palsy (Schuengel et al., 2009).

In families of children with autism, the period between noticing deviant development in the child and receiving the ASD-diagnosis may be critical to parental grief and parents' coping strategies. Parents who report first suspicions of their child's developmental problems are more likely to receive early diagnosis than parents without concerns (Glascoe & Dworkin, 1995; Mandell et al., 2005) and therefore may be more able to cope with the diagnosis. Parents benefit from early diagnosis, especially when the delay between parents' first suspicions and time of receiving a final ASD-diagnosis is minimal (Renty & Roeyers, 2006). However, a significant diagnostic delay of more than two years

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is found (Dietz et al., 2007; Young et al., 2003). It is not clear how this diagnostic delay contributes to parents' reactions to the diagnosis. After screening, the parental compliance to professional advice varies greatly from early compliance (when the child is about two years old) to late and non-compliance (when the child is younger than 18 months) (Dietz et al., 2007). These findings suggest that some parents need time to accept the possibility of developmental problems.

The aims of the present study were to explore associations between RDI-classifications and parent and child characteristics as well as prediagnostic circumstances, as soon as possible after receiving the child's ASD-diagnosis. We hypothesized that severe autism symptomatology may obstruct parents in their resolution to the child's diagnosis, especially at the critical moment of receiving it. In addition, we expected that parents who identified their child's autism characteristics in early childhood themselves may be more likely to cope in an adequate, supportive way, than parents who did not recognize their child's special needs until receiving the ASD-diagnosis. Secondary, in a subsample of parents who continued to use regular care after the diagnostic phase, we explored stability of parental reaction to diagnosis over time.

Method

Procedure
Parents were invited to participate directly after receiving their child's diagnosis of ASD, and as part of a larger, randomized controlled intervention study. Parental reaction to diagnosis was assessed at baseline before randomization. Parents were asked to complete questionnaires to collect data on parent and child characteristics. Demographic data were collected during the diagnostic phase (e.g. parental age, marital status, postal area, education and occupation). After seven months, a follow up assessment of reactions to their children's diagnosis was acquired from parents who participated under control conditions, receiving usual care. We excluded longitudinal data of parents who received the experimental intervention, because of potential intervention effects. The experimental intervention was

a standardized attachment-based program, which might influence parents' reaction to the child's diagnosis, because of improved parent-child interactions. Participants were included from June 2008 to April 2012. Both parents of the child signed written informed consent before participation. The Medical Ethics Review Board of the University Medical Center Utrecht in the Netherlands (UMC Utrecht) approved the study protocol.

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Care as Usual

After parents received an ASD diagnosis of their child at the Department of Psychiatry of the UMC Utrecht, nursing care is usually offered. Nursing care consists of home training, regarding parents' questions about practical issues, and simultaneously parental guidance at the hospital. The number of home-visits and sessions at the hospital averaged about five visits, but they vary based on parent's preferences. In addition, group meetings with parents were provided for psychoeducation about ASD in general. The psychiatrist could prescribe medication to treat comorbid problems by indication. Parents can also receive support from external care providers.

Participants

Seventy-seven primary caregivers (90% mothers) of a child diagnosed with ASD (86% boys) at the UMC Utrecht were interviewed. Their employment ranged from 0-50 weekly hours (M = 19.36; SD = 13.97). The majority of families (96%) were assigned a moderate to high level of SES. Of the parents 82% were married, 8% were divorced and 10% were a single mother. Children's age ranged from 16 to 61 months (M = 42.87; SD = 9.99). More than half of the children were first born (60%). Children's developmental level ranged from mental retardation to high functioning. In 53% of this sample, the interval between child's chronological age and age equivalent of language development were found more than 6 months. Twelve percent of the children were raised bilingually. Children were diagnosed as having ASD by a board certified child psychiatrist according to extensive developmental history, all medical files, a semi structured observation (Autism Diagnostic Observation Schedule – Generic) and classification

of the Diagnostic and Statistical Manual of Mental Disorders 4th Edition-Text Revision (DSM-IV-TR; APA, 2000). The DSM-IV-TR includes Pervasive Developmental Disorders; Autistic Disorder (AD) and Pervasive Developmental Disorders Not Otherwise Specified (PDD-NOS), and other ASDs; Asperger's Disorder, Rett's Disorder and Childhood Disintegrative Disorder. Of the children, 67% were diagnosed with AD, and 33% were diagnosed with PDD-NOS (see Table1 and Table 2).

At the first assessment, the duration of time since receiving the diagnosis was five weeks on average (M = 5.19; SD = 4.14). The majority of children received ASD (with or without comorbid mental retardation) as the first and only diagnosis. Next to the ASD diagnosis, some children (36%) received a diagnosis or a combination of diagnoses (11% of the 36%) regarding somatic conditions; lung diseases (21%), sensory deficits (12% auditory and 6% visually problems), epilepsy (6%), growth disorder (4%), congenital heart muscles disease (3%), metabolic disease (1%).

Table 1 Child and Parent Characteristics

Variable (N=77)	Range	Mean	SD
Child characteristics			
Child's autistic characteristics	8-47	26.12	9.80
Child's developmental level	49-124	73.69	22.03
Child's language comprehension age equivalent (months)	7-75	32.45	16.91
Child's language production age equivalent (months)	7-75	32.08	16.07
Parent characteristics			
Mother's age (years)	25-52	36.03	4.83
Father's age (years)	25-56	38.39	6.19
Primary caregiver's age (years)	25-52	36.55	5.04
Child and parent questionnaires outcomes			
Child's challenging behaviors (ABC)	7-136	52.64	25.13
Mother's autistic traits (AQ)	2-39	13.31	7.48
Father's autistic traits (AQ)	4-35	14.63	6.20
Primary caregiver's autistic traits (AQ)	2-39	13.62	7.13
Parental personal efficacy	-6-44	22.58	10.61
Parental stress (PDH)	3- 53	23.18	13.10

Note. ABC: Aberrant Behavior Checklist; AQ: Autism Quotient; PDH: Parental Daily Hassles.

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Table 2 Sociodemographics

	Mother % (n)	Father % (n)	Primary caregiver % (n)
Dutch nationality	87% (67)	91% (70)	90% (69)
Low level of education (< 2nd degree)	14% (11)	23% (18)	14% (11)
Moderate level of education (2nd degree)	44% (34)	40% (31)	48% (37)
High level of education (bachelor/ master degree)	42% (32)	33% (25)	38% (29)
Education unknown		4% (3)	

Note. Not Dutch nationality: Chinese, Colombian, Israeli, Irish, French, Moroccan, Nigerian, Russian, Somalia, Turkish.

Instruments

Autism Diagnostic Observation Schedule - Generic (ADOS – G)
The ADOS-G (Lord et al., 2000) offers the opportunity to quantify deficits across the autism spectrum, controlling for effects of language and cognitive delay, in individuals with significant impairments. ADOS-G total score is the cumulative score of five test domains: communication, social reciprocity, play, stereotypic behavior and other problems. A high ADOS-G total score refers to severe autistic symptoms. The ADOS-G consists of four modules. In the current study, children were assessed using module 1 ($n = 42$) and module 2 ($n = 35$), based on their individual level of expressive language. The child psychiatrist, who was certificated for ADOS-reliability, administered the ADOS-G, during the diagnostic phase, prior to this study.

Mullen Scales of Early Learning (MSEL)
The MSEL (Mullen, 1995) is a standardized developmental test that yields a mental age score for children between three and 68 months of age. The MSEL consists of five test domains, one measuring cross motor skills (not administered in the current study), and four cognitive domains: 1) visual reception, 2) fine motor skills, 3) receptive language and 4) expressive language. The final early learning composite standard

score is a combination of non-verbal (domain 1 and 2) and verbal composites (domain 3 and 4), which ranges from low to high cognitive functioning. The test was administered by a trained clinical psychologist at baseline assessment.

Language development
The Reynell test for Dutch language comprehension (Van Eldik et al. 1995) and the Schlichting test for Dutch language production (Schlichting et al., 1995) were used in individual tests, administered by a trained psychologist or speech language therapist at baseline and follow-up assessment. The Reynell and Schlichting tests evaluate receptive and expressive language respectively, for children between 14 and 75 months of age. If children were not able to cooperate in the individual tests (37% of this sample), parental reports were collected by Dutch versions (Zink & Lejaegere, 2002; Zink & Lejaegere, 2007) of the MacArthur-Bates Communicative Developmental Inventories (CDIs) (Fenson et al., 1993; Fenson et al., 2007). CDIs consist of three forms, corresponding with different age groups; 1) 'Word and Gestures' (8-16 months of age), 2) 'Words and Sentences' (16-30 months of age) and 3) 'CDI-level III' (30-37 months of age), assessing vocabulary comprehension and production. The inventories were also used for children with impaired language development beyond the specified age ranges (Fenson et al., 1994). Its validity was shown in children with and without language delay (Heilmann et al., 2005) and confirmed in parental reports of early language development in children with autism, using CDIs (Charman et al., 2003; Luyster et al., 2007). In the current study, the level of children's language development was based on the converted age equivalents of total language comprehension scores of the Reynell or N-CDI, and total word-production scores of the Schlichting or N-CDI.

Aberrant Behavior Checklist (ABC)
The ABC (Aman et al., 1985) is a 58- item questionnaire to assess children's challenging behavior. On a 4-point rating scale, outcome ranges from no to severe challenging behaviors (scores 0 to 174). It comprises the following five factors: (1) irritability, agitation, crying; (2)

lethargy, social withdrawal; (3) stereotypic behavior; (4) hyperactivity, noncompliance; (5) inappropriate speech. In an ASD sample, factor analysis indices a moderate fit for the five factor solution (Brinkley et al., 2007).The ABC was found promising for assessing the severity of autism behaviors in early childhood (Karabekiroglu & Aman. 2009). Parents completed the questionnaire at baseline and follow-up. Internal consistency of the scale (Cronbach's alpha) was .94 ($n = 75$). Missing values were randomly scattered across items and subjects and no variable missed more than 10% of values. Mean scores within the observed group were imputed before the total set was entered for analyses.

Socio-economic status (SES)
SES was based on status scores 2010 of postal areas ($M = 0.17$; $SD = 1.16$). Status scores are derived from level of education, employment and income (The Netherlands Institute for Social Research/SCP, 2010). A higher status score refers to higher SES. In this sample, SES ranges from -1.84 to 2.23 ($M = 0.94$; $SD = 0.84$).

Parental Efficacy Questionnaire (PEQ)
The PEQ is a 22-item, self-report questionnaire on a 5-point rating scale, which ranges from low to high efficacy (scores -44 to 44). The PEQ is based on Bandura's general theory of personal efficacy (Bandura, 1997) and has been adapted for parents of young children (Van IJzendoorn et al., 1999). The instrument assesses parents' feelings of competence in child rearing, particularly when parents meet stressful circumstances (Rutgers et al., 2007). The PEQ was administered at baseline and follow-up. Cronbach's alpha for internal consistency was .86.

Parental Daily Hassles (PDH)
The PDH is a 20-item instrument developed by Crnic and Greenberg (1990) to assess the strains and stresses accompanying child rearing. It contains descriptions of typical everyday life events in parent-child interactions, which are rated by the parent on a 5-point rating scale for frequency of occurrence of the hassle (scores 0 to 80). The PDH was administered at baseline and follow-up. Cronbach's alpha for internal consistency was .90.

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Symptom Checklist (SCL-90.
The SCL-90 (Arrindell & Ettema, 1986) is a 90 item, self-report questionnaire, rated on a 5-point scale to indicate mental health status of the parents. The instrument is designed to measure agoraphobia, anxiety, depression, somatic complaints, insufficiency in thinking and acting, distrust and interpersonal sensitivity, hostility, sleeping problems and a rest-category. The total score for psychoneuroticism is based on the sum of the sub- and rest-categories. Psychoneuroticism indicates a general level of psychoneurotic-somatic complaints. In this study, the norm table of the typical population was used; ranging from very low (score 90) to very high level of complaints (score > 183) (*M* typical population = 118.28, *SD* = 32.38 (Arrindel & Ettema, 1986, pg. 107). Internal consistency of the total scale (Cronbach's alpha) was .97. Missing items on the total scale were less than 5% (a maximum of two items per subscale and no items on the subscale sleeping problems). In these cases, the (estimated) scores on each subscale were computed based on the real scores (Arrindell & Ettema, 1986, pg. 97).

Autism Quotient (AQ)
The AQ (Baron-Cohen et al., 2001) is a brief, self-administered questionnaire to screen and quantify autistic traits in adults with normal intelligence, including the following domains; 1) social skill; 2) communication; 3) imagination; 4) attention to detail and 5) attention and switching. It comprises 50 items, regarding personal preferences and habits, which are rated on a 4-point Likert scale. A high AQ score (> 32) suggests clinical levels of traits associated with the autistic spectrum. An above average score (23-31) shows some autistic traits. An average score (11-22) is based on the scores of most women (score 15) and most men (score 17) in a typical population. The Dutch translation of the AQ was found to have satisfactory internal consistency (Cronbach's alpha of the total AQ score was .71 - .81) and test-retest reliability was .78 for the total AQ score (Hoekstra et al., 2008). Cronbach's alpha for internal consistency in our sample was .88. There were no missing values (< 0.03%).

Reaction to Diagnosis Interview (RDI)

The RDI (Marvin & Pianta, 1996) is a semi-structured narrative interview intended for research use on populations of parents with chronically ill or handicapped children. This interview is designed to measure parents' reactions to and coping strategies for dealing with the diagnosis of their child with a disabling condition or illness. The interview consists of a series of questions eliciting the parent's beliefs, memories and emotional reactions to the news of their child having an illness or disability (see Table 3). The interview takes approximately 10-15 minutes. The outcome is classified in Resolved with three subclassifications: action, thinking or feeling oriented or Unresolved with six subclassifications: emotionally overwhelmed, angrily preoccupied, neutralizing, depressed/passive, cognitive distortions or disorganized/confused.

According to the RDI manual, the interviews were videotaped, transcribed and coded (Marvin & Pianta, 1996; Dutch version J. Stolk, unpublished manual). Clinimetric studies revealed adequate intercoder reliability of RDI classifications (Barnett et al., 2006; Pianta et al., 1996). In this study, the first authors (IP and FN) were trained in the coding system by a certified trainer. Intercoder agreement was 100% on the main classifications and 80 - 90% on the subclassifications. Based on 18% of random selected interviews of the current study, the inter-rater reliability on the main classifications was kappa = .87, *p* < .01 and on the subclassifications was kappa = .75, *p* < .01. Additionally, all disagreements were conferenced to unanimity of opinion, prior to entering into the dataset.

Table 3 Reaction to Diagnosis Interview based on Marvin & Pianta (1996) and specified to diagnosis of Autism Spectrum Disorders

1. Looking back, when did you first realize that (child's name) had a medical problem?

2. What were your feelings at the time of this realization?

3. Later you visited the child psychiatrist. Tell me exactly what happened when you learned of (child's name) ASD-diagnosis? Where were you, who else was there, what were your thoughts and feelings at that moment?

4. Have these thoughts and feelings changed since receiving this diagnosis?

5. Did you, to your opinion, get a clear picture of (child's name) ASD characteristics, at the moment when you received the diagnosis?

6. Can you describe (child's name) ASD characteristics at this moment? And what are your expectations of (child's name) future?

7. Parents sometimes wonder or have ideas about why they have a child with ASD. Do you have anything like that that you wonder about? (prompt if necessary: for example, some parents feel that they might have done something to contribute to their child's condition, others have a religious contemplation or considering raising a child with ASD as a special task in life. What do you wonder about?)

8. Have these considerations changed over time?

Note. ASD: Autism Spectrum Disorder.

Results

Since we included the primary caregiver of the child, our sample comprised a minority of 10% fathers ($n = 8$). Independent sample t-tests (two-sided) and chi-squared tests were performed to detect differences between male and female primary caregivers. Distributions of employment (weekly hours) revealed a difference (χ^2 (18, n = 77) = 44.29, $p < .01$) between fathers ($M = 37.04$, $SD = 11.29$) and mothers ($M = 18.34$, $SD = 13.22$). No gender differences were found in other parental characteristics and parental reports (all $p > .05$). In this sample, 8% of the primary caregivers as well as 8% of all parents reported an above average level or high level of autistic traits. Parental mental health showed an above average level of complaints, including a high level of depression

and above average levels of somatic complaints and insufficiency in thinking and acting compared to typical population levels (see table 4).

Table 4 Parental Mental Health by SCL-90

(Sub)category (N = 77)	Mean	SD	Norm
Agoraphobia	7.57	1.14	low to average
Anxiety	13.84	4.29	average
Depression	25.47	8.79	high
Somatic complaints	18.96	6.64	above average
Insufficiency in thinking and acting	15.26	6.36	above average
Distrust and interpersonal sensitivity	24.08	7.08	average
Hostility	7.93	2.32	average
Sleeping problems	4.75	2.17	average
Psychoneuroticism	129.34	35.58	above average

Note. SCL-90: Symptom Checklist, norm based on typical population.

Reaction to diagnosis and cross-sectional associations

The first assessment classified 75% (n = 58) of the parents as resolved. Proportion tests computed with Medical Calculation revealed 95% confidence interval of 64% to 84%. The proportion of parents classified as resolved was significantly higher than in previous studies: 40% of the parents in ASD-studies, $p < .01$ and 60% of the parents in all studies, $p < .01$ (see table 5).

Table 5 RDI-main classifications of various studies at first assessments

Study (1ste author& year)	Child's diagnosis	Sample	N	Resolved n	Unresolved n	Resolved %
Milshtein 2010	ASD	Mothers	61	22	39	36
Milshtein 2010	ASD	Fathers	60	30	30	50
Oppenheim 2009	ASD	Mothers	45	15	30	33
Total ASD studies				67	99	40*
Marvin 1996	CP	Primary caregivers	70	33	37	47
Pianta 1996	CP	Primary caregivers	68	31	37	46
Rentinck 2010	CP	Parents	38	31	7	82
Schuengel 2009	CP	Parents	255	208	47	82
Lord 2008	PKU	Mothers	52	36	16	69
Lord 2008	PKU	Fathers	47	36	11	77
Pianta 1996	Epilepsy	Primary caregivers	23	13	10	57
Sheeran 1997	CP or epilepsy	Mothers	97	47	50	48
Barnett 2006	Congenital disorder	Mothers	44	20	24	45
Kearney 2011	Psychiatric disorder	Mothers	33	18	15	55
Total studies				540	353	60*

Note. ASD: Autism Spectrum Disorder; CP: Cerebral Palsy; PKU: phenylketonuria; RDI: Reaction to Diagnosis Interview.
*$p < .01$ compared with 75% resolved classifications of primary caregivers in this study (proportion test with Medical Calculation).

Parents with a resolved RDI-classification were characterized mostly with an action oriented strategy (62%, *n* = 36), followed by thinking oriented (26%, *n* = 15) and feeling oriented strategies (12%, *n* = 7). Parents classified as unresolved (*n* = 19) were subclassified as neutralizing (32%, *n* = 6), emotionally overwhelmed or depressed/passive (both 26%, *n* = 5) and angrily preoccupied (16%, *n* = 3).

Independent sample *t*-tests (two-sided) and chi-squared tests demonstrated associations between children's autism severity and parental origin. Unresolved RDI-classifications were overrepresented in parents of children with higher levels of autistic symptoms, based on the ADOS-G total score (F (72, -2.00) = 0.13, p = .050). Also parents of other than Dutch nationality were classified more often unresolved than Dutch parents (χ^2 (1, n = 77) = 6.87, p = .02). Hierarchical regression was employed to determine whether child autism severity or nationality of the primary caregiver predicted parental resolution status. In the first step child autism severity contributed significantly to the prediction (5%) of parental resolution status F (1, 73) = 3.99, p = .050. At step two adding nationality of the primary caregiver to the equation, 13% of the variance was explained, F (2, 73) = 5.28, p < .01. No other associations were found between RDI-outcomes and parents' characteristics (age, level of education, marital status, SES, traits of broad autism phenotype and mental health), parental efficacy, parental daily hassles, and child characteristics (age, gender, birth order, challenging behaviors, developmental level, language age equivalents, and bilingualism). Furthermore, prediagnostic circumstances, including parental recognition (83% of the parents detected their child's problems by themselves), children's age of primary ASD-suspicions (*M* = 16.99 months, *SD* = 9.20) and the time since diagnosis revealed no associations with resolution/nonresolution of diagnosis either.

Stability of RDI-classification
After seven months, we analyzed data of parents who received care as usual and completed both RDIs (*N* = 35). A total of 83% of parents versus 77% at baseline, demonstrated resolved RDI-outcome over time. Repeated measures revealed that the majority of parents (83%, *n*= 29) continued the same RDI-classifications since the first assessment (RDI

at time 1: $M = 1.23$, $SD = 0.43$; RDI at time 2: $M = 1.17$, $SD = 0.38$, $p > .05$) (see figure 1). Four parents (11%) switched from unresolved to resolved classifications (all action oriented). Two parents (6%) switched from a resolved classification into an unresolved, depressive/passive category.
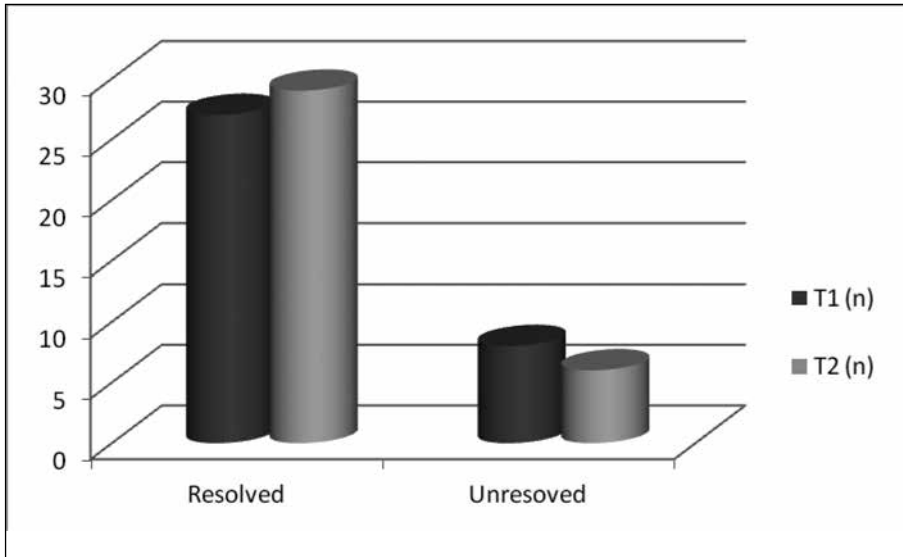
Figure 1 *Number of parents classified as resolved or unresolved at 5 weeks (T1) and 30 weeks (T2) after diagnosis*

Half of the parents ($n = 18$) at stable main category level, whether resolved or unresolved, changed subclassifications. Within the group of parents with resolved RDI-outcomes ($n = 25$) classifications of action oriented strategies lessened in favor of feeling oriented classifications (see figure 2).
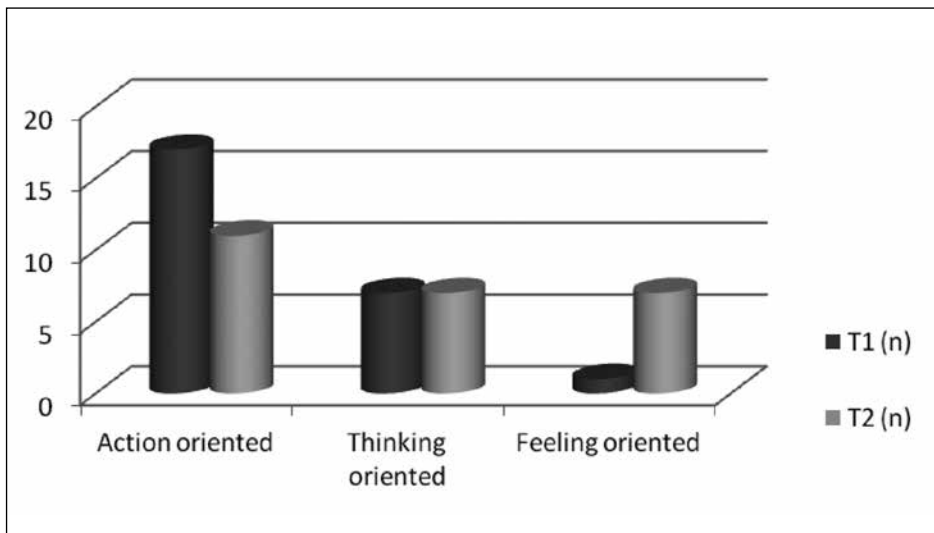
Figure 2 *Number of parents in subclassifications of continued resolved classifications (n =25), at 5 weeks (T1) and 30 weeks (T2) after diagnosis*

At the subcategory level of unresolved reports, all three parents with angrily preoccupied subclassifications changed into a different subclassification, i.e. one into 'emotionally overwhelmed', one into 'neutralizing' and one into 'cognitive distortions'. Although these subclassification-fluctuations within the classification of stable resolved/unresolved reaction to diagnosis are of interest, the number of changes in this sample is too small for further exploration.

To explore potential associations of parental coping strategies as measured by (un)changed RDI-subclassifications with severity of the child's ASD and age as well as the differences between assessments over time in parental daily hassles and reported feelings of efficacy, we divided the group in four categories: 1) remaining resolved, 2) remaining unresolved, 3) changing from unresolved into resolved, and 4) changing from resolved into unresolved. Using one-way analysis of variance, we found that the group of parents who changed from unresolved into resolved resolution showed the highest increase in parental efficacy $F(3,31) = 3.92$, $p = .02$ (see table 6).

Table 6   One way ANOVA of effects of (un)changed RDI-classifications on child autism severity, child age and differences over time of parental daily hassles and reported feelings of efficacy (N= 35)

	Group									
T1: T2:	Resolved Resolved		Unresolved Unresolved		Unresolved Resolved		Resolved Unresolved			
	Mean	SD	Mean	SD	Mean	SD	Mean	SD	F	p
ADOS-G total score	25.80	9.72	29.75	09.61	31.75	10.24	25.50	20.51	0.51	.68
Age child in months	43.40	9.92	40.12	18.23	44.41	09.14	41.49	20.22	0.12	.95
PEQ de- or increase at T2	01.75	6.77	07.62	10.00	15.00	07.94	-2.00	04.24	3.29	.02
PDH de- or increase at T2	01.71	9.19	-1.42	07.53	-9.00	11.36	0.79	08.19	1.27	.30

Note. ADOS-G: Autism Diagnostic Observation Schedule–Generic; ANOVA: analysis of variance; PDH: Parental Daily Hassles; PEQ: Parental Efficacy Questionnaire; RDI: Reaction to Diagnosis Interview; T1: baseline assessment; T2: 7-months follow-up assessment.

Discussion

In the present study, we explored associations of parents' and children's characteristics and prediagnostic circumstances with parents' reactions to diagnosis. We found that the majority of parents came to terms with their young child's ASD soon after receiving the diagnosis. Nevertheless parents of children with more severe autistic symptoms and parents with nationalities other than Dutch were more often classified as unresolved. No other associations between resolution/nonresolution and parental or child characteristics were found. Furthermore, administration of the RDI directly after receiving the diagnosis, as well as prediagnostic circumstances revealed no associations with parental resolution to diagnosis.

Additionally, we focused on short-term stability of reaction to diagnosis of parents who received usual care after the diagnostic phase. About seven months after receiving the diagnosis, most of the

parents demonstrated stable RDI-classifications. Changes of resolved RDI-outcome were found in advance of changes into unresolved classifications. Stability of parents' reaction to diagnosis seemed to be influenced by the level of parental efficacy. Overall, the main RDI-classifications remained stable while subclassifications changed over time, comparable with previous longitudinal findings of Barnett et al. (2006) and Rentinck et al. (2010).

In this study, we found a majority of parents classified as resolved. This is in line with Kandel and Merrick (2007), who report that parents of children with developmental disabilities experience positive emotions towards child rearing, despite frequent confrontations with higher levels of distress. Our finding is also consistent with broader research examining coping mechanisms in parents of children with ASD. Parents are generally capable to adapt the challenges of raising a child with ASD, using for example, social support (Rutgers et al., 2007; Twoy et al., 2007). Mobilizing family members and community members is one of the effective coping strategies parents use to manage daily stressors (Luther et al., 2005; Twoy et al., 2007). However, some parents may need more time to benefit from (in)formal support. Independently of SES, we found that parents of newly diagnosed children with other than Dutch nationality showed more often unresolved RDI-classifications. This finding cannot be explained by a specific cultural background, since this group of parents consisted of a diversity of nationalities (Chinese, Colombian, Israeli, Irish, French, Moroccan, Nigerian, Russian, Somalia and Turkish). Generally, accessibility to resources varies among countries, because of cultural and language differences, influencing parental stress experiences, related to RDI-outcomes (Lord et al., 2008; Schuengel et al., 2009). Otherwise, the level of family support may be an influencing factor, because parents originating from foreign countries may be limited in seeking informal support due to mental and physical distance to their relatives. However, the amount of non-Dutch parents in this sample is too small for scientific interpretation.

Surprisingly, the brief time-interval since parents received the diagnosis of the child and the RDI, did not result in a high proportion of unresolved RDI-scores. On the contrary, the percentage of parents classified as resolved in this study was significantly higher compared

to other studies regarding ASD (Milshtein et al., 2010; Oppenheim et al., 2009), but consistent with studies of other diagnoses of Lord et al. (2008), Rentinck et al. (2010) and Schuengel et al. (2009) (see table 5). Inconsistencies in findings between this study and other studies regarding ASD can be explained by heterogeneous samples (both parents, only mothers or primary caregivers). Also, proportions of children with AD versus PDD-NOS differ between these studies. AD is considered a more severe form of ASD than PDD-NOS. Previous studies included about 80% parents of children with AD (Milshtein et al., 2010; Oppenheim et al., 2009) in contrast with 67% parents of children with AD in our sample. Fewer children with AD may probably decrease the number of parents with nonresolution states to diagnosis. This is in line with our finding of significantly more parents who expressed unresolved feelings and thoughts of their child's newly diagnosis when ASD-symptoms were more severe. Nevertheless, regarding the association between autism severity and parental resolution, in this study the nationality of the primary caregiver seems to be a stronger predictor for the risk of coping problems with the child's ASD-diagnosis. Besides acknowledgement of autism severity or parent's nationality, to be able to detect parents at risk for unresolved reactions to their child's diagnosis, the RDI may be an important and useful instrument in clinical practice. Knowledge of parental reaction to diagnosis may advance parental support, including early intervention programs to promote adequate caregiving.

Similar to prior research (Kearney et al., 2011; Milshtein et al., 2010; Wachtel & Carter, 2008), this study revealed no other associations between parental reactions to diagnosis and (demographic) parental and child characteristics. Although parents reported above average levels of psychoneuroticism, including high levels of depression, no significant correlation was found between parent's mental health condition and reaction to ASD-diagnosis. This result confirmed findings of Wachtel and colleagues (2008), despite their modified RDI-coding system into a continuous variable with multiple dimensions versus a dichotomous variable in this study. Contrary, another study (Kearney et al., 2011) showed child rearing stress in combination with maternal depression to be predictive of unresolved RDI-classifications.

This however, was found in a heterogeneous sample of school-age children with various psychiatric disorders.

Analogous to the study of Milshtein et al. (2010), no association was found between reaction to ASD-diagnosis and parental autistic traits. Mild behavioral characteristics that resemble the deficits as seen in ASD, are often found in non-affective first-degree relatives of individuals with ASD, the so called 'broad autism phenotype' (Ghaziuddin, 2005; Klusek et al., 2012; Micali et al., 2004; Pickles et al., 2000). In particular, parents of children with ASD scored higher on AQ-scores, compared to parents of typically developing children (Bishop et al., 2004; Kose et al., 2013; Ruta et al., 2012; Wheelwright et al., 2010). In this study, the majority of parents revealed an average AQ-score. An explanation may be that the time of measuring influences this finding. During administering the RDI, parents often explained that they did not acquire a full understanding of their child's symptomatology when they received the diagnosis, despite long lasting suspicions and challenging behaviors of the child. The process of recognizing the child's autistic characteristics completely and then to extrapolate these characteristics to one's own, may need some time. In contrast, parents may underreport autistic traits, because they can interpret the AQ-items clinically (Sheeren & Stauder, 2008; Lau et al., 2013). One way or the other, parents might have slightly misjudged their own characteristics related to ASD as a usual risk of self-reporting. Also, psychometric properties of the AQ were recently discussed. Although the Cronbach's alpha of .72 for internal consistency of the AQ was found satisfactory, the values of two other instruments to assess broad autism phenotype were found somewhat higher (Cronbach's alpha of .90 and .95) (Ingersoll et al., 2011). However, the use of the AQ in our study was consistent with the previous study regarding associations between parents' broad autism phenotype and RDI-outcome (Milshtein et al., 2010). Furthermore, low AQ-scores in the current sample could be explained by the majority of mothers. Previously, mothers of children with ASD were found to report lower total AQ-scores than fathers (Hoekstra et al., 2008; Lau et al., 2013; Ruta et al., 2012; Scheeren & Stauder, 2008) and even lower or similar total AQ-scores than mothers of typical developing children (Lau et al., 2013; Scheeren & Stauder, 2008).

In a subsample of parents who received general care at the university hospital, a longitudinal pilot of parental reaction to children's ASD-diagnosis was accomplished. In line with studies of populations with other neurological disorders (Barnett et al., 2006; Rentinck et al., 2010), the majority of parents showed stability of their reactions to diagnosis. Changes of the main classification shifted to higher percentages in resolved categories than to the opposite. Furthermore, our study replicates past findings regarding changes in subclassifications with respect to children's diagnosis (Rentinck et al., 2010). The predominant prevalence of action oriented strategies may be explained by a diagnostic delay of two years on average. When parents finally receive the child's diagnosis, they may be eager to act, resulting in an action oriented classification. Additionally, several months later, when (practical) support is more balanced, parents have the opportunity to cope according to different strategies. Coping strategies may synchronize with parents' characters, which results in higher levels of thinking and feeling orientations. Other parental characteristics, such as parental efficacy are also important to consider. We found a substantial increase of parental efficacy associated with a shift from unresolved into resolved RDI-classification, while a decrease in parental efficacy over time showed an opposite change of resolved into unresolved classification. Variances of RDI-main and -subclassifications over time are of interest to detect phases within the adaptation process of parental coping with children's ASD, though the frequency of classification-changes in this study is too low for analysis. In order to understand the mechanism behind this process a qualitative study design regarding parental experience with respect to diagnosis over time may be appropriate. Qualitative research allows for in-depth interviewing (Holloway & Wheeler, 2002), which may identify important themes of parents' hopes and believes for their child's development. Also, several follow-up assessments may reveal more insight in the course of parents' reaction to diagnosis over time and to what extent parental characteristics (e.g. parental efficacy) and other factors, such as social support, may be of influence.

We studied RDI-classifications of parents in a moderate sample of 77 young children with ASD. The mean time-interval of five weeks since

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parents received their child's diagnosis and RDI-administration contrasted with the larger time-intervals of previous studies in ASD-samples. This early time-point of assessment contributes to existing findings with respect to the dynamic process of adaptation. However, several limitations of this study should be addressed. Response and selection bias may underlie the lack of association between several parental characteristics and the reaction of parents to ASD-diagnosis. Firstly, self-report questionnaires were used, which can evoke politically correct answers. Secondly, parents were invited to the RDI as part of an intervention study, which recruited generally motivated and active parents, resulting in an overrepresentation of parents with an 'action oriented' subclassification. Action oriented coping emphasizes orientation to activities and caregiving towards the child's special needs, such as the use of interventions. Finally, the majority of included parents had a moderate to high SES. Future longitudinal research is needed in a larger sample with more heterogeneous SES and demographics to examine parental reaction to their child's ASD diagnosis and its associations with aspects of adjustment, such as the level of (informal) support and phases within the process of adaptation.

Summary

Parents of children with ASD showed high levels of acceptance and active adaptation to the recent diagnosis of their child. This parental coping does not depend on experienced levels of parental stress or the presence of depressive moods. Parental reaction to diagnosis of their child is associated with the severity of children's autistic symptomatology and parent's nationality. To identify families at risk for unresolved reactions to their child's diagnosis can be of clinical relevance. Knowledge of parent's resolution/nonresolution to diagnosis can contribute to tailored parental support and intervention programming. Whether parents show resolved or unresolved RDI-classifications, the subtype categories appear to be dynamic. Future (qualitative) research is needed on parental experiences with respect to their child's ASD-diagnosis over time.

References

Aman, M.G., Singh, N.N., Stewart, A.W., & Field, C.J. (1985). Psychometric characteristics of the aberrant behavior checklist. *American Journal of Mental Deficiency, 89*(5), 492-502.

Arrindell, W.A. & Ettema, J.H.M. (1986). SCL-90. Lisse: Swets & Zeitlinger.

American Psychiatric Association (2013). Diagnostic and Statistical Manual of Mental Disorders (5th ed.). Washington DC.

American Psychiatric Association. (2000). *Diagnostic and statistical manual of mental disorders* (*4th ed., Tekst Revision).* Washington DC

Bandura, A. (1995). Self-efficacy in Changing Societies, Chapter 1: 1-45. Cambridge University Press, Chapter doi: http://dx.doi.org/10.1017/CBO9780511527692.003.

Bandura, A. (1997). Self-efficacy: The exercise of control. Freeman, New York.

Bakermans-Kranenburg, M. J., Van IJzendoorn, M. H., & Juffer, F. (2003). Less is more: meta-analyses of sensitivity and attachment interventions in early childhood. *Psychological Bulletin, 129*(2), 195-215.

Barnett, D., Clements, M., Kaplan-Estrin, M., McCaskill, J.W., Hunt, KH, Butler, C.M., et al. (2006). Maternal resolution of child diagnosis: stability and relations with child attachment across the toddler to preschooler transition. *Journal of Family Psychology : JFP : Journal of the Division of Family Psychology of the American Psychological Association (Division 43), 20*(1), 100-107.

Baron-Cohen, S., Wheelwright, S., Skinner, R., Martin, J., & Clubley, E. (2001). The autism-spectrum quotient (AQ): evidence from Asperger syndrome/high-functioning autism, males and females, scientists and mathematicians. *Journal of Autism and Developmental Disorders, 31*(1), 5-17.

Bekhet, A.K., Johnson, N.L., & Zauszniewski, J.A. (2012). Resilience in family members of persons with autism spectrum disorder: a review of the literature. *Issues Mental Health Nursing,* 33: 650-656.

Benjak, T., Vuletic Mavrinac, G., & Pavic Simetin, I. (2009). Comparative study on self-perceived health of parents of children with autism spectrum disorders and parents of non-disabled children in Croatia. *Croatian Medical Journal, 50*(4), 403-409.

Bishop, D., Maybery, M., Maley, A., Wong, D., Hill, W., & Hallmayer, J. (2004). Using self-report to identify the broad phenotype in parents of children with autistic spectrum disorders: a study using the Autism-Spectrum Quotient. *Journal of Child Psychology and Psychiatry, 45:* 1431-1436.

Bowlby, J., (1969). *Attachment and Loss: vol 1. Attachment.* New York: Basic Books.

Brinkley, J., Nations, L., Abramson, R.K., Hall, A., Wright, H.H., Gabriels, R., et al. (2007). Factor analysis of the aberrant behavior checklist in individuals with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 37*(10), 1949-1959.

Cassidy, J. & Shaver, Ph.R. (2008). *Handbook of Attachement: theory, research, and clinical implications.* (2nd ed.). New York: The Guilford Press.

Charman, T., Drew, A., Baird, C., & Baird, G. (2003). Measuring early language development in preschool children with autism spectrum disorder using the MacArthur Communicative Development Inventory (Infant Form). *Journal of Child Language, 30*(1), 213-236.

Crnic, K.A., & Greenberg, M.T. (1990). Minor parenting stresses with young children. *Child Development,* 61, 1628–1637.

Dabrowska, A., & Pisula, E. (2010). Parenting stress and coping styles in mothers and fathers of pre-school children with autism and Down syndrome. *Journal of Intellectual Disability Research, 54*, 266-280.

Davis, N.O., & Carter, A.S. (2008). Parenting stress in mothers and fathers of toddlers with autism spectrum disorders: Associations with child characteristics. *Journal of Autism and Developmental Disorders, 38*(7), 1278-1291.

De Haan, A.D., Prinzie, P., & Dekovic, M. (2009). Mothers' and fathers' personality and parenting: the mediating role of sense of competence. *Developmental Psychology, 45*: 1695-1707.

De Wolff, M. S., & Van IJzendoorn, M. H. (1997). Sensitivity and attachment: a meta-analysis on parental antecedents of infant attachment. *Child Development, 68*(4), 571-591.

Dietz, C., Swinkels, S.H., Van Daalen, E., Van Engeland, H., & Buitelaar, J.K. (2007). Parental compliance after screening social development in toddlers. *Archives of Pediatrics & Adolescent Medicine, 161*(4), 363-368.

Dunn, M.E., Burbine, T., Bowers, .C.A., & Tantleff-Dunn, S. (2001). Moderators of stress in parents of children with autism. *Community Mental Health Journal, 37*(1), 39-52.

Eisenhower, A.S., Baker, B.L., & Blacher, J. (2005). Preschool children with intellectual disability: syndrome specificity, behavior problems, and maternal well-being. *Journal of Intellectual Disability Research : JIDR, 49*(Pt 9), 657-671.

Fenson, L., Dale, P.S., Reznick, J.S., Bates E., Thal, D., Hartung, J., et al. (1993). *Guide and technical manual for the MacArthur Communicative Development Inventories.* San Diego, CA: Singular Press.

Fenson, L., Dale, P.S., Reznick, J.S., Bates, E., Thal, D.J., & Pethick, S.J. (1994). Variability in early communicative development. *Monographs of the Society for Research in Child Development, 59*(5), 1-173; discussion 174-85.

Fenson L., Marchman V.A., Thal D., Dale P.S., Bates, E., & Reznick, J.S. (2007). *The MacArthur-Bates Communicative Development Inventories: User's Guide and technical Manual (2nd edition).* Baltimore: Paul H. Brookes.

Ghaziuddin, M. (2005). A family history study of Asperger syndrome. *Journal of Autism and Developmental Disorders, 35*(2), 177-182.

Glascoe, F.P. & Dworkin, P.H. (1995). The role of parents in the detection of developmental and behavioral problems. *Pediatrics* 95: 829-836

Green, V.A., Pituch, K.A., Itchon, J., Choi, A., O'Reilly, M., & Sigafoos, J. (2006). Internet survey of treatments used by parents of children with autism. *Research in Developmental Disabilities, 27*(1), 70-84.

Heilmann, J., Weismer, S.E., Evans, J., & Hollar, C. (2005). Utility of the MacArthur-Bates communicative development inventory in identifying language abilities of late-talking and typically developing toddlers. *American Journal of Speech-Language Pathology / American Speech-Language-Hearing Association, 14*(1), 40-51.

Higgins, D.J., Bailey, S.R., & Pearce, J.C. (2005). Factors associated with functioning style and coping strategies of families with a child with an autism spectrum disorder. *Autism : The International Journal of Research and Practice, 9*(2), 125-137.

Hoekstra, R.A., Bartels, M., Cath, D.C., & Boomsma, D.I. (2008). Factor structure, reliability and criterion validity of the Autism-Spectrum Quotient (AQ): a study in Dutch population and patient groups. *Journal of Autism and Developmental Disorders, 38*(8), 1555-1566.

Holloway, I., & Wheeler, S. (2002). *Qualitative Research in Nursing (Second Edition).* Blackwell Science, Oxford.

Ingersoll, B., Hopwood, C. J., Wainer, A., & Brent Donnellan, M. (2011). A comparison of three self-report measures of the broader autism phenotype in a non-clinical sample. *Journal of Autism and Developmental Disorders, 41*(12), 1646-1657.

Karabekiroglu, K., & Aman, M.G. (2009). Validity of the aberrant behavior checklist in a clinical sample of toddlers. *Child Psychiatry and Human Development, 40*(1), 99-110.

Kandel, I., & Merrick, J. (2007). The child with a disability: parental acceptance, management and coping. *TheScientificWorldJournal, 7*, 1799-1809.

Kearney, J. A., Britner, P. A., Farrell, A. F., & Robinson, J. L. (2011). Mothers' resolution of their young children's psychiatric diagnoses: associations with child, parent, and relationship characteristics. *Child Psychiatry and Human Development, 42*(3), 334-348.

2

Klusek, J., Losh, M., & Martin, G. E. (2012). Sex differences and within-family associations in the broad autism phenotype. *Autism : The International Journal of Research and Practice,* doi:10.1177/1362361312464529.

Koegel, R.L., Schreibman, L., Loos, L.M., Dirlich-Wilhelm, H., Dunlap, G., Robbins, F.R., et al. (1992). Consistent stress profiles in mothers of children with autism. *Journal of Autism and Developmental Disorders, 22*(2), 205-216.

Kose, S., Bora, E., Erermis, S., Ozbaran, B., Bildik, T., Aydin, C. (2013). Broader autistic phenotype in parents of children with autism: Autism Spectrum Quotient-Turkish version. *Psychiatry and Clinical Neurosciences, 67*: 20-27.

Lau, W.Y., Gau, S.S., Chiu, Y.N., Wu, Y.Y., Chou, W.J., Liu, S.K. et al (2013). Psychometric properties of the Chinese version of the Autism Spectrum Quotient (AQ). *Research in Developmental Disabilities, 34:* 294-305.

Lord, B., Ungerer, J., & Wastell, C. (2008). Implications of resolving the diagnosis of PKU for parents and children. *Journal of Pediatric Psychology, 33*(8), 855-866.

Lord, C., Risi, S., Lambrecht, L., Cook, E.H., Jr., Leventhal, B.L., Dilavore, P.C., et al. (2000). The autism diagnostic observation schedule-generic: a standard measure of social and communication deficits associated with the spectrum of Autism. *Journal of Autism and Developmental Disorders, 30*, 205-223.

Luong, J., Yoder, M.K., & Canham, D. (2009). Southeast Asian parents raising a child with autism: a qualitative investigation of coping styles. *Journal of School Nursing, 25:* 222-229.

Luther, E., Canham, D., & Young Cureton, V. (2005). Coping and social support for parents of children with autism. *Journal of School Nursing, 21:* 40-47.

Luyster, R., Lopez, K., & Lord, C. (2007). Characterizing communicative development in children referred for autism spectrum disorders using the MacArthur-Bates Communicative Development Inventory (CDI). *Journal of Child Language, 34*(3), 623-654.

Mandel,l D.S., Novak, M.M.., & Zubritsky, C.D. (2005). Factors associated with age of diagnosis among children with autism spectrum disorders. *Pediatrics, 116:* 1480-1486.

Marvin, R. S., & Pianta, R. C. (1996). Mothers' reactions to their child's diagnosis: Relations with security of attachment. *Journal of Clinical Child Psychology, 25*(4), 436-445.

Micali, N., Chakrabarti, S., & Fombonne, E. (2004). The broad autism phenotype: findings from an epidemiological survey. *Autism : The International Journal of Research and Practice, 8*(1), 21-37.

Milshtein, S., Yirmiya, N., Oppenheim, D., Koren-Karie, N., & Levi, S. (2010). Resolution of the diagnosis among parents of children with autism spectrum disorder: associations with child and parent characteristics. *Journal of Autism and Developmental Disorders, 40*(1), 89-99.

Mullen, E.M. (1995). *Mullen Scales of Early Learning.* Circle Pines, Minn: American Guidance Service.

Oppenheim, D., Koren-Karie, N., Dolev, S., & Yirmiya, N. (2009). Maternal insightfulness and resolution of the diagnosis are associated with secure attachment in preschoolers with autism spectrum disorders. *Child Development, 80*(2), 519-527.

Osborne, LA, McHugh, L, Saunders, L, & Reed, P (2008). A possible contra-indication for early diagnosis of Autistic Spectrum Conditions: Impact on parenting stress. *Research in Autism Spectrum Disorders, 2*(4), 707-715.

Pakenham, K.I., Sofronoff, K., & Samios, C. (2004). Finding meaning in parenting a child with Asperger syndrome: correlates of sense making and benefit finding. *Research in Developmental Disabilities, 25:* 245-264.

Pianta, R.C., Marvin, R.S., Britner, P.A., & Borowitz, K.C. (1996). Mothers' resolution of their children's diagnosis: Organized patterns of caregiving representations. *Infant Mental Health Journal, 17*(3), 239-256.

Pickles, A., Starr, E., Kazak, S., Bolton, P., Papanikolaou, K., Bailey, A., et al. (2000). Variable expression of the autism broader phenotype: findings from extended pedigrees. *Journal of Child Psychology and Psychiatry, and Allied Disciplines, 41*(4), 491-502.

Rentinck, I.C., Ketelaar, M., Schuengel, C., Stolk, J., Lindeman, E., Jongmans, M.J., & Gorter, J.W. (2010). Short-term changes in parents' resolution regarding their young child's diagnosis of cerebral palsy. *Child: Care, Health and Development, 36*(5), 703-708.

Renty, J., & Roeyers, H. (2006). Satisfaction with formal support and education for children with autism spectrum disorder: the voices of the parents. *Child: Care, Health and Development, 32*(3), 371-385.

Ruta, L., Mazzone, D., Mazzone, L., Wheelwright, S., & Baron-Cohen, S. (2012). The Autism-Spectrum Quotient—Italian version: a cross-cultural confirmation of the broader autism phenotype. *Journal of Autism and Developmental Disorders, 42:* 625-633.

Rutgers, A.H., Van IJzendoorn, M.H., Bakermans-Kranenburg, M.J., Swinkels, S.H., Van Daalen, E., Dietz, C. et al. (2007). Autism, attachment and parenting: a comparison of children with autism spectrum disorder, mental retardation, language disorder, and non-clinical children. *Journal of Abnormal Child Psychology, 35*(5), 859-870.

2

Scheeren, A.M., & Stauder, J.E. (2008). Broader autism phenotype in parents of autistic children: reality or myth? *Journal of Autism and Deelopmental Disorders, 38:* 276-287.

Schieve, L.A., Blumberg, S.J., Rice, C., Visser, S.N., & Boyle, C. (2007). The relationship between autism and parenting stress. *Pediatrics, 119 Suppl 1*, S114-21.

Schlichting, J.E.P.T., Van Eldik, M.C.M., Spelberg, H.C., Van der Meulen, S., & Van der Meulen, B.F. (1995). *Schlichting Test voor Taalproductie*. Nijmegen: Berkhout.

Schuengel, C., Rentinck, I. C., Stolk, J., Voorman, J. M., Loots, G. M., Ketelaar, M., et al. (2009). Parents' reactions to the diagnosis of cerebral palsy: associations between resolution, age and severity of disability. *Child: Care, Health and Development, 35*(5), 673-680.

Sheeran, T., Marvin, R.S., & Pianta, R.C. (1997). Mothers' resolution of their child's diagnosis and self-reported measures of parenting stress, marital relations, and social support. *Journal of Pediatric Psychology, 22:* 197-212.

Sivberg, B. (2002). Family system and coping behaviors: a comparison between parents of children with autistic spectrum disorders and parents with non-autistic children. *Autism : The International Journal of Research and Practice, 6*(4), 397-409.

The Netherlands Institute for Social Research/SCP (2010), Retrieved from: http://www.scp.nl (accessed 20 December 2012)

Twoy, R., Connolly, P. M., & Novak, J. M. (2007). Coping strategies used by parents of children with autism. *Journal of the American Academy of Nurse Practitioners, 19*(5), 251-260.

Van Eldik, M.C.M., Schlichting, J.E.P.T., Spelberg, H.C., Van der Meulen, S. & Van der Meulen, B.F. (1995). *Reynell Test voor Taalbegrip.* Nijmegen: Berkhout.

Van IJzendoorn, M.H., Bakermans-Kranenburg, M.J., & Juffer, F. (1999). The parental efficacy questionnaire. Leiden University, Center for Child and Family Studies (Unpublished Manuscript).

Wachtel, K., & Carter, A.S. (2008). Reaction to diagnosis and parenting styles among mothers of young children with ASDs. *Autism : The International Journal of Research and Practice, 12*(5), 575-594.

Warfield, M.E., Krauss, M.W., Hauser-Cram, P., Upshur, C.C., & Shonkoff, J.P. (1999). Adaptation during early childhood among mothers of children with disabilities. *Journal of Developmental Behavioral Pediatrics 20:* 9-16.

Weiss, M.J. (2002). Hardiness and social support as predictors of stress in mothers of typical children, children with autism, and children with mental retardation. *Autism: The International Journal of Research and Practice, 6*(1), 115-130.

Wheelwright, S., Auyeung, B., Allison, C., & Baron-Cohen, S. (2010). Defining the broader, medium and narrow autism phenotype among parents using the Autism Spectrum Quotient (AQ). *Molecular Autism, 1*(1), 10.

Young, R.L., Brewer, N., & Pattison, C. (2003). Parental identification of early behavioral abnormalities in children with autistic disorder. *Autism : The International Journal of Research and Practice, 7*(2), 125-143.

Zink, I., & Lejaegere, M. (2002). *N-CDI's: Lijsten voor Communicatieve Ontwikkeling. Aanpassing en hernormering van de MacArthur CDIs van Fenson et al.* Leuven/ Leusden: ACCO.

Zink, I. & Lejaegere, M. (2007). *N-CDI 3; Aanpassing en hernormering van de MacArthur CDI: Level III van Dale et al. (2000).* Culemborg: Centraal Boekhuis BV.

2

Chapter 3

Development of a Video feedback Intervention to promote Positive Parenting for Children with Autism (VIPP-AUTI)

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Abstract

In this paper we describe the development and content of Video feedback Intervention to promote Positive Parenting for Children with Autism (VIPP-AUTI). VIPP-AUTI is an adapted version of the evidence-based intervention VIPP. The lack of social responsiveness in children with autism often lowers the quality of the parent-child interaction. A wide range of early interventions exist to cope with the disorder. The majority of early interventions for children with autism focus on their deficits of (social) skills, while the number of evidence-based interventions to improve early parent-child interaction patterns is limited. The aim of VIPP-AUTI is to enhance parental sensitivity to children's (autistic) characteristics, in order to improve child developmental outcome by increased parental support.

Introduction

Video feedback Intervention to promote Positive Parenting (VIPP) is an evidence-based intervention protocol, rooted in attachment theory and research (Juffer et al., 2008) originally developed for children at risk for insecure attachment. Over the past twenty years VIPP has been adapted and validated to be used in various (non-clinical and clinical) families, settings, and cultures (Juffer et al., 2009). For example, VIPP with an additional focus on sensitive discipline (VIPP-SD) supports parents to interact with their child not only in a sensitive way, but also to cope with difficult, oppositional child behavior (Van Zeijl et al., 2006). In adapting the VIPP for children with autism, in the range from infancy to preschool age, we complemented the original program with key ingredients for understanding and responding appropriately to symptoms of autism in parent-child interactions.

Autism and early interventions

Autism or Autism Spectrum Disorders (ASD) is a neurodevelopmental disorder characterized by impairments of social interaction and communication, combined with repetitive or restricted behaviors and interests. (American Psychiatric Association, 2013). Autism is a chronic disorder. Currently, the prevalence is estimated at 1% (Baird et al., 2006; Fombonne, 2009; Rice, 2009), though recently higher rates of ASD

up to 2% have been discussed (Charman, 2011; Kim et al., 2011; Lord, 2011). More boys than girls are affected, with a sex ratio of about four to one. Comorbidity with intellectual disability ranges from 12% to 68% (Chakrabati & Fombonne , 2005).

Parenting young children with autism did not show large differences compared to parenting in other clinical and nonclinical groups, although the challenge for parents to interpret their child's attachment needs and signals may be more difficult because of the child's deficits in reciprocal interaction (Rutgers et al., 2007). In studies involving older children with autism, their parents reported more stress than parents of typically developing children or children with mental retardation, resulting in higher levels of depression, anxiety, somatic complaints, family dysfunction and burnout (Benjak et al., 2009; Eisenhower et al., 2005; Higgins et al., 2005; Weiss, 2002). Therefore, early support for parents is needed. The aims of early intervention are to reduce ASD symptom severity, to prevent secondary (behavioral) problems, and to support family functioning. A wide range of interventions are available, using psychosocial, pharmacological and other approaches but well-designed randomized controlled studies are limited (Green et al., 2006; Ospina et al. 2008; Rogers & Vismara, 2008).

The most well-researched and effective early interventions are based on applied behavior analysis (ABA; see meta-analyses of Makrygianni & Reed, 2010; Peters-Scheffer et al., 2011; Reichow et al., 2012; Virués-Ortega 2010). Children in ABA-based groups show significantly better outcomes on cognition, language and adaptation than control groups. ABA interventions are based on operant conditioning approaches. Parent-implemented ABA interventions can improve the child's social communication behavior and parent-child interaction (McConachie & Diggle, 2007; Oono et al., 2013). For example, an ABA-based intervention showed increased joint attention and play skills together with improved parent-child interaction (Kasari et al., 2006). However, comprehensive ABA programs are expensive and the intensity of ABA programs may increase family stress, especially when parents are heavily involved (Schwichtenberg & Poehlmann, 2007).

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The VIPP-AUTI program

In a collaborative project of the Center for Child and Family Studies, Leiden University and the University Medical Center, Utrecht, we designed the intervention program Video-feedback Intervention to promote Positive Parenting for Children with Autism (VIPP-AUTI). We adapted the original VIPP to the specific needs in families of a young child with autism using knowledge of the manifest core symptoms of ASD (stereotypical or rigid behavior, deficits in early development of communication and play), together with clinical experience and knowledge of attachment-based interventions.

The original VIPP program comprises a short-term interaction-focused intervention at home, using recent videotaped parent-child interactions in daily situations such as playing together and during mealtimes (Juffer et al., 2008). The intervener does not interfere during filming to ensure that a natural parent-child interaction is observed, including the child's entire behavioral repertoire. Intervention in a natural environment is recommended to facilitate the social interactions of young children with autism (McConnell, 2002; Wallace & Rogers, 2010). The aim of VIPP-AUTI is to reduce the child's symptomatology by enhancing parental sensitivity to the autistic traits of the child. Showing parents their own interaction patterns may enhance their insight into the specific individual needs of the child with autism, and result in more optimal parental responses.

The VIPP-AUTI protocol consists of five sessions, four of which are devoted to the VIPP themes (*Attachment and Exploration, 'Speaking for the child', 'Sensitivity chain'*, and *Sharing emotions*) with an additional autism-related component for each session. The fifth session is a booster session. The additional themes for autism are (1) mastery motivation and play, (2) joint attention, (3) daily problems and routine, and (4) emotions and (stereotypical) behavior (See Table1). These themes are based on pivotal skills which in children with autism show deviant development (play and joint attention), behavior that may hinder the overall development (stereotypic behavior) and challenging behavior that parents described (daily problems). By discussing these themes based on specific video-fragments, the parents will learn to 'read' the signals of their child and will learn about the child's strengths and weaknesses.

Table 1 Themes of the VIPP-AUTI program

	Sensitivity themes of VIPP	*Adapted themes of autism*
Session 1	Attachment and Exploration	Mastery motivation and Play
Session 2	'Speaking for the child'	Joint attention
Session 3	'Sensitivity chain'	Daily problems and routine
Session 4	Sharing emotions	Emotions and (stereotypical) behavior
Session 5	Booster session	Booster session

Note. VIPP-AUTI: Video feedback Intervention to promote Positive Parenting adapted to Autism

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Session 1: Attachment and Exploration, Mastery motivation and Play

The theme of the first session of the original VIPP is exploration versus attachment behavior. During this session the focus is on showing the difference between the child's contact-seeking behavior and play, and on explaining the differential responses needed from the parent (Juffer et al., 2008). Recognizing the difference between the child's seeking proximity and exploring activities can be considered as the first step towards parental sensitivity during play. The intervener may suggest that the parent follows the child's lead in play behavior to gain contact and attention. Once engagement is established, the parent can try to move the child's interest on to other, more varied or advanced play behavior. The parent may also respond explicitly to the child's (subtle) signals of proximity/contact seeking, e.g. looking back when the child looks at the parent or commenting positively when the child prefers to play close to the parent.

In this session, video clips show the parent and child playing together with toys for pretend play, constructive play, and social play. In our study we included, in order: (1) a doll and tea set; (2) bricks for building; and (3) a book. Before starting with a new activity, the parent and the child are asked to clear up the toys from the previous activity.

Using these videotaped play interactions and clean up fragments, the video feedback focuses on the child's exploration and

attachment behavior as well as the child's autistic traits in motivation and play skills. Positive, successful interaction fragments are highlighted to demonstrate the child's orientation to the parent. Moreover typical play behaviors of the individual child are reviewed. The often limited or restricted play repertoire of children with autism is a challenge to many parents (Naber et al., 2008). Video feedback aims at increasing the parents' acceptance of the child's play behavior, and their support of the child's play, as well as at enhancing parents' recognition and accurate interpretation of the child's subtle signals to seek contact.

Vignette session 1

Session 1: attachment and exploration, mastery motivation and play

Lisa is a three-year-old girl with autism and severe delays in cognitive and speech / language development.

The first film used for video feedback showed parent and child playing together with a doll and tea set. It was divided into 14 fragments of 10 to 30 seconds each. The intervener showed the fragments while naming Lisa's behavior in a neutral way. First, Lisa focused on the tea set and together with her mother she poured pretend tea in a cup. The 18-month old brother was also interested in the toys, and he enjoyed the game as part of Lisa's natural environment. After one minute, Lisa finished playing with the doll and tea set. She watched her mother from a distance while mother and son continued playing with the tea set. Lisa started playing with a pillow, sitting next to her mother. She listened when her mother asked her to join in the tea ceremony, but she preferred to play on her own. She turned the pillow and she used her foot to roll over another toy on the floor. These pictures showed the way Lisa was seeking contact (sitting next to her mother, while watching and listening) and explored (with the pillow and her own toy). A topic of discussion was Lisa's brief attention for new toys. The mother told the intervener about Lisa's repetitive interests in the same, well- known toys. However, her mother was surprised to see Lisa's attention towards her. Perhaps because Lisa was not speaking yet and played on a modest developmental level, her mother had thought her daughter's quality of interaction was lower than she displayed on the film clips. The intervener discussed how the mother could improve her contact with Lisa by joining her in her play behavior.

Session 2: 'Speaking for the child' and Joint attention

In the original VIPP, the theme of the second session is 'speaking for the child'. The intervener comments on video fragments by verbalizing the child's nonverbal signals in doing, thinking, trying, etc., and invites the parent to join in to speak for the child. Accurate perception of children's signals by verbalizing their facial expressions and non-verbal cues shown on the video is promoted. In children with autism, the non-verbal cues can be rare or subtle, and video feedback offers the opportunity to view and review brief fragments. Joint attention, which is one of the earliest signs of social interaction (Mundy et al., 2000), refers to the coordination of the child's visual attention with the visual attention of another person; it starts to develop before the age of six months (Mundy et al., 2009). The development of joint attention is strongly related to the quality of the interaction between children with autism and their parents, including parental sensitive caregiving (Bruinsma et al., 2004; Naber et al., 2007; Siller & Sigman, 2002).

Three video fragments of three minutes each are used in this session. First, the child plays alone with a toy camera, with the parent close. The parent is instructed to join in only at the child's initiative. In the second fragment, parent and child build a tower of blocks. The third fragment involves an unexpected event. While the child explores a toy, the intervener unexpectedly sounds a peep behind her back, and repeats it several times before the source of the peep (a little bath duck) is shown to the child. Reviewing the video fragments, the intervener provides subtitles (by speaking for the child) for the child's play behavior and joint attention signals, including signals of attachment and exploration that were highlighted in the previous session. The parent is invited to join in the subtitling of the child's behaviors. If applicable, limited eye contact of the child is discussed in the context of alternative communication strategies, such as using a glove puppet. Finally, general language development is discussed. The intervener encourages the parent to speak as much as possible with the child, even - or especially - when the child does not respond verbally. The importance of guiding the child's behavior with comments (McDuffie et al., 2006), explanation and compliments is emphasized.

Vignette session 2

Session 2: speaking for the child, joint attention

Robert is a four-year-old boy with autism spectrum and below average cognitive and speech / language functioning.

Robert and his mother were building a tower in the shape of a clown. The film was divided into 18 fragments of on average 18 seconds. The mother collected the blocks out of the box, which Robert was strongly opposed to. After six seconds though, Robert became curious, especially when his mother showed him a block. The intervener explained Robert's joint attention behavior that during this video clip took the form of looking together at the same object. Robert needed time to get used to the new toy. His mother showed patience and invited Robert repeatedly to join in, by asking him how the tower worked. Gradually, Robert became at ease and accepted the building project. He pointed to a block to start building the tower. Again, this fragment showed the joint attention of Robert and his mother. Clearly, Robert understood the game and he constructed the tower perfectly. At the end Robert protested about placing the last block on the tower (the head of the clown), because its shape differed from the other blocks. Robert needed more time to get used to unexpected objects and situations. This behavior was also seen in the previous session. By inviting the mother to speak for her child, Robert's joint attention behavior became more obvious.

Session 3: 'Sensitivity chain', Daily problems and routine

The video-feedback focuses on sensitivity chains according to the original VIPP, and on the dyad's daily patterns and routines. The intervener introduces parental responses to the child's behavior within 'sensitivity chains'. The child may show positive (smiling, attending, pointing) or negative (crying, screaming, dashing) signals. Adequate, i.e. timely and positive, responses by the parent (smiling back, consoling, explaining expectations) are highlighted, and their relevance to gaining the child's trust and feeling understood are demonstrated by the positive reactions of the child (continuing eating, stopping crying, helping). Making these chains-of-events explicit improves the parent's awareness of how the child's and their own behavior interact. In addition, parents learn to adapt their responses to the (perhaps impaired) level at which the child processes social information (Noens & Van Berckelaer-Onnes, 2005).

Two aspects of daily problems for many families with a child with autism are addressed: (1) managing challenging behavior and (2) support of communication. Various recurrent problems are prevalent in children with autism, including eating problems (Laud et al., 2009), sleep problems (Couturier et al., 2005) and challenging behavior (e.g. Matson et al., 2011). To manage child-non-compliance, elements of VIPP–Sensitive Discipline are included, focusing on non-coercive responses to disruptive behavior, positive reinforcement, and using time-out procedures (Juffer et al., 2008, pg. 17-18).

In this third session, a 20-30 minute videotape of an everyday meal (snack or dinner) is used. Standardization of meal-time video pictures is limited because of the varying and personal daily patterns in families. In general, the feedback focuses on the organization of a predictive, daily structure. In addition, we introduce the use of augmentative communication devices such as pictograms, photographs or written texts in order to advance communication in everyday circumstances. The use of such communication devices is based on visually structured components of evidence-based interventions; the Treatment and Education of Autistic and Communication Handicapped Children (TEACCH) program (Mesibov & Shea, 2010; Erba, 2000) and the Picture Exchange Communication System (PECS) (Bondy & Frost, 2001).

Session 4: Sharing emotions, Emotions and (stereotypical) behavior

During the fourth session, the original VIPP focuses on sharing emotions – showing and encouraging parents' affective attunement to the positive and negative emotions of their child. Sharing emotions requires the recognition of emotions. In children with autism emotions are often paired with stereotypic behavior, due to their problematic regulation of emotions. Stereotypical behavior varies in degree and performance from simple behaviors such as stereotypical movements, tics, and repetitive manipulation of objects, to complex behaviors such as restricted interests or preoccupations, rigid rituals, repetitive language and specific object attachments (Lam et al., 2008; Turner, 1999); and it may be difficult for parents to 'read'.

The following three-minute play fragments are used: (1) the dyad's making music together using a xylophone, (2) singing songs

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Vignette session 3

Session 3: sensitivity chains, daily problems and routine

Ana is a two-year-old girl with autism spectrum and cognitive as well as speech / language development on average levels.

The complete family sat at the table (mother, father, five-year-old sister and Ana), eating lunch, which was filmed for 18 minutes. A total of 41 video clips were highlighted (average stop every 26 seconds), including five chains of sensitivity. At the beginning of the meal, the first chain comprised the following fragment. *Ana showed her own knife proudly/ the mother confirmed to notice Ana's showing behavior/ Ana started to butter her sandwich with the knife.* Ana enjoyed the meal. She was interested in the food and was able to remain sitting at the table for a long time. Her parents created a calm ambiance. Another example of a sensitive chain was as follows:
Ana intended to take another piece of cheese/ the mother looked into Ana's eyes as she explained to Ana that she already had a piece and therefore she could not have any more cheese/ Ana accepted this clear message and started to eat. The mother's response to Ana's signal was timely, together with firm limitation setting regarding the behavior she expected from her daughter. During other fragments, the intervener discussed topics of daily routines, e.g., how to discipline Ana's behavior sensitively. For example, taking leadership with steady agreements, limiting the choice of sandwich filling, limiting Ana's inattention with fewer toys and less food on the table, enhancing daily structure by initiating and finishing the meal more clearly (confirming when Ana is allowed to leave the table) and guiding with compliments all the little steps of eating bites, drinking sips and sitting at the table quietly. After the video feedback, the importance of structure and routines in other daily situations was acknowledged, including the use of pictograms as a supporting communication device. The mother recognized Ana's anger and noncompliance when unexpected situations occurred. The intervener discussed sensitive discipline strategies, such as preparing Ana for alterations in advance, involving Ana in daily activities (e.g., to set out the dish), showing empathy for Ana's mood, naming and explaining parental rules, giving alternatives - all using positive reinforcement strategies. Temper tantrums may be dealt with by the use of a sensitive time-out, in duration corresponding to her young age (never longer in minutes than the child's age in years). As the mother noted that Ana takes language literally -which is often the case in children with autism - the intervener advised Ana's mother to avoid the use of jokes and metaphors.

with gestures to provoke imitation, and (3) blowing soap bubbles. For many children with autism these play materials challenge their emotion processing and communication skills. The parents are invited to name the emotional state of their child by interpreting his or her stereotypical behavior.

The preparation of this session includes the screening of all previous tapes to identify the child's stereotypical behaviors. The parent is invited to review the meaning or context of such behaviors in order to recognize the function or underlying emotions. Basically, stereotypical behaviors are signals that the child is excited and may need the parent's help in structuring the situation, or the parent's help in dealing with emotions.

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Vignette session 4

Session 4: sharing emotions, emotions and stereotypic behavior

Daniel is a four-year-old boy with autism. He has average cognitive development and below average speech / language functioning.

Daniel was excited about blowing soap bubbles. The film was stopped at 9 intervals of about 30 seconds each. It included one sensitivity chain. Daniel immediately responded to his mother's clear instruction to create more distance in order to give room for the bubbles. He liked to catch and extinguish the bubbles. Daniel looked expectantly at his mother when she started to blow new bubbles, and together they laughed a lot during this game. The following sensitivity chain was shown. *Daniel asked for a big bubble/ so his mother told him she would try to blow a very big bubble/ Daniel jumped joyfully as he expected a big bubble.* After a while, his mother started to count the bubbles, but Daniel did not cooperate. He just wanted to play without counting. His mother showed Daniel how to blow bubbles himself and both enjoyed the game again. His mother learned to distinguish different aims of play; education and entertainment. By following her son in the way he preferred to play, the interaction remained positive. During the current and previous films, Daniel started to jump when he became enthusiastic. This stereotypical behavior is partly age-related. By jumping, Daniel showed feelings of excitement. His mother was relieved to interpret Daniel's stereotypical jumping as an expression of positive feelings, without the need to interfere. Based on the video feedback, his mother practiced waiting longer for Daniel's responses. She noticed the need for Daniel to process information at his own pace as she watched him listening and understanding her messages very well.

Session 5: Booster session

During the final home visit, the other parent is invited to join the feedback. During this session, we review all themes. The three previously-taped film fragments are (1) playing with a wooden fruit set, (2) reading a book, and (3) playing with a marble run.

The intervener shows the film fragments as before, inviting the parents to speak for the child and highlighting sensitivity chains. The intervener discusses with both parents the positive parent-child interactions, daily routines, coping patterns and the degree of the child's autism features seen in play, communication and stereotypical behavior.

Written information

After each session, the parent can practice the individual pieces of advice from the video-feedback, supported by a brief, standard written explanation corresponding to the theme of the session. This standard dossier comprises background information on the topic and general tips for handling children's behavior. At the end of the program, the parent has collected the full dossier, which can serve as a store of advice for the future. Additionally in the last session, the parent will receive an individualized written summary. This brief summary describes topics with respect to the individual competences of the child and developmental issues the parent is encouraged to attend to.

Intervener criteria and training

The standardization of VIPP-AUTI allows professionals of various disciplines to act as interveners. Professionals who are experienced in care for children with autism, including nurses, pedagogical or social workers, home trainers, and professionals in daycare settings, might be in the position to learn to apply VIPP-AUTI rather quickly and effectively. Working according to the intervention protocol can be learned in a workshop with additional supervision on the job. Also, an online professional network is accessible on www.linkedin.com/groups/ VIPP-SD international.

Current findings

In a randomized controlled trial with the VIPP-AUTI (N=78) we found that parents who received VIPP-AUTI (n = 40) were more likely to respect child's autonomy and exploring behavior, without interfering or dominating children's behavior too much. Parents also reported stronger personal feelings of competence in child rearing when they finished VIPP-AUTI. Satisfaction of parents after receiving VIPP-AUTI as measured with the Dutch version of the Client Satisfaction Questionnaire (CSQ-8; Attkinson & Zwick, 1982; De Brey, 1983; De Wilde & Hendriks, 2005) ranged from mostly satisfied to very satisfied with the program. In a separate paper, we will report quantitative details of the findings (Poslawsky et al., under review; see chapter 4).

The following quotes of three parents illustrated training responsiveness to the training several months after receiving VIPP-AUTI.

The mother of Alice (48 months old and diagnosed with AD nine months ago)*:*

"I have learned what Alice does and why she does that. It was interesting. I saw myself ordering Alice to play with me, for instance, but that way I wouldn't want to play either. It can be asked in a much more playful way. The home trainer pointed out that Alice puts her head back sometimes. I didn't see that at first, because I am always with her. But a little while ago I did notice it, and she does do that. I also learned not to give all the food at once, but rather just one plate at a time, with the right amount. The home trainer has explained to me that I should talk to Alice a lot to increase her language abilities. It is true that I never used to say much, because Alice didn't speak."

The mother of Thom (58 months old and diagnosed with AD nine months ago):

> *"I have learned to watch Thom in order to understand him better. I need to spend more time with him and that is a point of attention for me. I am not used to structure my life but I need to. I am working hard at changing myself."*

The mother of Vincent (46 months old and diagnosed with AD eight months ago):

> *"I have learned that I should talk a lot, explain everything, basically guide all that Vincent does, in a word: talk. The footage has shown me a lot. What I saw was confronting, but not in a negative way. It made me realise a lot. For instance, when I turn around, I am busy thinking about what I am going to do, and take no notice of the fact that Vincent is still following me. He is focused on me. Apart from that, the written instructions were nice to read, for instance on how to play together – I really try to follow the tips."*

One of the nurses who provided VIPP-AUTI reported also educational effectiveness:

> *"The first two sessions were aimed at getting to know each other and at getting used to the filming and talking about the footage. It was surprising to see that very few parents minded being filmed. Some parents wanted practical advice pretty soon. I would tell them that I wanted to get to know their child better first, because the point was to think of solutions together - solutions that fit with their child in particular, because every child is unique. In my experience this works well."*

Discussion

VIPP-AUTI is an adaptation of the attachment-based VIPP, specifically targeted to families with young children with autism. Early intervention, directly after the diagnosis of autism, enables parents to recognize and manage the autistic traits of their child better. VIPP-AUTI can be provided very early, even during the first year of life, because of its focus on parental sensitivity to children's autistic characteristics. Although parents of children with autism are found to be sensitive to children's needs such sensitivity was found unrelated to attachment security in the children (Van IJzendoorn et al., 2007). Increased understanding of the child's behaviors associated with autism may elevate the level of autism-specific parental sensitivity that in its turn may enhance the children's attachment security. Furthermore, the individualized video method acknowledges factors such as cultural background, autism-related traits of the parent, and co-morbidity of the child's diagnosis, which may partly explain its feasibility.

In contrast to long-term evidence-based, comprehensive intervention programs for young children with autism, VIPP-AUTI is short-term, comprising only five home-visits over three months that can be implemented on top of care-as-usual. Interaction-focused interventions have shown to be effective on infant attachment security when they include fewer than 16 sessions (Bakermans-Kranenburg et al., 2003). A short-term program limits the intervention burden. Greater parental insight into the meaning of their child's autism can indicate a direction for additional long-term intervention, if necessary. Evidently VIPP-AUTI is not meant to cure autism but to help optimize the developmental niche created by the parent for the child with autism.

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References

American Psychiatric Association (2013). Diagnostic and Statistical Manual of Mental Disorders (5th ed.). Washington DC.

Attkisson, C. C., & Zwick, R. (1982). The client satisfaction questionnaire: Psychometric properties and correlations with service utilization and psychotherapy outcome. *Evaluation and Program Planning, 5*(3), 233-237.

Baird, G., Simonoff, E., Pickles, A., Chandler, S., Loucas, T., Meldrum, D., & Charman, T. (2006). Prevalence of disorders of the autism spectrum in a population cohort of children in South Thames: the Special Needs and Autism Project (SNAP). *Lancet, 368*(1474-547; 9531), 210-215.

Bakermans-Kranenburg, M. J.,Van IJzendoorn, M. H., & Juffer, F. (2003). Less is more: meta-analyses of sensitivity and attachment interventions in early childhood. *Psychological Bulletin, 129*(2), 195-215.

Benjak, T., Vuletic Mavrinac, G., & Pavic Simetin, I. (2009). Comparative study on self-perceived health of parents of children with autism spectrum disorders and parents of non-disabled children in Croatia. *Croatian Medical Journal, 50*(4), 403-409.

Bondy, A., & Frost, L. (2001). The picture exchange communication system. *Behavior Modification, 25*(5), 725-744.

Bruinsma, Y., Koegel, R. L., & Koegel, L. K. (2004). Joint attention and children with autism: a review of the literature. *Mental Retardation and Developmental Disabilities Research Reviews, 10*(3), 169-175.

Chakrabarti, S., & Fombonne, E. (2005). Pervasive developmental disorders in preschool children: confirmation of high prevalence. *The American Journal of Psychiatry, 162*(6), 1133-1141.

Charman, T. (2011). The highs and lows of counting autism. *The American Journal of Psychiatry, 168*(9), 873-875.

Couturier, J. L., Speechley, K. N., Steele, M., Norman, R., Stringer, B., & Nicolson, R. (2005). Parental perception of sleep problems in children of normal intelligence with pervasive developmental disorders: prevalence, severity, and pattern. *Journal of the American Academy of Child and Adolescent Psychiatry, 44*(8), 815-822.

De Brey, H. (1983). A cross-national validation of the Client Satisfaction Questionnaire: The Dutch experience. *Evaluation and Program Planning, 6*(3–4), 395-400.

De Wilde, E.F., & Hendriks, V.M. (2005). The Client Satisfaction Questionnaire: psychometric properties in a Dutch addict population. *European Addiction Research, 11*(4), 157-62.

Eisenhower, A. S., Baker, B. L., & Blacher, J. (2005). Preschool children with intellectual disability: syndrome specificity, behavior problems, and maternal well-being. *Journal of Intellectual Disability Research : JIDR, 49*(Pt 9), 657-671.

Erba, H. W. (2000). Early intervention programs for children with autism: Conceptual frameworks for implementation. *American Journal of Orthopsychiatry, 70*(1), 82-94.

Fombonne, E. (2009). Epidemiology of pervasive developmental disorders. *Pediatric Research, 65*(6), 591-598.

Green, V. A., Pituch, K. A., Itchon, J., Choi, A., O'Reilly, M., & Sigafoos, J. (2006). Internet survey of treatments used by parents of children with autism. *Research in Developmental Disabilities, 27*(1), 70-84.

Higgins, D. J., Bailey, S. R., & Pearce, J. C. (2005). Factors associated with functioning style and coping strategies of families with a child with an autism spectrum disorder. *Autism : The International Journal of Research and Practice, 9*(2), 125-137.

Juffer, F., BakermansKranenburg, M. J., & van IJzendoorn, M. H., ed. (2008). *Promoting Positive Parenting. An Attachment-Based Intervention.* New York/ London: Lawrence Erlbaum Associates/ Taylor & Francis Group.

Juffer, F., BakermansKranenburg, M. J., & Van IJzendoorn, M. H. (2009). Attachment-Based Interventions: Heading for Evidence-Based Ways to Support Families. *ACAMH Occasional Papers, 29,* 47-57.

Kasari C, Freeman S, & Paparella T. (2006). Joint attention and symbolic play in young children with autism: a randomized controlled intervention study. *J Child Psychol Psychiatry*, *47*(6):611-620.

Kim, Y. S., Leventhal, B. L., Koh, Y. J., Fombonne, E., Laska, E., Lim, E. C., et al. (2011). Prevalence of autism spectrum disorders in a total population sample. *The American Journal of Psychiatry, 168*(9), 904-912.

Lam, K. S., Bodfish, J. W., & Piven, J. (2008). Evidence for three subtypes of repetitive behavior in autism that differ in familiality and association with other symptoms. *Journal of Child Psychology and Psychiatry, and Allied Disciplines, 49*(11), 1193-1200.

Laud, R. B., Girolami, P. A., Boscoe, J. H., & Gulotta, C. S. (2009). Treatment outcomes for severe feeding problems in children with autism spectrum disorder. *Behavior Modification, 33*(5), 520-536.

Lord, C. (2011). Epidemiology: How common is autism? *Nature, 474*(7350), 166-168.

Makrygianni, M. K., & Reed, P. (2010). A meta-analytic review of the effectiveness of behavioral early intervention programs for children with Autistic Spectrum Disorders. *Research in Autism Spectrum Disorders, 4*(4), 577-593.

3

Matson, J. L., Kozlowski, A. M., Worley, J. A., Shoemaker, M. E., Sipes, M., & Horovitz, M. (2011). What is the evidence for environmental causes of challenging behavior in persons with intellectual disabilities and autism spectrum disorders? *Research in Developmental Disabilities, 32*(2), 693-698.

McConachie, H., & Diggle, T. (2007). Parent implemented early intervention for young children with autism spectrum disorder: a systematic review. *Journal of Evaluation in Clinical Practice, 13*(1), 120-129.

McConnell, S. R. (2002). Interventions to facilitate social interaction for young children with autism: review of available research and recommendations for educational intervention and future research. *Journal of Autism and Developmental Disorders, 32*(5), 351-372.

McDuffie, A. S., Yoder, P. J., & Stone, W. L. (2006). Labels increase attention to novel objects in children with autism and comprehension-matched children with typical development. *Autism, 10*(3), 288-301.

Mesibov, G. B., & Shea, V. (2010). The TEACCH Program in the Era of Evidence-Based Practice. *Journal of Autism and Developmental Disorders, 40*(5), 570-579.

Mundy, P., Card, J., & Fox, N. (2000). EEG correlates of the development of infant joint attention skills. *Developmental Psychobiology, 36*(4), 325-338.

Mundy, P., Sullivan, L., & Mastergeorge, A. M. (2009). A parallel and distributed-processing model of joint attention, social cognition and autism. *Autism Research : Official Journal of the International Society for Autism Research, 2*(1), 2-21.

Naber, F. B., Bakermans-Kranenburg, M. J., Van IJzendoorn, M. H., Swinkels, S. H., Buitelaar, J. K., Dietz, C., et al. (2008). Play behavior and attachment in toddlers with autism. *Journal of Autism and Developmental Disorders, 38*(5), 857-866.

Naber, F. B., Swinkels, S. H., Buitelaar, J. K., Dietz, C., Van Daalen, E., Bakermans-Kranenburg, M. J., et al. (2007). Joint attention and attachment in toddlers with autism. *Journal of Abnormal Child Psychology, 35*(6), 899-911.

Noens, I. L., & van Berckelaer-Onnes, I. A. (2005). Captured by details: sense-making, language and communication in autism. *Journal of Communication Disorders, 38*(2), 123-141.

Oono, I., Honey, E., & McConachie, H. (2013). Parent-mediated early intervention for young children with autism spectrum disorders (ASD). *Cochrane Database of Systematic Reviews, 4*, CD009774-CD009774. doi:10.1002/14651858.CD009774.pub2

Ospina, M. B., Krebs Seida, J., Clark, B., Karkhaneh, M., Hartling, L., Tjosvold, L., et al. (2008). Behavioral and developmental interventions for autism spectrum disorder: a clinical systematic review. *PloS One, 3*(11), e3755.

Peters-Scheffer, N., Didden, R., Korzilius, H., & Sturmey, P. (2011). A meta-analytic study on the effectiveness of comprehensive ABA-based early intervention programs for children with Autism Spectrum Disorders. *Research in Autism Spectrum Disorders, 5*(1), 60-69.

Rice ,C. (2009). Prevalence of autism spectrum disorders - Autism and Developmental Disabilities Monitoring Network, United States, 2006. *MMWR.Surveillance Summaries : Morbidity and Mortality Weekly Report.Surveillance Summaries / CDC, 58*(10), 1-24.

Reichow, B., Barton, E.E., Boyd, B. A., & Hume, K. (2012). Early intensive behavioral intervention (EIBI) for young children with autism spectrum disorders (ASD). *Cochrane Database of Systematic Reviews, 10*, CD009260-CD009260. doi:10.1002/14651858.CD009260.pub2

Rogers, S. J., & Vismara, L. A. (2008). Evidence-based comprehensive treatments for early autism. *Journal of Clinical Child and Adolescent Psychology : The Official Journal for the Society of Clinical Child and Adolescent Psychology, American Psychological Association, Division 53, 37*(1), 8-38.

Rutgers, A., Van IJzendoorn, M., Bakermans-Kranenburg, M., Swinkels S., Van Daalen E., Dietz C., et al. (2007). Autism, attachment and parenting: a comparison of children with autism spectrum disorder, mental retardation, language disorder, and non-clinical children. *Journal of Abnorm Child Psychology*, 35 (5), 859-870.

Schwichtenberg, A., & Poehlmann, J. (2007). Applied behavior analysis: Does intervention intensity relate to family stressors and maternal well-being? *Journal of Intellectual Disability Research, 51*(8), 598-605.

Siller, M., & Sigman, M. (2002). The behavior of parents of children with autism predict the subsequent development of their children's communication. *Journal of Autism and Developmental Disorders, 32*(2), 77-89.

Turner, M. (1999). Annotation: Repetitive behavior in autism: a review of psychological research. *Journal of Child Psychology and Psychiatry, and Allied Disciplines, 40*(6), 839-849.

Van IJzendoorn, M. H., Rutgers, A. H., Bakermans-Kranenburg, M. J., Swinkels, S. H., Van Daalen, E., Dietz, C., et al. (2007). Parental sensitivity and attachment in children with autism spectrum disorder: comparison with children with mental retardation, with language delays, and with typical development. *Child Development, 78*(2), 597-608.

3

Van Zeijl, J., Mesman, J., Van IJzendoorn, M. H., BakermansKranenburg, M. J., Juffer, F., Stolk, M. N., et al. (2006). Attachment-based intervention for enhancing sensitive discipline in mothers of 1- to 3-year-old children at risk for externalizing behavior problems: A randomized controlled trial. *Journal of Consulting and Clinical Psychology, 74*(6), 994-1005.

Virues-Ortega, J. (2010). Applied behavior analytic intervention for autism in early childhood: meta-analysis, meta-regression and dose-response meta-analysis of multiple outcomes. *Clinical Psychology Review, 30*(4), 387-399.

Wallace, K. S., & Rogers, S. J. (2010). Intervening in infancy: implications for autism spectrum disorders. *Journal of Child Psychology and Psychiatry, and Allied Disciplines, 51*(12), 1300-1320.

Weiss, M. J. (2002). Harrdiness and social support as predictors of stress in mothers of typical children, children with autism, and children with mental retardation. *Autism: The International Journal of Research and Practice, 6*(1), 115-130.

Chapter 4

**Enhancing Parental Efficacy and Sensitivity to
Children with Autism Spectrum Disorder:
A Randomized Controlled Trial with
Video-feedback to promote Positive Parenting
adapted to Autism (VIPP-AUTI)**

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(Under review)

Abstract

In a randomized controlled trial we tested the home-based intervention program Video feedback Intervention to promote Positive Parenting adapted to Autism (VIPP-AUTI) with 78 primary caregivers and their young child diagnosed with Autism Spectrum Disorder (ASD). VIPP-AUTI is an attachment-based intervention program, focusing on parental sensitive responsiveness and the child's individual ASD-symptomatology in five home visits. VIPP-AUTI, as compared with usual home training, demonstrated effectiveness in enhancing parental sensitive responsiveness by reducing intrusiveness. Moreover, parents in the VIPP-AUTI group showed increased feelings of efficacy in child rearing post-intervention. Implementation of VIPP-AUTI in clinical practice is facilitated by the use of a detailed manual, a relatively brief training of interveners, and the features of a short-term interaction-focused intervention.

Introduction

Parent training might be effective in promoting parental feelings of efficacy in taking care of a child with Autism Spectrum Disorder (ASD), and in elevating the level of parental sensitive responsiveness. Parent training might thus help to optimize cognitive and socio-emotional development of children with ASD within the boundaries of their disorder. In this study we test the effects of an early attachment-based intervention adapted for families of a child with ASD on parental attitudes and sensitive parenting in a randomized controlled trial.

ASD is a neurodevelopmental disorder, including core deficits of stereotypic behavior, social and communication impairments (American Psychiatric Association [APA], 2013). Parental sensitive responsiveness is the extent to which parents adequately respond to the needs of their child and it is found to be associated with the quality of a child's attachment relationship (Ainsworth, 1978; Bakermans-Kranenburg et al. 2003). An attachment relationship is the affective bond between an infant and its primary caregiver, which is formed for protection and survival (Bowlby, 1969, Cassidy & Shaver, 2008). In young children with ASD, a high prevalence of insecure and disorganized attachment relationships has been observed (Naber et al. 2007; Rutgers et al. 2004). The development of a secure attachment relationship may be at risk in children with ASD

because parental sensitive responsiveness could be negatively affected by the unusual affective behavior of children with ASD (Dawson et al. 1990). Furthermore, children's deficits in processing social information (Gervais et al. 2004; Noens & Van Berckelaer-Onnes, 2005; Ploog, 2010) complicate parent-child interactions. Parents need to be aware that they have to stimulate the child more explicitly, both verbally and non-verbally (Kasari et al. 1988; Doussard-Roosevelt et al. 2003), and yet not to be intrusive.

Parenting a child with ASD is generally found to be more stressful than parenting typically developing children or even children with mental retardation, resulting in higher levels of depression, anxiety, somatic complaints, family dysfunction and symptoms of burnout (Benjak et al. 2009; Eisenhower et al. 2005; Higgins et al. 2005; Koegel et al. 1992; Weiss, 2002). Parental distress while raising their child with ASD is also negatively associated with parental efficacy (Bekhet et al. 2012; Giallo et al. 2011; Kuhn & Carter, 2006; Pakenham et al. 2004). Parental efficacy refers to parents' perceived confidence in their competence to child rearing (Bandura, 1995). In an ASD sample, greater parental efficacy was found to increase parents' positive active engagement with the child (Kuhn & Carter, 2006).

Previous studies based on parent-implemented early interventions for children with ASD demonstrated improvement of the child's social communication behavior, parental performance and parent-child interaction (McConachie & Diggle 2007; Oono et al., 2013). Furthermore, Wallace and Rogers (2010) recommended individualized interventions at home for infants and toddlers with ASD, focusing on responsive, sensitive parent-child interactions. However, the effectiveness of most of these interventions is based on studies with intensive programming, such as applied behavior analysis (ABA) interventions (see meta-analyses of Makrygianni & Reed 2010; Peters-Scheffer et al. 2011; Reichow et al., 2012; Virués-Ortega 2010;). Treatment intensity of ABA programs was found to be a risk for increased parental stress (Schwichtenberg & Poehlmann, 2007) and is accompanied with substantial costs, which could discourage the use of interventions in common clinical practice.

Randomized trials conducted in families dealing with various developmental complexities, although not previously ASD, have shown effectiveness of interaction-focused interventions on parental sensitivity to

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infant's signals, especially when video feedback was used and the duration was short term (Bakermans-Kranenburg et al. 2003). Based on these meta-analytic findings and inspired by attachment theory a brief home training Video feedback Intervention to promote Positive Parenting (VIPP) was developed and tested, and variants for specific issues were examined and found to be effective in families of infants, toddlers and preschoolers (Juffer et al. 2008; Juffer et al. 2009). Providing VIPP offers several advantages. Firstly, VIPP programs allow additional foci which correspond to the children's special needs. These programs, with and without additional modules, demonstrated effectiveness of parental sensitivity in clinical and non-clinical groups across various countries (e.g. Juffer et al. 2009; Moss et al. 2011). Secondly, low intensity programming of VIPP could limit family burden and costs. Finally, VIPP is both standardized and individualized, meaning that interveners use a general protocol but attune specific themes and guidelines to individual parent-child dyads. It should be realized that children with ASD have complex and multifaceted disorders, with varying severity. Therefore, any parent training should be attuned to the individual needs.

We modified VIPP into Video feedback Intervention to promote Positive Parenting adapted to Autism (VIPP-AUTI), an intervention program to support parents of children with ASD in interacting with the child. VIPP-AUTI aims to enhance parental sensitivity for the child's signals, taken its autistic features into account, by means of video feedback. The intervener video-tapes actual parent-child interactions at home, and in a next session watches and discusses carefully selected videotaped episodes of parent-child interactions together with the parent. Video feedback provides the parent with an opportunity to reflect on his or her interactions with the child and the responses of the child to these interactive efforts, with an emphasis on positive, successful interaction sequences. VIPP-AUTI thus enables the parent to have a look in the mirror as a stimulus to intensify parental self-reflection and mentalization. VIPP-AUTI focuses on stimulating parents' understanding and managing of the manifest core symptoms of young children with ASD, including stereotypical or rigid behavior and deficits in early development of communication and play. The development and content of the VIPP-AUTI program is described in detail in Poslawsky, et al. (under review; see chapter 3).

In this paper we focus on the effects of VIPP-AUTI on the parents involved. We hypothesized that VIPP-AUTI improves parental sensitive responsiveness in the interaction with the child. Secondly, VIPP-AUTI may enhance parental feelings of efficacy and relieve some of the burden they experience in child rearing.

Method

The VIPP-AUTI study is a collaboration project between University Medical Center Utrecht (UMC Utrecht, Department of Psychiatry) and Leiden University (Center for Child and Family Studies). The randomized controlled trial investigated the effectiveness of the VIPP-AUTI program aimed at improving parent-child interaction in the presence of ASD by enhancing parental sensitive responsiveness and by enhancing parental feelings of competence and reducing burden in child rearing.

Participants

Seventy-eight children with ASD (86% boys) and their primary caregivers (90% mothers) participated in this study. Middle to high level of SES (96%) was characteristic of the majority of the participating families. Parental age ranged from 25 to 52 years ($M = 36.6$, $SD = 5.04$). Most parents were married couples (82%) with Dutch nationality (90%). Children's age ranged from 16 to 61 months ($M = 43.0$, $SD = 9.96$). More than half of the children were first born (59%). Children's developmental level ranged from mental retardation to above average functioning ($M = 73.7$, $SD = 22.03$). Age equivalent of children's language development was 32 months on average (M language comprehension= 32.8 months, $SD = 17.08$, and M language production = 32.1 months, $SD = 16.07$). In 53% of the children, the interval between the children's chronological age and the age equivalent of their language development was more than 6 months. Twelve percent of the children were raised bilingually.

Diagnosis

Children were diagnosed as having ASD by a board-certified child psychiatrist according to extensive developmental history, all medical files, a semi structured observation (Autism Diagnostic Observation Schedule – Generic) and classification of the Diagnostic and Statistical Manual of

Mental Disorders 4th Edition-Text Revision (DSM-IV-TR; APA, 2000). Sixty-eight percent of the children were diagnosed with Autistic Disorder (AD), and 32% were diagnosed with Pervasive Developmental Disorders Not Otherwise Specified (PDD-NOS). The majority of the children received ASD (with or without comorbid mental retardation) as the first and only diagnosis. In addition to the ASD diagnosis, some children (36%) received a diagnosis regarding somatic conditions, e.g. epilepsy, lung diseases or sensory deficits.

Procedure

Parents of newly ASD-diagnosed children under the age of five years were invited to participate in the study. Of the invited families ($N = 157$) 50% did not participate. Main reasons for non-participation were receiving other formal support of external facilities, non-preference for home training or expected burden of research assessments (see Figure 1). The primary caregiver of participating families was included, regardless of gender. Another inclusion criterion was that parent and child had to live at the same address. Exclusion criteria were parents who did not care for their child themselves and children with interfering comorbid medical problems. Comorbid medical problems were considered as interfering when current therapy aimed at other than ASD-related concerns, e.g. a hospital admission to treat severe epileptic seizures. The inclusion period was from June 2008 to April 2012. After randomization all but one family remained involved in the trial until the first post-test.
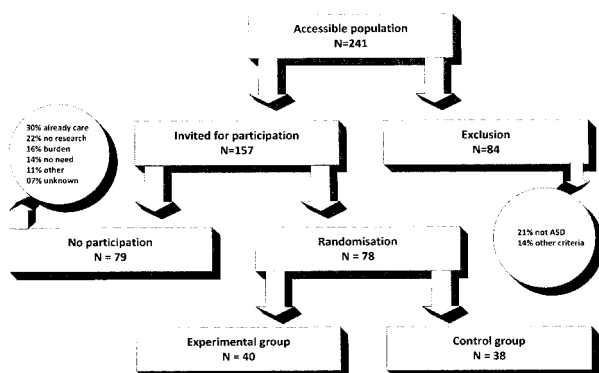
Figure 1 *Flowchart of inclusion.*

After baseline assessments, participants were randomly divided into the experimental group (VIPP-AUTI) or control group (Care-as-Usual, CU). Randomization by computer generated tables was done by a staff manager, who was not involved in the research project. Since both groups received home visits, parents were not aware whether they received the experimental intervention or care as usual. Before randomization and after providing the home-based intervention, parent-child interactions were videotaped at the university hospital and at home. Parents were also asked to complete questionnaires. Demographic data (e.g. parental age, marital status, postal area, education and occupation) were collected during the diagnostic phase prior to the study. The study protocol was approved by the ethics committee of the UMC Utrecht. Both parents gave written informed consent before participation.

Treatment

Care-as-Usual (CU)
After parents received an ASD diagnosis of their child at the Department of Psychiatry of the UMC Utrecht, nursing care is usually offered. Over a period of at most six months, the participants in the CU group received this usual nursing home training, with a mean of five home visits. Based on parents' preferences, a variation of the number of home visits was restricted to at least three and at most seven visits. The care, based on behavioral and family therapeutic interventions, was meant to support parents individually with respect to practical issues of parenting a child with ASD.

Experimental group (VIPP-AUTI)
Over a period of three months, participants received VIPP-AUTI, comprising five home visits of 60-90 minutes each at a two-weekly frequency. During the home visits, video feedback was provided, using film fragments of parent-child interactions filmed in the previous session. According to the intervention protocol, issues regarding the child's behavior and interaction with the parent were discussed with the parent. The autistic traits and the unique performance of the child were highlighted. The last home visit was a booster session, in which partners of the primary caregivers were invited to join the video feedback. VIPP-AUTI was performed by comprehensively

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trained nurses, using a detailed manual and with intensive monitoring of therapy fidelity (by IP, FN, MB-K, MvIJ).

In addition of the nursing home training, whether parents received CU or VIPP-AUTI, multi-disciplinary staff members provided two group meetings with parents for psychoeducation about ASD in general. Furthermore, the psychiatrist could prescribe medication to treat comorbid problems by indication. Parents could also receive support from external care providers and most of the children attended (special) day-care or school. The nurses who provided VIPP-AUTI or CU had similar levels of education and experience in caring for children with ASD.

Instruments

Baseline measures

Autism diagnostic observation schedule - generic (ADOS – G). The ADOS-G (Lord et al. 2000) quantifies deficits across the autism spectrum, controlling for effects of language and cognitive delay, in individuals with significant impairments. The ADOS-G total score is the cumulative score of five test domains: communication, social reciprocity, play, stereotypic behavior and other problems. A high ADOS-G total score refers to severe autistic symptoms. The ADOS-G consists of four modules. In the current study, children were assessed using module 1 ($n = 42$) and module 2 ($n = 36$), based on their level of expressive language. The child psychiatrist, who was certified for ADOS-reliability, administered the ADOS-G during the diagnostic phase, prior to this study.

Mullen scales of early learning (MSEL)
The MSEL (Mullen, 1995) is a standardized developmental test that yields a mental age score for children between 3 and 68 months of age. The MSEL assesses five domains, one measuring cross motor skills (not used in the current study), and four cognitive domains: 1) visual reception, 2) fine motor skills, 3) receptive language, and 4) expressive language. The early learning composite standard score is a combination of non-verbal (domain 1 and 2) and verbal composites (domain 3 and 4), ranging from low (<70) to high cognitive functioning (>70). The test was administered by a certified clinical psychologist.

Language development
The Reynell test for Dutch language comprehension (Van Eldik et al. 1995) and the Schlichting test for Dutch language production (Schlichting et al. 1995) were administered individually by a certified psychologist or speech language therapist. The Reynell and Schlichting tests evaluate receptive and expressive language, respectively, for children between 14 and 75 months of age. If children were not able to do these tests (n= 26) the psychologist or speech language therapist collected parental reports using the Dutch versions of the MacArthur-Bates Communicative Developmental Inventories (CDIs; Fenson, 1993; Fenson et al. 2007; N-CDIs; Zink & Lejaegere, 2002; Zink & Lejaegere, 2007) (n= 22). The CDIs consist of three forms, corresponding with different age groups; 1) 'Word and Gestures' (8-16 months of age), 2) 'Word and Sentences' (16-30 months of age) and 3) 'CDI-level III' (30-37 months of age), assessing vocabulary comprehension and production. The inventories were also used for children beyond the specified age ranges when they had impaired language development (Fenson et al. 1994). The validity of the measure has been shown in children with and without language delay (Heilmann et al. 2005), and for children with ASD (Charman et al. 2003; Luyster et al. 2007).

In the current study, language development was based on the converted age equivalents of total language comprehension scores of the Reynell or N-CDI, and total word-production scores of the Schlichting or N-CDI.

Socio-economic status (SES). SES was based on status scores 2010 of postal areas. Status scores were derived from level of education, employment and income per postal area by a national organization (The Netherlands Institute for Social Research/SCP, 2010). Higher status scores refer to higher SES.

Symptom checklist (SCL-90)
The SCL-90 (Arrindell & Ettma, 1986) is a 90 item self-report questionnaire, rated on a 5-point scale to indicate mental health status of the parents. The instrument measures agoraphobia, anxiety, depression, somatic complaints, insufficiency in thinking and acting, distrust and interpersonal

sensitivity, hostility, sleeping problems and a rest-category. The total score on psychoneuroticism is based on the sum of all sub-categories. Psychoneuroticism indicates a general level of psychoneurotic-somatic complaints. In this study, the norm table of the typical population was used; ranging from very low (score 90) to very high non-wellbeing (score > 183). In this sample, internal consistency of the total scale (Chronbach's alpha) was .97. Missing items on the total scale were less than 5% (a maximum of two items per subscale and no items on the subscale sleeping problems). In these cases, the (estimated) scores on each subscale were computed based on the real scores (Arrindell & Ettma, 1986, pg. 97).

Primary outcome measure
Emotional availability scales. The Emotional availability scales (EAS; Biringen et al., 2000) were used to assess parental emotional responsiveness to their child's needs from a transactional and systemic perspective (Biringen, 2000; Biringen & Easterbrooks, 2012). The scales consist of four subscales of parenting behavior: 1) sensitivity, 2) structuring, 3) non-intrusiveness, and 4) non-hostility; and two subscales for child behavior: 1) child involvement and 2) child responsiveness. The subscale 'sensitivity' of the parent is coded on a 9-point rating scale. High sensitivity refers to a parent's responsiveness to the child's emotional signals and flexible communication. Parental 'structuring' is coded on a 5-point rating scale, and refers to parental ability to support learning and exploration with respect for the child's autonomy. High scores of structuring are assigned when parents not only respond to the child's verbal cues, but also to non-verbal cues. Parental 'non-intrusiveness' is coded on a 5-point rating scale with high scores for parents who are available for the child without interfering, overprotective, or overwhelming behaviors. An intrusive parent controls the interaction and shows lack of respect for the child's autonomy. 'Non-hostility' is also coded on a 5-point rating scale. Parents receive high scores when no negative emotions (e.g. impatience, discontent, rolling the eyes, etc.) are shown. The EAS also includes rating scales for child responsiveness and involvement, but since we aimed at testing the effects of the intervention program on parenting behaviors, we focused the analyses on the parenting scales. The scale for non-hostility was extremely skewed

(*Skewness* = -2.36, *SE*= .27) and thus excluded from further analysis, leaving observations of parental sensitivity, structuring, and non-intrusiveness.

Parent-child interactions were video-taped during 15 minutes of a semi-structured play-situation. Parents were asked to play with the child as they normally do, using three different sets of toys. The play sessions were coded by trained observers, who were unaware of the intervention type parents received (VIPP-AUTI or CU). Intercoder reliability for randomly chosen observations (10% of all data) was adequate (mean intra-class correlations: sensitivity *r* = .77, structuring *r* = .78, non-intrusiveness *r* = .77). Pretest and posttest sessions of the same dyad were coded by different observers.

Secondary outcome measures
Parental efficacy questionnaire (PEQ). The PEQ is a 22-item, self-report questionnaire on a 5-point rating scale which ranges from low to high efficacy (possible scores from -44 to 44). The PEQ is based on Bandura's general theory of personal efficacy (Bandura, 1997), and adapted to parenting by Van IJzendoorn and colleagues (1999, unpublished manuscript). The instrument assesses parents' feelings of competence in child rearing, particularly under stressful circumstances (Rutgers et al. 2007). In this sample, Cronbach's alpha for internal consistency was .87.

Parental daily hassles (PDH). The PDH is a 20-item instrument developed by Crnic and Greenberg (1990) to assess the strains and stresses accompanying child rearing. It contains descriptions of typical everyday life events in parent-child interactions, rated by the parent on a 5-point rating scale for frequency of occurrence of the hassle (scores 0 to 80). In this sample, Cronbach's alpha for internal consistency was .90.

Client satisfaction questionnaire (CSQ-8). The CSQ-8 (Attkisson & Zwick, 1982) is a self-report questionnaire to assess satisfaction to treatment of clients in mental health services. The Dutch version of the CSQ-8 showed the same satisfactory psychometric properties as the original, English questionnaire and was found suitable to use in Dutch populations (De Brey, 1983; De Wilde & Hendriks, 2005). The CSQ-8 contains 8 items scored

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on a Likert scale with 1 = quite dissatisfied, 2 = indifferent or mildly dissatisfied, 3 = mostly satisfied, 4 = very satisfied. Examples of response options include "To what extent has our program met your needs?", and "Have the treatment you received helped you to deal more effectively with your problems?' We administered the CSQ-8 three months after the posttest. In this sample, Cronbach's alpha for internal consistency was .92 (N = 71).

Results

Group differences
To check the equivalence of the experimental and control group at pretest, independent sample t-tests and chi-squared tests were performed. No significant differences between the VIPP-AUTI and CU group were found for parent and child gender or age, sociodemographics, diagnostic delay, time-interval of receiving ASD-diagnosis, parental mental health, parental efficacy, parental daily hassles, or parental emotional responsiveness at pretest (see Table 1). However, significant group differences were found on father's employment and child's school attendance. Fathers in the VIPP-AUTI group worked fewer hours per week (M = 34; SD =11.5) than fathers of the CU group (M = 41; SD =10.0), t (70) = 2.79, $p < .01$). Finally, regardless of age, more children in the CU group visited school (n = 10) than children in the VIPP-AUTI group (n = 4), χ^2 (1, N = 76) = 3.99, $p < .05$. Father's employment and children's school attendance were included as covariates in further analyses. Three months post intervention, all parents reported to be satisfied or very satisfied with both VIPP-AUTI and CU, without any group difference, t (69) = -0.75, p = 0.46 (VIPP-AUTI group M = 24.6; SD = 4.5; CU group M =25.4; SD = 4.7)

Intervention effects on parental sensitive responsiveness
A repeated measures multivariate analysis of variance (MANOVA) was performed to assess intervention effects on parental sensitive responsiveness (based on the parenting scales of the EAS) with VIPP-AUTI or CU as a between-subjects factor and time as a within subject factor, and father's hours of work and child school attendance as covariates. A significant time by group effect was found for non-intrusiveness,

F (1, 72) = 4.30, p = .04, η^2 = .06 (d = 0.49). After the intervention, the parents who received the VIPP-AUTI program showed an increase in non-intrusiveness, whereas non-intrusiveness decreased in the CU group. Sensitivity (F [1, 72] = 0.34, p = .56, η^2 = .00) and structuring (F [1, 72] = 0.30, p = .59, η^2 = .01) did not show significant intervention effects (see Table 2).

Table 1 Background and pretest variables in experimental and control groups.

Primary Caregiver Characteristics	Group					
	VIPP-AUTI (n = 40)		CU (n = 38)			
	M	SD	M	SD	t	p
Age	36.80	04.84	36.42	05.30	0.33	.74
Parental employment (hours p/w)	20.70	15.10	17.45	12.85	1.02	.31
SES (< -.99 = low status; >1.33 = high status)	00.99	00.71	00.95	00.97	0.22	.83
Parental efficacy	23.39	09.11	21.62	12.21	0.71	.48
Parental stress (PDH)	23.71	12.62	22.57	13.81	0.37	.71
Psychoneuroticism (SCL-90)	127.70	36.33	131.17	35.16	0.42	.67
Child Characteristics						
Age (months)	42.16	09.02	43,80	10.92	0.72	.47
Autistic characteristics	25.03	10.55	26.35	10.10	0.56	.58
Developmental level	74.63	23.50	72.68	20.61	0.39	.70
Language comprehension age equivalent (months)	32.77	16.41	32.88	18.04	0.03	.98
Language production age equivalent (months)	33.71	15.89	30.22	16.35	0.85	.40

Note. PDH: Parental Daily Hassles; SCL-90: Symptom Checklist; SES: Social Economic Status.

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Table 2 Repeated measures MANOVA of parental sensitive responsiveness, perceived efficacy and daily hassles

Sensitive responsiveness	Group X Time			
	F	df	p	η^2
Parental non-intrusiveness	4.30	1	.04	.06
Parental structuring	0.30	1	.59	.01
Parental sensitivity	0.34	1	.56	.00
Perceived efficacy and burden				
Parental efficacy	5.66	1	.02	.08
Parental daily hassles	0.00	1	.98	.00

Note. MANOVA: multivariate analysis of variance.

Intervention effects on parental efficacy and daily hassles

We also applied a repeated measures MANOVA with VIPP-AUTI or CU as a between-subjects factor and time as a within subject factor to assess intervention effects on parental efficacy and daily hassles. Again, father's hours of work and child school attendance were entered as covariates. A significant time by group interaction effect was found for parental efficacy, $F (1, 62) = 5.66$, $p = .02$, $\eta^2 = .08$ ($d = 0.61$). Parents in the VIPP-AUTI group showed an increase in their feelings of competence after the intervention, whereas parents in the CU group did not show change over time. No interaction effect between time and group was found for daily hassles, $F (1, 62) = 0.00$, $p = .98$, $\eta^2 = .00$ (see also Table 2). Daily hassles showed no significant correlations with parental efficacy or sensitive responsiveness

Discussion

In this randomized controlled trial with families raising a child with ASD, the attachment-based intervention VIPP-AUTI demonstrated to be more effective as compared to CU. The program, based on a combination of

insights from attachment theory and behavioral- and developmental problems in children with ASD, improved aspects of observed parental sensitive responsiveness towards the child with ASD, in particular by decreasing intrusiveness, and it increased parental efficacy feelings in parenting a child with ASD.

Parents demonstrated significantly increased non-intrusiveness after receiving VIPP-AUTI, resulting in a medium effect size, which is in the same range as effect sizes of VIPP for other populations in previous studies (Juffer et al. 2009). Intrusiveness refers to qualities of overdirectiveness, overstimulation, interference, or overprotectiveness, undermining the child's autonomy (Biringen et al. 2000). There are several reasons for parents to become intrusive, but one plausible explanation for parents of a child with ASD might be that the parent is emphasizing too much his or her role as a teacher who should help the child to recover from cognitive and language delays. The parent may feel that the child needs to catch up with its age mates, and that large part of the parental role should be educational (Safe et al. 2012; Tsai et al. 2008). The parent teaches the child rather than allowing the child to play and learn from its own experiences in his or her own tempo (Jordan, 2003; Pajareya & Nopmaneejumruslers, 2011).

Although, previously, no differences in sensitivity between parents of children without and with ASD has been found (Van IJzendoorn et al. 2007), some studies demonstrated that parents of children with ASD tended to use more physical control to stimulate their child in task performance, especially when the child's engagement was limited, than parents do in the case of children or siblings without ASD (Kasari et al. 1988; Doossard-Roosevelt et al. 2003). Whereas this parental attitude may be functional to activate the child, it could also be a risk for over-stimulation and intrusiveness. Children with ASD often need more time to perform tasks because of impairments in social information processing (e.g. Gervais et al. 2004; Ploog, 2010). Increased parental awareness of these needs (e.g., more time to explore; exploration in another way than expected) could result in less intrusive parental behavior, as is shown in this study.

Parents of children with ASD were found to be less authoritative than parents of children without ASD (Rutgers et al. 2007). An authoritative parenting style is characterized by high responsiveness

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and encouraging independence of the child. The VIPP-AUTI program supports parents to understand the ASD characteristics of their child, and focuses on the child's competences. This empathic understanding may offer parents room to step back in order to guide the child in its own pace, in other words to become less intrusive. In addition, low parental intrusiveness enhances relaxed and joyful parent-child interactions and thereby contributes to the child's exploring behavior. Parental non-intrusiveness is also associated with toddlers' self-regulation (Feldman et al. 2011). In children with ASD, emotion regulation is limited (Pouw et al. 2013), leading to temper tantrums and frustration. This could elicit more intrusive parental behavior in an attempt to avoid child temper tantrums. The VIPP-AUTI program aims at increasing parental insight in the causes and functions of specific frustration related child behavior, and enhances their attentiveness to child-signs of overstimulation. By recognizing and understanding early signs of frustration, parents may downregulate intrusive interactions and stop pushing the child too hard, thereby preventing negative child behavior.

In the current study, parents who received the VIPP-AUTI intervention also reported increased parental efficacy compared to parents receiving care as usual. During the video feedback, some parents explicitly mentioned regaining confidence in their own parenting because the videotaped episodes showed their child's unexpected (often subtle) responsiveness to parental initiatives and responses. VIPP-AUTI aims at increasing parental awareness of the autistic characteristics of their child, including the child's communication patterns, which are often delayed or atypical. Poor quality of social interaction and (expressive) language in children with ASD was found to be associated with challenging behavior (Chiang, 2008; Murphy et al. 2005), and could undermine feelings of adequate parenting. Increased interaction quality may affect feelings of parental efficacy. Positive effects of VIPP-AUTI on both non-intrusiveness and parental efficacy may amplify its influence on positive parent-child relationships.

The feasibility of the VIPP-AUTI intervention is facilitated by its standardization, using a detailed manual, relatively brief training for implementation and short duration, with only five sessions. Intervention compliance was high; all participants received the five complete home

visits in a period of three months. It should be noted that we found positive effects of the VIPP-AUTI intervention compared to care as usual, which included fairly extensive home training and rather robust parenting support system. In both groups parents were satisfied to very satisfied with the treatment. Effectiveness of VIPP-AUTI on parent's non-intrusiveness and parental efficacy can perhaps be explained by differences between VIPP-AUTI and care as usual. The standard care offered to families of a child with ASD is question based, which might elicit a focus on problematic parenting situations, whereas VIPP-AUTI aims at highlighting positive parent-child interactions. Moreover, using video feedback of parent-child interactions in the VIPP-AUTI program may allow parents to interpret and mentalize their own interactions with their child more intensively than through verbal communication as in care as usual.

Some limitations need to be addressed. Since this study was mono-center, intervention effectiveness cannot be generalized to centers with other care as usual programs. Care as usual in the current child psychiatry center consisted of rather intensive home-visits, so the effects of VIPP-AUTI may be larger in settings with less intensive care as usual. Furthermore, the majority of included parents have middle to high SES, and were married couples. The homogeneity of this sample requires further research of intervention effectiveness in families with more heterogeneous sociodemografic characteristics. Thirdly, we only examined short-term effects on parenting. Long-term effects should be established, as well as effects of VIPP-AUTI on child outcomes such as social and communicative abilities. Lastly, at baseline parental sensitive responsiveness in both groups was moderate to high, which might have created a ceiling effect preventing significant improvements on all parenting subscales.

In sum, VIPP-AUTI was tested in a randomized controlled trial using a detailed intervention manual, an equally intensive care as usual program for the control group, and extensive pretests and posttests. VIPP-AUTI was shown to be effective for families of children with ASD. Parents receiving the VIPP-AUTI program demonstrated increased sensitive responsiveness toward their child, as apparent from lower levels of intrusiveness in the interaction with their child. Moreover, they reported increased feelings of efficacy in child rearing compared to parents who received care as usual.

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References

Ainsworth, M.D.S. (1978). Bowlby-Ainsworth Attachment Theory. *Behavioral and Brain Sciences, 1*(3), 436-438.

American Psychiatric Association (2013). Diagnostic and Statistical Manual of Mental Disorders (5th ed.). Washington DC.

American Psychiatric Association (2000). *Diagnostic and Statistical Manual of Mental Disorders (4th ed., Text Revision)*. Washington DC.

Arrindell, W.A. & Ettema, J.H.M. (1986). SCL-90. Lisse: Swets & Zeitlinger.

Attkisson, C.C. & Zwick, R. (1982). The client satisfaction questionnaire: Psychometric properties and correlations with service utilization and psychotherapy outcome. *Evaluation and Program Planning, 5*(3), 233-237.

Bakermans-Kranenburg, M.J., Van IJzendoorn, M.H. & Juffer, F. (2003). Less is more: meta-analyses of sensitivity and attachment interventions in early childhood. *Psychological Bulletin, 129*(2), 195-215.

Bandura, A. (1995). *Self-efficacy in Changing Societies.* Cambridge University Press. Chapter 1, 1-45.

Bandura, A. (1997). *Self-efficacy: The exercise of control*. New York: Freeman.

Benjak, T., Vuletic Mavrinac, G. & Pavic Simetin, I. (2009). Comparative study on self-perceived health of parents of children with autism spectrum disorders and parents of non-disabled children in Croatia. *Croatian Medical Journal, 50*(4), 403-409.

Bekhet, A.K., Johnson, N.L. & Zauszniewski, J.A. (2012). Resilience in family members of persons with autism spectrum disorder: a review of the literature. *Issues in Mental Health Nursing, 33*(10), 650-656.

Biringen, Z. (2000). Emotional availability: conceptualization and research findings. *The American Journal of Orthopsychiatry, 70*(1), 104-114.

Biringen, Z. & Easterbrooks, M.A. (2012). Emotional availability: concept, research, and window on developmental psychopathology. *Development and Psychopathology, 24*(1), 1-8.

Biringen, Z., Robinson, J.L., & Emde, R.N. (2000). Appendix B: The emotional Availability Scales (3rd ed.; abridged infancy/ early childhood version). *Attachment and Human development,* 2, 256-270.

Bowlby, J. (1969). *Attachment and Loss: vol 1. Attachment.* New York: Basic Books.

Cassidy, J. & Shaver, Ph.R. (2008). *Handbook of Attachement: theory, research, and clinical implications.* (2nd ed.). New York: The Guilford Press.

Charman, T., Drew, A., Baird, C. & Baird, G. (2003). Measuring early language development in preschool children with autism spectrum disorder using the MacArthur Communicative Development Inventory (Infant Form). *Journal of Child Language, 30*(1), 213-236.

Chiang, H.M. (2008). Expressive communication of children with autism: the use of challenging behavior. *Journal of Intellectual Disability Research : JIDR, 52*(11), 966-972.

Crnic, K.A. & Greenberg, M.T. (1990). Minor parenting stresses with young children. *Child Development,* 61, 1628–1637.

Dawson, G. Hill, D. Spencer, A. Galpert, L., & Watson, L. (1990). Affective exchanges between young autistic children and their mothers. *Journal of Abnormal Child Psychology, 18*(3), 335-345.

De Brey, H. (1983). A cross-national validation of the Client Satisfaction Questionnaire: The Dutch experience. *Evaluation and Program Planning, 6*(3–4), 395-400.

De Wilde, E.F. & Hendriks, V.M. (2005). The Client Satisfaction Questionnaire: psychometric properties in a Dutch addict population. *European Addiction Research, 11*(4), 157-62.

Doussard-Roosevelt, J.A., Joe, C.M., Bazhenova, O.V. & Porges, S.W. (2003). Mother-child interaction in autistic and nonautistic children: characteristics of maternal approach behaviors and child social responses. *Development and Psychopathology, 15*(2), 277-295.

Eisenhower, A.S., Baker, B.L. & Blacher, J. (2005). Preschool children with intellectual disability: syndrome specificity, behavior problems, and maternal well-being. *Journal of Intellectual Disability Research : JIDR, 49*(Pt 9), 657-671.

Feldman, R., Dollberg, D. & Nadam, R. (2011). The expression and regulation of anger in toddlers: relations to maternal behavior and mental representations. *Infant Behavior & Development, 34*(2), 310-320.

Fenson, L., Dale, P.S., Reznick, J.S., Bates, E., Thal, D., Hartung, J., et al. (1993). *Guide and technical manual for the MacArthur Communicative Development Inventories.* San Diego, CA: Singular Press.

Fenson, L., Dale, P.S., Reznick, J.S., Bates, E., Tha,l D.J. & Pethick, S.J. (1994). Variability in early communicative development. *Monographs of the Society for Research in Child Development, 59*(5), 1-173; discussion 174-85.

Fenson, L., Marchman, V.A., Thal, D., Dale, P.S., Bates, E. & Reznick, J.S. (2007). *The MacArthur-Bates Communicative Development Inventories:User's Guide and technical Manual (2nd edition).* Baltimore: Paul H. Brookes.

4

Gervais, H., Belin, P., Boddaert, N., Leboyer, M., Coez, A., Sfaello I., et al. (2004). Abnormal cortical voice processing in autism. *Nature Neuroscience, 7*(8).

Giallo, R., Wood, C.E., Jellett, R. & Porter, R. (2011). Fatigue, Wellbeing and Parental Self-Efficacy in Mothers of Children With an Autism Spectrum Disorder. *Autism : The International Journal of Research and Practice,* doi:10.1177/1362361311416830

Heilmann, J., Weismer, S.E., Evans, J. & Hollar, C. (2005). Utility of the MacArthur-Bates communicative development inventory in identifying language abilities of late-talking and typically developing toddlers. *American Journal of Speech-Language Pathology / American Speech-Language-Hearing Association, 14*(1), 40-51.

Higgins, D.J., Bailey, S.R. & Pearce, J.C. (2005). Factors associated with functioning style and coping strategies of families with a child with an autism spectrum disorder. *Autism : The International Journal of Research and Practice, 9*(2), 125-137.

Jordan, R. (2003). Social play and autistic spectrum disorders: a perspective on theory, implications and educational approaches. *Autism : The International Journal of Research and Practice, 7*(4), 347-360.

Juffer, F., Bakermans-Kranenburg, M.J. & Van IJzendoorn, M.H., ed. (2008). *Promoting Positive Parenting. An Attachment-Based Intervention.* New York/ London: Lawrence Erlbaum Associates/ Taylor & Francis Group.

Juffer, F., Bakermans-Kranenburg, M.J. & Van IJzendoorn, M.H. (2009). Attachment-Based Interventions: Heading for Evidence-Based Ways to Support Families. *ACAMH Occasional Papers, 29,* 47-57.

Kasari, C., Sigman, M., Mundy, P. & Yirmiya, N. (1988). Caregiver interactions with autistic children. *Journal of Abnormal Child Psychology, 16*(1), 45-56.

Koegel, R.L., Schreibman, .L, Loos, L.M., Dirlich-Wilhelm, H., Dunlap, G., Robbins, F.R. & Plienis, A.J. (1992). Consistent stress profiles in mothers of children with autism. *Journal of Autism and Developmental Disorders, 22*(2), 205-216.

Kuhn, J.C. & Carter, A,S, (2006) Maternal self-efficacy and associated parenting cognitions among mothers of children with autism. *The American Journal of Orthopsychiatry, 76*(4), 564-575.

Lord, C., Risi, S., Lambrecht, L., Cook, E.H. Jr., Leventhal, B.L., Dilavore, P.C., et al. (2000). The autism diagnostic observation schedule-generic: a standard measure of social and communication deficits associated with the spectrum of Autism. *Journal of Autism and Developmental Disorders, 30,* 205-223.

Luyster, R., Lopez, K. & Lord, C. (2007). Characterizing communicative development in children referred for autism spectrum disorders using the MacArthur-Bates Communicative Development Inventory (CDI). *Journal of Child Language, 34*(3), 623-654.

Makrygianni, M.K. & Reed, P. (2010). A meta-analytic review of the effectiveness of behavioral early intervention programs for children with Autistic Spectrum Disorders. *Research in Autism Spectrum Disorders, 4*(4), 577-593.

McConachie, H. & Diggle, T. (2007). Parent implemented early intervention for young children with autism spectrum disorder: a systematic review. *Journal of Evaluation in Clinical Practice, 13*(1), 120-129.

Moss, E., Dubois-Comtois, K., Cyr, C., Tarabulsy, G.M., St-Laurent, D. & Bernier, A. (2011). Efficacy of a home-visiting intervention aimed at improving maternal sensitivity, child attachment, and behavioral outcomes for maltreated children: a randomized control trial. *Development and Psychopathology, 23*(1), 195-210.

Mullen, E.M. (1995). *Mullen Scales of Early Learning.* Circle Pines, Minn: American Guidance Service.

Murphy, G.H., Beadle-Brown, J., Wing, L., Gould, J., Shah, A. & Holmes, N. (2005). Chronicity of challenging behaviors in people with severe intellectual disabilities and/or autism: a total population sample. *Journal of Autism and Developmental Disorders, 35*(4), 405-418.

Naber, F.B., Swinkels, S.H., Buitelaar, J.K., Bakermans-Kranenburg, M.J., Van IJzendoorn, M.H., Dietz, C., et al. (2007). Attachment in toddlers with autism and other developmental disorders. *Journal of Autism and Developmental Disorders, 37*(6), 1123-1138.

Noens, I.L. & Van Berckelaer-Onnes, I.A. (2005). Captured by details: sense-making, language and communication in autism. *Journal of Communication Disorders, 38*(2), 123-141.

Oono, I., Honey, E., & McConachie, H. (2013). Parent-mediated early intervention for young children with autism spectrum disorders (ASD). *Cochrane Database of Systematic Reviews, 4*, CD009774-CD009774. doi:10.1002/14651858.CD009774.pub2.

Pajareya, K. & Nopmaneejumruslers, K. (2011). A pilot randomized controlled trial of DIR/Floortime parent training intervention for pre-school children with autistic spectrum disorders. *Autism : The International Journal of Research and Practice, 15*(5), 563-577.

4

Pakenham, K.I., Sofronoff, K. & Samios, C. (2004). Finding meaning in parenting a child with Asperger syndrome: correlates of sense making and benefit finding. *Research in Developmental Disabilities, 25*(3), 245-264.

Peters-Scheffer, N., Didden, R., Korzilius, H. & Sturmey, P. (2011). A meta-analytic study on the effectiveness of comprehensive ABA-based early intervention programs for children with Autism Spectrum Disorders. *Research in Autism Spectrum Disorders, 5*(1), 60-69.

Ploog, B.O. (2010). Stimulus overselectivity four decades later: a review of the literature and its implications for current research in autism spectrum disorder. *Journal of Autism and Developmental Disorders, 40*(11), 1332-1349.

Pouw, L.B., Rieffe , C., Oosterveld, P., Huskens, B. & Stockmann, L. (2013) Reactive/proactive aggression and affective/cognitive empathy in children with ASD. *Research in Developmental Disabilities, 34*(4), 1256-1266.

Reichow, B., Barton, E.E., Boyd, B. A., & Hume, K. (2012). Early intensive behavioral intervention (EIBI) for young children with autism spectrum disorders (ASD). *Cochrane Database of Systematic Reviews, 10*, CD009260-CD009260. doi:10.1002/14651858.CD009260.pub2.

Rutgers, A., Bakermans-Kranenburg, M.J., Van IJzendoorn, M.H. & Van Berckelaer-Onnes, I. (2004). Autism and attachment: a meta-analytic review. *Journal of Child Psychology and Psychiatry, 45*(6), 1123-1134.

Rutgers, A.H., Van IJzendoorn, M.H., Bakermans-Kranenburg, M.J., Swinkels, S.H., Van Daalen, E., Dietz, C., et al. (2007). Autism, attachment and parenting: a comparison of children with autism spectrum disorder, mental retardation, language disorder, and non-clinical children. *Journal of Abnormal Child Psychology, 35*(5), 859-870.

Safe, A., Joosten, A. & Molineux, M, (2012). The experiences of mothers of children with autism: managing multiple roles. *Journal of Intellectual & Developmental Disability, 37*(4), 294-302.

Schlichting, J.E.P.T., Van Eldik, M.C.M., Spelberg, H.C., Van der Meulen, .S & Van der Meulen, B.F. (1995). *Schlichting Test voor Taalproductie*. Nijmegen: Berkhout.

Schwichtenberg, A. & Poehlmann, J. (2007). Applied behavior analysis: Does intervention intensity relate to family stressors and maternal well-being? *Journal of Intellectual Disability Research, 51*(8), 598-605.

The Netherlands Institute for Social Research/SCP (2010), Retrieved from: http://www.scp.nl (accessed 20 December 2012)

Tsai, W.C., Tsai, J.L. & Lotus Shyu, Y.I. (2008). Integrating the nurturer-trainer roles: Parental and behavior/symptom management processes for mothers of children with autism. *Social Science & Medicine (1982), 67*(11), 1798-1806.

Van Eldik, M.C.M., Schlichting, J.E.P.T., Spelber,g H.C., Van der Meulen, S. & Van der Meulen, B.F. (1995). *Reynell Test voor Taalbegrip.* Nijmegen: Berkhout.

Van IJzendoorn, M.H., Rutgers, A.H., Bakermans-Kranenburg, M.J., Swinkels, S.H., Van Daalen, E., Dietz, C. et al. (2007). Parental sensitivity and attachment in children with autism spectrum disorder: comparison with children with mental retardation, with language delays, and with typical development. *Child Development, 78*(2), 597-608.

Virues-Ortega, J. (2010). Applied behavior analytic intervention for autism in early childhood: meta-analysis, meta-regression and dose-response meta-analysis of multiple outcomes. *Clinical Psychology Review, 30*(4), 387-399.

Wallace, K.S. & Rogers, S.J. (2010). Intervening in infancy: implications for autism spectrum disorders. *Journal of Child Psychology and Psychiatry, and Allied Disciplines, 51*(12), 1300-1320.

Weiss, M.J. (2002). Hardiness and social support as predictors of stress in mothers of typical children, children with autism, and children with mental retardation. *Autism: The International Journal of Research and Practice, 6*(1), 115-130.

Zink, I. & Lejaegere, M. (2002). *N-CDI's: Lijsten voor Communicatieve Ontwikkeling. Aanpassing en hernormering van de MacArthur CDIs van Fenson et al.* Leuven/ Leusden: ACCO.

Zink, I. & Lejaegere, M. (2007). *N-CDI 3; Aanpassing en hernormering van de MacArthur CDI: Level III van Dale et al. (2000).* Culemborg: Centraal Boekhuis BV.

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Chapter 5

Improving Joint Attention of Children with Autism Spectrum Disorder: A Randomized Controlled Trial of a Parent-Mediated Intervention

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Abstract

We studied the effectiveness of the parent-mediated intervention program Video feedback Intervention to promote Positive Parenting adapted to Autism (VIPP-AUTI) on children's joint attention and play. VIPP-AUTI is a short-term home training with demonstrated effectiveness on parent sensitivity. Currently, we tested the effects of VIPP-AUTI on children's joint attention and play behavior in a randomized controlled trial with a sample of 78 children and their primary caregivers. Results demonstrated that VIPP-AUTI, as compared with usual home training, increased children's initiating joint attention behavior. No intervention effects were found on children's responding to joint attention and their play behavior. The positive effect on children's initiating joint attention skills supports the use of VIPP-AUTI in families of young children with ASD.

Introduction

Parent training aimed at enhancing the quality of parent-child interaction might be effective in decreasing symptomatology in children with Autism Spectrum Disorder (ASD), in particular with respect to their communication and socio-emotional development. In a randomized controlled trial, we demonstrated the effectiveness of a parent-mediated intervention program (VIPP-AUTI) in enhancing parental sensitive responsiveness toward their child with ASD. The intervention program reduced parents' intrusiveness (promoting reciprocal interaction with respect to their child's autonomy) and enhanced their feelings of efficacy in child rearing (Poslawsky et al., under review; see chapter 4). In the current study, we tested intervention effects on joint attention and play behavior in young children with ASD.

ASD is a neurodevelopmental disorder characterized by social and communication deficits, combined with repetitive or restricted behaviors and interests with an onset before 36 months of age (American Psychiatric Association [APA], 2013). One of the core deficits in young children with ASD is absent or limited joint attention (Lord et al. 2000; Luyster et al. 2007). Joint attention refers to a child's dyadic or triadic relation between self, other and object (Bakeman & Adamson, 1984; Leekam et al., 2000) and is related to prelinguistic social development (e.g. Charman, 2003;

Dawson et al., 2004) as part of the neurological maturation rooted in earliest infancy (Mundy et al., 2009).

Neurological development is sensitive to environmental influences. Children with ASD were found to benefit from early intervention targeting at improvement of joint attention skills (Patten & Watson, 2011), and joint attention skills have been found associated with the quality of parent-child interaction (Bruinsma et al. 2004; Kasari et al. 2010; Claussen et al. 2002; Siller & Sigman, 2002). Two types of joint attention are distinguished: (1) responding to joint attention (RJA); and (2) initiating joint attention (IJA). RJA is defined as the child's ability to follow and share another person's visual attention leads (e.g. gazing, pointing, etc.). IJA refers to the child's ability to create or indicate spontaneously another person's visual attention to share (e.g. Mundy et al., 2009). The level of joint attention of children with ASD is associated with their quality of play behavior (Charman, 1997) and with caregivers' sensitive responsiveness to the child during play interaction (sharing children's toy-directed attention in an undemanding and continuing way) (Siller & Sigman, 2002).

Play behavior reflects many developmental aspects of children, such as cognitive, linguistic, emotional and social development (Jordan, 2003) and is also influenced by the quality of parent-child interaction (Marcu et al., 2009; Naber et al., 2008). During the first two years of life, play development can be divided into the following stages: (1) simple object manipulation; (2) relational, non-functional play with two or more objects; (3) functional (conventional) play; and (4) symbolic (make-believe or pretend) play (Ungerer & Sigman, 1981). When children play with objects they intend playful engagement, sensorimotor exploration and symbolic functions through physical interaction with materials (Baranek et al., 2005). Social play refers to interactive play with other persons (Luckett et al., 2007). Infants with ASD showed atypical play development (Jarrold et al., 1993; Jarrold et al., 1996; Rutherford & Rogers, 2003), corresponding with deviations in their use of toys or being stuck in restricted interests (Ungerer & Sigman, 1981; Van Berckelaer-Onnes, 2003; Wulff, 1985; Williams, 2003).

In the current study, we investigated joint attention and play behavior of children with ASD after families received Video feedback

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Intervention to promote Positive Parenting adapted to Autism (VIPP-AUTI: Poslawsky et al., under review; see chapter 3) or care as usual. Although the intervention was not aimed at the children, but at their parents, we examined the effects of VIPP-AUTI on the children. We hypothesized that after parents received VIPP-AUTI, joint attention and play skills of children with ASD would improve via enhanced quality of parent-child interaction. Improvements of children's joint attention and play skills may be mediated by increased parental sensitive responsiveness. We assessed parental sensitive responsiveness before and after the intervention using the Emotionally Availability Scales (EAS; Biringen et al., 2000) with the subscales for sensitivity, structuring and non-intrusiveness, and found increased non-intrusiveness in parents who received the VIPP-AUTI intervention (Poslawsky et al., under review; see chapter 4). We expected to find positive effects of VIPP-AUTI on child-outcomes at the follow-up assessment (but not at the immediate post-test), based on findings in previous VIPP-related intervention studies (Bakermans-Kranenburg, et al., 2008; Klein Velderman et al., 2006) which show that changes in parental attitude and behavior may need some time to settle to stimulate observable changes in the child's behavior.

Method

The VIPP-AUTI study is a collaboration project between University Medical Center Utrecht (Department of Psychiatry) and Leiden University (Center for Child and Family Studies). As part of a single blind randomized controlled trial, in which parental sensitive responsiveness to children with ASD improved after receiving VIPP-AUTI (Poslawsky et al., under review; see chapter 4), we tested for therapeutic effects of VIPP-AUTI on joint attention and play skills of children with ASD.

Participants

Seventy-eight children with ASD (86% boys) and their primary caregivers (90% mothers) participated in this study. Children's age ranged from 16 to 61 months (M = 43.0, SD = 10.0). Children's developmental level ranged from mental retardation to above average functioning (range 49-124, M = 73.7, SD = 22.0). Age equivalent of children's language development was 32 months on average (M language comprehension= 32.8, SD = 17.1 and

M language production = 32.1, *SD* = 16.1). In 53% of the sample, the gap between the child's chronological age and age equivalent of language development was more than 6 months. Twelve percent of the children were raised bilingually. More than half of the children were first born (59%). Parental age ranged from 25 to 52 years (*M* = 36.6, *SD* = 5.0) and the majority of parents were Dutch (90%) and married (82%). The social economic status of the participating families was medium on average (*M* = 0.97, *SD* = 0.84 on a scale ranging from -1.84 to 2.23).
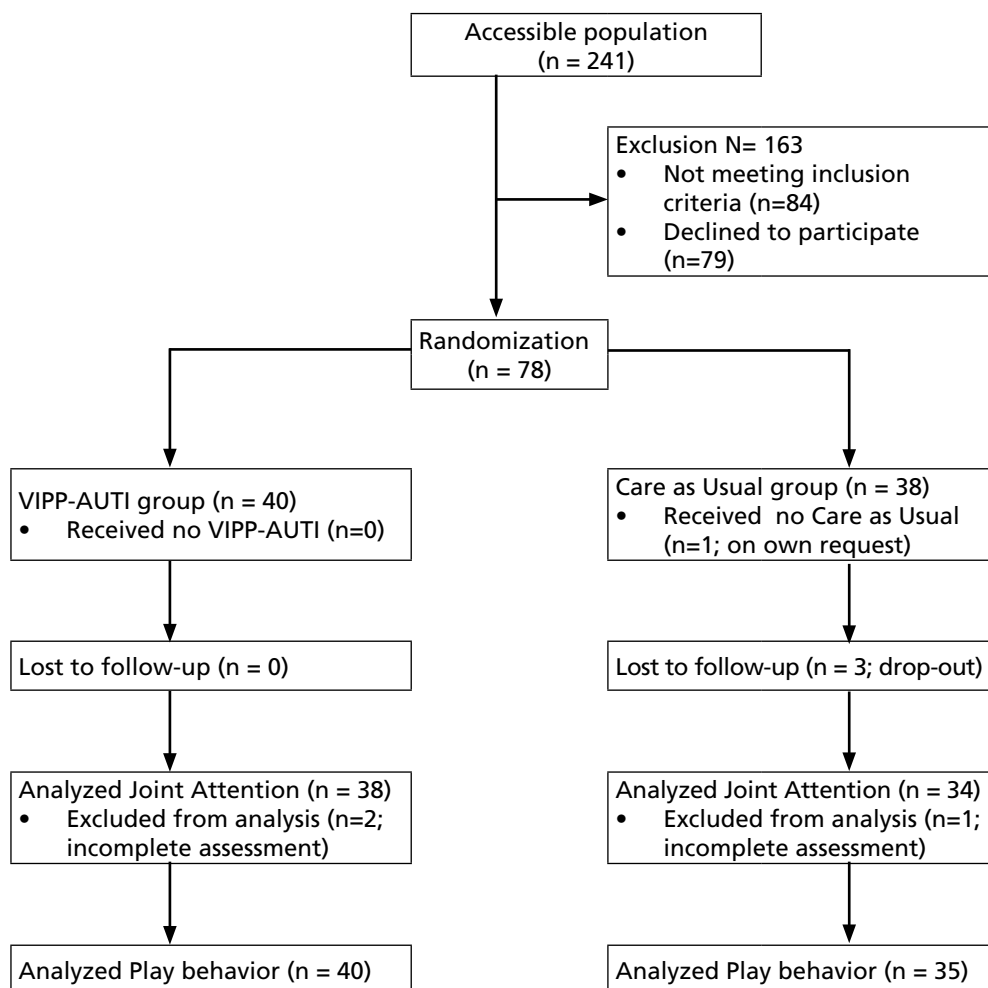
Diagnosis

Children were diagnosed as having ASD by a board-certified child psychiatrist according to extensive developmental history, all medical files, a semi structured observation (Autism Diagnostic Observation Schedule – Generic) and classification of the Diagnostic and Statistical Manual of Mental Disorders 4th Edition-Text Revision (DSM-IV-TR; APA, 2000). Of the children, 68% were diagnosed with Autistic Disorder (AD), and 32% were diagnosed with Pervasive Developmental Disorder (PDD-NOS). The majority of the children received ASD (with or without comorbid mental retardation) as the first and only diagnosis. In addition to the ASD diagnosis, some children (36%) received a diagnosis or a combination of diagnoses regarding somatic conditions; lung diseases (21%), sensory deficits (12% auditory and 6% visually problems), epilepsy (6%), growth disorder (4%), congenital heart muscles disease (3%), or metabolic disease (1%).

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Procedure

Newly ASD-diagnosed children under the age of five years and their primary caregivers were invited to participate in this intervention study at the university hospital. The inclusion period was from June 2008 to April 2012 (see Figure 1). After baseline assessments, participants were randomly divided into the experimental group (VIPP-AUTI) or control group (Care as Usual; CU). Randomization by computer generated tables was done by a staff manager who was not involved in the research project. Since both groups received home visits, families were not aware whether they received the experimental intervention or care as usual. Before randomization, at post-test, and after a no-treatment 3-month

follow-up period, children's joint attention was assessed at the university hospital and children's play behavior was observed at home. At the same time points, parents were asked to complete questionnaires, including assessments of the child's challenging behavior, because of its possible impact on parent-child interaction. Demographic data were administered prior to the study during the diagnostic phase. The study protocol was approved by the ethics committee of the University Medical Center Utrecht. Both parents gave written informed consent before participation.

Accessible population (n = 241)

Exclusion N= 163
- Not meeting inclusion criteria (n=84)
- Declined to participate (n=79)

Randomization (n = 78)

VIPP-AUTI group (n = 40)
- Received no VIPP-AUTI (n=0)

Care as Usual group (n = 38)
- Received no Care as Usual (n=1; on own request)

Lost to follow-up (n = 0)

Lost to follow-up (n = 3; drop-out)

Analyzed Joint Attention (n = 38)
- Excluded from analysis (n=2; incomplete assessment)

Analyzed Joint Attention (n = 34)
- Excluded from analysis (n=1; incomplete assessment)

Analyzed Play behavior (n = 40)

Analyzed Play behavior (n = 35)

Figure 1 *Flow chart of inclusion*

Care-as-Usual (CU)

All participants were offered usual care after receiving the child's ASD-diagnosis, including parental group meetings of psycho-education about ASD in general, pharmacological treatment when indicated or support from external care providers. Participants in the CU group received usual nursing home training consisting of five visits on average over a period of at most six months. The care, based on behavioral and family therapeutic interventions, was meant to support the parents and it focused on practical issues of parenting a child with ASD.

Experimental Group (VIPP-AUTI)

VIPP-AUTI was provided during three months, consisting of five home visits of 60-90 minutes each at a two-weekly interval (Poslawsky et al., under review; see chapter 3). As part of the VIPP-AUTI program, child and parent were videotaped in daily situations, particularly when they were playing together and during mealtimes. The intervener did not interfere during filming to ensure that a natural parent-child interaction was observed, including the child's entire behavioral repertoire. During each home visit, the previously filmed fragments of the individual parent-child interactions were discussed with the parent according to the intervention protocol. Each session was devoted to a theme regarding the quality of interaction related to observable ASD-characteristics, e.g. play behavior, joint attention, daily routines and stereotypical behavior. The autistic traits and the unique behavior of the child were highlighted, mirroring positive parent-child interaction patterns. The partners of the primary caregivers were invited to join the video feedback at the fifth home visit (a booster session).VIPP-AUTI was performed by comprehensively trained nurses, using a detailed manual and with intensive monitoring of therapy fidelity (by IP, FN, MB-K, MvIJ).

Measures

Autism diagnostic observation schedule - generic (ADOS – G)
The ADOS-G (Lord et al., 2000) quantifies deficits across the autism spectrum, controlling for effects of language and cognitive delay, in individuals with significant impairments. ADOS-G total score is the

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cumulative score of five test domains: communication, social reciprocity, play, stereotypic behavior and other problems. Higher ADOS-G total scores refer to more severe autistic symptoms. The ADOS-G consists of four modules. In the current study, children were assessed using module 1 ($n = 42$) and module 2 ($n = 36$), based on their level of expressive language. The child psychiatrist, who was certified for ADOS-reliability, administered the ADOS-G during the diagnostic phase prior to the study.

Mullen scales of early learning (MSEL)
The MSEL (Mullen, 1995) is a standardized developmental test that yields a mental age score for children between 3 and 68 months of age. The MSEL assesses five domains, one measuring cross motor skills (not used in the current study), and four cognitive domains: (1) visual reception, (2) fine motor skills, (3) receptive language, and (4) expressive language. The early learning composite standard score is a combination of non-verbal (domain 1 and 2) and verbal composites (domain 3 and 4), which ranges from low (<70) to high cognitive functioning (>70). The test was administered by a certified clinical psychologist at pretest.

Language development
The Reynell test for Dutch language comprehension (Van Eldik et al., 1995) and the Schlichting test for Dutch language production (Schlichting et al., 1995) were administered individually by a certified psychologist or speech language therapist at pretest and follow-up. The Reynell and Schlichting tests evaluate receptive and expressive language, respectively, for children between 14 and 75 months of age. If children were not able to do these tests (n = 26), the assistants collected parental reports using the Dutch version of the MacArthur-Bates Communicative Developmental Inventories (CDIs; Fenson, 1993; Fenson et al., 2007; N-CDIs; Zink & Lejaegere, 2002; Zink & Lejaegere, 2007) (n = 22). The CDIs consist of three forms, corresponding with different age groups; (1) 'Word and Gestures' (8-16 months of age), (2) 'Word and Sentences' (16-30 months of age) and (3) 'CDI-level III' (30-37 months of age), assessing vocabulary comprehension and production. The inventories were also used for children beyond the specified age ranges when they had impaired language development (Fenson et al.,

1994). The validity of the measure has been shown in children with and without language delay (Heilmann et al., 2005), and for children with autism (Charman et al. 2003; Luyster et al., 2007).

In the current study, language development score was based on the converted age equivalents of total language comprehension scores of the Reynell or N-CDI, and total word-production scores of the Schlichting or N-CDI.

Aberrant behavior checklist (ABC)
The ABC (Aman et al., 1985) is a parent report questionnaire to assess children's atypical and challenging behavior. The ABC contains 58 items scored on a Likert scale with 0 = not true, 1 = somewhat or sometimes true, 2 = regularly true and 3 = very (often) true. The ABC comprises five factors: (1) irritability, agitation, crying; (2) lethargy, social withdrawal; (3) stereotypic behavior; (4) hyperactivity, noncompliance; (5) inappropriate speech. The ABC was reported to be adequate for assessing the severity of autistic behaviors in early childhood (Karabekiroglu & Aman, 2009). In this sample, internal consistency of the scale (Cronbach's alpha) was .94.

Socio-economic status (SES)
SES was based on status scores 2010 of postal areas. Status scores were derived from level of education, employment and income per postal area by The Netherlands Institute for Social Research (SCP, 2010). Higher status scores refer to higher SES.

Early social and communication scales (ESCS)
The ESCS (Mundy et al., 2003) scores are based on videotaped semi-structured interaction to measure joint attention. Joint attention behaviors, behavioral requests and social interaction behaviors are evoked by different types of play tasks initiated by the experimenter. The assessment was performed according to protocol. The child was seated opposite to the investigator at a small table. The experimenter presented the child with an array of novel toys. The toys included three small wind-up toys, three hand operated toys including a balloon, a small car, a ball, a picture book, a comb, cap and glasses.

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Posters were positioned on the walls to the left, right and behind the child. Throughout the session, only one toy at a time was presented to the child. The original scoring methods as described in the manual were followed with lower scores for lower level joint attention behavior (for example makes eye contact, reaches for a toy) and higher scores for higher level joint attention behavior (points at something while maintaining eye contact, offers a toy while maintaining eye contact). A combination of 16 tasks and a score for verbalization allowed a child to reach a maximum total score of 143 points. IJA (8 of the 16 tasks), which refers to the frequency with which the child uses eye contact, pointing and showing to initiate shared attention to objects or events, and RJA (5 of the 16 tasks), which refers to the child's skill in following the tester's line of vision and pointing gestures, were rated. Based on 13% of randomly chosen cases, the intra-class correlation coefficient of two independent coders, blind to each child's experimental condition, was .92 for IJA and .94 for RJA.

Play behavior
Children's play behavior was observed during a 15-minute videotaped free-play session. The children were provided with a standardized set of toys including a tea set (cups, saucers, teapot and spoons), a doll and related attributes, cars, a garage, a puzzle, a pop-up toy, a spinner and a book. The parent was instructed to passively monitor while the child was playing. When the child was seeking contact or interaction, the parent was allowed to respond in a natural way. Play behavior was coded using the ethogram of Naber et al., (2008), based on an ethogram previously developed by Ungerer and Sigman (1981). The video segments were watched and coded by trained students for toy preference and level of play category; (1) manipulative, (2) relational, (3) functional, and (4) symbolic play, every 10 seconds during 15 minutes. The highest level of play shown (ranging from 1 to 4) and variation in play by toy preference were used in the analyses. Based on 16% of randomly chosen cases, intra-class correlation coefficients of three independent coders, unaware of the child's experimental condition, for level of play and variation in toy preference were .71 and .99, respectively.

Results

To check the equivalence of the experimental and control group at pretest after the random group assignment, independent sample *t*-tests and chi-squared tests were performed. No significant differences between the VIPP-AUTI and CU group were found for child's gender, age, autism characteristics, cognitive and language developmental level and challenging behavior at pretest (see Table 1). However, significant group differences were found on child's school attendance. Regardless of age, more children in the CU group visited school ($n = 10$) than children in the VIPP-AUTI group ($n = 4$), χ^2 (1, $N = 76$) = 3.99, $p < .05$. School attendance was thus included as a factor in further analyses.

Table 1 Background and pretest variables in experimental and control groups

Child Characteristics	Group					
	VIPP-AUTI (n = 40)		CU (n = 38)			
	M	*SD*	*M*	*SD*	*t*	*p*
Age (months)	42.16	09.02	43.80	10.92	0.72	.47
Autistic characteristics	25.03	10.55	26.35	10.10	0.56	.58
Developmental level	74.63	23.50	72.68	20.61	0.39	.70
Language comprehension age equivalent (months)	32.77	16.41	32.88	18.04	0.03	.98
Language production age equivalent (months)	33.71	15.89	30.22	16.35	0.85	.40
Child's challenging behaviors (ABC)	53.96	26.82	51.08	23.30	0.49	.63

Note. ABC: Aberrant Behavior Checklist.

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Bivariate correlations among post-intervention parental sensitivity, structuring and non-intrusiveness (EAS scores) and follow-up joint attention and play behavior were computed. As expected, IJA and RJA were significantly correlated (r (72) = .62, $p < .01$). Parental sensitivity and structuring were significantly related to child joint attention; sensitivity and IJA (r (72) = .35, $p < .01$), sensitivity and RJA (r (72) =.35, $p < .01$), structuring and IJA (r (72) = .27, $p < .05$), structuring and RJA (r (72) =.34, $p < .01$). More parental sensitivity and structuring were associated with better joint attention performance of the child. No significant relations were found between parental non-intrusiveness and child joint attention. Child play behavior did not show significant correlations with parental sensitivity or non-intrusiveness. Parental structuring was associated with higher levels of child play (r (72) =.26, $p < .05$), but not significantly correlated with child variation in toy preference. Level of play was significantly related to RJA (r (72) = .37, $p < .01$), but not to IJA (r (72) = .19, $p = .10$) (see Table 2).

Table 2 Pearson correlation matrix among parental sensitive, structuring and non-intrusiveness by scores on the Emotional Availability Scales at posttest, Joint Attention, and Play behavior at follow-up (N=72)

	Non-intrusiveness	Sensitivity	Structuring	IJA[a]	RJA[b]	Play level	Play variation
Non-intrusiveness		.70**	.59**	.12	.14	.14	.23
Sensitivity			.72**	.35**	.35**	.21	.16
Structuring				.27*	.34**	.26*	.17
IJA[a]					.62**	.19	.04
RJA[b]						.37**	.17
Play level							.10

Note. * $p < .05$, ** $p < .01$, [a]IJA: Initiating Joint Attention sum scores, [b]RJA: Responding Joint Attention sum scores

Intervention Effect on Children's Joint Attention

A repeated measures multivariate analysis of variance (MANOVA) with VIPP-AUTI versus CU and school attendance as between-subjects factor and time as within-subjects factor was performed to assess the development of joint attention behavior across pretest and follow-up assessment. The three-way interaction of group by school attendance by time was significant for IJA, $F (8, 61) = 2.35$, $p = .03$, partial $\eta^2 = .24$ (see Table 3). The three-way interaction of group by school attendance by time for RJA was not significant, $F (5, 64) = 1.84$, $p = .12$, partial $\eta^2 = .13$.

In particular, for children who did not attend school, the intervention was effective in increasing IJA behavior between pretest and follow-up, $F (8, 49) = 2.41$, $p = .03$, partial $\eta^2 = .28$ (see Figure 2). Because VIPP-AUTI demonstrated only effectiveness in enhancing parental non-intrusiveness (Poslawsky et al., under review; see chapter 4) and parental non-intrusiveness at post-test was not significantly related to child joint attention at follow-up, parental non-intrusiveness could not mediate the effect of VIPP-AUTI on children's IJA skills.

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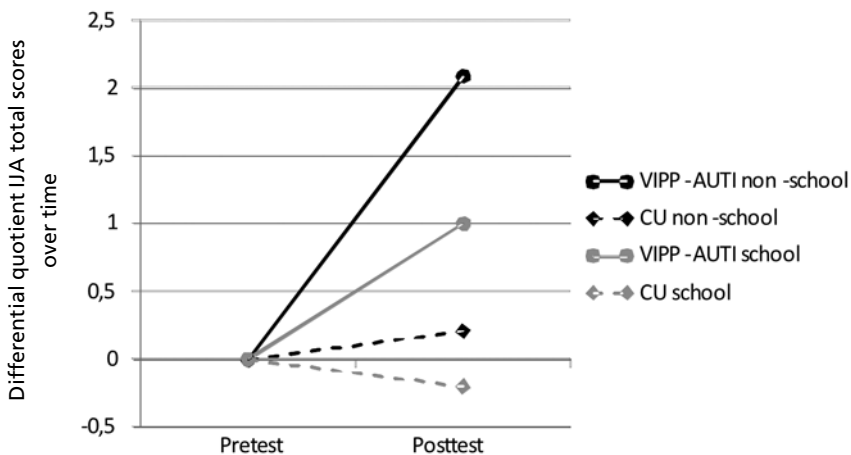
Figure 2 *Development of initiating joint attention (as indicated by scores on the Early Social Communication Scales) across pretest and follow up for experimental (VIPP-AUTI) and control (CU) groups with and without school attendance*

Table 3 Mean values of Joint Attention (as indicated on scores by the Early Social Communication Scales) and Play behavior of groups at pretest and follow-up

Joint Attention	Group			
	VIPP-AUTI (n=38)		Care as Usual (n=34)	
	M	SD	M	SD
Pretest				
IJA[a]	31.45	07.12	34.32	05.68
RJA[b]	48.05	17.03	50.56	14.93
Follow-up				
IJA[a]	33.42	06.86	34.41	05.70
RJA[b]	51.68	17.38	52.94	17.95
Play behavior	(n=40)		(n=35)	
Pretest				
Play level	3.13	0.69	3.20	0.68
Play variation	6.88	2.14	6.91	2.11
Follow-up				
Play level	3.28	0.60	3.29	0.62
Play variation	5.83	2.28	6.46	2.09

Note. .[a]IJA: Initiating Joint Attention sum scores, [b]RJA : Responding Joint Attention sum scores

Intervention Effect on Children's Play Behavior

A repeated measures MANOVA with VIPP-AUTI or CU and school attendance as between-subjects factor and time as within-subjects factor revealed no significant three-way interaction of group by school attendance by time for level of play and play variation across

pretest and follow-up assessment, $F(2,70) = 0.44$, $p = .65$ partial $\eta^2 = .01$ (see Table 3). For children who did not attend school no significant interaction of group by time was found either, $F(2, 58) = 0.15$, $p = .86$, partial $\eta^2 = .01$.

Discussion

In this randomized controlled trial the parent-mediated VIPP-AUTI program proved to be effective on improving child initiating joint attention behavior. Three months post-intervention, the children of parents who received the VIPP-AUTI intervention demonstrated higher levels of IJA than children whose parents received usual nursing home training. In particular, children who did not yet attend school demonstrated enhanced IJA skills after the VIPP-AUTI program. Intervention effects on responding to joint attention and play behavior were however not significant.

As mentioned before, IJA refers to the child's ability to create or indicate spontaneously another person's visual attention to share, while RJA is defined as the child's ability to follow and share another person's visual attention leads (e.g. Mundy et al., 2009). VIPP-AUTI demonstrated to have a therapeutic effect on the child's IJA but not on RJA, despite significant associations between IJA and RJA. An explanation may be that IJA and RJA follow different pathways in social neurocognitive development (Mundy et al, 2009), accompanied by differences in intervention susceptibility. Furthermore, an association between the quality of parent-child relationship and infants' IJA rather than RJA was found previously (Claussen et al., 2002), which is in line with our results.

In our sample, school attendance was a potentially confounding factor, because more children in the control group visited school than did children in the VIPP-AUTI group. The effectiveness of VIPP-AUTI on children's IJA was demonstrated especially in children who attended day-care instead of school. It is possible that the underlying factor explaining the results is the amount of time the parent and child spent together. Children who visited day-care spent fewer hours per week outside their home environment than children attending primary school. Thus, children who did not attend school spent more hours at home, and potentially profited more from the effects of the

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intervention on their parents. Our study included however only 14 children attending school, and replication of the moderating effect of school attendance in a larger sample is badly needed.

VIPP-AUTI demonstrated effectiveness in enhancing parental non-intrusiveness (Poslawsky et al., under review; see chapter 4). Given the absence of an association between parental non-intrusiveness at post-test and children's joint attention at follow-up, the effect of VIPP-AUTI on children's IJA could not be explained by parental non-intrusiveness as a mediating variable. Additional parent-centered factors, beyond our study focus, might have acted as mediators. VIPP-AUTI aims at promoting parents' understanding of the autistic traits of their child, in order to optimize parent-child interaction. The intervention addresses aspects of parental states of mind, such as mind-mindedness or insightfulness. Mind-mindedness refers to parents' attuned comments to the child's state of mind (Meins et al., 2003), and is associated with social-cognitive development of the child (Meins et al., 2013). Parental insightfulness focuses somewhat broader on the relation between positive parenting and parents' empathic insight in their children's motives underlying their behavior (Oppenheim & Koren-Karie, 2002). VIPP-AUTI might have enhanced maternal insightfulness of their children. In future studies with VIPP-AUTI, the potential mediating role of mind-mindedness and insightfulness in changing children's developmental outcome should be examined.

No significant intervention effect was found on children's play behavior. Children of both groups showed the same levels of play and variation in play. Play behavior was measured during unstructured, solitary play of the children. Although the primary caregiver was in the same room, the instruction was that he or she stays passive. As part of the VIPP-AUTI intervention, parents were shown how to motivate their child, challenging the child to engage in more active play. However, during free play, the child has to motivate itself, which may result in its familiar and often restricted play behavior. An assessment of children's play level in a more structured session might show evidence of experimentally enhanced quality of parent-child interaction and child play as a result. It should be noted that other studies aimed at improving play behavior of children with ASD provided treatment at

the level of the child (Jung & Sainato, 2013), while in our study VIPP-AUTI focused on the parent in interaction with the child. Whereas VIPP-AUTI addressed enjoyable engagement and awareness of the child's interests to improve children's play development, more active, direct and systematic teaching strategies may be needed, including a play context with peers (Jung & Sainato, 2013). Optimizing parent-child interaction, which is the aim of VIPP-AUTI, may serve as a base for additional treatment targeted at improving play behavior of children with ASD.

We compared the effects of VIPP-AUTI with an intensive type of care as usual. Therefore, the finding of children's enhanced IJA can be interpreted as treatment specific, which supports the use of VIPP-AUTI in clinical practice. However, a limitation of the study is the relatively brief follow-up period of three months for the assessment of child outcomes. Another limitation is the potentially restricted generalizability due to the rather homogeneous sample of families' sociodemografics and the mono-center study design. Therefore, future research including multicenter and longitudinal studies is needed.

In sum, this randomized controlled trial demonstrated the effectiveness of VIPP-AUTI, a short term parent-mediated program, in influencing initiating joint attention in children with ASD. We suggest that VIPP-AUTI can provide early and effective support in parenting children with ASD to promote some of these children's crucial social skills even on top of specialized care as usual.

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References

Aman, M. G., Singh, N. N., Stewart, A. W., & Field, C. J. (1985). Psychometric characteristics of the aberrant behavior checklist. *American Journal of Mental Deficiency, 89*(5), 492-502.

American Psychiatric Association (2013). *Diagnostic and Statistical Manual of Mental Disorders (Fifth Edition)*. Washington DC.

American Psychiatric Association (2000). *Diagnostic and Statistical Manual of Mental Disorders (Fourth Edition, Text Revision)*. Washington DC.

Bakeman, R., & Adamson, L. B. (1984). Coordinating attention to people and objects in mother-infant and peer-infant interaction. *Child Development, 55*(4), 1278-1289.

Bakermans-Kranenburg, M. J., Van IJzendoorn, M. H., Pijlman, F. T., Mesman, J., & Juffer, F. (2008). Experimental evidence for differential susceptibility: dopamine D4 receptor polymorphism (DRD4 VNTR) moderates intervention effects on toddlers' externalizing behavior in a randomized controlled trial. *Developmental Psychology, 44*(1), 293-300.

Baranek, G. T., Barnett, C. R., Adams, E. M., Wolcott, N. A., Watson, L. R., & Crais, E. R. (2005). Object play in infants with autism: methodological issues in retrospective video analysis. *The American Journal of Occupational Therapy.: Official Publication of the American Occupational Therapy Association, 59*(1), 20-30.

Biringen, Z., Robinson, J.L., & Emde, R.N. (2000). Appendix B: The emotional Availability Scales (3rd ed.; and abridged infancy/ early childhood version). *Attachment and Human development,* 2, 256-270.

Bruinsma, Y., Koegel, R. L., & Koegel, L. K. (2004). Joint attention and children with autism: a review of the literature. *Mental Retardation and Developmental Disabilities Research Reviews, 10*(3), 169-175.

Charman, T. (1997). The relationship between joint attention and pretend play in autism. *Development and Psychopathology, 9*(1), 1-16.

Charman, T. (2003). Why is joint attention a pivotal skill in autism? *Philosophical Transactions of the Royal Society of London.Series B, Biological Sciences, 358*(1430), 315-324.

Charman, T., Drew, A., Baird, C., & Baird, G. (2003). Measuring early language development in preschool children with autism spectrum disorder using the MacArthur Communicative Development Inventory (Infant Form). *Journal of Child Language, 30*(1), 213-236.

Claussen, A. H., Mundy, P. C., Mallik, S. A., & Willoughby, J. C. (2002). Joint attention and disorganized attachment status in infants at risk. *Development and Psychopathology, 14*(2), 279-291.

Dawson, G., Toth, K., Abbott, R., Osterling, J., Munson, J., Estes, A., et al. (2004). Early social attention impairments in autism: social orienting, joint attention, and attention to distress. *Developmental Psychology, 40*(2), 271-283.

Fenson, L, Dale, PS, Reznick, JS, Bates E, Thal, D, Hartung, J, et al. (1993). *Guide and technical manual for the MacArthur Communicative Development Inventories.* San Diego, CA: Singular Press.

Fenson, L., Dale, P. S., Reznick, J. S., Bates, E., Thal, D. J., & Pethick, S. J. (1994). Variability in early communicative development. *Monographs of the Society for Research in Child Development, 59*(5), 1-173; discussion 174-85.

Fenson, L., Marchman, V. A., Thal, D., Dale, P. S., Bates, E., & Reznick, J. S. (2007). *The MacArthur-Bates Communicative Development Inventories:User's Guide and technical Manual (2nd edition).* Baltimore: Paul H. Brookes.

Heilmann, J., Weismer, S. E., Evans, J., & Hollar, C. (2005). Utility of the MacArthur-Bates communicative development inventory in identifying language abilities of late-talking and typically developing toddlers. *American Journal of Speech-Language Pathology / American Speech-Language-Hearing Association, 14*(1), 40-51.

Jarrold, C., Boucher, J., & Smith, P. (1993). Symbolic play in autism: a review. *Journal of Autism and Developmental Disorders, 23*(2), 281-307.

Jarrold, C., Boucher, J., & Smith, P. K. (1996). Generativity deficits in pretend play in autism. *British Journal of Developmental Psychology, 14*, 275-300.

Jordan, R. (2003). Social play and autistic spectrum disorders: a perspective on theory, implications and educational approaches. *Autism : The International Journal of Research and Practice, 7*(4), 347-360.

Jung, S., & Sainato, D. M. (2013). Teaching play skills to young children with autism. *Journal of Intellectual & Developmental Disability, 38*(1), 74-90.

Karabekiroglu, K., & Aman, M. G. (2009). Validity of the aberrant behavior checklist in a clinical sample of toddlers. *Child Psychiatry and Human Development, 40*(1), 99-110.

Kasari, C., Gulsrud, A. C., Wong, C., Kwon, S., & Locke, J. (2010). Randomized controlled caregiver mediated joint engagement intervention for toddlers with autism. *Journal of Autism and Developmental Disorders, 40*(9), 1045-1056.

5

Klein Velderman, M., Bakermans-Kranenburg, M.J., Juffer, F., Van IJzendoorn, M.H., Mangelsdorf, S., & Zevalkink, J. (2006). Preventing preschool externalizing behavior problems through video-feedback intervention in infancy. Infant Mental Health Journal, 27, 466-493.

Leekam, S. R., Lopez, B., & Moore, C. (2000). Attention and joint attention in preschool children with autism. *Developmental Psychology, 36*(2), 261-273.

Lord, C., Risi, S., Lambrecht, L., Cook, E. H., Jr., Leventhal, B. L., Dilavore, P. C, et al. (2000). The autism diagnostic observation schedule-generic: a standard measure of social and communication deficits associated with the spectrum of Autism. *Journal of Autism and Developmental Disorders, 30*, 205-223.

Luckett, T., Bundy, A., & Roberts, J. (2007). Do behavioral approaches teach children with autism to play or are they pretending? *Autism, 11*(4), 365-388.

Luyster, R, Lopez, K, & Lord, C (2007). Characterizing communicative development in children referred for autism spectrum disorders using the MacArthur-Bates Communicative Development Inventory (CDI). *Journal of Child Language, 34*(3), 623-654.

Marcu, I., Oppenheim, D., Koren-Karie, N., Dolev, S., & Yirmiya, N. (2009). Attachment and symbolic play in preschoolers with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 39*(9), 1321-1328.

Meins, E., Fernyhough, C., Wainwright, R., Clark-Carter, D., Das Gupta, M., Fradley, E., & Tuckey, M. (2003). Pathways to understanding mind: construct validity and predictive validity of maternal mind-mindedness. *Child Development, 74*(4), 1194-1211.

Meins, E., Fernyhough, C., Arnott, B., Leekam, S. R., & De Rosnay, M. (2013). Mind-Mindedness and Theory of Mind: Mediating Roles of Language and Perspectival Symbolic Play. *Child Development,* doi:10.1111/cdev.12061; 10.1111/cdev.12061

Mullen, E. M. (1995). *Mullen Scales of Early Learning.* Circle Pines, Minn: American Guidance Service.

Mundy, P. Delgado, C., Block, J., Venezia, M., Hogan, A. & Seibert, J. (2003) *A Manual for the Abridged Early Social Communication Scales (ESCS)*. University of Miami. Retrieved from: http://www.ucdmc.ucdavis.edu/mindinstitute/ourteam/faculty_staff/escs.pdf Accessed 14 may 2008 and 23 November 2012.

Mundy, P., Sullivan, L., & Mastergeorge, A. M. (2009). A parallel and distributed-processing model of joint attention, social cognition and autism. *Autism Research : Official Journal of the International Society for Autism Research, 2*(1), 2-21.

Naber, F. B., Bakermans-Kranenburg, M. J., Van IJzendoorn, M. H., Swinkels, S. H., Buitelaar, J. K., Dietz, C., et al. (2008). Play behavior and attachment in toddlers with autism. *Journal of Autism and Developmental Disorders, 38*(5), 857-866.

Oppenheim, D., & Koren-Karie, N. (2002). Mothers' insightfulness regarding their children's internal worlds: The capacity underlying secure child-mother relationships. *Infant Mental Health Journal, 23*(6), 593-605.

Patten, E., & Watson, L. R. (2011). Interventions Targeting Attention in Young Children With Autism. *American Journal of Speech-Language Pathology, 20*(1), 60-69.

Rutherford, M. D., & Rogers, S. J. (2003). Cognitive underpinnings of pretend play in autism. *Journal of Autism and Developmental Disorders, 33*(3), 289-302.

Sigman, M., & Ungerer, J. A. (1984). Attachment behaviors in autistic children. *Journal of Autism and Developmental Disorders, 14*(3), 231-244.

Siller, M., & Sigman, M. (2002). The behaviors of parents of children with autism predict the subsequent development of their children's communication. *Journal of Autism and Developmental Disorders, 32*(2), 77-89.

Schlichting,, J. E. P. T. , Van Eldik, M. C. M., Spelberg, H. C., Van der Meulen, S., & Van der Meulen, B. F. (1995). *Schlichting Test voor Taalproductie*. Nijmegen: Berkhout.

The Netherlands Institute for Social Research/SCP (2010), Retrieved from: http://www.scp.nl Accessed 4 December 2012.

Ungerer, J. A., & Sigman, M. (1981). Symbolic play and language comprehension in autistic children. *Journal of the American Academy of Child Psychiatry, 20*(2), 318-337.

Van Berckelaer-Onnes, I. A. (2003). Promoting early play. *Autism: The International Journal of Research and Practice, 7*(4), 415-423.

Van Eldik,, M. C. M., Schlichting, J. E. P. T., Spelberg, H. C., Van der Meulen, S. & Van der Meulen, B. F. (1995). *Reynell Test voor Taalbegrip.* Nijmegen: Berkhout.

Williams, E. (2003). A comparative review of early forms of object-directed play and parent-infant play in typical infants and young children with autism. *Autism : The International Journal of Research and Practice, 7*(4), 361-377.

Wulff, S. B. (1985). The symbolic and object play of children with autism: a review. *Journal of Autism and Developmental Disorders, 15*(2), 139-148.

Zink, I. & Lejaegere, M. (2002). *N-CDI's: Lijsten voor Communicatieve Ontwikkeling. Aanpassing en hernormering van de MacArthur CDIs van Fenson et al.* Leuven/Leusden: ACCO.

Zink, I. & Lejaegere, M. (2007). *N-CDI 3; Aanpassing en hernormering van de MacArthur CDI: Level III van Dale et al. (2000).* Culemborg: Centraal Boekhuis BV

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Chapter 6

**Oxytocin Enhances Paternal Sensitivity
to a Child with Autism:
A Double-Blind Within-Subject Experiment with
Intranasally Administered Oxytocin.**

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Abstract

Oxytocin seems associated with parenting style, and experimental work showed positive effects of intranasally administered oxytocin on parenting style of fathers. Here, the first double-blind, placebo-controlled, within-subject experiment with intranasal oxytocin administration to fathers of children with autism spectrum disorder (ASD) is presented. Fathers with their typically developing toddler (n=18), and fathers of toddlers diagnosed with ASD (n=14), were observed in two play sessions of 15 minutes each with an intervening period of 1 week. In all fathers oxytocin elevated the quality of paternal sensitive play: fathers stimulated their child in a more optimal way, and they showed less hostility which suggests the positive effects of oxytocin on paternal sensitive play irrespective of clinical status of their child.

Introduction

The neuropeptide oxytocin creates warm feelings for offspring (Carter, 1998; Feldman et al., 2007; Galbally et al., 2011; Insel, 1992) and support empathic concern for conspecifics (MacDonald & MacDonald, 2010) through better recognition of emotional facial expression (Bartz et al., 2010). Moreover, it would elevate the level of trust in other human beings, in particular when they belong to the in-group (De Dreu et al., 2010; Kosfeld et al., 2005). Oxytocin administration was also shown to enhance the ability of adults with autism to understand emotions in speech (Hollander et al., 2007), and to help children with autism to better recognize people's intentions by reading their eyes (Guastella et al., 2010).

In a recent meta-analysis on experiments with intranasally administered oxytocin in non-clinical samples it was concluded that a sniff of oxytocin indeed changes emotion perception and behavior in trusting relationships (Van IJzendoorn & Bakermans-Kranenburg, 2012). In one of the experiments it was shown that fathers of typically developing toddlers showed more sensitive structuring of their play after they were administered oxytocin (Naber et al., 2010). Here we test through a double-blind within-subject experiment whether sensitive responsiveness of fathers for their child with autism can be enhanced by intranasally administered oxytocin similar to fathers of typically developing children.

Oxytocin is produced in the hypothalamus, in particular in the

paraventricular nucleus of the hypothalamus, and sent down into the pituitary to reach the bloodstream as a hormone, or up into the limbic system and cortex as a neurotransmitter. Oxytocin promotes lactation (Panksepp, 1998) and is also involved in enhancing the sensitivity for social and emotional signals, and lowering feelings of anxiety and stress (Carter, 1998). In a study with functional magnetic resonance imaging (fMRI) it was demonstrated that oxytocin administration reduced right amygdala activation and enhanced insula and inferior frontal gyrus activation when subjects were exposed to infant crying compared to scrambled control sounds. Reduced amygdala activation may point to decreased aversive feelings to cry signals whereas increased activation of the insula and inferior frontal gyrus may reflect more empathic processing of emotional stimuli (Riem et al., 2011).

Oxytocin is not only involved in contractions during parturition and in milk production, but also in the development of parent-infant attachment, in mothers as well as in fathers. In a pioneering set of studies Feldman and her team (2007; 2010) examined oxytocin across pregnancy and the early postpartum period in relation to subsequent maternal and paternal interactive behavior to the infant. Elevated oxytocin levels were found to be associated with more sensitive parental interactions with the infant. Examining oxytocin in mothers and fathers engaged in a 15-min play-and-contact interaction session with their 4-6 month old infants Feldman et al. (2010) found similar levels of oxytocin in mothers and fathers which were associated with parent-specific modes of interaction. Whereas higher maternal oxytocin levels were associated with more affectionate touch, higher paternal oxytocin levels were uniquely associated with more stimulatory play, but not affectionate touch. Furthermore, highly affectionate mothers showed oxytocin increases following playful interactions with their infants while similar increases were observed in highly stimulating fathers.

In an experimental study with fathers of typically developing toddlers we showed that intranasal oxytocin administration leads to more responsive interactions of fathers with their child during play (Naber et al., 2010). In the oxytocin condition fathers stimulated their child's exploration and autonomy in a better way than in the placebo condition. The fathers also tended to show less hostility to the child in the oxytocin condition

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showing less impatience and discontent. This experimental finding nicely converges with the Feldman et al. (2010) study that found elevated oxytocin levels in fathers who were asked to engage in a "play-and touch" interaction session with their infant. Both studies combined may indicate that fathers who are more responsive when interacting with their infants during play may produce more oxytocin, which in turn may increase the pleasure of the father in the interaction with the child and contribute to the father's ability to provide responsive care of a stimulating nature.

Parenting a child requires many social skills of the parent. Parenting a child with autism seems even more complicated. For example, when a typical developing child is crying, one usually soothes the child by holding it. However, in the case of autism, holding can be experienced by the child as even more stressful. The signals of children with autism are not always easy to understand, or they trigger parental responses that are adequate for typically developing children, but not for children with autism. The difficulties in social interaction of a child with autism are often reported by parents from the first months of the child's life, long before the child receives the diagnosis of autism (Lord, 1995). It seems that the parents of a child with autism need to interact in different ways compared to parents of typically developing children. They need to be sensitive to the autistic characteristics of their child, in order to avoid intrusiveness or overstimulation.

Because fathers of children with autism are challenged even more than fathers of typically developing children in finding sensitive ways of playful interactions we examine here the effects of intranasally administered oxytocin on paternal sensitivity and structuring during play. We expect to find an increase in paternal sensitivity during play with a child with autism to a similar extent as in case of fathers of a typically developing child.

Method

Procedure
Eighteen fathers of typical developing children (mean age 37.4 years, *SD* = 4.23, range 31-45) and fourteen fathers of a child with ASD (mean age 39.4 years, *SD* = 6.46, range 33-45) participated in the double-blind,

placebo-controlled, within-subject design. Child psychiatrists diagnosed the children as defined by the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV) and the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2002). Seventeen of the 18 fathers of the typically developing children were included in a previous oxytocin experiment (see Naber et al., 2010). All participants received once intranasal oxytocin and once a placebo to investigate the effects of oxytocin on the interaction with their child. Neither the experimenter nor the participant knew during which visit the participant received oxytocin; roughly half of them (n=15) received oxytocin during the first visit. The study protocol was approved by the ethics committee of the University Medical Center Utrecht. All fathers gave written informed consent before their participation. Written informed consent for the children was given by both parents. The participants were all healthy volunteers with at least one child in the age between 1.5 and 6 years of age (mean age 44.0 months, SD = 14.51). Children with ASD were older (mean age 56.14 months, SD = 7.20) compared to the typically developing children (mean age 33.41 months, SD = 10.72), t = -6.77, p <.01. A single dose of 24 IU oxytocin nasal spray (Syntocinon spray, Novartis, Basal, Switzerland) or placebo, nasal spray without oxytocin, was administered intranasally 45 minutes before the start of the play session. In a recent study we showed that oxytocin levels remain elevated even more than 2 hours after intranasal administration (Huffmeier et al., 2012). In the 45 minutes before the play session the fathers performed (computer) tasks without their child. Participants underwent both the oxytocin and the placebo conditions with an interval of one week in a balanced within-subject design.

The play sessions lasted 15 minutes and were slightly different on both occasions. During the first visit they played a game in which father and child in turn placed tiny dolls on a rocking tower until the tower fell over, they played with a doctor-kit, and they made a large floor-puzzle. During the second visit, father and child were invited to build a tower of magnets, after which they played with a doll and a tea-set, and then painted a coloring picture. Fathers were instructed to play with their child as they usually did. Both sessions took place at home and were videotaped for later analysis.
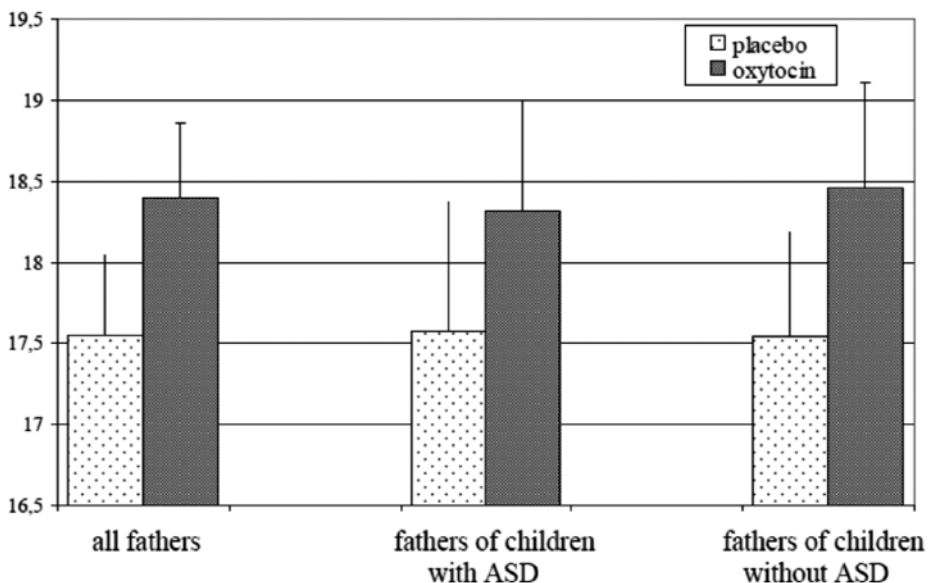
Measures

Paternal Responsiveness

The Emotional Availability Scales (EAS; Biringen et al., 2000) were used to assess paternal sensitivity. The scales consist of four subscales for parenting behavior; sensitivity, structuring, non-intrusiveness, and non-hostility. Sensitivity of the parent was coded on a 9-point rating scale. High sensitivity refers to a parent's responsiveness to the child's emotional signals and communication in a flexible way. A parent was coded insensitive when interaction between parent and child was rare or inflexible. Parental structuring was coded on a 5-point rating scale. It refers to the parental ability to support learning and exploration with respect for the child's autonomy. When a parent does not offer structure or does not involve the child in play, low scores on structuring are assigned. High scores for structuring are assigned when parents not only respond to the child's verbal cues, but also to non-verbal cues. Non-intrusiveness was coded on a 5-point rating scale with high scores for parents who are available for the child without being interfering, overprotective, or overwhelming. An intrusive parent controls the interaction and shows lack of respect for the child's autonomy. Non-hostility was also coded on a 5-point rating scale. Parents received high scores when no negative emotions (e.g. impatience, discontent, rolling the eyes, etc.) were shown.

The EAS also includes rating scales for child responsiveness and involvement. Responsiveness is rated as the intensity and the quality of the child's reactions to the parent's bids, while involvement reflects the child's attempts to engage the parent in the interaction. Child responsiveness and child involvement were coded on 7-point rating scales.

The play sessions were coded by three trained observers, who were unaware of the condition of the fathers (oxytocin or placebo). Two of them coded the fathers of typically developing children (mean intra-class correlation for the parenting scales $r = .89$, $n = 11$); and two of them (one overlapping) coded the fathers of children with autism (mean intra-class correlation $r = .78$, $n = 7$). We used one observer's ratings of all first visit play sessions and the other's ratings of all second visit play sessions.

Results

A repeated measures analysis of variance with the overall parenting scale as dependent variable, condition (oxytocin or placebo) as a within-subject factor, and child gender and group (typically developing children or children with ASD) as between-subject factors showed a significant effect for condition, $F(1, 29) = 4.18$, $p = .050$, $\eta^2 = .13$. Group and gender were no significant predictors (group, $F(1, 29) = 0.02$, $p = .879$, $\eta^2 < .01$; gender, $F(1, 29) = 0.06$, $p = .813$, $\eta^2 < .01$). The interaction effects of condition and group, $F(1, 29) < 0.01$, $p = .99$, $\eta^2 < .01$, and of condition and gender, $F(1, 29) = 0.20$, $p = .66$, $\eta^2 = .01$, were not significant, implying that the effect of oxytocin was similar in fathers of normally developing children and in fathers of children with ASD, and similar in fathers of boys and fathers of girls. Figure 1 shows the scores on the overall parenting scale in the placebo and oxytocin conditions for the total group, for fathers of children with ASD, and for fathers of typically developing children.

Figure 1 *Scores (M, SE) on the overall parenting scale in the placebo and oxytocin conditions for the total group, for fathers of children with ASD, and for fathers on typically developing children.*

A multivariate repeated measures analysis of variance on the four parenting scales with condition (oxytocin or placebo) as a within-subject factor showed an overall effect of oxytocin administration on parenting behavior $F(4, 28) = 2.74$, $p = .048$, $\eta^2 = .28$. Univariate analyses showed significant effects for *Structuring*, $F(1, 31) = 8.33$, $p = .007$, $\eta^2 = .21$, and for *Hostility*, $F(1, 31) = 6.13$, $p = .019$, $\eta^2 = .17$. Effects for *Sensitivity*, $F(1, 31) = 0.93$, $p = .34$, $\eta^2 = .03$, and *Intrusiveness*, $F(1, 31) = 1.94$, $p = .17$, $\eta^2 = .06$, were not significant. In the oxytocin condition fathers of normally developing and ASD children showed more structuring sensitivity and less hostility than in the placebo condition.

In a multivariate repeated measures analysis of variance on child responsiveness and child involvement with condition (oxytocin or placebo) as a within-subject factor, and child gender and group (typically developing children or children with ASD) as between-subject factors, we found a significant effect for group, $F(2, 27) = 4.75$, $p = .017$, $\eta^2 = .25$. Children with ASD were significantly less involved than typically developing children (oxytocin: ASD $M = 4.00$, $SD = 2.38$, typically developing $M = 5.78$, $SD = 0.97$; placebo: ASD $M = 3.77$, $SD = 2.22$, typically developing $M = 5.64$, $SD = 0.76$), and children with ASD showed lower levels of responsiveness (oxytocin: ASD $M = 4.18$, $SD = 2.11$, typically developing $M = 5.75$, $SD = 0.86$; placebo: ASD $M = 4.30$, $SD = 1.91$, typically developing $M = 5.69$, $SD = 0.66$). There were no significant effects for gender ($F(2, 27) = 0.02$, $p = .976$, $\eta^2 < .01$) or condition ($F(2, 27) = 0.62$, $p = .547$, $\eta^2 = .04$), nor were interaction effects between group and condition or between group and gender significant.

Discussion

This is the first experimental study showing that intranasal oxytocin administration enhances the quality of playful interactions of fathers with their child with an autism spectrum disorder. In the oxytocin condition, fathers of children with ASD stimulated their child in a more optimal way, and they showed less hostility. The effects of oxytocin administration were similar for both fathers of typically developing children and fathers of children with ASD. Children's involvement and responsiveness were not affected by oxytocin administered to their

parent. As expected children with ASD showed lower involvement and responsiveness compared to typically developing children. Taken together these findings suggest the positive effects of oxytocin on paternal sensitive play irrespective of clinical status of their child.

The role of fathers in parenting responsibilities has increased in the last few decades (Yeung et al., 2001). In particular, family sociologists report an increased participation in positive engagement activities for fathers (Pleck, 2010). However, father-child interaction seems different from mother-child interaction. Fathers are more likely to play when they interact with their child, whereas mothers spend more time in caregiving activities (Roggman et al., 2004), and this 'specialization' seems reflected in the oxytocin effects we found for paternal stimulatory play. Traditionally fathers have been described as focused on stimulating play, with less emphasis on emotional support and warmth (Grossmann et al., 2008). Mothers might relate to their infants with sensitive warmth, whereas fathers might choose sensitive stimulation as a way to promote feelings of security in their infants (Lucassen et al., 2012), and it is this playful stimulation that was enhanced by oxytocin administration.

Children with ASD render their parents' task of deciphering their signals more difficult because they may not express their emotions in explicit ways (Van IJzendoorn et al., 2007). This, together with the passivity of the child during interaction, may also confuse parents about their child's intellectual abilities and potentials (Bouma & Schweitzer, 1990; Koegel et al., 1992), which may trigger mothers of children with autism to use more physical contact, more high intensity behaviors and fewer social verbal approaches (Doussard-Roosevelt et al. 2003). Moreover, parents of children with autism more often use control strategies than parents of typically developing children (Kasari et al., 1988). When this happens in a sensitive way, attuned to the needs of the child, this may improve the developmental outcomes of the child. Our results, showing an improvement of fathers' sensitive structuring of play interactions after oxytocin administration, may thus have important implications for the developmental outcomes of the child.

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Several intervention studies have been conducted with children with autism, using a wide range of therapies (Rogers & Vismara, 2008; Ospina et al., 2008). The potential benefits of parent training are increased parenting skills, renewed confidence and reduced stress for parents as well as children (McConachie & Diggle, 2007). Evidence for the potentially important role of oxytocin in human parenting may be derived from experimental studies administering oxytocin to individuals with autism, which enhanced their social cognitions and empathic feelings (Bartz & Hollander, 2006; Bartz et al., 2011), and in studies relating autism or parental sensitivity to functional variations in the oxytocin receptor gene (Bakermans-Kranenburg & Van IJzendoorn, 2008; Jacob et al., 2007; Wu et al., 2005; Ylisaukko-oja et al., 2006). In non-clinical individuals intranasal oxytocin administration increased feelings of interpersonal trust (Heinrichs et al., 2003; Kosfeld et al., 2005), and improvement of "mind-reading" capacities (Domes et al., 2007). In the study of Hurlemann et al., (2010) it was demonstrated that oxytocin can facilitate socially reinforced learning as well as emotional empathy in men. Although our sample size was rather small we present here a first proof of principle that oxytocin levels positively influence the parenting skills of fathers of children with ASD. The effect size was substantial; note that the effect of oxytocin administration on parenting was of the order of the difference in child involvement and responsiveness between children with and without ASD. Although this finding does not necessarily imply any pharmacotherapeutic consequence (Van IJzendoorn & Bakermans-Kranenburg, 2012), we suggest that the use of oxytocin as a catalyst of interaction-focused parent training might be worthwhile to examine in more detail in the next future.

References

Bakermans-Kranenburg, M.J., & Van IJzendoorn, M.H. (2008). Oxytocin receptor (OXTR) and serotonin transporter (5-HTT) genes associated with observed parenting. *Social Cognitive and Affective Neuroscience,* 3, 128-134.

Bartz, J.A., & Hollander, E. (2006). The neuroscience of affiliation: Forging links between basic and clinical research on neuropeptides and social behavior. *Hormones and Behavior*, 50, 518–528.

Bartz, J. A, Zaki, J., Bolger, N., Hollander, E., Ludwig, N. N., Kolevzon, A., & Ochsner, K. N. (2010). Oxytocin selectively improves empathic accuracy. *Psychological Science,* 21, 1426-1428.

Bartz, J. A., Zaki, J., Bolger, N., & Ochsner, K. N. (2011). Social effects of oxytocin in humans: Context and person matter. *Trends in Cognitive Sciences,* 15, 301-9.

Biringen, Z., Robinson, J.L., & Emde, R.N. (2000). Appendix B: The emotional Availability Scales (3rd ed.; abridged infancy/ early childhood version). *Attachment and Human development,* 2, 256-270.

Bouma, R., & Schweitzer, R. (1990) The impact of chronic childhood illness on family stress: a comparison between autism and cystic fibrosis. *Journal of Clinical Psychology*, 46(6), 722-30.

Carter, C.S. (1998). Neuroendocrine perspectives on social attachment and love. *Psychoneuroendocrinology*, 23 (8), 779-818.

De Dreu, C.K.W., Greer, L.L., Handgraaf, M.J.J., Shalvi, S., Van Kleef, G.A., Baas, M., et al. (2010). The neuropeptide oxytocin regulates parochial altruism in intergroup conflict among humans. *Science,* 328, 1408–1411.

Domes, G., Heinrichs, M., Michel, A., Berger, C., & Herpertz, S.C. (2007). Oxytocin improves "mind-reading" in humans. *Biological Psychiatry*. 61(6), 731-3.

Doussard-Roosevelt, J. A., Joe, C. M., Bazhenova, O. V., & Porges, S. W. (2003). Mother-child interaction in autistic and nonautistic children: Characteristics of maternal approach behaviors and child social responses. *Development and Psychopathology,* 15, 277-295.

Feldman, R., Weller, A., Zagoory-Sharon, O., & Levine, A. (2007). Evidence for a neuroendocrinological foundation of human affiliation: plasma oxytocin levels across pregnancy and the postpartum period predict mother-infant bonding. *Psychological Science*. 18(11), 965-70.

6

Feldman, R., Gordon, I., Schneiderman, I., Weissman, O., & Zagoory-Sharon, O. (2010). Natural variations in maternal and paternal care are associated with systematic changes in oxytocin following parent-infant contact. *Psychoneuroendocrinology,* 35(8), 1133-1141.

Galbally, M., Lewis, A.J., Van IJzendoorn, M.H., & Permeel, M. (2011). The role of oxytocin in mother-infant relations, a systematic review on human studies. *Harv. Rev. Psychiatry*, 19 (1), 1-14.

Guastella, A.J., Einfeld, S.L., Gray, K.M., Rinehart, N.J., Tonge, B.J., Lambert, T.J. et al. (2010). Intranasal oxytocin improves emotion recognition for youth with autism spectrum disorders. *Biological Psychiatry,* 67, 692–694.

Grossmann, K. E., Grossmann, K., Kindler, H. & Zimmermann, P. (2008). A wider view of attachment and exploration: The influence of mothers and fathers on the development of psychological security from infancy to young adulthood. In J. Cassidy & P. Shaver (Eds.), Handbook of attachment theory and research. New York: Guilford.

Heinrichs, M., Baumgartner, T., Kirschbaum, C., & Ehlert, U. (2003). Social support and oxytocin interact to supress cortisol and subjective responses to psychosocial stress. *Biological psychiatry*, 54, 1389-1398.

Hollander, E., Bartz, J., Chaplin, W., Phillips, A., Sumner, J., Soorya, L., et al. (2007). Oxytocin increases retention of social cognition in autism. *Biological Psychiatry,* 61, 498–503.

Huffmeijer, R., Alink, L.R.A., Tops, M., Grewen, K.M., Light, K.C., Bakermans-Kranenburg, M.J. et al. (2012). Salivary levels of oxytocin remain elevated for more than two hours after intranasal oxytocin administration. *Neuroendocrinological Letters*, 33(1), 101-105.

Hurlemann, R., Patin, A., Onur, O.A., Cohen, M.X., Baumgartner, T., Metzler, S. et al. (2010). Oxytocin enhances Amygdala-dependent, socially reinforced learning and emotional empathy in humans. *Journal of Neuroscience*, 30(14), 4999-5007.

Insel, T.R. (1992). Oxytocin – a neuropeptide for affiliation: evidence from behavioral, receptor autoradiographic, and comparative studies. *Psychoneuroendocrinology*, 17, 3-35.

Kasari, C., Sigman, M., Mundy, P., & Yirmiya, N. (1988). Caregiver interactions with autistic children. *Journal of Abnormal Child Psychology,* 16, 45–56.

Kosfeld, M., Heinrichs, M., Zak, P.J., Fischbacher, U., & Fehr, E. (2005). Oxytocin increases trust in humans. *Nature*, 435, 673-676.

Koegel, L.K., Koegel, R.L., Hurley, C., & Frea,W.D. (1992). Improving social skills and disruptive behavior in children with autism through self-management. *Journal of Applied Behavior Analysis*, 25, 341-353.

Lord, C. (1995). Follow-up of two year olds referred for possible autism. *Journal of Child Psychology and Psychiatry*, 36, 1365-1382.

Lord, C., Rutter, M., DiLAvore, P.C., & Risis, S. (2002). *Autism Diagnostic Observation Schedule.* Los Angelos, CA: Western Psychology Services.

Jacob, S., Brune, C.W., Carter, C.S., Leventhal, B.L., Lord, C., & Cook, E.H. (2007). Association of the oxytocin receptor gene (OXTR) in Caucasian children and adolescents with autism. *Neuroscience Letters*, 417, 6–9.

Lucassen, N., Tharner, A., Van IJzendoorn, M.H., Bakermans-Kranenburg, M.J., Volling, B.L., Verhulst, F.C., et al. (2011). The association between paternal sensitivity and infant-father attachment security: A meta-analysis of three decades of research. *Journal of Family Psychology,* 25 (6), 986-992.

MacDonald, K., & MacDonald, T.M. (2010). The peptide that binds: a systematic review of oxytocin and its prosocial effects in humans. *Harv Rev Psychiatry,* 18(1), 1-21.

McConachie, H.M.A. & Diggle, T. (2007). Parent implemented early intervention for young children with autism spectrum disorder: a systematic review. *Journal of Evaluation in Clinical Practice*, 13, 120-129.

Naber, F.B.A.,Van IJzendoorn, M.H., Deschamps, P., Van Engeland, H., & Bakermans-Kranenburg, M.J. (2010). Intranasal oxytocin increases fathers' observed responsiveness during play with their children: a double-blind within-subject experiment. *Psychoneuroendocrinology,* 35, 1583–1586.

Ospina, M. B., Krebs Seida, J., Clark, B., Karkhaneh, M., Hartling, L., Tjosvold, L. et al. (2008). Behavioral and developmental interventions for autism spectrum disorder: a clinical systematic review. *PloS One, 3*(11).

Panksepp, J. (1998). *Affective neuroscience: The foundation of human and animal emotions.* New York: Oxford University Press.

Pleck, J.H. (2010). Paternal involvement: Revised conceptualizations and theoretical linkages with child outcome. In M. E. Lamb (Ed.), *The role of the father in child development* (5th ed., pp. 58-93). Hoboken, NJ: Wiley.

Riem, M.M.E., Bakermans-Kranenburg, M.J., Pieper, S., Tops, M., Boksem, M.A.S., Vermeiren, R.R.J.M., et al. (2011). Oxytocin modulates: amygdala, insula and inferior frontal gyrus responses to infant crying: a randomized control trial. *Biological Psychiatry,* 70, 291–297.

Rogers, S.J., & Vismara, L.A. (2008). Evidence-based comprehensive treatments for early autism. *Journal of Clinical Child and Adolescent Psychology,* 37(1), 8-38.

6

Roggman, L.A., Boyce, L.R., Cook, G.A. Christiansen, K., & Jones, D. (2004). Playing with daddy; social toy play, Early head start, and developmental outcomes. *Fathering*, 2(1), 83-108.

Van IJzendoorn, M.H., & Bakermans-Kranenburg, M.J. (2012). A sniff of trust: meta-analysis of the effects of intranasal oxytocin administration on face recognition, trust to in-group, and trust to out-group. *Psychoneuroendocrinolog,* 37(3), 438-443.

Van IJzendoorn, M.H., Rutgers, A.H., Bakermans-Kranenburg, M.J., Van Daalen, E., Dietz, C., Buitelaar, J., et al. (2007). Parental sensitivity and attachment in children with autism spectrum disorder: comparisons with children with mental retardation, with language delays, and with typical developing children. *Child Development,* 78(2*),* 597-608.

Wu, M., Jia, Y., Ruan, J., Liu, Y., Guo, M., Shuang, X., et al. (2005). Positive association of the oxytocin receptor gene (OXTR) with autism in the Chinese Han population, *Biological Psychiatry,* 58,74–77.

Yeung, W. J.**,** Sandberg, J.**,** Davis-Kean, P. E.**, &** Hofferth, S. L. (2001). Children's time with fathers in intact families**.** *Journal of Marriage and Family,* 63**,** 136–154**.**

Ylisaukko-oja, T., Alarcon, M., Cantor, R.M., Auranen, M., Vanhala, R., Kempas, E. et al.(2006). Search for autism loci by combined analysis of Autism Genetic Resource Exchange and Finnish families. *Annals of Neurology*, 1, 145-155.

Chapter 7

Summary and General Discussion

Summary

The present thesis focused on the competences of parents when rearing a child with ASD and the way nurses and other care providers can effectively empower parents of children with ASD. Coping-related parental competences were explored in a study of parental reactions to their child's ASD diagnosis (**chapter 2**). The results demonstrated that the majority of 77 parents came to terms with their child's ASD soon after receiving the diagnosis. The parents' reaction to the diagnosis was associated with the severity of the ASD characteristics and the parents' nationality. Parents predominantly reported an action-oriented coping style, which implies the adaptation of the parents' caregiving routines to the child's impairments, generally resulting in activating special services for their child. With the availability of a reliable diagnosis of ASD in children younger than three years of age (Van Daalen et al., 2009), nurses and other care providers are challenged to adapt intervention strategies to toddlers in order to meet the needs of parents of young children with ASD. The quality of the parent-child interaction is considered a critical foundation of children's developmental process (Ainsworth, 1978; Bowlby, 1969; Cassidy & Shaver, 2008) and has been the focus of many intervention programs (see Bakermans-Kranenburg et al., 2003). However, early interventions for families of children with ASD with the explicit aim of improving parent-child interactions are limited. Therefore, the greater part of this thesis concerns the development (**chapter 3**) and testing (**chapters 4 and 5**) of an interaction-focused intervention program: VIPP-AUTI (Video feedback Intervention to promote Positive Parenting adapted to Autism). The aim of the VIPP-AUTI program is to enhance parental sensitive responsiveness to the special needs of children with ASD. In a randomized control trial with a sample of 78 families we tested and demonstrated the effectiveness of the VIPP-AUTI program on parents' sensitive and adequate reactions to their child's signals as well as their increased confidence in their own competence to raise their child (**chapter 4**). Furthermore, VIPP-AUTI was found to be effective in enhancing children's initiating joint attention behavior (**chapter 5**). Our experimental within-subject study using intranasally administered oxytocin with fathers of children with and without ASD (**chapter**

6) demonstrated that oxytocin enhanced paternal sensitive play-interaction with their child, regardless the children's condition. The aim of this thesis is to contribute to the empowerment of parents in rearing their child with ASD from the perspective of attachment theory.

General discussion

Parental reaction to their child's ASD diagnosis

Our first aim was to explore parents' reactions to the ASD diagnosis of their child using the Reaction to Diagnosis Interview (RDI; Marvin & Pianta, 1996). Positive or negative reactions to diagnosis are classified as 'resolved' or 'unresolved' and the corresponding successful or unsuccessful coping strategies are distinguished. Previous studies with the RDI demonstrated associations between parental resolution and the quality of parent-child interactions in samples of children with developmental disabilities (Barnett et al., 2006; Marvin & Pianta, 1996), including ASD (Oppenheim et al., 2009; Wachtel & Carter, 2008). The RDI could function as a baseline assessment of parents' current state of resolution to diagnosis and their corresponding coping strategy. This information offers care providers the opportunity to tailor their interventions to the parent's individual competences and needs. For example, when parents report an action-oriented strategy, meaning that they are willing to do something for their child and to modify their caregiving routines, the care provider can focus on changing parenting routines. Also, when parents show unresolved resolution strategies, an intervention can focus on relieving the experienced burden. Furthermore, the easy administration of the RDI facilitates its use in clinical practice. Nevertheless, within the field of ASD-research and clinical practice, the use of the RDI is limited. To our knowledge, only three previous studies used the RDI in parents of children with an ASD-diagnosis (Milshtein et al., 2010; Oppenheim et al, 2009; Wachtel & Carter, 2008). Our study, together with those previous investigations, revealed strongly diverging proportions of resolved versus unresolved states of resolution, ranging from 33% to 75% of parents classified as resolved. The variety of the results from these studies can be explained by the heterogeneity of samples, study objectives, RDI-coding,

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outcome measurements, and time-intervals between diagnosis and RDI assessment. As an extension of previous studies using the RDI with parents of children with ASD, we explored the stability of the RDI classification and examined correlations between reaction to diagnosis and parent-child characteristics somewhat sooner after the ASD diagnosis of the child had been received (five weeks on average in contrast to more than six months in previous studies). Only six out of 35 parents changed for better (unresolved into resolved) or for worse (resolved into unresolved) in the main RDI classifications over a period of seven months. Changes from an unresolved to a resolved RDI classification were correlated with an increase of parental efficacy feelings towards child rearing, while the opposite change from a resolved to an unresolved classification indicated a decrease of parental efficacy. This finding is in line with a study by Kuhn and Carter (2006) demonstrating a relation between parental efficacy and the mothers' positive or negative feelings about their child's ASD. However, in most cases, parental resolution was stable over time, as in samples of children with other neurological disorders (Barnett et al., 2006; Rentinck et al., 2010). In more detail, we found fluctuations in parents' coping strategies, suggesting a dynamic process in the acceptance of their child's ASD diagnosis. This instability of coping strategy over time is consistent with findings in the recent nursing research literature on parents of children with complex and special needs (Rehm, 2013). Rehm reviewed 22 studies and found unbalanced emotional feelings in parents going from distress to rewards derived from child rearing. Furthermore, evidence-based nursing interventions to support these parents were found to be lacking (Rehm, 2013). Regarding the ASD population, it could be important to tailor interventions to the specific rewarding experiences arising from parents' early interactions of children with ASD, in order to alleviate some of the inevitable stress experiences. VIPP-AUTI could contribute to such support and help parents to adapt constructively to the ASD diagnosis of their child. VIPP-AUTI could also fill some of the gap in the nursing literature with respect to evidence-based nursing care provision.

Video feedback intervention adapted to ASD

The second aim of this thesis addressed the development of VIPP-AUTI. Generally, in clinical mental health practice, nurses are involved briefly after the diagnostic phase to provide psycho-education and professional support to patients. In accordance with a systematic approach to developing evidence-based nursing interventions (Van Meijel et al., 2004), we modified the short-term and attachment-based intervention program Video feedback Intervention to promote Positive Parenting (VIPP) (Juffer et al., 2008). First, we studied the nature of the problems and care needs of VIPP populations in relation to the target population of families with a child with ASD, and we analyzed the use of early interventions for children with ASD (current practice). Integrating these findings in VIPP (see chapter 3), we designed the manual for the VIPP-AUTI program. In addition, we carried out a brief pilot study of intervention feasibility involving four families of children with various degrees of ASD severity and cognitive functioning. On the basis of this pilot, VIPP-AUTI was considered feasible in clinical practice and the intervention was judged to be ready for a stringent test of effectiveness.

VIPP-AUTI includes five home visits in which film fragments of daily situations of individual parent-child interactions are discussed with the parent in order to enhance the parent's sensitive responsiveness to the (often atypical) signals of their child with ASD. The program comprises elements of self-management approaches (Barlow et al., 2002), e.g. delivery at home, focus on the specific family, the use of video feedback, discussing parental experiences, provision of (written) information, promoting reinterpretation of the child's (ASD) symptoms and cognitive reframing of parent-child interactions. Promoting self-care and involving family support within the natural environment of patients (or caregivers) are also prevailing policy priorities in the health care system of the Netherlands (Ministry of Health Welfare and Sport; VWS, Kamerbrief, 2013), in order to reduce the growth of society's health expenditures. Chronic childhood disabilities were indeed found to entail high economic costs, especially for children with mental health problems (Stabile & Allin, 2012) including childhood ASD (Järbrink, 2007). Enhancing the quality of

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early parent-child interactions by VIPP-AUTI could empower parents to manage the ASD symptoms of their child adequately. Understanding the child's behavior, and confidence in their own parental abilities in child rearing, could also limit the use of inappropriate therapy. For these reasons, VIPP-AUTI could contribute to early and cost-effective treatments.

Effectiveness of VIPP-AUTI on parent-child interaction

To our knowledge, VIPP-AUTI is the first standardized, interaction-focused intervention for parents and children with ASD, including evidence of effectiveness on aspects of parenting and child development. Our aims were to investigate whether VIPP-AUTI was effective on parental sensitive responsiveness to children with ASD and their feelings about child rearing as well as on children's joint attention and play behavior. We compared the effects of VIPP-AUTI with the usual nursing care at the university hospital. Data were collected at baseline (T1), after finishing the intervention (T2), and after a three month follow-up period without treatment (T3). Parent-child interaction was assessed at home and the parents completed questionnaires. Data analysis at T2 revealed the short-term outcomes of this trial on aspects of parental interaction style and reported feelings about child rearing (see chapter 4). The finding that parents became less intrusive after receiving VIPP-AUTI supported its effectiveness. The method of video feedback made parents aware of the way their child with ASD responded to their interactions. For example, the time the child needed to explore, to change to another situation, or to understand and subsequently answer a message, were presented and discussed using film fragments. This feedback appeared to change some of the parents' prior (educational) behaviors and made them less intrusive. Wan et al. (2012) found aspects of parental intrusive interaction patterns already present in the prediagnostic phase of infants at risk of ASD (siblings of a child with ASD) versus low-risk controls. The quality of early parent-infant interactions has been found to be associated with a later ASD-diagnosis of infants who were at risk (Wan et al. 2013). Therefore, interaction-focused intervening could be of utmost importance. Green et al. (2013) conducted a preventive,

interaction-focused intervention for infants at risk of developing ASD with elements of the VIPP program and demonstrated intervention feasibility. Consistent with the findings of Green et al. (2013), the parents involved in our study adjusted remarkably well to being filmed and they reported high levels of satisfaction with the treatment. Also, the 100% adherence to the VIPP-AUTI program (see chapter 5, figure 1) pointed to its applicability. The nurses who provided the intervention evaluated the use of the comprehensive manual as positive, and they considered the challenge of tailoring the video feedback to the individual family to be feasible.

At T3, we investigated children's joint attention and play behavior as observable core symptoms in young children with ASD. We found that the initiation of joint attention (IJA) was significantly greater in children whose parents received VIPP-AUTI than in the control group. This finding suggests that the child's IJA is especially susceptible to changes in parental behavior, such as improved parental sensitive responsiveness. A meta-analysis of Bakermans-Kranenburg et al. (2003) revealed empirical evidence that interventions targeted at enhancing parental sensitivity were also effective in improving the quality of parent-child interaction. After VIPP-AUTI was received, enhanced parental sensitive responsiveness could have activated the development of IJA skills by the increased quality of parent-child interactions. However, we could not test for mediation, because we found no association between parental non-intrusiveness at post-test and children's joint attention at follow-up. Other parental variables beyond our study focus could have mediated between the effects of VIPP-AUTI and children's IJA skills. For example, the VIPP-AUTI program promoted the accuracy of parents' perceptions of their child's signals by 'speaking for the child', that is verbalizing the child's facial expressions and non-verbal cues (see chapter 3). 'Speaking for the child' includes parents' attuned comments to their child's state of mind and refers to parental mind-mindedness (Meins et al., 2003). Meins et al. (2013) found mind-mindedness to be associated with the social-cognitive development of children. Furthermore, VIPP-AUTI targeted parents' adequate responses to children's signals as well as their affective attunement to the child's emotions, which could have enhanced

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parental insightfulness. Oppenheim and Koren-Karie (2002) found empirical support for the view that parents' insightfulness regarding the child's inner world and motives for its behavior is related to positive parenting and the quality of parent-child interaction. In future studies with VIPP-AUTI it would be interesting to examine various potential mediating, parent-directed factors for changing children's IJA, such as mind-mindedness and insightfulness.

Although we could not establish that a mediating effect of VIPP-AUTI explained the children's increased IJA skills, the VIPP-AUTI effects on children's IJA extends previous findings. Patten and Watson (2011) described the effectiveness of treatment on children's joint attention in a review, including four small randomized control trials (see Field et al., 1997; Gulsrud et al., 2007; Kasari et al., 2006; Yoder & Stone et al., 2006 in Patten & Watson, 2011). Most of the effective treatments were intensive (more than 16 weekly sessions) and child-directed, whereas the VIPP-AUTI is brief and interaction-focused.

In conclusion, VIPP-AUTI demonstrated effectiveness on important parental attitudes and behavior, and on child interactive skills, which promote the quality of parent-child interaction. Together with the feasibility of the program, our study offers support for implementation of the VIPP-AUTI program in clinical practice.

Intranasal oxytocin administration to fathers of children with ASD
A final aim was to investigate the influence of oxytocin, a neuropeptide and attachment-related hormone, on parent-child interactions (see chapter 6). In previous studies, oxytocin was found to influence emotional empathy (by enhanced emotional face recognition) and trust in close relationships (see meta-analysis by Van IJzendoorn and Bakermans-Kranenburg, 2012). These findings point to the potential use of oxytocin as a pharmacological adjunct to treatment, especially for individuals with ASD (Bakermans-Kranenburg & Van IJzendoorn, 2013). There is an emerging research focus on the influence of oxytocin in the ASD population (Green & Hollander et al., 2010). For example, oxytocin administration was found to enhance emotional face recognition in the presence of ASD in youth (Guastella et al., 2010) and adults (Domes et al., 2013). For young children with ASD, the

focus of oxytocin effects could be directed at parent-child interactions. Social and emotional development is grounded in the attachment relationship between an infant and its primary caregiver (Bowlby, 1969; Cassidy & Shaver, 2008; Feldman, 2012). This relationship is formed not only by a close interpersonal bond, but also through an active interaction process of synchrony between parent and child (Feldman, 2007). Synchrony refers to the coordination of parental behavior to the child's signals and reciprocal infant responses. Oxytocin was found to mediate stress and reward-related neurological mechanisms and synchronous parenting (Atzil et al., 2011). Moreover, Feldman (2012) described parallel responses in one attachment partner to physiological changes in the other. Thus, if the parent is influenced, the child's behavior, including underlying physiological processes, can change through mutual interaction. In other words, the level of oxytocin can be influenced in young children via the parent, and this could be very important for intervention purposes. In our study of children with and without ASD, we found that a father's sensitive responsiveness was enhanced during playful interaction with their child after oxytocin administration. This finding was consistent with previous studies that demonstrated similar results with fathers of typically-developing children (Naber et al., 2010) and elevated oxytocin levels in fathers during play-and-contact interaction with infants (Feldman et al., 2010). In our study, all fathers (of children with and without ASD) demonstrated improvements in their stimulation skills directed at exploration activities of the child after oxytocin administration, though children with ASD responded less to their fathers than children without ASD, and in the short run their responsiveness did not change because of the oxytocin administration in their fathers. This finding suggests that increased paternal sensitivity through oxytocin administration could advance the effectiveness of treatment to promote adequate parent-child interactions. It will be particularly interesting in future research to examine whether oxytocin administration when combined with the VIPP-AUTI program enhances the effectiveness of treatment. The next step in research should focus on the parallel physiological response of children with ASD. The question could be whether oxytocin administration to parents simultaneously elevates the level of oxytocin

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in the child with ASD during interaction. Such an effect could enhance child responsiveness if not immediately after the treatment then maybe in the long run.

Limitations and implications for future research

The effectiveness of VIPP-AUTI was proven in a single blind randomized control trial and compared with an intervention of equal intensity. However, some limitations should be noted when the findings of chapters 2, 4 and 5 are interpreted.

First, the monocenter study design reduced generalizability. VIPP-AUTI was compared with the usual care provision at the University Medical Center Utrecht. The usual care programs in other care settings could differ in content and intensity. Therefore, a multicenter approach is needed to compare the effects of VIPP-AUTI with various mental health services.

Second, most of the parents in our sample had middle to high SES and most were married couples with Dutch nationality. This could have caused some bias to our results. Other studies have demonstrated that families in more disadvantaged circumstances, such as financial problems or divorce, benefited less from treatment (Karst & Van Hecke, 2012). However, the findings of previous studies of VIPP programs in other than ASD populations have demonstrated the effectiveness of intervention in a demographically broader range of participants, including families with multiple problems (Juffer et al., 2009; Moss et al., 2011). VIPP-AUTI could be beneficial for parents regardless their sociodemographics. Future research should include parents with lower levels of SES and various cultural backgrounds in order to answer the hypothesis that the effectiveness of VIPP-AUTI is generalizable. Another selection bias could have been the recruitment of parents to participate in an intervention study, resulting in a sample of highly active and motivated parents. Especially in the study of parental reaction to children's ASD-diagnosis, this could explain the high number of parents with active oriented coping strategies, in contrast to the results of previous studies.

Other limitations concern the measurements we applied. To assess parents' internal states (e.g. self-efficacy, daily hassles, broad

autism phenotype, mental health) we used self-report instruments. These reports could have been biased by socially desirable answers and by expectations about treatment effects, especially during post-intervention and follow-up assessments, although the high intensity of the care-as-usual program might have mitigated differences between intervention and control group.

Finally, the relatively brief period of follow-up assessments (at T2 and T3) could have captured only a few of the complex and dynamic factors related to the impact of rearing a child with ASD and parent-child interactions. Future longitudinal studies are needed to examine parental adaptation processes to their child's ASD diagnosis and the long-term effects after receiving VIPP-AUTI.

Implementation of VIPP-AUTI in clinical practice

VIPP-AUTI was provided after the diagnostic phase at the university hospital. When parents received an ASD diagnosis of their child, they were often confronted with waiting lists for additional services such as specialized day care or comprehensive therapy programs. Other parents were at risk of shopping around because they strived for all the interventions that were available in the short term. We therefore recommend evidence-based intervention at the university hospital during the critical period after an ASD-diagnosis, using VIPP-AUTI.

VIPP-AUTI turned out to be feasibly implementable in clinical practice because the training of professionals is relatively brief, the manual is detailed and the programming is short-term. The VIPP-AUTI program can be implemented in settings other than the university hospital with respect to protocol fidelity. Of course, resources have to be taken into account: for example, budgets for home visits, (video) materials, logistic arrangements and competent staff members who are facilitated to receive adequate training, support and supervision, but most of these are also needed for the care as usual that these parents receive, and amount to less than the more intensive, long-term interventions offered to children with ASD. Professionals in various disciplines are in a position to act as intervener when they are experienced in caring for children with ASD, supporting the potential of VIPP-AUTI in clinical practice and health care for children with ASD.

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References

Ainsworth, M. D. S. (1978). Bowlby-Ainsworth Attachment Theory. *Behavioral and Brain Sciences, 1*(3), 436-438.

Atzil, S., Hendler, T., & Feldman, R. (2011). Specifying the neurobiological basis of human attachment: brain, hormones, and behavior in synchronous and intrusive mothers. *Neuropsychopharmacology : Official Publication of the American College of Neuropsychopharmacology, 36*(13), 2603-2615.

Bakermans-Kranenburg, M. J., Van IJzendoorn, M. H., & Juffer, F. (2003). Less is more: meta-analyses of sensitivity and attachment interventions in early childhood. *Psychological Bulletin, 129*(2), 195-215.

Bakermans-Kranenburg, M. J., & Van IJzendoorn, M. H. (2013). Sniffing around oxytocin: review and meta-analyses of trials in healthy and clinical groups with implications for pharmacotherapy. *Translational Psychiatry, 3*, e258.

Barlow, J., Wright, C., Sheasby, J., Turner, A., & Hainsworth, J. (2002). Self-management approaches for people with chronic conditions: a review. *Patient Education and Counseling, 48*(2), 177-187.

Barnett,, D., Clements, M., Kaplan-Estrin, M., McCaskill, J. W., Hunt, K. H., Butler, C. M. et al. (2006). Maternal resolution of child diagnosis: stability and relations with child attachment across the toddler to preschooler transition. *Journal of Family Psychology: Journal of the Division of Family Psychology of the American Psychological Association (Division 43), 20*(1), 100-107.

Bowlby J, (1969). *Attachment and Loss: vol 1. Attachment.* New York: Basic Books

Cassidy J., & Shaver Ph. R. (2008). *Handbook of Attachment: theory, research, and clinical applications*.2nd ed. New York: The Guilford Press.

Domes, G., Heinrichs, M., Kumbier, E., Grossmann, A., Hauenstein, K., & Herpertz, S. C. (2013). Effects of Intranasal Oxytocin on the Neural Basis of Face Processing in Autism Spectrum Disorder. *Biological Psychiatry,* doi:10.1016/j.biopsych.2013.02.007; 10.1016/j.biopsych.2013.02.007

Feldman, R. (2007). Parent-infant synchrony and the construction of shared timing; physiological precursors, developmental outcomes, and risk conditions. *Journal of Child Psychology and Psychiatry, and Allied Disciplines, 48*(3-4), 329-354.

Feldman, R. (2012). Oxytocin and social affiliation in humans. *Hormones and Behavior, 61*(3), 380-391.

Feldman, R., Gordon, I., Schneiderman, I., Weisman, O., & Zagoory-Sharon, O. (2010). Natural variations in maternal and paternal care are associated with systematic changes in oxytocin following parent-infant contact. *Psychoneuroendocrinology, 35*(8), 1133-1141.

Green, J. J., & Hollander, E. (2010). Autism and oxytocin: New developments in translational approaches to therapeutics. *Neurotherapeutics, 7*(3), 250-257.

Green, J., Wan, M., Guiraud, J., Holsgrove, S., McNally, J., Slonims, , et al. (2013). Intervention for infants at risk of developing autism: A case series. *Journal of Autism and Developmental Disorders,* doi:10.1007/s10803-013-1797-8

Guastella, A. J., Einfeld, S. L., Gray, K. M., Rinehart, N. J., Tonge, B. J., Lambert, T. J. et al. (2010). Intranasal Oxytocin Improves Emotion Recognition for Youth with Autism Spectrum Disorders. *Biological Psychiatry, 67*(7), 692-694.

Jarbrink, K. (2007). The economic consequences of autistic spectrum disorder among children in a Swedish municipality. *Autism : The International Journal of Research and Practice, 11*(5), 453-463.

Juffer, F., Bakermans-Kranenburg, M. J., & Van IJzendoorn, M. H., ed. (2008). *Promoting Positive Parenting. An Attachment-Based Intervention.* New York/ London: Lawrence Erlbaum Associates/ Taylor & Francis Group

Juffer, F., Bakermans-Kranenburg, M. J., & Van IJzendoorn, M. H. (2009). Attachment-Based Interventions: Heading for Evidence-Based Ways to Support Families. *ACAMH Occasional Papers,* 29, 47-57.

Karst, J. S., & Van Hecke, A. V. (2012). Parent and family impact of autism spectrum disorders: A review and proposed model for intervention evaluation. *Clinical Child and Family Psychology Review, 15*(3), 247-277.

Kuhn, J. C., & Carter, A. S. (2006). Maternal self-efficacy and associated parenting cognitions among mothers of children with autism. *The American Journal of Orthopsychiatry, 76*(4), 564-575.

Marvin, R. S., & Pianta, R. C. (1996). Mothers' reactions to their child's diagnosis: Relations with security of attachment. *Journal of Clinical Child Psychology, 25*(4), 436-445.

Meins, E., Fernyhough, C., Wainwright, R., Clark-Carter, D., Das Gupta, M., Fradley, E., et al. (2003). Pathways to understanding mind: construct validity and predictive validity of maternal mind-mindedness. *Child Development, 74*(4), 1194-1211.

7

Meins, E., Fernyhough, C., Arnott, B., Leekam, S. R., & de Rosnay, M. (2013). Mind-Mindedness and Theory of Mind: Mediating Roles of Language and Perspectival Symbolic Play. *Child Development,* doi:10.1111/cdev.12061; 10.1111/cdev.12061

Milshtein, S., Yirmiya, N., Oppenheim, D., Koren-Karie, N., & Levi, S. (2010). Resolution of the diagnosis among parents of children with autism spectrum disorder: associations with child and parent characteristics. *Journal of Autism and Developmental Disorders, 40*(1), 89-99.

Ministry of Health Welfare and Sport; VWS, Kamerbrief 2013. Retrieved from http://www.rijksoverheid.nl/ministeries/vws/documenten-en-publicaties/kamerstukken/2013/02/08/kamerbrief-gezamenlijke-agenda-vws-van-systemen-naar-mensen.html (accessed June 2013)

Moss, E., Dubois-Comtois, K., Cyr, C., Tarabulsy, G. M., St-Laurent, D., & Bernier, A. (2011). Efficacy of a home-visiting intervention aimed at improving maternal sensitivity, child attachment, and behavioral outcomes for maltreated children: a randomized control trial. *Development and Psychopathology, 23*(1), 195-210.

Naber, F., Van IJzendoorn, M. H., Deschamps, P., Van Engeland, H., & Bakermans-Kranenburg, M. J. (2010). Intranasal oxytocin increases fathers' observed responsiveness during play with their children: A double-blind within-subject experiment. *Psychoneuroendocrinology,35*, 1583-1586.

Oppenheim, D., & Koren-Karie, N. (2002). Mothers' insightfulness regarding their children's internal worlds: The capacity underlying secure child-mother relationships. *Infant Mental Health Journal, 23*(6), 593-605.

Oppenheim, D., Koren-Karie, N., Dolev, S., & Yirmiya, N. (2009). Maternal insightfulness and resolution of the diagnosis are associated with secure attachment in preschoolers with autism spectrum disorders. *Child Development, 80*(2), 519-527.

Patten, E., & Watson, L. R. (2011). Interventions Targeting Attention in Young Children With Autism. *American Journal of Speech-Language Pathology, 20*(1), 60-69.

Rehm, R. (2013). Nursing's contribution to research about parenting children with complex chronic conditions: An integrative review, 2002 to 2012. *Nursing Outlook,* doi:10.1016/j.outlook.2013.03.008

Rentinck, I. C., Ketelaar, M., Schuengel, C., Stolk, J., Lindeman, E., Jongmans, M.J., et al. (2010). Short-term changes in parents' resolution regarding their young child's diagnosis of cerebral palsy. *Child: Care, Health and Development, 36*(5), 703-708.

Stabile, M., & Allin, S. (2012). The economic costs of childhood disability. *The Future of Children / Center for the Future of Children, the David and Lucile Packard Foundation, 22*(1), 65-96.

Van Daalen, E., Kemner, C., Dietz, C., Swinkels, S. H., Buitelaar, J. K., & Van Engeland, H. (2009). Inter-rater reliability and stability of diagnoses of autism spectrum disorder in children identified through screening at a very young age. *European Child & Adolescent Psychiatry, 18*(11), 663-674.

Van Meijel, B., Gamel, C., Van Swieten-Duijfjes, B., & Grypdonck, M. H. (2004). The development of evidence-based nursing interventions: methodological considerations. *Journal of Advanced Nursing, 48*(1), 84-92.

Van IJzendoorn, M. H., & Bakermans-Kranenburg, M. J. (2012). A sniff of trust: meta-analysis of the effects of intranasal oxytocin administration on face recognition, trust to in-group, and trust to out-group. *Psychoneuroendocrinology, 37*(3), 438-443.

Wachtel, K, & Carter, AS (2008). Reaction to diagnosis and parenting styles among mothers of young children with ASDs. *Autism : The International Journal of Research and Practice, 12*(5), 575-594.

Wan, M., Green, J., Elsabbagh, M., Johnson, M., Charman,T., & Plummer, F. (2012). Parent-infant interaction in infant siblings at risk of autism. *Research in Developmental Disabilities, 33*(3), 924-932.

Wan, M., Green, J., Elsabbagh, M., Johnson, M., Charman,T., & Plummer, F. (2013). Quality of interaction between at-risk infants and caregiver at 12-15 months is associated with 3-year autism outcome. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 54*(7), 763-771.

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Chapter 8

Nederlandse samenvatting

Dankwoord

Curriculum Vitae

Publications

Inleiding

Autisme of een Autisme Spectrum Stoornis (ASS) is een aandoening van de hersenen, waarbij de verwerking van informatie uit de omgeving atypisch verloopt. ASS ontstaat voor het derde levensjaar door een combinatie van erfelijke en omgevingsfactoren. Er is een grote variatie in de mate en ernst van symptomen. Algemene kenmerken zijn beperkingen op het gebied van sociale relaties en (non)verbale communicatie, in combinatie met de aanwezigheid van stereotiepe en rigide gedragspatronen. Uit epidemiologische studies blijkt dat ASS bij ongeveer één op de 150 kinderen voorkomt (Fombonne 2009). Door eerdere en betere diagnostiek wordt een diagnose ASS tegenwoordig al op jonge kinderleeftijd gesteld. Behandeling van deze chronisch verlopende aandoening richt zich op de drie volgende aspecten: (1) stimulatie van de ontwikkeling, (2) preventie van secundaire (gedrags) problemen en (3) ondersteuning van het gezin. Vooral de sociale beperkingen van een kind met ASS kunnen voor ouders een extra zorg en opvoedingsbelasting betekenen. Veel van deze ouders doen dan ook een beroep op hulpverlening binnen (kinderpsychiatrische) zorgvoorzieningen. Dit proefschrift richt zich op ouderlijke competenties voor de opvoeding van een kind met ASS en de wijze waarop verpleegkundigen en andere zorgverleners deze competenties effectief kunnen versterken.

Belangrijkste bevindingen

In een exploratie studie naar de reactie van ouders op de ASS-diagnose van hun kind beschrijven we competenties van ouders in relatie tot hun copingstijl (**hoofdstuk 2**). De bevindingen laten zien dat de meerderheid van 77 ouders de ASS-diagnose van het kind positief aanvaardde. De reactie van ouders op de diagnose was afhankelijk van de ernst van de ASS-kenmerken en de nationaliteit van ouders; ouders hadden meer moeite met de diagnose van het kind bij een ernstiger vorm van ASS of als ouders een andere dan de Nederlandse nationaliteit bezaten. De ouders rapporteerden hoofdzakelijk een actie-georiënteerde copingstijl, wat betekent dat ouders opvoedingsgewoonten aanpassen aan de beperkingen van het kind en meestal (gespecialiseerde) hulpverlening inschakelen. Omdat

tegenwoordig een ASS-diagnose bij kinderen jonger dan het vijfde levensjaar gesteld wordt, is het belangrijk voor verpleegkundigen en andere hulpverleners om interventiestrategieën aan te passen aan de peuter- en kleuterleeftijd, zodat deze aansluiten bij de hulpvraag van ouders met jonge kinderen. Op grond van de gehechtheidstheorie en hierop gebaseerd onderzoek (Cassidy & Shaver, 2008) is bekend dat de kwaliteit van ouder-kind interacties een kritieke rol speelt in het ontwikkelingsproces van een kind. In het algemeen richten veel interventieprogramma's zich dan ook op de ouder-kind interactie. Voor gezinnen van kinderen met ASS zijn echter weinig vroege interventies beschikbaar met het expliciete doel om ouder-kind interacties te verbeteren. Om die reden is het grootste gedeelte van dit proefschrift gericht op de ontwikkeling (**hoofdstuk 3**) en het toetsen (**hoofdstuk 4 en 5**) van een interactie-gericht interventieprogramma: VIPP-AUTI (*Video feedback Intervention to promote Positive Parenting adapted to Autism*). Doel van het VIPP-AUTI programma is om de sensitiviteit van ouders voor de behoeften van kinderen met ASS te vergroten. In een gerandomiseerde, gecontroleerde studie met 78 gezinnen onderzochten en vonden we effectiviteit van het VIPP-AUTI programma met betrekking tot sensitieve en adequate reacties van ouders op de signalen van het kind. Ook vonden we een toename van het zelfvertrouwen van ouders in hun competenties om hun kind op te voeden (**hoofdstuk 4**). In **hoofdstuk 5** bleek vervolgens dat na VIPP-AUTI het initiatief vanuit het kind om aandacht te delen (een vorm van communicatie waarbij twee personen op hetzelfde reageren) meer verbeterd was dan in de controlegroep. Tot slot beschrijven we in **hoofdstuk 6** een experimentele, placebo-gecontroleerde studie met 32 vaders van kinderen met en zonder ASS. Ongeacht de conditie van het kind, vonden we na toediening van oxytocine een verbetering van sensitiviteit van de vaders voor de signalen van het kind tijdens spel.

Reactie van ouders op de ASS-diagnose van hun kind

Ten eerste onderzochten we de reactie van ouders op de ASS-diagnose van hun kind met het 'Reactie op de Diagnose Interview' (RDI; Marvin & Pianta, 1996)). Dit instrument classificeert een positieve of een negatieve reactie van ouders op de diagnose van hun kind,

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die onderverdeeld wordt in respectievelijk adequate of inadequate copingstrategieën. Deze informatie is van belang voor hulpverleners om interventies aan te kunnen passen aan de individuele competenties en behoeften van ouders. Als ouders bijvoorbeeld een actie-georiënteerde copingstijl hebben, wat inhoudt dat ouders iets willen doen voor hun kind en hun opvoedingsgewoonten willen aanpassen, kan de hulpverlener zich richten op verandering van patronen. Als ouders echter moeite hebben met de diagnose kan een interventie zich richten op vermindering van de ervaren belasting. Daarnaast bleek het RDI, door de eenvoudige afname, een bruikbaar instrument in de praktijk. Toch wordt in de praktijk en op het gebied van ASS-onderzoek nog weinig gebruik gemaakt van het RDI. In onze studie namen we het interview af bij de ouders kort na ontvangst van de ASS-diagnose en herhaalden we het interview na zeven maanden om de stabiliteit van reacties over tijd te meten. Slechts zes van 35 ouders veranderden van RDI-hoofdclassificatie van een negatieve naar een positieve reactie of omgekeerd van een positieve naar een negatieve reactie op de diagnose. We vonden een samenhang van deze veranderingen met respectievelijk toegenomen of afgenomen zelfvertrouwen van ouders in de opvoeding. Bij de meerderheid van de ouders vonden we echter, net als in voorgaande studies met steekproeven van kinderen met andere neurologische beperkingen (Barnett et al., 2006; Rentinck et al., 2010), stabiele reacties op de ASS-diagnose van het kind. Meer gedetailleerd, lieten de onderliggende copingstijlen (RDI-subclassificaties) van ouders wel wisselingen zien. Deze veranderingen van copingstijl suggereerden een dynamisch proces van ouders in het omgaan met de ASS-diagnose van hun kind. In verplegingswetenschappelijke literatuur worden eveneens instabiele copingstijlen gevonden bij ouders van kinderen met complexe, chronische ziekten (Rehm, 2013). Daarbij schommelen emoties van ouders tussen gevoelens van stress en beloning tijdens het opvoeden. Evidence-based verpleegkundige interventies om deze ouders te begeleiden blijken echter schaars. Met het oog op de ASS-populatie kan het belangrijk zijn om hulpverlening op maat te bieden met extra aandacht voor de belonende interactiepatronen tussen ouder en kind ter compensatie van onvermijdelijke stresservaringen. VIPP-AUTI kan

als interactie-gerichte interventie bijdragen aan dergelijke hulp voor
ouders van jonge kinderen met ASS en het aanbod van evidence-based
verpleegkundige interventies aanvullen.

Aanpassing van de video feedback interventie aan ASS

Het tweede doel van het onderzoek beschreven in dit proefschrift
betreft de ontwikkeling van VIPP-AUTI. Na de diagnostische fase in
de kinder- en jeugdpsychiatrie bieden verpleegkundigen vaak een
beperkte periode psycho-educatie en professionele hulpverlening aan
(familie van) patiënten. Volgens een systematische werkwijze voor
de ontwikkeling van verpleegkundige interventies hebben we het
basisprogramma van de kortdurende en gehechtheidsgerelateerde
interventie 'Video feedback Intervention to promote Positive Parenting
(VIPP)' aangevuld. VIPP is een evidence-based programma gericht op
het bevorderen van de kwaliteit van ouder-kind interacties (Juffer
et al., 2008). Ten eerste vergeleken we de aard van problemen en
zorgbehoeften van VIPP-populaties met die van gezinnen met
kinderen met ASS en we onderzochten het huidig gebruik van vroege
interventies voor kinderen met ASS (de huidige praktijk). Met de
integratie van deze bevindingen in VIPP, ontwierpen we de handleiding
voor het VIPP-AUTI-programma (zie hoofdstuk 3). Vervolgens
onderzochten we in een korte pilotfase de praktische uitvoerbaarheid
van het programma bij vier gezinnen met kinderen met uiteenlopende
gradaties van ASS en cognitief functioneren. Op basis van deze
pilotstudie vonden we VIPP-AUTI geschikt om in de klinische praktijk
uit te voeren en te toetsen op effectiviteit.

VIPP-AUTI bestaat uit vijf huisbezoeken waarin filmfragmenten
van ouder-kind interacties in dagelijkse situaties met de ouders
besproken worden om ouderlijke sensitiviteit voor de (vaak ongewone)
signalen van het kind met ASS te vergroten. Het programma bevat
elementen van zelfmanagement benaderingen, zoals implementatie
bij het gezin aan huis, gebruik van video feedback, geven van
(schriftelijke) informatie, bevorderen van begrip voor de (ASS)
symptomen van het kind en verandering van denkpatronen tijdens
ouder-kind interacties. Het bevorderen van zelfredzaamheid, zo veel
mogelijk thuis en gericht op steun van het eigen sociale netwerk zijn

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huidige speerpunten in het politieke beleid van langdurige (jeugd)zorg om kosten in de gezondheidszorg beheersbaar te maken. VIPP-AUTI kan van meerwaarde zijn in het aanbod van vroege en kosteneffectieve behandelingen. VIPP-AUTI, kan ouders ondersteunen in een adequate omgang met hun kind met ASS en daarmee de kwaliteit van ouder-kind interacties in een vroeg stadium bevorderen. Door begrip van ouders voor het gedrag van het kind en zelfvertrouwen in eigen opvoedingsvaardigheden is het ook mogelijk om het gebruik van onnodige en/of ongeschikte behandelingen te voorkomen.

Effectiviteit van VIPP-AUTI op ouder-kind interacties
Voor zover wij weten is VIPP-AUTI de eerste geprotocolleerde, interactie-gerichte interventie voor ouders en kinderen met ASS met aantoonbare effectiviteit op aspecten van ouder- en kinduitkomsten. Onze doelstellingen waren om te onderzoeken of VIPP-AUTI effectief was om de sensitiviteit van ouders voor de signalen van kinderen met ASS en hun gevoelens van competentie in de opvoeding te verhogen. Bij het kind keken we naar het spelgedrag en de vaardigheden om aandacht te delen. Wij vergeleken VIPP-AUTI met de gebruikelijke vrij intensieve verpleegkundige zorg die vanuit het academisch ziekenhuis gegeven wordt. De gegevens werden voor de interventie (T1) en na de interventie (T2) verzameld en ook drie maanden later na een periode zonder interventie (T3). De ouder-kind interactie werd thuis geobserveerd en de ouders vulden vragenlijsten in. De bevinding dat ouders minder intrusief (een vorm van overstimulatie) werden na behandeling met VIPP-AUTI in vergelijking met de controlegroep, is veelbelovend voor de effectiviteit van de interventie. De methode van video feedback maakte ouders bewust van de manier waarop hun kind met ASS in interactie reageerde. Met de filmfragmenten toonden en bespraken we bijvoorbeeld hoeveel tijd het kind nodig had om nieuwe dingen te ontdekken, zich aan te passen aan een verandering van situatie of om een boodschap te begrijpen en vervolgens te beantwoorden. Deze feedback leek bepaalde gebruikelijke (onderwijzende) gedragingen van ouders te veranderen in minder intrusief gedrag. Daarnaast raakten de deelnemende ouders in onze studie opmerkelijk snel gewend aan de ervaring om gefilmd te

worden en zij rapporteerden een hoge mate van tevredenheid met de behandeling. De doelmatigheid van VIPP-AUTI werd ook geïllustreerd doordat alle ouders de behandeling voltooiden. De verpleegkundigen die de interventie uitvoerden vonden de uitgebreide handleiding en het 'op maat' aanpassen van de video feedback aan het individuele gezin werkzaam.

Bij T3 onderzochten we gedeelde aandacht en spelgedrag van de kinderen als zichtbare kernsymptomen van ASS op jonge leeftijd. We vonden het initiatief om aandacht te delen (IJA) sterker toegenomen bij kinderen van ouders die VIPP-AUTI kregen dan in de controlegroep. Deze bevinding suggereert een speciale gevoeligheid van IJA bij kinderen voor gedragsveranderingen van de ouder, waaronder een verbetering van ouderlijke sensitiviteit. Wij vonden echter geen verband tussen de mate van ouderlijke intrusiviteit na de interventie en de vaardigheid om aandacht te delen van de kinderen drie maanden later, zodat mediatie van het effect via verminderde intrusiviteit niet aan de orde was. Het is mogelijk dat andere variabelen, die wij buiten beschouwing lieten, medieerden tussen het effect van VIPP-AUTI op IJA-vaardigheden van het kind. VIPP-AUTI bevat bijvoorbeeld de instructie voor de hulpverlener en ouders om te 'spreken voor het kind', dat betekent het verwoorden van gezichts- en non-verbale uitdrukkingen van het kind (zie hoofdstuk 3). 'Spreken voor het kind' houdt in dat ouders zich kunnen inleven in de gedachtewereld van hun kind, wat ook wel 'mind-mindedness' wordt genoemd (Meins et al., 2003). Ook richtte VIPP-AUTI zich op het inzicht van ouders in de beweegredenen van het kind voor zijn/haar gedrag, 'insightfulness' geheten. Voor toekomstig onderzoek kan het interessant zijn om diverse mogelijke mediatiefactoren, zoals mind-mindedness (Meins et al., 2013) en insightfulness (Oppenheim & Koren-Karie, 2002), te onderzoeken die IJA van kinderen kunnen beïnvloeden.

Hoewel wij geen mediatie-effect via veranderingen in ouderlijk gedrag konden aantonen als verklaring voor de verbetering van IJA-vaardigheden bij de kinderen, komt het therapeutisch effect van VIPP-AUTI op IJA overeen met resultaten van andere interventies die IJA bij kinderen met ASS verbeteren (Patten & Watson, 2011). De meeste effectieve behandelingen waren echter kindgericht en intensief (meer

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dan 16 uur wekelijkse sessies), terwijl VIPP-AUTI interactie-gericht en kortdurend is.

Concluderend vonden wij effectiviteit van VIPP-AUTI op enkele belangrijke aspecten van ouderlijk gedrag en interactieve vaardigheden van het kind, die de kwaliteit van ouder-kind interacties bevorderen. Samen met de werkzaamheid van het programma biedt onze studie perspectief om het VIPP-AUTI programma te implementeren in de klinische praktijk.

Oxytocine voor vaders van kinderen met ASS

Tot slot onderzochten we de invloed van oxytocine, een neuropeptide en gehechtheidsgerelateerd hormoon, op vader-kind interacties. Uit eerdere studies is gebleken dat oxytocine het emotionele inlevingsvermogen (door toegenomen herkenning van gezichtsuitdrukkingen) en het vertrouwen in anderen beïnvloedt (Van IJzendoorn & Bakermans-Kranenburg, 2012). Oxytocine kan daarom wellicht relevant zijn als aanvullende farmaceutische behandeling voor mensen met ASS. Voor jonge kinderen met ASS kan onderzoek naar de potentiële werkzaamheid van oxytocine zich ook richten op ouder-kind interacties. Sociale en emotionele ontwikkeling is gebaseerd op de gehechtheidsrelatie tussen baby's en hun primaire verzorger. Deze relatie wordt niet alleen gevormd door een vertrouwde persoonlijke nabijheid, maar ook door een actief interactieproces van synchronie tussen ouder en kind (Feldman, 2007). Synchronie is de overeenkomst van het gedrag van ouders met de signalen van het kind en de gelijktijdige reactie van het kind daarop (afstemming van gedrag). Eerdere studies lieten een mediatie-effect zien van oxytocine op het verband tussen de mate van synchronie van ouders en neurologische stress- en beloningsmechanismen (Atzil et al., 2011). Het bleek zelfs dat binnen een gehechtheidsrelatie verandering in fysiologische processen in de ene persoon eenzelfde verandering teweeg bracht in de ander (Feldman, 2012). Dit kan betekenen dat door ouders te beïnvloeden ook het gedrag van het kind en onderliggende fysiologische processen kunnen veranderen tijdens wederzijdse interactie. Met andere woorden, het oxytocinegehalte in jonge kinderen kan mogelijk beïnvloed worden via de ouder, wat relevant kan zijn

voor behandeldoelen. In onze studie vonden we na toediening van oxytocine een verhoogde sensitiviteit tijdens spel-interactie bij vaders van kinderen met en zonder ASS. Alle vaders gingen na toediening van oxytocine het kind meer stimuleren om nieuwe dingen te ontdekken, ondanks het gegeven dat de kinderen met ASS minder op acties van hun vader reageerden dan de kinderen zonder ASS. Onze bevinding kan betekenen dat oxytocine kan bijdragen aan de effectiviteit van interventies die adequate ouder-kind interacties bevorderen door het verhogen van sensitiviteit bij vaders. Vragen die opkomen naar aanleiding van de resultaten van deze studie zijn of het mogelijk is dat toediening van oxytocine aan ouders in combinatie met het VIPP-AUTI programma de effectiviteit van behandeling vergroot en, misschien nog belangrijker, of gelijktijdig tijdens interactie het oxytocinegehalte in het kind wordt beïnvloed. Een dergelijk effect kan mogelijk de wederkerige reactie van kinderen met ASS op anderen verhogen, op korte of op lange termijn.

Beperkingen en implicaties voor vervolgonderzoek

De effectiviteit van het VIPP-AUTI programma is aangetoond in een gerandomiseerde, gecontroleerde studie en is vergeleken met een behandeling van gelijke duur en doelstelling. De resultaten van de studies die beschreven zijn in de hoofdstukken 2, 4 en 5 dienen echter geïnterpreteerd te worden in het licht van een aantal beperkingen.

Ten eerste vond de studie in één centrum plaats waardoor de generaliseerbaarheid van de bevindingen beperkt wordt. VIPP-AUTI is vergeleken met het standaard zorgaanbod in het academisch ziekenhuis. Het zorgaanbod in andere instellingen kan verschillen wat betreft inhoud en intensiviteit. Om die reden is vervolgonderzoek nodig om het effect van VIPP-AUTI te vergelijken met het behandelaanbod in diverse (kinderpsychiatrische) zorgvoorzieningen.

Ten tweede hadden de meeste ouders in onze steekproef een gemiddelde tot hoge sociaal economische status en de Nederlandse nationaliteit. Ook waren de meeste ouders getrouwd. Ouders in nadelige omstandigheden, zoals financiële problemen of echtscheiding, zijn soms minder ontvankelijk voor therapie. Het VIPP programma in andere dan ASS-populaties is echter met succes toegepast in gezinnen

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met diverse demografische achtergronden en omstandigheden (Juffer et al., 2009; Moss et al., 2011). Vervolgonderzoek met steekproeven van ouders met een lager sociaal economische status en diverse culturele achtergronden is nodig om de generaliseerbaarheid van VIPP-AUTI, net als van andere VIPP programma's, te bevestigen. Een andere selectiebias is mogelijk de werving geweest van deelnemers voor een interventiestudie. De steekproef bestond uit vooral actieve en gemotiveerde ouders. Dit kan verklaren waarom wij in de studie naar de reactie van ouders op de ASS-diagnose van hun kind een groot aantal ouders vonden met een actie-georiënteerde copingstijl.

Andere beperkingen betreffen de gebruikte meetinstrumenten. De ouders vulden zelf vragenlijsten in over bijvoorbeeld zelfvertrouwen, dagelijkse belasting, breder autisme fenotype en psychische klachten. Deze vragenlijsten kunnen sociaal wenselijke antwoorden bevatten of tijdens de vervolgmetingen ingevuld zijn naar verwachtingen over behandeleffecten. De betrouwbaarheid van respons kan daardoor worden beperkt. Met het enkelblinde karakter van de studie en de gelijkwaardige vormen van thuisbegeleiding is echter getracht de kans op verschillen tussen de experimentele en controle groep te verkleinen.

Ten slotte konden we met de vervolgmetingen in de vrij korte periode na de interventie slechts enkele van de complexe en dynamische factoren onderzoeken die ouder-kind interacties en de opvoeding van kinderen met ASS beïnvloeden. Vervolgonderzoek is nodig om te kijken hoe reactieprocessen van ouders met betrekking tot een ASS-diagnose van hun kind verlopen en om inzicht te krijgen in lange termijn effecten van een behandeling met VIPP-AUTI.

Implementatie van VIPP-AUTI in de klinische praktijk

VIPP-AUTI werd in aansluiting op de diagnostische fase vanuit het academisch ziekenhuis gegeven. Nadat kinderen een ASS-diagnose ontvingen, werden ouders vaak geconfronteerd met wachtlijsten voor vervolgbehandeling, zoals plaatsing op een medisch kinderdagverblijf of intensieve behandelprogramma's. Andere ouders liepen het risico om allerlei interventies tegelijk uit te proberen, die op korte termijn beschikbaar waren. In de kritieke fase na een diagnose in

het academisch ziekenhuis bevelen we daarom evidence-based behandeling met VIPP-AUTI aan.

VIPP-AUTI bleek eenvoudig te implementeren in de klinische praktijk vanwege de relatief korte training van zorgverleners, de gedetailleerde handleiding en het korte programma. Het VIPP-AUTI programma is geprotocolleerd en kan daardoor ook in andere voorzieningen dan het academisch ziekenhuis gebruikt worden. Natuurlijk zijn daar faciliteiten voor nodig, zoals budget voor huisbezoeken, (video)materialen, logistieke organisatie en getrainde zorgverleners, die voldoende tijd en supervisie kunnen krijgen. De meeste van deze faciliteiten zijn echter ook nodig in de huidige zorgverlening voor deze gezinnen, maar dan met het voordeel van minder kosten en belasting dan de intensieve en langdurige interventies die gegeven worden aan kinderen met ASS. De brede inzetbaarheid van het VIPP-AUTI programma door zorgverleners van verschillende disciplines, die ervaring hebben met kinderen met ASS, draagt bij aan de relevantie van dit programma voor de klinische praktijk en gezondheidszorg voor kinderen met ASS.

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Referenties

Atzil, S., Hendler, T., & Feldman, R. (2011). Specifying the neurobiological basis of human attachment: brain, hormones, and behavior in synchronous and intrusive mothers. *Neuropsychopharmacology: Official Publication of the American College of Neuropsychopharmacology, 36*(13), 2603-2615.

Barnett,, D., Clements, M., Kaplan-Estrin, M., McCaskill, J. W., Hunt, K. H., Butler, C. M. et al. (2006). Maternal resolution of child diagnosis: stability and relations with child attachment across the toddler to preschooler transition. *Journal of Family Psychology : JFP : Journal of the Division of Family Psychology of the American Psychological Association (Division 43), 20*(1), 100-107.

Cassidy J., & Shaver Ph. R. (2008). *Handbook of Attachment: theory, research, and clinical applications*.2nd ed. New York: The Guilford Press.

Feldman, R. (2007). Parent-infant synchrony and the construction of shared timing; physiological precursors, developmental outcomes, and risk conditions. *Journal of Child Psychology and Psychiatry, and Allied Disciplines, 48*(3-4), 329-354.

Feldman, R. (2012). Oxytocin and social affiliation in humans. *Hormones and Behavior, 61*(3), 380-391.

Fombonne, E. (2009). Epidemiology of pervasive developmental disorders. *Pediatric Research, 65*(6), 591-598.

Juffer, F., Bakermans-Kranenburg, M. J., & Van IJzendoorn, M. H., ed. (2008). *Promoting Positive Parenting. An Attachment-Based Intervention.* New York/ London: Lawrence Erlbaum Associates/ Taylor & Francis Group

Juffer, F., Bakermans-Kranenburg, M. J., & Van IJzendoorn, M. H. (2009). Attachment-Based Interventions: Heading for Evidence-Based Ways to Support Families. *ACAMH Occasional Papers,* 29, 47-57.

Marvin, R. S., & Pianta, R. C. (1996). Mothers' reactions to their child's diagnosis: Relations with security of attachment. *Journal of Clinical Child Psychology, 25*(4), 436-445.

Meins, E., Fernyhough, C., Wainwright, R., Clark-Carter, D., Das Gupta, M., Fradley, E., et al. (2003). Pathways to understanding mind: construct validity and predictive validity of maternal mind-mindedness. *Child Development, 74*(4), 1194-1211.

Meins, E., Fernyhough, C., Arnott, B., Leekam, S. R., & de Rosnay, M. (2013). Mind-Mindedness and Theory of Mind: Mediating Roles of Language and Perspectival Symbolic Play. *Child Development,* doi:10.1111/cdev.12061; 10.1111/cdev.12061

Moss, E., Dubois-Comtois, K., Cyr, C., Tarabulsy, G. M., St-Laurent, D., & Bernier, A. (2011). Efficacy of a home-visiting intervention aimed at improving maternal sensitivity, child attachment, and behavioral outcomes for maltreated children: a randomized control trial. *Development and Psychopathology, 23*(1), 195-210.

Oppenheim, D., & Koren-Karie, N. (2002). Mothers' insightfulness regarding their children's internal worlds: The capacity underlying secure child-mother relationships. *Infant Mental Health Journal, 23*(6), 593-605.

Patten, E., & Watson, L. R. (2011). Interventions Targeting Attention in Young Children With Autism. *American Journal of Speech-Language Pathology, 20*(1), 60-69.

Rehm, R. (2013). Nursing's contribution to research about parenting children with complex chronic conditions: An integrative review, 2002 to 2012. *Nursing Outlook,* doi:10.1016/j.outlook.2013.03.008

Rentinck, I. C., Ketelaar, M., Schuengel, C., Stolk, J., Lindeman, E., Jongmans, M.J., & Gorter, J.W. (2010). Short-term changes in parents' resolution regarding their young child's diagnosis of cerebral palsy. *Child: Care, Health and Development, 36*(5), 703-708.

Van IJzendoorn, M. H., & Bakermans-Kranenburg, M. J. (2012). A sniff of trust: meta-analysis of the effects of intranasal oxytocin administration on face recognition, trust to in-group, and trust to out-group. *Psychoneuroendocrinology, 37*(3), 438-443.

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Chapter 8

Nederlandse samenvatting

Dankwoord

Curriculum Vitae

Publications

Dankwoord

Allereerst dank ik alle ouders en kinderen hartelijk voor deelname aan de studies. Ondanks het drukke leven van alledag maakten jullie ruimte voor de diverse onderzoeksmetingen. Jullie ontvingen ons thuis en kwamen (soms met het hele gezin) naar het ziekenhuis. Ook had niemand bezwaar om studenten te laten meekijken. Zo boden jullie hen de beste leersituatie. Wij hebben het onderzoek met veel plezier uitgevoerd, dankzij jullie vertrouwen als ouders in de begeleiding en het (vaak grote) enthousiasme van jullie kinderen tijdens de spelsituaties.

Dit proefschrift is naast de betrokkenheid van deelnemende gezinnen tot stand gekomen met de inzet van vele mensen vanuit het UMC Utrecht en de Universiteit Leiden. Ik ben dan ook dankbaar dat ik van 'twee (zo belangrijke) walletjes heb mogen eten'; kinder- en jeugdpsychiatrie en pedagogiek.

Geachte promotor prof. dr. van Engeland, beste Herman. Het is ruim zes jaar geleden dat je mij het vertrouwen gaf om me te kunnen ontwikkelen in wetenschappelijk onderzoek. Ik ben je zeer dankbaar daarvoor en voor je begeleiding in al die jaren. Ondanks of juist dankzij je huidige emeritaat was je makkelijk bereikbaar en toegankelijk. Ik heb veel geleerd van je rijke ervaring en heldere beschouwingen. De warme ontvangst van jou en Herma in Montfoort maakten voor mij de 'laatste loodjes' dragelijk. Jouw relativeringsvermogen, je humor en interesse waren voor mij een inspiratie om te kunnen schrijven en een baken tijdens pieken van stress. Ook veel dank voor de koppeling van het oxytocine-onderzoek aan ons project. Het is verrijkend om van diverse kanten te zoeken naar positieve invloeden op de ontwikkeling van kinderen met autisme.

Geachte promotor prof. dr. Bakermans-Kranenburg, beste Marian en geachte prof. dr. van IJzendoorn, beste Rien. Vanuit de Universiteit Leiden hebben jullie mij gezamenlijk begeleid bij dit project. In juni 2008 begonnen we met de uitvoering van de thuisbegeleidingen die vervolgens tot augustus 2012 zouden duren. De bijeenkomsten met jullie waarin we de voorbereiding van diverse huisbezoeken

bespraken, waren bijzonder leerzaam. Met jullie feedback leerde ik de toepassing van de interventie verbeteren. Verder dank ik jullie voor het overbrengen van wetenschappelijke en statistische kennis. Ook dankzij jullie grondige en snelle reacties op de talloze concepten voor artikelen ligt hier nu dit proefschrift. Het is voor mij een eer dat jullie mij wilden begeleiden.

Geachte copromotor dr. Naber, beste Fabiënne. Het begon in de wandelgangen. Zoals altijd, zat je vol inspirerende ideeën over nieuwe onderzoeken en tot mijn verrassing zag je mij wel een onderzoek uitvoeren. De plannen werden snel serieus, er kwam een goedgekeurd protocol en we konden starten. Er brak een geweldige periode aan. Met jou en door jou is dit proefschrift tot stand gekomen. Samen volgden we cursussen, bezochten symposia, vroegen (tevergeefs) subsidies aan, voerden het onderzoek uit, vormden een projectteam met de thuisbegeleiders, hielden presentaties, begeleidden studenten, maakten keuzes, discussieerden en deelden allerlei emoties. Nu zitten we in de afbouwfase en vervaagt de intensiviteit van onze samenwerking. Terugkijkend is het vooral jouw kennis en ervaring die me gebracht hebben waar ik nu sta. Een betere copromotor had ik me niet kunnen wensen. Dank voor alles!

De leden van de leescommissie, prof. dr. M.J. Schuurmans, prof. dr. R.S. Kahn, prof. dr. W.C.H.J. Matthys, prof. dr. F. Juffer en prof. dr. J. Schenk wil ik hartelijk danken voor het kritisch beoordelen van het manuscript.

I acknowledge dr. D. Wheatley, BioMedes ltd office, United Kingdom for editing some parts of the manuscript.

Zonder studenten was de uitvoering van dit project onmogelijk geweest. Zij verzamelden en codeerden data en assisteerden bij allerlei onderzoekstaken. Graag wil ik daarvoor danken Elsbeth, Else, Nikki, Maike, Irene, Veerle, Sandra, Lisa, Valerie, Maaike, Leeanna, Aniek, Marjolein, Christien en Miranda. Christien, extra dank voor de taaltesten. De snoeppot tovert niemand plotseling meer vol, maar blijft populair. Aniek en Marjolein, jullie kwamen zelfs na je studie terug

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om te coderen en te assisteren, extra dank daarvoor. Marjolein jouw perfecte data-organisatie is nog steeds van onschatbare waarde.

Beste Carolien Wasmus en Renske Houben, jullie hebben de experimentele thuisbegeleiding met groot enthousiasme en inzet gegeven en gevormd. Ik dank jullie voor de fantastische samenwerking en jullie zorgvuldigheid om volgens het protocol te handelen. Nog leuker dat jullie nu mijn paranimfen willen zijn, zodat we ook deze promotie als een gezamenlijk resultaat kunnen vieren.

De thuisbegeleiders van de controlegroep Saskia Boelen-Wieckardt, Rosalie Visser en Marieke de Rijke wil ik hartelijk danken voor de betrokkenheid bij de studie. Het lukte jullie om binnen twee weken na randomisatie te starten met de thuisbegeleiding; een nieuwe structuur. Dank zij jullie hebben we ook veel ingevulde vragenlijsten van ouders retour kunnen krijgen.

Dr. van Daalen, beste Emma, graag bedank ik je voor je betrokkenheid bij dit onderzoek en alle adviezen. Ook dr. de Jonge, beste Maretha dank ik zeer. Jouw bijdrage aan de ontwikkeling van VIPP-AUTI en later de informatie over de nieuwe diagnostische opvattingen zorgden voor kwaliteit. Emma en Maretha, het is een luxe om experts zo dichtbij te hebben.

Alle overige disciplines van het voormalig team 'Jonge kinderen met ontwikkelingsstoornissen, of wel de 'peuterpoli', wil ik hartelijk danken. In het bijzonder dank ik Karlijn Steggehuis voor de afname en supervisie van psychologische en taaltesten bij de deelnemende kinderen (en tijdens de 1ste fase dank aan Sanne Haspels). Natuurlijk ook dank aan alle arts-assistenten die samen met de thuisbegeleiders de schriftelijke informatie over het onderzoek aan ouders gaven.

Veel taaltesten namen logopedisten af buiten het UMC. Ik dank jullie in het bijzonder voor de prettige samenwerking. Het was fantastisch dat jullie tijd namen om een (soms extra) test af te nemen, zodat de kinderen niet speciaal daarvoor naar het ziekenhuis hoefden te komen.

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De verpleegkundige wetenschappelijke onderzoeksgroep was (en blijft gelukkig) een platform van leren en kennis uitwisseling. Vanaf het 1ste uur dank ik Han, Julia, Floor en Astrid voor het samen schrijven van een 1ste artikel. Daarna volgde het IJsland-project, met heel veel dank aan Thora (ook voor een ontspannen wijntje als buurvrouw), Marijke, Martine en Janneke (met jou sparren Janneke blijft een genot). Nu staat de onderzoeksgroep onder leiding van Marieke Schuurmans en Jaap Trappenberg en dank ik hen en alle leden voor de inspiratie die zij geven.

Een niet te onderschatten inspiratiebron is het docentschap bij KGW met dank aan Claudia, Irene (of is het Airien?), Harmieke, alle mededocenten en vooral de kritische en leergierige studenten.

Ook dank ik de inzet van de 'jonge gezinnen'. Tibor, bedankt voor de ingeleverde tijd, koerierdiensten en meelezen. Victor, jij was de allereerste vader die zich liet filmen om ons protocol uit te proberen. Hartelijk dank daarvoor en voor alle tips die je vanuit je uitgebreide vakkennis aan ons gaf. En natuurlijk onze kleine vriendinnetjes (en soms proefkonijntjes) Imani, Olivia, Lieve en Bibi. Leuk dat jullie met mama meekwamen en bedankt voor de mooie tekeningen die ik van jullie kreeg. Extra dank aan Lieve, dat je speciaal een tekening 'moeder en kind' hebt gemaakt voor de omslag van dit boekje. Je bedacht er ook een speelkleed bij; precies wat bij onze thuisbegeleiding hoorde.

Verder dank aan alle (oud)collega's van Psychiatrie en daarbuiten voor jullie medeleven en belangstelling voor dit onderzoekstraject. Jullie zijn met te veel om op te noemen, maar voor enkelen maak ik graag een uitzondering. Coby, Hanneke, Ciska, Lidewij, Els, Wieske en Annerieke, nog steeds vind ik het jammer dat jullie niet meer in het UMC werken. Ook dank voor het regelmatige contact uit Amsterdam met Ria W., Ria v M., Riet en Ellen (wie is er aan de beurt?). Heel belangrijk is natuurlijk de dagelijkse gezelligheid en steun van mijn huidige kamergenoten: Martijn, Annemarie, Floor, (en flexplekgebruikers) Marianne, Desiree, Margo en Rita. En de stimulerende samenwerking met Laura, Jos, Manon, Paula, Liesbeth, Carry, Antoinette (dank voor het inschakelen van je netwerk) en verpleegkundigen/ groepsleiders van de afdeling

Psychiatrie. Bijzondere dank ook aan Iris Sommer en Sarah Durston voor jullie vertrouwen door mij al een jaar voor de promotiedatum als onderzoekscoördinator aan te nemen. Een uitdagende taak, waar ik de komende tijd graag aan verder werk.

Om de werkdruk een beetje de baas te blijven is het UMC koor een uitkomst gebleken. Veel dank aan Erwin van Geenen en alle koorleden voor het heerlijk samen zingen.

Beste Everlien, Roland, Angela, Marie-Louise, Bianca, Nico, Anke, Jopke en Jo het was een welkome afleiding en leerzame ervaring om met jullie het gezamenlijke symposium van Rho Chi Chapter-at-Large en V&VN te organiseren.

Lieve buren, dank voor de gezellige BBQ's, kerstborrels, verjaardagen, praatjes op straat en in de tuin. Zo dichtbij zoveel mensen om op te rekenen is echt geweldig.
Lieve vrienden, we kennen elkaar al zolang; vanaf de kleuter- en middelbare school, padvinderij of via Kees (Tiel Utrecht en Docete). Gelukkig hebben jullie het ook druk, maar we pakken op elk moment de draad weer op alsof de tijd niet verstrijkt. Dank voor jullie vriendschap.
Lieve (schoon)familie, familie kies je niet, maar anders had ik voor jullie gekozen. Het is fijn om samen lusten en lasten te delen. Dank dat jullie er zijn. Bijzondere dank aan mijn schoonmoeder, die mijn carrière mogelijk maakte door op te passen (en nog veel meer) toen onze kinderen nog klein waren. Zonder dat ma, lag er nu geen proefschrift. Lieve schoondochters Whitney, dank voor je vrolijke gezelligheid en Trynke, extra dank voor je hulp bij de statistiek.
Lieve Ruth, Arnout en Jasper, het laatst komen de personen die het allerbelangrijkste zijn. Jullie richten als volwassenen je eigen leven in, daarvoor ben ik dankbaar. Door mijn focus op werk en onderzoek heb ik jullie minder kunnen bieden dan ik wilde. Ik hoop dan ook snel dit te verbeteren door ruimte te creëren voor meer gezamenlijke belevenissen. Dank voor jullie geduld. Ik ben heel trots op jullie en hoe jullie in het leven staan.

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Allerliefste Kees, jij hebt je moeiteloos aangepast aan dit jarenlange promotieonderzoek. Je ving en vangt al mijn stemmingen op, beleeft alles mee en betekent alles voor mij. Onze liefde duurt al zoveel jaren, het is dus echte liefde en ja, 'ik ook van jou…'.

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Nederlandse samenvatting

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Curriculum Vitae

Irina Poslawsky werd geboren op 27 januari 1954 te Utrecht. In 1972 deed zij eindexamen HBS-B aan het 2de Vrijzinnig Christelijk Lyceum te Den Haag. Aan de Hogeschool Midden-Nederland Utrecht behaalde zij in 1976 haar HBO-V diploma en in 1991 het getuigschrift van de Voortgezette Opleiding Sociaal Psychiatrisch Verpleegkundige. Na drie jaar deeltijdstudie aan de Universiteit Utrecht, studeerde zij in 2003 af in de Algemene Gezondheidswetenschappen, richting Verplegingswetenschap.

Irina begon haar loopbaan in 1976 als verpleegkundige in een algemeen ziekenhuis te Utrecht. Van 1977 tot 1999 werkte zij in Amsterdam als sociaal psychiatrisch verpleegkundige (spv) in de ambulante Kinder- en Jeugdpsychiatrie bij de GG & GD (Speciaal Onderwijsteam), de Centrale Riagg Dienst en RIAGG Zuid/Nieuw-West. Van 1991 tot 1999 combineerde zij de functie spv met teamleider. Daarnaast was Irina van 1990 tot 1995 projectleider en coördinator van het VTO-Samenwerkingsverband te Amsterdam (Vroegtijdige onderkenning Ontwikkelingsstoornissen). Ook was zij van 1996 tot 2001 lid van de Raad van Toezicht van het Sociaal Agogisch Centrum, het Burgerweeshuis te Amsterdam. Vanaf 1999 vervolgde Irina haar loopbaan aan het Universitair Medisch Centrum te Utrecht (UMC Utrecht), tot 2005 als coördinerend hoofdverpleegkundige van de afdeling Kinderpsychiatrie en vanaf 2005 tot heden als verpleegkundig beleidsmedewerker bij de afdeling Psychiatrie. Drie jaar tot 2006 was zij lid van de Medisch Ethische Toetsingscommissie Utrecht. Daarnaast is zij vanaf 2003 cursusdocent aan de masteropleiding Klinische Gezondheidswetenschappen. In 2007 startte Irina in deeltijd met het promotietraject op de afdeling Psychiatrie en Hersencentrum Rudolf Magnus van het UMC Utrecht, waarvan de onderzoeksresultaten in dit proefschrift zijn beschreven.

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Curriculum Vitae

Publications

Publications

Poslawsky I.E., Schuurmans M.J., Lindeman E., & Hafsteinsdóttir T.B. 2010. A systematic review of nursing rehabilitation of stroke patients with aphasia. *Journal of Clinical Nursing,* 19; 17-32

Beentjes T.A.A., Goossens P.J.J., & **Poslawsky I.E.** 2011. Caregiver Burden in Bipolar Hypomania and Mania: a systematic review. *Perspectives in Psychiatric Care,* doi: 10.1111/j.1744-6163.2012.00328.x

Naber F.B.A., **Poslawsky I.E.,** Van IJzendoorn M.H., Van Engeland H., & Bakermans-Kranenburg M.J. 2013 Brief report: Oxytocin enhances paternal sensitivity to a child with autism: A double blind within-subject experiment with intranasally administered oxytocin. *Journal of Autism and Developmental Disorders,* 43; 224-229

Poslawsky I.E., Naber F.B.A., Van Daalen, E., & Van Engeland H. 2013. Parental reaction to early diagnosis of their children's Autism Spectrum Disorder: an exploratory study. *Child Psychiatry and Human Development,* doi: 10.1007/s10578-013-0400-z

Abstract presentations (oral)
8th Quadrennial Congress of the European Association of Neuroscience Nurses, Iceland May-June 2007. Title: Communication Problems in Patients with Stroke: A Systematic Review of the Literature.

7[de] Vlaams-Nederlands Wetenschappelijk Congres, Amsterdam November 2007. Poster presentation. Title: Spraak-taalproblemen na een beroerte: een systematische literatuurstudie.

Symposium 'Ongemerkt Autistisch', Ede October 2008. Title: De Utrechts-Leidse Interventie-methode: de behandeling van jonge kinderen met Autisme Spectrum Stoornissen.

4th European Nursing Congress Older persons: the future of care, Rotterdam October 2010. Title: Patients with aphasia after stroke and the role of nurses.

12[de] Nationaal Autisme Congres, Rotterdam March 2012. Poster presentation. Title: Oxytocin Enhances Paternal Sensitivity to a Child with Autism.

Symposium 'Vroege interventie bij jonge kinderen met autisme', Ede April 2012. Title: Videofeedback interventie voor ouders van jonge kinderen met autisme.

Book chapter
Poslawsky I.E., Schuurmans M., & Hafsteinsdóttir T.B. 2009. Communi-catiestoornissen na een beroerte. Hoofdstuk 9 in V*erpleegkundige revalidatierichtlijn beroerte*. Hafsteinsdóttir T.B. & Schuurmans M. ed. Maarssen. Elsevier Gezondheidszorg.

Publication for clinical practice
Poslawsky I.E., Van Wakeren A., Gooskens F., Machielsen J.E.H.M., & Boter H. 2004. De time-outruimte bij agressie in de kinderpsychiatrie. *Tijdschrift voor Verpleegkundigen,* 114 (7/8); 56-62

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