

Unravelling Challenging Behaviour

A study into the factors that influence the occurrence of challenging behaviour of children with an Autistic Spectrum Disorder

Yvette M. Dijkhoorn

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challenging behaviour of children with an Autistic Spectrum
Disorder

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Voorwoord

Het moeilijke gedrag van kinderen die door hun autisme grotendeels afhankelijk zijn van hun opvoeders, maar wier gedrag ook juist die opvoeders ernstig kan kwetsen, is fascinerend en vraagt om nader onderzoek om de opvoeders en de kinderen wetenschappelijk verantwoorde interventies te bieden. Ik prijs mezelf gelukkig dat juist deze problemen onderwerp van mijn proefschrift konden worden. Ik heb het altijd als logisch ervaren om eerst als clinicus met deze groep ervaring op te doen en vervolgens onderzoek te doen. Juist deze klinische ervaringen hebben tot de vraagstelling geleid. Onderzoek doen is interessant, maar weten welk praktijkdoel het dient, maakt het nog interessanter.

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Pap en mam, 'het boek' is af!

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Yvette Dijkxhoorn

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1 Introduction

Parents, caretakers and teachers of children with autism are faced with behaviour that hinders the upbringing and education of these children. Some behaviour is specific to autism, other behaviour is not. Both the specific and non-specific behaviour problems can be so extreme that they become dangerous to the health of the child or that of others. Especially the non-specific behaviour problems receive relatively little attention in the literature on autism, but everyone who encounters these children on a daily basis knows that these problems are by no means rare and can pose serious problems for caretakers.

In the last decades the amount of research carried out on autism, especially the origin of the disorder and the cognitive functioning of people with autism is impressive. At the same time we see an increased awareness of the prevalence of behaviour problems among people with Mental Retardation. However, research into the comorbidity of the Pervasive Developmental Disorders and Mental Retardation is relatively scarce; in particular research into behaviour problems of these double handicapped individuals is limited. This is surprising, even more so if we consider that research on people with Mental Retardation and the most extreme forms of behaviour problems, such as self-injurious behaviour, violence towards others and destruction, has shown that a high percentage of these people have a Pervasive Developmental Disorder (De Vos, Buysse & Van Berckelaer-Onnes, 1996). The comorbidity of a Pervasive Developmental Disorder and Mental Retardation seems to be an important risk factor for the development of behaviour problems. However, little is known about the emergence and course of this behaviour, which is a major problem when we are faced with the challenge of treating these children and their behaviour problems.

This leads us to our general research question:

Which factors predispose to the emergence of the extreme challenging behaviour of children with a Pervasive Developmental Disorder and Mental Retardation?

Children develop through interactions with their environment. According to the transactional model of child development, the continuous interaction of the child with his communicative partners provides the feeding grounds for healthy development (Wetherby, Prizant & Schuler, 2000). This interplay of the child with its possibilities and shortcomings, influenced by neurophysiological variables and the ability of the caregiver to respond to the child's needs and the environment, determine the outcome of the development. Bronfenbrenner and Ceci (1994) propose in their bio-ecological model that proximal processes influence effective development and determine the outcome of genetic predispositions. With this model individual differences between people with identical genetic traits can be explained. O'Brien (2000) has divided the factors that could play a role in the emergence and maintenance of the challenging behaviour of children with Mental Retardation into *predisposing factors* (e.g. the severity, the cause and the nature of the disability of the child and the personal history of the child), *precipitating factors* (e.g.

environmental change and physical morbidity) and *perpetuating factors* (e.g. biochemical behavioural modulators, learning theory, interpersonal factors and social insights). These factors can be risk factors but may also function as protective factors. So according to the bio-ecological model, these factors could both maximize and minimize the outcome.

For the purpose of our research we needed to assess various factors that influence the development of children with a Pervasive Developmental Disorder and Mental Retardation. Wenar & Kerig (1999) proposed a developmental framework that incorporates factors within the child, the **intrapersonal context**, (e.g. behaviour, cognition, personality); factors concerned with the interaction, the **interpersonal context** (e.g. parent-child relationships, teacher-child relationships) and the **super-ordinate context** (e.g. family, group, socio-economic status); and biological factors, the **organic context** (e.g. genetic predisposition, illness).

The bio-ecological model of Bronfenbrenner and Ceci (1994) and the developmental framework of Wenar & Kerig (1999) combined with the factors of O'Brien (2000) to explain the emergence of challenging behaviour provide us with a model for our research. They can be integrated into a framework to assess children with a Pervasive Developmental Disorder and Mental Retardation.

We carried out a literature study on behaviour problems among children with a Pervasive Developmental Disorder and Mental Retardation first. We then designed a research project which consisted of two phases (see chapter three). In the first phase we carried out an analysis of existing data, to explore the main intrapersonal factors. We want to know:

Which behaviour problems do children with a Pervasive Developmental Disorder display?

Which intrapersonal factors play a crucial role in the emergence of these problems?

The results of this phase, which will be described in chapter four, provided us with some basic information which gave us the opportunity to explore the specifics of the behaviour problems of children with a Pervasive Developmental Disorder and Mental Retardation more thoroughly. The goal of the second phase was to design a framework with which 'children at risk' can be assessed. The results of this phase will be described in chapter five.

We will end with conclusions and recommendations.

2 Challenging behaviour of children with a Pervasive Developmental Disorder and Mental Retardation

2.1 Introduction

This chapter will begin with a discussion on the terminology and the classification of challenging behaviour. Furthermore the Pervasive Developmental Disorders and Mental Retardation and the divergence and convergence of the two will be discussed. Various factors that could explain the differences between individuals with these disorders will be analysed. Possible causes and implications of behaviour problems as displayed by children with a Pervasive Developmental Disorder and Mental Retardation will be described. We have also collected information about the behaviour problems of children with Mental Retardation and adults with a Pervasive Developmental Disorder and Mental Retardation.

2.2 Challenging Behaviour

Behaviour problems, behaviour disorder, maladaptive behaviour, antisocial behaviour, aberrant behaviour and challenging behaviour are just a few of the terms used to describe behaviour that is found to be abnormal in children. In developmental psychopathology a continuum from normal behaviour to severe psychopathology has been proposed (Achenbach, 1997). According to developmental psychopathologists, behaviour problems are the result of psychopathology (Burack, 1997). The main question remains however: at which point should the behaviour of children be considered problematic. To answer this question an objective measure is needed to assess whether the behaviour is problematic and to whom it is problematic.

The interactions of the child with or without a disability with its environment determine healthy development (Bronfenbrenner & Ceci, 1994), but these interactions also determine psychopathology, which can lead to behaviour problems. Moreover, we share the view of Došen (1990) who states that the behaviour of children with Mental Retardation is not only determined by the surroundings and organic factors, but is also the result of an interaction between a biological child and a child with its own unique experiences with its social and material surroundings. Thus behaviour always takes place in an interaction with others. Therefore the people in that interaction determine whether the behaviour is problematic.

From a pedagogical point of view the behaviour of children has to be considered problematic when their caregivers experience problems in dealing with the behaviour. In addition Kars (1995) states that the behaviour of mentally retarded subjects is problematic when there is a problem both for the person with Mental Retardation and for his or her surroundings. Young children and children with autism with or without Mental Retardation hardly ever consider their own behaviour to be

problematic. The labelling is almost exclusively carried out by the significant others in their surroundings. We have adapted the following factors from Evans & Meyer (1985), which have to be taken into consideration when assessing the problematic behaviour of people with Mental Retardation:

- the context: behaviour takes place in a certain context, depending on the values, norms and expectations of the system; behaviours are seen as problematic or not. Emerson (2001) points out that social rules in a setting, the ability of the person with MR to explain his behaviour, the beliefs of others about MR and behaviour problems, and the capacity of the setting to deal with these behaviours together define behaviour as challenging.
- the age of the child: expected behaviours change with age, some behaviours are considered 'normal' in toddlers but not in teenagers;
- the developmental age of the child: age and developmental age are comparable in normal development, but developmental delay defines MR. However, children with a Pervasive Developmental Disorder and Mental Retardation have a highly irregular developmental profile, which implies that one cannot speak of a developmental age;
- the sex: girls are expected to exhibit different behaviours than boys;
- the interaction with the surroundings: behaviours occur in an interaction: this is a very important factor, especially for children with disorders (such as autism), which by definition include problems with interaction and communication.

Thus, if behaviour is defined by the interaction, factors within the child (biological and intrapersonal factors) and the surroundings (super-ordinate), including parent-child and child-child interactions, and child rearing (interpersonal factors) have to be considered. We will use the term 'challenging behaviour', because this term incorporates both the influence of the surroundings and the observable behaviours. The question of 'who is to blame' is not taken into consideration. Emerson (2001) gives two major advantages of this term. Firstly the term is free from assumptions about the psychological characteristics of the behaviour. Challenging behaviour can be very functional and even adaptive to the person with MR. Secondly the term challenging behaviour refers specifically to behaviours that are dangerous and not only unusual or culturally abnormal. Challenging behaviour literally poses a challenge for both the person with Mental Retardation and his or her surroundings.

2.2.1 Classifying Challenging Behaviour

Labelling behaviour as problematic is a normative process. To make a comparison possible, it is necessary to reformulate the problems in a more objective way, even if total objectivity is not possible. The research described below involves children with an Autistic Spectrum Disorder and Mental Retardation and extreme challenging behaviour. These children are often referred to regional advisory teams, which were established by the Dutch government for the purpose of helping people with Mental Retardation and extreme challenging behaviour and their caretakers. The Dutch Care Institute (NzI) carried out some research to determine which people with Mental

Retardation, who live in residential care facilities, needed the extra help (Schuring et al., 1990). They defined a mentally handicapped person with challenging behaviour as follows: someone with Mental Retardation who shows behaviours that are dangerous, a serious threat or unmanageable for the person himself, or other people or objects in his surroundings. They also included the group of people with Mental Retardation for whom the above-mentioned behaviours would reoccur if specific treatment or measures were to be withdrawn.

We will use the definition as proposed by Emerson (2001, p. 5) who states that challenging behaviour is: ‘culturally abnormal behaviour(s) of such an intensity, frequency or duration that the physical safety of the person or others is likely to be placed in serious jeopardy, or behaviour which is likely to seriously limit use of, or result in the person being denied access to, ordinary community facilities.’

Classification of challenging behaviour is not an easy task. Some challenging behaviour can be categorized under psychiatric conditions, but lots of behaviour will not fit a specific category. Challenging behaviour is not a synonym for a psychiatric disorder, neither do all psychiatric problems lead to challenging behaviour (Emerson, 2001).

We are faced with another problem when we apply psychiatric criteria to mentally retarded people, especially those with a more severe mental handicap. Sovner & Hurley (in Došen, 1990, p. 22) have formulated four factors that make it difficult to diagnose psychiatric disorders in people with MR:

1. The lower level of functioning, especially the difficulties with communication, make it very difficult for the person with MR to make others aware of his feelings and thoughts.
2. Lack of imagination causes certain specific symptoms to be presented inadequately (e.g. delusions in schizophrenia).
3. When people with MR face stress, they will often disintegrate to a lower level of functioning; this phenomenon often disguises other symptoms (e.g. dementia, psycho-organic pathology).
4. Even at a low level of emotional stress, people with MR can react with significant behavioural changes, both in intensity and quality.

These phenomena complicate the classification of psychiatric conditions of children with Mental Retardation. Dykens (2000) emphasizes these problems and adds that the role of these factors increases as the level of functioning decreases. These problems probably cause even greater difficulties classifying co-morbid psychiatric conditions in children with Pervasive Developmental Disorders. After all, impairments in socialization, communication and imagination occur by definition in these children. In addition to the third point one could argue that the autistic symptoms easily disguise other problems.

Einfeld & Tonge (1996c) have described problems in the use of multi-axis classification systems, specifically the ICD-10 guidelines (WHO, 1992) for people with Mental Retardation. When applying this guideline to people with autism who also display other problems, e.g. hyperactivity, the classification autism precedes the classification of the hyperkinetic disorder. The hyperkinetic disorder therefore cannot

be diagnosed as a co-morbid disorder. Although most clinicians claim to be able to distinguish between the two, and medication is often helpful for clients with both autism and hyperkinetic problems, classifying both disorders is not possible according to the ICD-10. Although the statement 'Attention Deficit/Hyperactivity Disorder is not diagnosed if the symptoms of inattention and hyperactivity occur exclusively during the course of a Pervasive Developmental Disorder or a Psychotic Disorder' in the DSM-IV-TR (APA, 2000, p. 91) often confuses people, according to the DSM-IV-TR (APA, 2000), co-morbidity of these two disorders can occur, but a thorough assessment of the source of the hyperactivity is required.

Therefore classifying behaviour by means of psychiatric labelling according to DSM-IV-TR or ICD-10 criteria is very difficult and, although helpful in explaining some behaviour, classifying challenging behaviour by means of these systems is not a very fruitful exercise.

Another way to classify behaviour has been proposed by Schuring et al. (1990). They have developed the Consensus Protocol Severe Behaviour problems (CEP) in order to distinguish between people with and without challenging behaviour who live in Dutch institutions for people with Mental Retardation. Through a consensus method several people who are involved in the care of a client (e.g. caretakers, parents or spokesmen) determine the level of challenging behaviour together. This method of classifying incorporates at least the view of people who are directly involved in taking care of the client with challenging behaviour and can therefore be considered a pedagogical view on challenging behaviour.

Another way of classifying challenging behaviour is to compare a score obtained with a norm. This procedure is applied with different instruments that can also be used for children, e.g. the 'Maladaptive Behavior Scale for Mentally Retarded individuals' (in Dutch SGZ) designed by Kraijer & Kema (1994) and the Developmental Behavior Checklist (DBC) designed by Einfeld & Tonge (1995). The SGZ measures the following behaviour factors: aggressive behaviours, verbal aggressive behaviours and mixed challenging behaviour. According to Kraijer a child must be called 'behaviourally disturbed' when both the professional opinion and the SGZ-score agree on the presence of disturbed behaviour.

The DBC is described as a questionnaire that has to be completed by the primary caretakers of children and adolescents with MR, to assess behavioural and emotional disturbance. The decision whether behaviour is within the clinical range or not is based on a 'cut-off score'. Although these instruments are helpful, Dykens (2000) has postulated that the diversity between these instruments is great and none seem to cover the range of problems that people with Mental Retardation show. It is always only a proportion. Another problem is that all of the above-mentioned instruments have specific norms for children with Mental Retardation, but not for children with a Pervasive Developmental Disorder. Because of the presence of items about behaviour specific to the Pervasive Developmental Disorders, children with a Pervasive Developmental Disorder and Mental Retardation always have a score on these instruments.

So, we really do not have valid instruments to determine the broad scope of the behaviour of children with a Pervasive Developmental Disorder and Mental Retardation. The available instruments only apply for a proportion of the clients or cover a limited range of behaviours or disorders. Therefore Dykens (2000) has suggested that the best way to gather information on the challenging behaviour of children with MR is to adopt a 'multi-method, multi-informant' approach. To collect an optimum amount of information, individual descriptive diagnostics should be carried out. Clinical assessments by a professional, together with standardized instruments using different informants, provide researchers with at least the most complete information about the challenging behaviour.

2.3 The Pervasive Developmental Disorders and Mental Retardation

In the currently used classification systems, DSM-IV-TR (APA, 2000) and ICD 10 (WHO, 1992), the Pervasive Developmental Disorders and Mental Retardation are considered to be two separate disorders. However, for people with the Autistic Disorder, the rate of incidence of Mental Retardation was found to be 75-80% (Bailey, Philips & Rutter, 1996; Kraijer, 1998; Van Berckelaer-Onnes & Van Engeland, 1997). In this chapter we will describe the divergence and convergence of the two disorders, starting with the terminology and the history and working up to recent descriptions of the two disorders. Different factors that could explain individual differences between children will be discussed.

2.3.1 Terminology

The term 'Pervasive Developmental Disorders', as used in the classification systems, is rather vague. It is supposed to be the umbrella term for all five classifications but, especially in the case of Asperger Syndrome and high functioning autism, it is debatable whether all developmental aspects are afflicted, as the term 'pervasive' would imply. Most clinicians and parent associations prefer the term 'Autistic Spectrum Disorders' as proposed by Lorna Wing (1993b). This term defines the disorders as the consequence of the triad of impairments in socialization, communication and imagination. In the following chapters we will continue to use this term and its abbreviation ASD.

The ASD's are found at all levels of intelligence. We will use the term Mental Retardation, or its abbreviation MR, to define the group with a significantly low level of intellectual and adaptive functioning. The American Association on Mental Retardation (AAMR) provides us with the following definition of Mental Retardation (Luckasson et al., 1992, p. 15). 'MR refers to substantial limitations in present functioning. It is characterized by significantly sub- average intellectual functioning, existing concurrently with related limitations in two or more of the following applicable adaptive skill areas: communication, self-care, home living, social skills, community use, self-direction, health and safety, functional academics, leisure and work. MR manifests before age 18'. This latest version has been

criticized, especially with regard to children, because most of the adaptive skills do not really apply to children, and the areas that are specifically applicable to children (e.g. school work) have not been included (Vig & Jedrysek, 1999). We will therefore use the criteria as defined by the DSM-IV-TR (APA, 2000). In the DSM-IV-TR, an IQ below 70 is mentioned specifically. This implies that in this operationalization of MR, the focus is primarily on sub-average intellectual functioning.

The DSM-IV-TR defines 4 subgroups, according to the level of functioning. As especially young and low functioning children with MR are often very difficult to test with regular IQ-tests, the level of functioning is also given as developmental age. This has been adapted from Došen (1990) and Kraijer & Plas (1997).

| DSM-IV-TR classification | IQ-score | Developmental Age | Cognitive phase |
|--------------------------|------------------|-----------------------------|---------------------|
| Mild MR | 50-55 till 70 | 6 - 7 to 11 years | Concrete operations |
| Moderate MR | 35-40 till 50-55 | 3;6 - 4 to 6 - 7 years | Prelogic |
| Severe MR | 20-25 till 35-40 | 1;6 - 2 to 3;6 till 4 years | Pre-operational |
| Profound MR | Below 20-25 | Below 1;6 - 2 years | Senso-motoric |

2.3.2 The history of the Autistic Spectrum Disorders and Mental Retardation

The first descriptions of eleven children who exhibited a condition that 'differs so markedly and uniquely from anything reported so far, that each case merits-and, I hope, will eventually receive- a detailed consideration of its fascinating peculiarities' (p. 217) was given by Leo Kanner (1943). He thought that social withdrawal was the primary feature and hence chose the name 'Autism' (derived from the Greek word *autos* (=self). Kanner, however, was not the first to use the term Autism. In Europe, Bleuler (1911) used the term 'Autismus' as a description of a symptom of schizophrenia, namely: 'Die Loslösung von der Wirklichkeit zusammen mit dem relativen und absoluten Überwiegen des Binnenslebens' (p. 52). The term 'autistic' can also be found in the annals of the Pedagogy Institute in Nijmegen between 1937 and 1940. Members of the staff of this institute described children with behaviour very similar to that of the children Kanner described a few years later (Van Berckelaer-Onnes, 1979). In both descriptions the stereotypical behaviour (e.g. hand-flapping, rocking, spinning), extreme anxieties, resistance to change, lack of interest in the environment including people and retarded development of communication were mentioned as the most striking features of these children.

Asperger (1944) published a finding similar to that of Kanner, but his article did not become widely known until the 1980's. He also described a group of boys with specific social problems. He named his group of boys 'autistischen Psychopathen im Kindesalter'. The key features noted in his boys were: an oldish appearance, an unnatural way of talking, high intelligence, limited interests and poor motor skills. He found that the boys made contact in an odd way, tending towards maliciousness. Wing (1981) mentioned Asperger syndrome for the first time in the Anglosaxon literature. Since 1994 it is considered a subgroup of the Pervasive Developmental Disorders (APA, 1994).

The above-mentioned revolutionary researchers in the field of autism did not describe a general cognitive delay in their cases. Kanner (1954) even stated that

autism could be clearly distinguished from schizophrenia due to the age of onset and from 'oligophrenia' due to the normal cognitive potential. Van Krevelen (1963), however, stated that it was almost impossible to distinguish autism from 'imbecilitas'.

It took quite some time before it was established that the often observed splinter skills were not always proof of normal cognitive potential and that the low IQ-scores were not the result of poor 'test-attitude' (Volkmar & Lord, 1998). The work of Wing & Gould (1978 & 1979) in particular provided ample evidence for the theory that the majority of people displaying the 'triad of impairments' function at a significantly lower level of intellectual functioning.

Mainly due to the work of Creak (1961), Rutter (1978), Wing & Gould (1979) and Wing (1993b) clear diagnostic criteria are nowadays available. Both major classification systems more or less agree on the basic diagnostic behavioural features. Both have included the category 'Pervasive Developmental Disorders', which incorporates the diagnoses Autistic Disorder, Asperger Disorder, Rett's Disorder, Childhood Disintegrative Disorder and the Pervasive Developmental Disorder Not Otherwise Specified (PDD-NOS) including Atypical Autism. In the ICD 10 (WHO, 1992) the last category is called Atypical Autism.

The level of functioning of individuals who belong to one of the five categories differs markedly. People with Asperger Disorder have intellectual capabilities within the normal range (IQ>70). On the other hand Rett's Disorder and Childhood Disintegrative Disorder are always associated with (severe) MR (Volkmar & Lord, 1998).

Most studies on intellectual functioning have been carried out within the population Autistic Disorder. Specific information about the presence of MR in the PDD-NOS category is scarce (Prior & Ozonoff, 1998).

Still open is the issue of age at onset. Whereas the diagnostic criteria are clear for variable age of onset of MR, which by definition has to occur before the age of 18 years, there seem to be differences of opinion about the ASD's. According to the DSM-IV-TR (APA, 2000), the first symptoms of the ASD have to become manifest before the age of 3 years. Rett's Disorder and Childhood Disintegrative Disorder (CDD) are characterized by a developmental regression, in the case of CDD even after the age of 3 years. Developmental regression is often reported in children with an ASD. Davidovitch et al. (2000) found regression in 47.5% of cases studied. Often regression in the ASD's is associated with more severe symptoms (Hoshino et al., 1987; Davidovitch et al., 2000).

In the following paragraph we will describe the ASD's in combination with Mental Retardation at the different levels.

2.3.3 Description of the Autistic Spectrum Disorders and Mental Retardation

For a long time the ASD's were described mainly as a set of clustered behaviours. Extensive research is still ongoing at different levels, namely the neurobiological, the

cognitive and the behavioural levels. However we have to take into account the fact that the majority of the studies investigated a population with the Autistic Disorder without MR. Below we will give a short summary of the state-of-the-art in research into both disorders.

2.3.3.1 The Biology of the Autistic Spectrum Disorders and Mental Retardation

Since the first systematic twin study carried out by Folstein & Rutter (1977) and research into the characteristics of the parents of autistic children (Freeman et al.; 1978), a bulk of information has been gathered on the biological origin of autism. So far, this has led to the conclusion that the ASD's are multifactorially determined biological disorders (Gillberg, 1999).

A strong genetic link has been established (Rutter, Bailey, Bolton & LeCoulteur, 1993; Bailey, Philips & Rutter, 1996), the heritability of an underlying liability to autism is estimated to be more than 90%. However, this liability involves not only the ASD's, but also a much broader spectrum of social and communication problems (Rutter, 2000). Kraijer (1998) has stated that for those with ASD and MR, the cause of the intellectual disability often coincides with the cause of the ASD. The lower the level of functioning, the higher the percentage of ASD's and the greater the number of known causes (Kraijer, 1998). Gillberg (2001) states that in 1:4 cases of Autistic Disorder an associated medical disorder, such as tuberous sclerosis or a chromosomal disorder, has been diagnosed. As far as abnormalities in the structure of the brain are concerned, strong associations have been found with underlying diseases: e.g. increased brain size in tuberous sclerosis, Sotos syndrome and neurofibromatosis. EEG abnormalities have been reported in over 50% of autistic individuals. The finding of epilepsy is closely related to the level of functioning. Within the population with syndromes associated with the more malignant forms of epilepsy (e.g. West syndrome, Lennox-Gastaut syndrome) a high prevalence of ASD's has been reported (Gillberg, 1991). Obstetric complications are considered to be another risk factor for severe psychopathology. Eaton, Mortensen, Thomsen & Frydenberg (2001) found a significant association between the presence of ASD's and lower birth weight as well as prematurity. They also found the age of the mothers and the number of children per mother to be significant risk factors. A more recent development in the search for the underlying problems of autism occurred in the field of physiology. Parents reported numerous additional health problems, such as bowel problems, eczema, asthma and migraines. These findings have led to the hypothesis of an 'immune connection' (Shattock & Whiteley, 2002). Whether these problems contribute to the symptoms of autism or are even the cause is still widely debated.

Concerning the biology of Mental Retardation, historically the aetiology of MR was divided into two groups (Dykens & Hodapp, 2001):

- 1) familial or socio-cultural MR, for which there is no apparent cause of the MR
- 2) organic MR, with a known pre-, peri- or postnatal cause.

The percentage of known genetically related conditions involving MR is rising all the time, due to increases in technology (Dykens & Hodapp, 2001). Two other

causes of MR are perinatal complications and environmental factors. On the association between the latter and the ASD's much less information is available.

2.3.3.2 Cognition in the Autistic Spectrum Disorders and Mental Retardation

On the cognitive level children with MR and children with an ASD differ markedly. In children with an ASD development in the various domains is scattered by definition. Rutter (1979) was the first to acknowledge specific strengths and weaknesses in the cognitive functioning of children with an ASD. Carter et al. (1998) have shown strengths and weaknesses in adaptive functioning. They even state that children with an ASD can be distinguished from children with other developmental disorders due to their relatively low score for the domain socialization on the Vineland Adaptive Behavior Scales, the relatively high score for daily living skills and the high scores for the maladaptive behaviour domain (Gillham, Carter, Volkmar & Sparrow, 2000). Baron-Cohen (1989), in his studies on Theory of Mind (ToM), has shown that children with an Autistic Disorder score significantly lower for ToM tasks than their peers with MR and the same developmental age. In the following paragraph we will discuss the specific cognitive deficits of children with an ASD.

These studies have proven that the cognition of children with an ASD does not follow the same developmental path as in normal children. Research into the development of cognition in children with MR is however not conclusive. Hodapp has described the development of children with MR by means of two hypotheses, namely the 'similar sequence hypothesis' and the 'similar structure hypothesis' (Hodapp, 1997, p. 192-193). Dykens & Hodapp (2001) state that the similar sequence hypothesis holds up reasonably well for both non-specific and organic types of MR, whereas the similar structure hypothesis has proven to be inaccurate for certain specific genetic aetiologies.

Kraijer & Plas (2002) conclude that both the developmental theories expounded by Zigler (1984) and the deficit-theories (e.g. Ellis) hold. The deficits occur at the 'molecular' level, whereas intelligence is such a molar entity that a deficit in one of the building stones of intelligence will lead to a decrease in general intelligence, hence Mental Retardation. The description of Mental Retardation in the DSM-IV-TR (APA, 2000) is a behavioural description and we will use that definition in this study. This does not imply that research into the cognition of the broad spectrum of people with Mental Retardation is not of major importance and we agree with Dykens (2000), who states research on this field should be carried out on aetiological subgroups.

2.3.3.3 Specific cognitive deficits in the Autistic Spectrum Disorders?

Hermelin & O'Connor (1970) were the first to acknowledge that a basic cognitive deficit might be the core problem of the Autistic Disorder. Prior & Ozonoff (1998), in their overview of the psychological factors of the ASD's, name the following areas of cognition disrupted by problems: sensor motor and perceptual development, attention, general intellectual functioning, academic functioning, splinter abilities, memory, emotion perception, Theory of Mind and executive functions.

Research into the origin of these cognitive problems can be divided into two groups. One group has been looking primarily into the social cognitive deficits. Baron-Cohen, Leslie & Frith (1985) explain the social and communication problems in the Autistic Disorder by the Theory of Mind (ToM) hypothesis. Strong correlations have been found between lack of ToM and abnormalities in social behaviour (Howlin, 1997) and pragmatic competence in communication (Eisenmajer & Prior, 1991). However in a review of literature on 'autistic features' in very young children, Vig & Jedrysek (1999) have stated that deficits in verbal communication and symbolic play cannot distinguish preschool children with autism from preschool children with MR; the same can be said for behaviours that involve a more complex organisation of behaviours, e.g. resistance to change and insistence on routines. Impaired socialization is found to be the best differentiating factor between children with MR and children with an ASD in combination with MR at a very young age. However, ToM does not occur before the age of four. Klin, Volkmar & Sparrow (1992) have even suggested that ToM-deficits seem to reflect developmental age and are therefore not a distinctive feature of autism rather of a cognitive delay. Because Mental Retardation is considered a cognitive delay, this is not a distinguishing feature.

Another explanation for the social cognitive deficit in the Autistic Disorder is the Personal Relatedness Perspective. Autistic children have different preferences when they look at people's faces (Hobson 1987; Hobson, Ouston & Lee, 1988) and listen to voices (Klin, 1991). Klin, Jones, Schultz & Volkmar (2003) propose a model using the enactive mind that explains the wrong-going of social cognition as the result of a reduced awareness of social stimuli and the focus of attention towards socially irrelevant stimuli. People with an ASD seem to pay more attention to the separate components than the overall configuration (Bailey et al. 1996), especially with regard to social stimuli. The results of experiments to test the ability of autistic children to recognise emotions (Hobson, 1986), represent a renewed view of autism as a defect in social relatedness. Hobson describes the Autistic Disorder primarily as an affective and interpersonal impairment (1989). He argues that the relationship of the autistic child with its caregivers is so disturbed that the child does not experience personal relatedness and as a result does not learn to understand other people. In other words, they do not acquire a concept of mind, because of an interpersonal impairment very early in the socio-emotional development. They display this as an inability to use protodeclarative gestures, e.g. pointing and showing things to others (shared attention). According to this developmental path they also have difficulty understanding 'I-You' relatedness and developing a concept of 'self' (Hobson, 1990).

Where the ToM explains the triad of impairments in the ASD's as a 'mentalization' deficit, the Personal Relatedness Perspective (PRP) explains these deficits in emotional recognition, the pragmatic deficits and the deficit in symbolic play as a lack of the innate ability to interact emotionally (Lucangeli, 1997). Several researchers have pointed out that joint attention seems to be a fundamentally social and affective deficit, because it occurs so early in life (Vig & Jedrysek, 1999). Both theories, however, only explain a proportion of the triad of impairments.

A second group of researchers has been looking into a broader concept of cognition, namely the general difficulties in high-level planning and control of behaviour (Bailey et al. 1996). Ozonoff (1995) implied that people with an Autistic Disorder have a deficient executive functioning (EF). Problems with overall EF are, however, not specific to the ASD's. Children with MR also exhibit problems with some executive functions, such as attention and generalization skills. The short span memory often seems to be intact in those with MR (Hodapp & Zigler, 1995). Executive functioning is an umbrella term; not all aspects have yet been found in those with ASD (Bailey et al., 1996), but studies have been carried out on the specificity of some aspects of EF in the ASD's, namely: working memory, inhibition and cognitive flexibility (Rogers & Benetto, 2000). As far as working memory is concerned the studies have as yet not provided us with a clear answer to the question whether problems with working memory capacity are specific for the ASD's or a sign of cognitive delay in general. Therefore for this specific point research is not at all conclusive about the differences between the ASD's and MR.

Several developmental disorders (e.g. ADHD, OCD) are associated with impaired inhibitory mechanisms. Some researchers have suggested that people with an Autistic Disorder perform similar to matched controls on simple inhibition tests, but have more problems with shifting attention (Ozonoff, 1994; Rogers & Benetto, 2000). Such problems with cognitive flexibility could explain the specific behaviour problems (e.g. resistance to change) of people with an ASD.

So, although problems with EF are not specific to the ASD's, research into some of the specific domains could provide information which could explain some of the specific deficits in the ASD's, especially concerning the problems of inflexibility.

Frith (1989; 2003) has presented another fruitful hypothesis concerning the cognitive deficit in the ASD's, especially the non-social aspects. She states that people with an ASD have a weak drive for central coherence. People with an ASD experience the world in fragments and are not able to see the parts in relation to the whole. Weak drive for central coherence not only explains poor performance of certain tasks but also superior performance of tasks like 'hidden figures' and 'block design' (Shah & Frith, 1983; Happé, Briskman & Frith, 2001). These integration problems affect all senses. Happé (1999) has even called it 'a cognitive style', rather than a 'cognitive deficit'. Thus far this theory has been regarded as the most specific for the ASD's.

In our view combining the four different theories provides us at present with the most comprehensive cognitive model to explain the triad of impairments (Lucangeli, 1997; Gillberg, 1999). In the light of challenging behaviour, the Personal Relatedness Perspective and to a lesser extent (because of the level of functioning) the Theory of Mind hypothesis present some useful explanations of the problems with socialization, whereas the Central Coherence Theory seems crucial for explanation of the problems with communication (Noens & van Berckelaer, 2002). The Executive Functions theory offers the most useful hypothesis for explanation of specific behaviour problems due to a lack of flexibility.

2.3.3.4 *The behavioural level of the ASD's and MR*

The DSM-IV-TR (APA, 2000) gives a description of the behavioural manifestations of the Pervasive Developmental Disorders and Mental Retardation (see Appendix A). In the previous paragraph we defined MR as a delay in cognitive functioning, with delays in adaptive functioning as a consequence and the ASD's as a specific cognitive deficit (Van Berckelaer-Onnes, 1996), which leads to the triad of impairments (Wing, 1993b). The associated behaviours, such as resistance to change, are considered a consequence of the triad of impairments.

In the paragraph below we will describe the triad of impairments with regard to the behaviours of children with an ASD in combination with MR throughout childhood.

Impairments in social interaction

Both Kanner (1943) and Asperger (1944) were struck by the severe social impairments in the boys they described. The problems in social interaction of people with an ASD represent the most severe handicap. Wing (2001) has described the social behaviour of babies, later diagnosed with an ASD. One group of babies seems to develop normally, a second group of babies is described as very squeamish and parents feel they are unable to soothe them. The third group of babies is the largest; they are often referred to as 'angel babies' (Wing, 2001, p. 12). In retrospect parents of the babies of the last group probably found them to be too easy.

Recent studies have focussed on attachment behaviour of children with an ASD. One of the first studies was carried out by Sigman and Ungerer (1984). Rutgers, Bakermans-Kranenburg, Van IJzendoorn and Van Berckelaer-Onnes performed a meta-analysis of 16 studies on attachment and autism (submitted). They found that approximately 50% of children with autism showed signs of attachment security in contrast to 65% in a normative sample (Van IJzendoorn, Goldberg, Kroonenberg & Frenkl, 1992). Rutgers et al. (submitted) also found that an increase in mental age has a positive effect on attachment behaviour. Therefore children with an ASD and MR show even less attachment security.

Because sample-sizes were too small the authors were not able to answer the question whether disorganized attachment is over-represented in samples of autistic children. Disorganized attachment is an important predictor for externalizing behaviour problems (Van IJzendoorn, Schuengel & Bakermans-Kranenburg, 1999). However, the current methods to assess attachment are hardly suitable for children with an ASS. The 'strange situation' is by definition a problem for children with an ASS.

When the babies grow up, the impairments in social interaction become more apparent. Wing (1996) describes three subtypes that occur in children with an ASD and MR, namely:

- the aloof type: children who belong to this type do not initiate and react to social interaction; they seem unresponsive to people and only interact to satisfy their basic needs;
- the passive type: children of the passive type do respond to social interaction, but do not initiate social contact; children of this type are willing to do as they are told and are considered 'easygoing';

- the active but odd type: children of this type initiate contact but the interaction is characterized by a lack of reciprocity and can be characterized as 'one-way'. Children of the active but odd type are often considered difficult and very demanding.

Children with an ASD and MR are more often of the aloof or passive type than high functioning children. There are no clear cut-off points between the groups and during development the individual with an ASD can shift from one subgroup to another. This sub-grouping also shows that the nature of the social impairment can differ immensely.

Because of the deviant developmental pathway, especially in social development, adolescents with a disorder in the autistic spectrum often fail to achieve full understanding of emotions, lack the ability to engage in reciprocal relationships in adulthood and also fail to reach full development of a conscience (Howlin, 1997).

Impairments in communication

All children with an ASD exhibit communication problems. The communication problems of people with an Autistic Spectrum Disorder become manifest in both the verbal and non-verbal components of communication (National Research Council, 2001). Gillberg (1999) has stated that the nonverbal communication deficit, as a result of impairment of mentalizing abilities, seems to be of more importance than the specific language problems. Children with autism seem to lack the capacity to acquire joint attention and symbol use (Wetherby, Prizant & Schuler, 2000). The lack of joint attention manifests itself in difficulties in orienting and paying attention to the communication partner, shifting and following gaze and drawing attention (National Research Council, 2001). Noens & Van Berckelaer (2002) state that the communication of people with autism and MR is characterized by the lack of intentionality and symbol use.

The level of communication and speech of people with an ASD determines to an important extent the predictive outcome (Wetherby et al., 2000; Howlin, Mawhood & Rutter, 2000). The National Research Council of Canada even states that development of communicational competence has proven to be the main predictor of outcome (2001).

The development of communication in children with an ASD undergoes problems from very early childhood. Babies with an ASD exhibit less variation in their babbling patterns and sounds than children with language disorders or MR (Wetherby, Prizant & Hutchinson, 1998). Their sounds are much less intentional and the reactions of parents do not become predictable to them. As mentioned before, children with an ASD have great difficulty acquiring joint attention behaviours as a result of this lack of intentionality. These include pointing in order to request objects (proto-imperative), and showing and giving objects to others to share interests (proto-declarative) (Lord & Magill, 1989). This deficit in the use of communication to acquire joint attention is considered to be a very important variable in the differential diagnosis between autism, specific language disorders and MR. Wetherby et al. (2000) even suggest that this failure in joint attention might be the core deficit in autism. They found specific scatter for children with an ASD. They

used similar amounts of communication utterances to request and protest but much less to change gaze and share attention. There were also fewer conventional gestures and less communication to achieve joint attention (Wetherby et al. 1998). Because of their lack of joint attention children with an ASD often do not reach the referential level. Therefore communication is mainly used to satisfy their needs.

Children with an ASD have great difficulty acquiring conventional and symbolic communication. Although the percentages differ, it is estimated that about 30-50% of those with an ASD never develop any useful speech (Wetherby, et al., 2000) and are called functionally mute. About one in four people with an ASD remain totally mute (Wing, 1996). Speech is only one of the means to communicate. Other means that are often used with children with an ASD are objects, photographs, pictograms and gestures. Children with an ASD do not develop alternatives spontaneously, as do children with language disorders or auditory problems (Wetherby et al., 2000). They mainly use primitive motor gestures, such as taking the other by the hand (NRC, 2001).

The group that does develop speech displays a variety of unusual features like echolalia, pronoun reversal, idiosyncrasies, neologisms, metaphorical language, and literal use of language (Van Berckelaer-Onnes, 1997). Wetherby et al. (2000) suggest that echolalia is a strategy used by children with an ASD to acquire language through linear repetition to produce an anticipated and learned response. Children with an ASD also have problems with language systems: language form (phonetics, prosody, and syntax) with semantics and pragmatics. Even high-functioning autistic persons have language problems, in particular with the pragmatic aspects (Fay & Schuler, 1980; Tager-Flusberg, 1989). The level of understanding is often much lower than expected, language is often only understood in familiar circumstances. The child with autism does not so much react on the language per se, but on daily routines or other cues. This poses an immense risk for over-demanding (Noens & Van Berckelaer, 2002).

Lots of people with an ASD use challenging behaviour as a means to communicate (Wetherby et al., 2000). Carr & Durand (1985) state that challenging behaviour might be the result of the lack of symbolic capacity, in other words a failure to reach the representational level. Teaching people with an ASD to communicate, with whatever means, helps diminishing challenging behaviour (Van Berckelaer-Onnes, Dijkxhoorn & van der Ploeg, 1996).

Impairments in imagination

The failure to develop joint attention and symbolisation also influences the development of imitation and play. Children with an ASD show significantly less imitation than their peers with MR (Rogers & Benetto, 2000). Children with an ASD do not develop symbolic play spontaneously. Their play is repetitive and the main goal often seems to be self-stimulating; e.g. spinning with objects, hitting two objects together etc. (Van Berckelaer-Onnes, 1994). Consequently lack of imagination leads to a rigid pattern of behaviour and lack of understanding others. Therefore, although this impairment in imagination is most striking in children, it remains a problem through life.

McCune Nicolich (1981) described the following stages in the development of play:

- simple manipulation;
- combination play;
- functional play;
- symbolic play.

Children with an ASD show very limited manipulation of objects. Their toy-play behaviour is very weak. They focus on one or two parts of the toy and do not further explore the toy. The chance of further developing their play, above the level of Simple Manipulation is therefore limited (Van Berckelaer-Onnes & Kwakkel, 1996). The stereotypical and perseverating actions are also visible in their combination play. They can be taught functional play, but they do not vary by themselves. Van Berckelaer-Onnes (1994) shows that they can learn functional play, but the quality is much less. Williams, Reddy & Costall (2001) found that autistic children show far less variety in their functional play than their developmental age controlled peers with MR. There is a strong relationship between functional and symbolic play skills and receptive and expressive language (Mundy, Sigman, Ungerer & Sherman, 1987). In contrast, children with an ASD do not show a delay in non-social constructive play (e.g. Lego). They can be very skilled at this (Wetherby & Prizant, 1998).

The lack of play and the lack of imagination in children with an ASD pose a serious threat to their development. As Lorna Wing (1996, p. 99) stated: 'The value of true imagination and creativity is in associating past and present experiences and making plans for the future, ranging from the mundane what to do tomorrow to the grand plans for the whole of life'.

2.3.3.5 *Associated Behaviour*

People with an Autistic Spectrum Disorder display a whole range of behaviours, related to the triad of impairments. Rutter (1985) has divided the observed problems into:

1. specific behaviour problems; defined as a direct result of the Autistic Spectrum Disorder, for example rigid patterns of behaviours, obsessions, ritualistic behaviours;
2. non-specific behaviour problems; those are not a direct result of the Autistic Spectrum Disorders but people with this disorder show a high percentage of these problems (e.g. sleeping problems, aggression, self injurious behaviours).

The specific behaviours, for example resistance to change, often occur very early in life. Parents of a child with an ASD often describe problems with sleeping routines, problems with the transition from bottle to spoon etc. Bodfish, Symons, Parker & Lewis (2000) have presented a study in which they compare repetitive behaviour in autism and MR. They found that repetitive behaviour is not specific for the ASD's, but the elevated pattern of occurrence and severity is. A similar finding has been described by Berkson, Tupa & Sherman (2001) in a study about the incidence of Self Injurious Behaviour of children with MR that under the age of three. They found eye poking and eye pressing almost exclusively in children with visual impairments and

complex hand movements (e.g. flapping) were more common in socially unresponsive children. These behaviours seem to be specific for the two different aetiologies.

Many of the non-specific behaviours seem to start at a later time, mainly because of a lack of communication (e.g. temper tantrums, self injurious behaviours).

Because not all the behaviours are clearly related to the triad of impairments we propose a division into three categories namely:

1. specific behaviours: behaviour that is a direct consequence of the triad of impairments (e.g. resistance to change, temper tantrums because of lack of communication);
2. related problems: behaviour that has a relation with the ASD (e.g. feeding problems as a result of hypersensitivity of the mouth);
3. additional or co-morbid problems.

In conclusion we find that people with Autistic Spectrum Disorders show numerous problems on all aspects of behaviour. Some are characteristic for the disorder and are used as criteria to classify the disorder (e.g. stereotypical behaviours, rigid patterns of behaviour), others are considered associated problems. The nature of the associated and additional problems is still unclear. This is one of the focuses of this research.

2.3.4 Prevalence of the Autistic Spectrum Disorders in combination with Mental Retardation

Statistics about the prevalence of the ASD's depend on the chosen definition. Almost all of the prevalence studies have been focussing on the prevalence of the Autistic Disorder itself and less on the other classifications. Fombonne carried out a meta-analysis on epidemiological studies of the AD. He found prevalence estimates ranging from 0.7 to 15.5 per 10.000 with a median of 4.8 (Fombonne, 1998). So even for the most strictly defined classification, the numbers vary immensely. In the study carried out by Wing & Gould (1979) on the prevalence of the 'triad of impairments' a ratio of 22 in 10.000 children was found. In 1991 Gillberg and his group found that 36 of the 10.000 7-16 year old in mainstream schools, fitted his criteria for Asperger's disorder. Recent reports of a rise in prevalence of the non-autism PDD's have surfaced. However, as long as different diagnostic criteria are used, comparing the data on prevalence is a difficult task. The recent attempt by Fombonne (1998) who compared data on the classification Autistic Disorder, showed that a rise in incidence could not be verified. Research into the prevalence on the broader category of PDD is still scattered and not definite. For example, a recent survey in Staffordshire, England (Chakrabarti & Fombonne, 2001) resulted in an estimated prevalence of 62.6:10.000 in this specific broad area, which is an immense figure that has to be explored further.

The estimation of the prevalence of MR is ranging from 1-2% of the population (Hodapp & Dykens, 1996). This prevalence rate is also depending on the definition

used by the researchers. As far as the author knows, no prevalence rates have been described yet, using the newest criteria.

The prevalence of the ASD's among people with MR varies across the level of functioning. The prevalence found among people with mild MR ranges from 1.8-25%; the prevalence found among people with moderate to profound MR however, ranges from 29.5-56.4% (Kraijer, 1998).

These prevalence rates are extremely high, and demand further elaboration. Such high prevalence rates are suggestive of considering MR as part of the ASD's. In the following we will elaborate on this subject.

2.3.5 Autistic Spectrum Disorders and Mental Retardation; co-morbidity?

To answer the question whether MR is a feature of the ASD's, or whether they are two separate disorders, we are obliged to look into the subject of co-morbidity.

Simonoff (2000) has described eight reasons why co-morbidity may occur, these are:

1. chance: it is possible to calculate the chance that two disorders occur together in the normal population. Co-morbidity only occurs at a greater than chance level, if the product of the frequencies of the single disorders is smaller than the prevalence of the co-morbid state;
2. sampling Bias: in the study of co-morbidity sample bias should always be considered. Bias could occur because people with a co-morbidity of disorders are more likely to search for treatment;
3. population Stratification: when searching for co-morbidity in a certain population, one risk factor could attribute to an increase in different disorders;
4. symptom overlap: two disorders can share symptoms;
5. correlated error variance: especially if the same person is rating both disorders, an association that is only based on an error of measurement may occur;
6. a distinct group: the two disorders should be viewed as a distinctive category, because the combination of the two disorders differs distinctly from the two single disorders, e.g. co-morbid conduct disorder and depression as opposed to depression not associated with conduct disorder (Volkmar & Woolston 1995);
7. shared risk factors: if the risk factors of two disorders overlap, the chance of co-morbidity rises;
8. phenotypic causality: this occurs if one disorder alters the risk for the second one, or reciprocally, e.g. the presence of Tuberosis sclerosis and epilepsy.

We will describe the factors that may contribute to the high prevalence of the co-occurrence of ASD and MR following the above-mentioned points.

Ad 1) Chance: if we calculate the chance of co-morbidity in the case of ASD and MR the chance of the two occurring together is according to the most prudent prevalence rates: 5:100.000 ($1/400 \times 2/100 = .00005$). In reality 75-80% of the people with an AD also have MR (Van Berckelaer-Onnes & Van Engeland, 1997), and the

prevalence of the ASD's among people with MR is estimated to be 1:5 (Kraijer, 1998). So the co-morbidity of the ASD's and MR is at least 4000 times higher than might be expected from chance alone.

Ad 2) Sampling Bias: the prevalence rates we have been discussing for the ASD's and MR are gathered through general population studies, so sampling bias can not explain the high prevalence of co-morbidity.

Ad 3) Population Stratification: when searching for co-morbidity in a certain population, one risk factor could attribute to an increase in different disorders. When we look at the biological factors, especially among the chromosomal disorders and the neurological causes of MR, we have to take population stratification into account. Especially in the more retarded subjects, the cause of the brain damage underlying MR, could well be the cause of the ASD (Bailey et al., 1996; Gillberg & Coleman, 1996; Kraijer, 1998). So one biological factor, could cause both disorders.

Ad 4) Symptom overlap: two disorders can share symptoms, e.g. delayed development in AD and MR. Only if there are differences in aetiology, response to intervention and outcome, separate classification is considered. As mentioned in the description of the two disorders, the ASD's and MR can clearly be distinguished. In people with a developmental age below 12 months, the distinction is much more difficult to make, due to a lack of social, communicative and imaginative behaviours (Došen, 1990; Mahler, 1965). The high incidence rates (over 40%) as mentioned by for example Kraijer (1998) in people with profound MR should in this light be prudently interpreted.

Ad 5) Correlated error variance: especially if the same person is rating both disorders, an association that is only based on an error of measurement, may occur. As mentioned before, the prudent way in which population studies have been carried out, should discard this argument.

Ad 6) A distinct group: the two disorders should be viewed as a distinctive category, because the combination of the two disorders differs clearly from the two single disorders. Most of the research into the ASD's has been carried out on High Functioning Autism (HFA), so without Mental Retardation. If we consider differences in aetiology, response to intervention and outcome, one could argue that HFA is distinct from the AD with MR (often called Low Functioning Autism or LFA). Outcome studies (for example Howlin & Goode, 1998; Rutter, 1985) have repeatedly shown that high IQ is a predictor for a more positive outcome. Rapin (1996) has also shown that HFA and LFA can be clearly distinguished regarding social and communicative behaviours. The argument of Frith and Happé (1998) that the delay in cognitive abilities is the consequence of the specific cognitive deficit, and therefore not co-morbid, undermines the notion that HFA and LFA are two distinct groups. However, it is questionable whether the arguments they pose, such as spiky IQ profiles, intact memory and normal scores on Inspection Time tasks, do apply to children with LFA. Furthermore the ASD's can also be diagnosed in children with levels of functioning that make the tests they describe, not applicable.

Children with an ASD in combination with MR show very different performances on the different domains and therefore very different scatters both in comparison with children with only MR and children with only ASD.

Ad 7) Shared risk factors: if the risk factors of two disorders overlap, the chance of co-morbidity raises, e.g. perinatal toxin-exposure. This so-called 'two-hit model', does not explain however the different prevalence rates among various causes of LFA (Frith & Happé, 1998).

Ad 8) Phenotypic causality: this occurs if one disorder alters the risk for the second one, or reciprocally, e.g. the presence of Tuberosis sclerosis and epilepsy. This argument could explain the high prevalence of ASD in some syndromes, although there is as yet neither a syndrome with a co-morbidity of 0%, nor one with a co-morbidity of 100% (Kraijer, 1998).

Concerning the behavioural and the cognitive levels the ASD's and MR are two different disorders. On the biological level the picture is still unclear. On the basis of the above-mentioned points it seems plausible to assume that the ASD's and MR are two separate disorders and the ASD without MR (HFA) differs from the ASD's with MR. Although the information is not complete on this issue, it seems plausible that phenotypic causality and shared risk factors could explain the high prevalence of the ASD's among people with MR.

Whether challenging behaviour differentiates between children with an ASD and MR and children with MR will be discussed in the next paragraphs.

2.4 Challenging behaviour of children with an ASD and/or MR

Challenging behaviour and psychopathology commonly occur in children with MR. One of the first epidemiological studies that made people aware of this increased risk, has been carried out by Rutter, Graham and Yule (1970) on the Isle of Wight. In the last 30 years several authors have found similar results (Benson, 1985; Bruininks, Hill & Morreau, 1988; Došen, 1990; Dykens, 2000; Dykens & Hodapp, 2001; Einfeld & Tonge, 1996a; 1996b; Volkmar & Woolston, 1995). The risk is estimated to be four to five times higher in people with MR than in people with normal intellectual capabilities. This estimation leads to a prevalence rate of additional psychopathology of between 30-50% (Došen, 1990; Emerson, 2001; O'Brien, 2000). Dykens & Hodapp (2001) state that as a rule of thumb 25% of the people with MR have significant psychiatric problems and the percentage of behaviour disorder is much higher. There are some indications that the prevalence is lower in the group with familial MR (Strømme & Diseth, 2000).

Romanczyk, Weiner, Lockshin and Ekdahl (1999) state that challenging and maladaptive behaviours in people with an ASD present huge challenges to service providers. These are often the behaviours that prevent these people from integrating in society and the behaviours that pose a threat to the education of children.

There is very little valid information on the prevalence of challenging behaviour in the MR group. Only a few population studies have been carried out. Most studies

look at a specific form of challenging behaviour (e.g. aggression), or at a specific group (e.g. adults with MR living in an institution) (Emerson, 2001). Very often challenging behaviour and psychopathology are not viewed separately. The problems in the study of co-morbid disorders that are mentioned previously, also apply to studies of challenging behaviour in the MR group. Other methodological sources of variability in the reported prevalence of co-morbid disorders in people with MR are according to Dykens (2000, p.408):

1. differences in IQ level;
2. different types of samples, e.g. outpatient, inpatient, epidemiological. Lots of the studies have been carried out in institutionalised adult populations, which are not representative for the entire population (Murphy, Hall, Oliver & Kissi-Debra, 1999; Emerson, 2001);
3. mixed etiologic groups;
4. different measures of psychopathology, e.g. measures assessing psychiatric illness and measures assessing maladaptive behaviours;
5. differences in diagnostic systems, e.g. DSM, ICD and different versions over the years;
6. different criteria for 'caseness', e.g. cut-off points, psychiatric diagnoses.

Dykens & Hodapp (2001) have stated that even though many professionals lack the knowledge to diagnose psychiatric disorders in people with MR and as a consequence diagnostic overshadowing is still a major issue, one of the most robust research findings in the field of MR in the last decade is the increased risk for psychiatric illness.

Another problem is that the challenging behaviour is often not specified. In most descriptions of the occurrence of challenging behaviour of people with MR, the presence of an ASD is not taken into account. There are, however, a few studies, that verify the assumption that challenging behaviour does occur more often in people with an ASD in combination with MR. For example, Didden, Duker and Korzilius (1997) published a meta-analysis on the effectiveness of treatment on problem behaviours. In this study they studied the aetiology of problem behaviours, including the presence of autism. In 152 of the 435 comparisons made there was an association with autism and in 20 with autistic-type. Kraijer (1998; 2000) has extensively researched the challenging behaviour of people with ASD in combination with MR using two screening instruments. The first gathers information about autistic behaviours (AVZ-R; Kraijer, 1994), the second gathers information about challenging behaviour (SGZ; Kraijer & Kema, 1994). Kraijer concludes that people with an ASD in combination with MR have a significantly higher risk of developing challenging behaviour than their peers with only MR do. This is especially true for stereotypical behaviours, masturbation, self-injurious behaviours, rumination, hyperactive behaviours, fears, sleeping problems, stealing food and pica (Kraijer, 1998). Dekker (1999) has also found that people with an ASD in combination with MR showed more problem behaviours than the group MR without an ASD. Not surprisingly, they do display more specific behaviour problems, but also more non-specific problems e.g. self-injurious behaviours, pica, destructive behaviours, hyperactivity, sleeping problems and fatigue.

In addition, evaluations of the advisory teams have shown an overrepresentation of people with an ASD in the referred clients with extreme challenging behaviour (de Vos, Buysse & Van Berckelaer-Onnes, 1996).

The above-mentioned amounts are not specific for children. Dunlap, Robbins, and Darrow (1994) interviewed parents about their perceptions of the challenging behaviour of their child with an ASD. They found that the parents reported a high frequency of challenging behaviour, when high is defined as more than once a day. The most frequently named were self-stimulation (61%) and destructive behaviour (40%). Volkmar et al., (1987) published a study in which they show that children with an ASD and MR score significantly higher on the Maladaptive Domain of the VABS (Sparrow et al., 1984) than the control group with only MR. Murphy et al. (1999) have carried out a study on the prevalence of potential self-injury and found that parents and caretakers were worried about self-injurious behaviour in 25% of the children with MR under 10 years of age. They found gender and sensory deficits and the presence of other behaviour problems and the degree of autism markers for the risk of self-injurious behaviour, although they acknowledge that these markers are not sufficient to predict the risk.

Stenfert Kroese & Fleming (1993) published a study on the prevalence of challenging behaviour in 5-14 year old with MR. They found 42.1% to have any form of challenging behaviour; 22% showed physical aggression. An even more important finding of this study is that the group with challenging behaviour still displayed these behaviours in the follow-up years. They state that occurrence of challenging behaviour in the early years is a very strong predictor for sustaining challenging behaviour through the lifespan. We did not find any indication why this should be different for children with MR and an ASD.

Summarizing: children with an ASD in combination with MR show both specific and non-specific behaviour problems. If challenging behaviour in the ASD's is considered a result of an inability to cope with the environmental demands and physical discomfort, as well as the result of disturbed neurochemical and neurophysiological functions (Tsai, 1999), it seems only logical that children with both an ASD and MR display an increased risk of developing these behaviours. However, these problems are not considered very problematic in all of the children and by all of the caretakers. In some children with an ASD and MR however, the challenging behaviour has already become almost unmanageable at a very young age. This group is often not specified in literature, because often just the frequency and not the severity of the problems have been taken into account. The prevalence rates of the challenging behaviour and the raising number of children referred to the regional advisory teams, bundled with the knowledge that these behaviours sustain through the lifespan, are strong arguments for the need for further research into this group.

There is little doubt about the increased prevalence of challenging behaviour, but there is still very little consensus about the reasons why people with MR have an increased risk and there is even less information on the protective factors.

2.5 Possible causes of challenging behaviour: models of psychopathology

Dykens & Hodapp (2001) have made an overview of recent insights into the risk factors of psychopathology in people with MR. They name:

- 1) distinctive aberrant personality styles;
- 2) atypical motivational styles;
- 3) increased risk of failure experiences;
- 4) more global and less differentiated self-concepts;
- 5) reinforcements of negative behaviours;
- 6) poor communication or assertiveness skills;
- 7) social strain;
- 8) social stigma;
- 9) peer rejection and ostracism;
- 10) comprised 'social intelligence';
- 11) heightened risk of exploitation and abuse;
- 12) family stress, parental psychopathology;
- 13) increased rates of seizure disorders;
- 14) abnormal neurological functioning;
- 15) high rates of sensory or motor impairments;
- 16) biochemical or neurological anomalies.

There are a variety of theories that provide models on the aetiology of challenging behaviour. Wenar & Kerig (1999) name the following: the medical model, the behavioural model, the cognitive model, the psychoanalytical model and the family systemic model. None of these models are specific for children with an ASD in combination with MR, but they can all provide explanations for the emergence of these problems in this group. The above-mentioned list of risk factors by Dykens & Hodapp also consists of factors derived from different theoretical models.

In the following we will describe the relevance of these models with regard to the explanation of challenging behaviour in children with an ASD and MR. We conclude with a proposal for an integrated model.

a) The medical model

The medical model will be described first. The model consists of two components. First the model assumes the general hypothesis that certain psychopathologies result from organic dysfunction. Second this model has provided a classification of psychopathological behaviours in terms of diagnostic entities.

As mentioned before, the aetiology of the ASD's is thought to be biological in origin. Biological problems are often contributing and facilitative for the occurrence of challenging behaviour. Swets (1995) names six medical causes for challenging behaviour:

- General medical causes: pain, itch, etc. can be causes of behaviour problems especially in those children who are not able to verbalize their discomfort. Research has shown that stereotypical and self-injurious behaviours can be the result of minor illnesses (e.g. dermatitis and otitis media) (Howlin, 1998).

The finding of Konstatareas & Homatidis (1987) that children with autism may suffer more frequently from ear infections only stresses the need for a medical screening when a sudden behavioural change occurs.

Also important is the finding of Steffenburg (1991) that 40% of the children with autism have an associated major medical condition, 10% of the children have a hereditary form of autism, but the remainder of the cases have unspecific signs of brain dysfunction.

Another important factor is sleep problems. Brylewski & Wiggs (1999) have shown that people with MR and sleep problems show more challenging behaviour, especially irritability, hyperactivity and stereotypy. Kiernan & Kiernan (1994) have noticed that challenging behaviour is more likely to occur in people with MR who show disturbed sleep patterns.

- Metabolical causes: some metabolic diseases have a very strong association with specific forms of challenging behaviour, e.g. self-injurious behaviour in Lesch-Nyhan syndrome and Prader-Willi syndrome.
- Dysfunction in liquor-circulation.
- Epilepsy: before grand-mal-seizures behavioural changes are often noticed. In some cases epilepsy does not manifest itself in seizures, but in challenging behaviour, e.g. aggression, fears, repetitive movements etc. Matson, Bamburg, Mayville & Khan (1999) state that individuals with seizure disorders show behavioural excesses and deficits. The prevalence of epilepsy in the ASD's is estimated to be 1:3 (Van Berckelaer-Onnes & Van Engeland, 1997).
- Medicines: side-effects of medications, especially those used for seizure disorders, are known to cause dizziness, headaches, depression, lethargy, fatigue, anxiety, irritability, excitability, hyperactivity, akathisia and mania. All of these symptoms can exacerbate challenging behaviour (Gardner & Sovner in Gardner & Whalen, 1996).
- Food and stimulants.

In addition Emerson (2001) names:

- Gender: boys are more likely to show challenging behaviour than girls. The relationship between gender and challenging behaviour is even stronger for the more severe forms of challenging behaviour (Kiernan & Kiernan, 1994) specifically aggression and property destruction (Borthwick-Duffy, 1994) and in institutional settings (Quereshi, 1994). Self-injurious behaviour seems to be the exception to this rule. There is some evidence that suggests that some forms of self-injury do occur more in girls and women with MR (Emerson, 2001).
- Specific syndromes and disorders
- Level of intellectual functioning: Several studies (Borthwick-Duffy, 1994; Kiernan & Kiernan, 1994; Quereshi, 1994) have shown positive correlations between level of functioning and the occurrence of one or more forms of challenging behaviour.
- Additional disabilities; especially problems in vision and hearing (Kiernan & Kiernan, 1994), problems in communication (Borthwick-Duffy, 1994;

Kiernan & Kiernan, 1994) and impairments of mobility (Kiernan & Kiernan, 1994) are often associated with challenging behaviour.

Gillberg & Coleman (1996) have stated that there are behavioural subtypes, typical of underlying medical conditions associated with autism. They found gaze avoidance to be typical for boys with fragile-X and boys with hypomelanosis of Ito. Other behaviours, namely hyperventilation, bruxism and severe self-destructive behaviours are more frequently associated with extremes of low cognitive ability. Harris (1992) noted a very high prevalence of self-injurious behaviour (especially hand-wringing) in Rett syndrome.

In the literature about self-injurious behaviours often a 'gate-model' is proposed. People who experience pain or itch, often experience relieve through inflicting other pain themselves. Especially ear banging, scratching etc. could be caused by this phenomenon. Another theory concerned with the causation of self-injurious and self-stimulatory behaviour is the opioid-excess theory. Stress or pain release opioids from the brain, which can relieve the person from that pain. Self-injurious behaviour is in this theory explained by an addiction to these opioid-effects (Romanczyk, Weiner, Lockshin & Ekdahl, 1999).

In practice not often a clear causal relationship between challenging behaviour and organic problems can be established. However, the presence of any medical condition can lead to challenging behaviour. O'Brien (2000) has stated that the nature of the disability, in our case the presence of an ASD and MR, with a biological origin is a predisposing factor, and an occurring disease can be a precipitating factor. Together with the inability of children with an ASD and MR to verbalize their problems, we can imagine the starting point of a never-ending circle of problems, therefore biological factors can also function as perpetuating factors.

b) The behavioural model

The classic behavioural view on challenging behaviour assesses behaviour on the base of the Antecedence-Behaviour-Consequence model. Precise delineation of the behaviour that needs to be modified is proposed through identifying the antecedents and consequences of that behaviour. Behaviour modification is the therapy used to redirect the consequences, so other behaviours will stop occurring (Duker & Didden, 1996). However, in the ASD's the focus on the observed behaviour does not necessarily lead to the most appropriate form of treatment (Emerson & Bromley, 1995). Furthermore, finding the antecedents and consequences as perceived by the child with an ASD will often prove to be extremely difficult (Howlin, 1998). A change in the aim of research has lead to look at the function or the message in the behaviour of the child with an ASD. Durand & Carr (1991) have identified five main functions of aggressive, self-injurious, stereotyped, or other disruptive behaviours, namely:

- indicating the need for help or attention;
- escaping from a stressful situation or activity;
- obtaining an object;
- protesting against unwanted events/activities;

- obtaining stimulation.

Although this line of explaining challenging behaviour looked promising, a study by Emerson & Bromley (1995) has shown that in 25% of the cases no function could be established and that a third of the behaviours proved to be multicausal. The scales designed to establish the function of challenging behaviour (e.g. the Motivation Assessment Scale by Durand & Crimmins, 1988) have all proven to be unsatisfying as far as the reliability and validity are concerned.

Feldman & Griffiths (1997) describe an ecobehavioural approach towards the assessment and treatment of challenging behaviour in children with developmental disorders. They state that challenging behaviour is learned behaviour and that a wide range of contextual factors can influence them. The child is motivated to learn both adaptive and maladaptive behaviours through positive and negative reinforcement. Positive reinforcers are for example attention, tangibles (e.g. food, favourite toy) and sensory stimulation. Through an analysis of the setting events, the antecedents -the events that precede the challenging behaviour- and the consequence -the reaction to that behaviour- a plan to change the targeted behaviours can be made. In their view the setting events can be of a biological, an environmental and a pedagogical origin. They describe three levels on which information must be gathered to obtain a reliable ecobehavioural assessment. The first level focuses mainly on the history, the second two are meant to define current skills.

- **Level 1: Informant Interviews;**
 - the nature and definition of the behaviour problems
 - client and family history and current caregiver status
 - biological and medical assessments of behaviour disorders
 - structural analysis of current environments
 - motivational analysis
 - client reinforcers
- **Level 2: Observations in Natural Environment**
 - baseline
 - scatterplot
 - antecedent-Behavior-Consequence Observations
- **Level 3: Experimental Analogue Assessments**
 - structural analogue assessments
 - analysis of consequences

This approach is not specific for the ASD's, but is often used and adapted for this group. Lovaas introduced intensive behavioural intervention programs to increase adaptive behaviours and decrease non-adaptive behaviours in young children with autism (Lovaas & Buch, 1997).

Another adaptation of behaviour therapy towards 'autism and related developmental disorders' is the TEACCH method. Schopler c.s. call their method psycho educational. Challenging behaviour is explained through the iceberg-metaphor. In this metaphor the underlying cognitive deficits of the ASD's are below the waterline

and challenging behaviour is depicted as an iceberg (Schopler, 1989). In this view the challenging behaviour can be changed by adaptations in the environment towards the underlying deficits (Schopler, 1995). The main focus of the TEACCH program is the enhancement of communication skills and the adaptation of the environment towards the needs of the child with an ASD.

According to O'Brien (2000) behavioural theories function mainly as a perpetuating factor and the above-mentioned schemata, provide us with a means to assess predisposing and perpetuating factors.

c) Cognitive Models

Piaget is considered to be the founder of developmental psychology and his ideas have been applied to psychopathology. According to Piaget children develop through fixed stages, driven by a need to adapt to the environment. This adaptation takes place through assimilation -incorporating new information into an existing scheme- and accommodation -the alteration of a scheme to incorporate new information (Wenar & Kerig, 1999). The specific cognitive impairment that is the core of the deficits of people with an ASD, severely threatens the development of children with this disorder. In Piagetian terms the challenging behaviour of children with an ASD and MR could be caused by both a delay in going through the stages and stagnation, at a certain stage because of their lack of assimilation and accommodation abilities. The specific cognitive deficits in the ASD's could be viewed as a predisposing factor for challenging behaviour.

d) Psychoanalytical Models

Wenar et al. (1999) describe three schools of psychoanalysis namely: classical psychoanalysis, ego psychology and object relations' theory.

Psychoanalysis is barely used with regard to the ASD's and MR. Maybe this can be explained by the damage done to parents due to the work of Bettelheim (1967). He thought the cause of autism was a disturbed relation between the mother and her child.

e) The Family Systemic Model

In the family systemic theory, the family is conceptualized as a system and this system is the unit of the analysis.

In the literature on challenging behaviour in children and adolescents the role of the surroundings e.g. upbringing, friends etc. is always considered to be extensive (Wenar et al., 1999). Also in the field of MR certain challenging behaviour and psychiatric disorders are often attributed to attachment problems, traumatic events in early life etc. (Došen, 1990). Stenfert Kroese & Fleming (1993) found no familial characteristics associated with challenging behaviour in their clinical group of children with MR, although more challenging children lived in one-parent families. These authors have also found that children with MR and challenging behaviour underwent more life changes in the early years.

Research into the environment of children with an ASD, especially the environment as a source of variability in outcome, is very rare (Wolery, 2000). One study that

investigated the effects of adult behaviour on the challenging behaviour of the child, showed that social stimuli and task stimuli affected the problems of the child but the level of attention of the adult did not (Taylor, Ekdahl, Romanczyk & Miller, 1994). Taylor and Carr (1992 a&b) found that socially avoidant or aloof children displayed more challenging behaviour when adult attention increased, but attention-seeking children showed less challenging behaviour when adult attention increased. Another measure of adult attention could be the staff: child ratio. Volkmar, Hoder & Cohen (1985) found that children with an ASD exhibited the greatest deviance when the staff: child ratio was the biggest. In particular stereotypes, self-injurious behaviours, direction of gaze and number of requests made to the children increased, when the staff: child ratio decreased. This would imply, from a pedagogical point of view, that adult sensitivity to the needs of children with an ASD diminishes challenging behaviour and optimal sensitivity cannot be obtained if the adult: child ratio is very high.

A major factor in the ability of parents to cope with the challenging behaviour of their child is the stress experienced. For a long time parents of children with MR were viewed as pathological families, but nowadays they are considered families under stress (Dykens & Hodapp, 2001). This notion led to a new area of research, which also included analysis of between-family and within-family differences as a source of variability. Van Berkum (1993) found that parental stress is mainly influenced by child factors, namely: extrovert behaviour problems, low adaptive skills and a poor physical condition. Quine & Paul (1985) found that high levels of maternal stress were closely associated with challenging behaviour. They found that the most stressful factor was nighttime disturbance.

Janssen, Schuengel & Stolk (2002) proposed a model in which the challenging behaviour of people with MR is considered to be the result of stress and a lack of coping mechanisms due to both child factors and parental coping mechanisms. They are especially interested in attachment behaviour as the main perpetuating factor in this model. The special features of children with an ASD and MR, especially the triad of impairments and problems with adaptation to stimuli, make this a very interesting model to study further. As far as this author knows, no such research has been carried out with children with an ASD and MR.

Therefore in the family systemic model we find both risk and protective factors for the emergence of challenging behaviour. According to O'Brien (2000) interpersonal factors function mainly as perpetuating factors.

f) The developmental dynamic model

For a long time the above-mentioned models were used independently and were thought to be incompatible. Evolution of the different models has brought them closer together, e.g. the medical model no longer considers hereditary vs. environment to be a major issue but states that an organism develops within an environment that interacts with organic variables.

A combination of the cognitive and the psychoanalytic models is the 'developmental dynamic model' as proposed by Došen (1990). He integrated the psychoanalytical

theories of Mahler and Erikson and the cognitive theory of Piaget to explain the psychiatric and behaviour problems of people with MR. In this view behaviour problems are described as a discrepancy between cognitive development, motor development and sensory development, on the one hand, and the level of socio-emotional functioning, on the other hand. Došen describes the socio-emotional development of children up to the age of three as follows (Došen, 1990, p. 47-49):

- 1) 0-6 months: first adaptation phase; sensory integration and integration of structure in space, time and individuals take place in this period;
- 2) 6-18 months: first socialization phase; attachment and basic security should be established in this phase;
- 3) 18-36 months: first individualization phase; in this phase distance from physical contact, communication at a distance, and the start of personality development should be established.

Challenging behaviour, according to this model, occurs when development stagnates because of deviant functioning or acquired psychological disease during the development in the first three years of life.

Došen's model is very comprehensive and has proven to be of great clinical value. However it does not use standard (e.g. DSM-IV) terms to describe the different problems or diseases, so the terminology is often considered to be confusing.

Wenar et al. (1999) proposed an integrated model or a developmental framework. This model provides us with the opportunity to incorporate intrapersonal (behaviour, cognition, personality, etc.), interpersonal (parent-child relationships, teacher-child relationships, etc.); super-ordinate (family, group, socio-economic status, etc.) and organic factors. We share the view of Wenar et al. (1999) that although there is divergence between the different models, there is also convergence. The convergence gives us the opportunity to combine them, and the divergence makes them complementary. The predisposing, precipitating and perpetuating within all four contexts proposed by Wenar et al. (1999).

When using the developmental framework, which incorporates the different models, a more eclectic view on the challenging behaviour of children with an ASD in combination with MR is possible. This provides us with the opportunity to include as much information as is available on the complex problems posed by these children.

In our view the choice of eclecticism is not a means to avoid a choice and a strong theoretical base (Van Gemert, 1985) but is a well-considered approach to strengthen the use of theory. We support the view of Van Strien (1986), who stated that in the practical sciences different theories should be used not only as deductive-nomological syllogisms but also as heuristic searchlights. In this way the different theories become sources that generate hypotheses. Because of the explorative character of our research, this view would seem to be the most useful.

2.6 Conclusion

In this chapter we have discussed the challenging behaviour of children with an ASD in combination with MR. Although challenging behaviour is by no means rare in this population, very little specific research has been carried out on the risk and protective factors. Mixed groups, different instruments and assessment strategies and diagnostic overshadowing are just a few of the explanations for the scarce information. This has led us to choose a research model with a broad view of the behaviours and the theories applied to explain these behaviours. Hence the choice for the framework described in the previous section. The research project, as described in the next chapters, was based on this model.

3 The design of the study

3.1 Introduction

The goal of the present study was to determine the factors that contribute to the emergence and development of challenging behaviour of children with an ASD and MR. Hopefully these factors can be combined into a model that can be used to identify children with an ASD and MR who are at risk for developing severe challenging behaviour. To find the factors that contribute to the development of severe challenging behaviour in children with an ASD and MR, we will use the developmental framework adapted from Wenar & Kerig (1999).

This framework consists of one dimension and four contexts, namely:

The **time dimension**; the expected behaviour of children changes over time; this can be viewed as a continuum, for example the chronological age. It can, however, also be conceptualized in terms of stages of development, e.g. Piaget's theory on cognitive development or Freud's theory on psychosexual development. This implies a spectrum, not a continuum.

The **intrapersonal context**; this encompasses the variables within the person, e.g. personality, level of functioning, behaviours, emotions, etc. This context provides the greatest amount of developmental data. Because we prefer an eclectic approach to psychopathology, we will not use one single theory but various individual theories that offer conflicting, but useful, information about the challenging behaviour at hand.

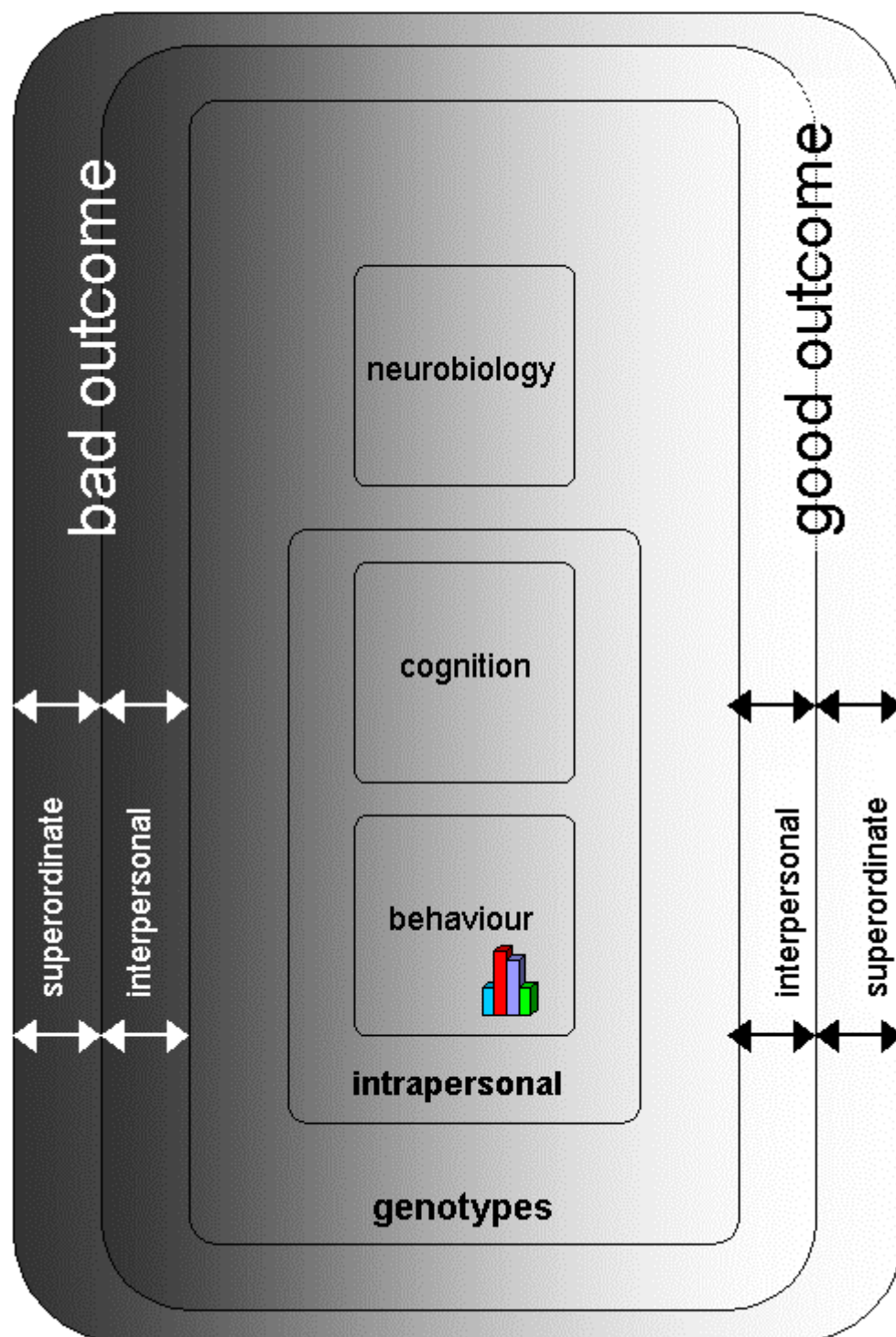
The **interpersonal context**; this context covers the interactions between individuals, e.g. interactions within the family, parent-child relationships and relationships with the peer group.

The **super-ordinate context**; this context deals with aggregates of individuals taken as a unit, e.g. the group, social class and culture.

The **organic context**; this context involves various aspects of the human body that are relevant for the understanding of deviant development, e.g. genetic information, functioning of the brain, etc.

Because of our general assumption that behaviour is determined not only by the surroundings and organic factors but also by the interaction between a biologically unique person with his own unique experiences within the social and material surroundings (Došen, 1990), we will gather information about the child through time and in all four contexts. Then again, all the above-mentioned contexts are in constant interaction with each other; therefore we will also look at the interaction between the different contexts. By combining the framework of Wenar & Kerig (1999) with the bio-ecological model (Bronfenbrenner & Ceci, 1994) we obtained a framework to assess the children and determine the predisposing, precipitating and perpetuating factors for the emergence of challenging behaviour over time.

Figure 3-1 The research model



It is generally acknowledged that the behaviours of children with an ASD change with age and depend on the level of functioning (Howlin, 1998). However, very little specific information is available about other general factors that could play a contributing role. To establish the general factors that influence challenging behaviour, we divided the research project into two phases. The first phase was designed to identify the factors needed for sample selection for the next phase of the research project (3.2). The set-up of the second phase will be explained in 3.3.

3.2 The first phase of the study

In this phase research was carried out by means of analysis of data that had already been collected using the Handicaps, Behaviour and Skills Schedule (Wing & Gould, 1982). The HBS was designed to gather information about both the development (social age) and pathology of children with an ASD. In the above-mentioned terminology the HBS gathers information about time and the intrapersonal context. The method of obtaining information is a semi-structured interview with the primary caregiver.

The data were collected by the research group ‘Severe Developmental Disorders’ of Leiden University and staff of the University Hospital in Utrecht. The sample consisted of 374 children with an ASD, ranging in age from 2 to 12 years.

The data allow us to investigate the following factors:

1. presence of mental retardation;
2. chronological age;
3. social age;
4. sex ;
5. level of functioning;
6. level of communication;
7. level of social interaction.

Therefore the time dimension (chronological age) and six intrapersonal factors can be studied.

3.2.1 The Handicaps Behaviours and Skills Schedule (HBS)

In this paragraph we will describe the Handicaps Behaviours and Skills Schedule (HBS), and discuss its use as a research instrument. First the history of the instrument is outlined, followed by a description of the instrument and the administration and scoring procedures. Furthermore the reliability and validity of the instrument will be discussed.

3.2.1.1 Development of the Instrument

Lorna Wing designed the Handicaps, Behaviour and Skills Schedule in the early nineteen seventies. She did this for the purpose of the epidemiological study then being carried out in the southeastern London borough of Camberwell. In this study Wing and her colleagues tried to identify children with the classic autistic syndrome and related syndromes (Wing & Gould, 1978).

The HBS has been described as an interview ‘to elicit, from parents and professional workers, details concerning the child's abilities and his behaviours as these are observed at home, or in school, nursery or other services’ (Wing & Gould, 1978: p. 79). It was designed as a research instrument, but it has proven to be of good clinical value over the years, because it offers a complete evaluation of a person with developmental delays. The authors claim that the instrument is suitable for both children and adults with mental retardation, autism and specific cognitive disorders (Wing, 1980). It is not a formal intelligence test and it is not suitable for people who are not retarded in any aspect of development (Wing, 1996).

Translations of the instrument into other languages were made, and studies were carried out in Denmark (Bernsen, 1980), Germany (Ort & Liepmann, 1981) and the Netherlands (Van Berckelaer & Van Duijn, 1993; Dijkxhoorn & Martens, 1992). The official Dutch translation, carried out by the Department of Orthopedagogics at Leiden University, became available in 1983.

3.2.1.2 Description of the Handicaps, Behaviour and Skills Schedule

The HBS is a semi-structured interview, not a questionnaire. The subscales have been arranged in an order that facilitates performance of the interview, the items according to the sequence of the developmental milestones.

There are 24 subscales of development and nine for abnormal or difficult behaviours. Most items of the developmental scale were derived from the Vineland Social Maturity Scale (Doll, 1965). The scores for these items can be added and converted into a social age equivalent.

Table 3-1 Contents of HBS

| DEVELOPMENTAL SCALE | BEHAVIOURAL SCALE |
|--|---|
| 1. Mobility (2 items) | 24. Abnormal Response to Visual Stimuli (5 items) |
| 2. Skilled Movements (3 items) | 25. Abnormal Proximal Sensory Stimulation (9 items) |
| 3. Feeding (4 items) | 26. Abnormal Bodily Movements (8 items) |
| 4. Washing (1 item) | 27. Routines and Resistance to Change (6 items) |
| 5. Dressing (4 items) | 28. Behaviour problems with Limited or no Social Awareness (12 items) |
| 6. Continence (4 items) | 29. Behaviour problems with Social Awareness (5 items) |
| 7. Comprehension of Speech (2 items) | 30. Sleeping Problems (2 items) |
| 8. Ability to use Speech (4 items) | 31. Practical Skills (3 items) |
| 9. Comprehension of Sign Language (2 items) | 32. Initiative and Perseverance (5 items) |
| 10. Ability to use Sign Language (2 items) | 33. Level of Independence (5 items) |
| 11. Abnormalities of Speech or Sign Language (7 items) | |
| 12. Comprehension of Nonverbal Communication (2 items) | |
| 13. Ability to use Nonverbal Communication (4 items) | |
| 14. Interest in Communication (3 items) | |

-
- 15. Educational Achievements (18 items)
 - 16. Entertainments (2 items)
 - 17. Imaginative Activities (2 items)
 - 18. Abnormal Imaginative Activities (2 items)
 - 19. Eye Contact(2 items)
 - 20. Social Responsiveness (2 items)
 - 21. Social Play (2 items)
 - 22. Social Interaction (1 item)
 - 23. Abnormal Response to Sounds (3 items)
-

3.2.1.3 Administration

As stated above, the HBS is not a standardized questionnaire. The interview should be adapted to the knowledge and insight of the respondent. Examples of introductory questions are provided. The abilities and disabilities of the individual should be considered in every question. The administrator can ask as many questions as he or she sees fit to obtain the required information to get correct ratings. Neutral questioning is obligatory to avoid denial of abnormality, especially with regard to the questions on abnormal behaviour (Wing, 1996).

The average duration of the interview is approximately 90 minutes.

All of these adaptations require skilled administrators. Only trained professionals administer the Dutch version. Lorna Wing, the designer of the instrument, was involved in the training of Dutch professionals.

3.2.1.4 Scoring

The items of the developmental scale are arranged in the order of the developmental milestones (ordinal). Some items are divided into twelve stages of development, others into only two. Sometimes indications of the developmental age are given; they can be used to compare the score of the handicapped child with that of a normal child. Often however, different stages have the same developmental age. The score for the sub-item that best describes the level of current performance must be put on the scoring sheet.

A total score can be obtained by adding the scores. A conversion table is available to obtain the equivalent social age.

The items concerning abnormal or difficult behaviour are scored on a 3-point scale (ordinal). The problem can be scored as a marked (the caretaker considers the problem to be a major one), a 'minor' or 'no' problem.

3.2.1.5 Reliability and Validity of the HBS

Several papers on the reliability (or consistency) of the HBS have been published.

The following forms of reliability must be taken into consideration (Neale & Liebert, 1986):

- test-Retest Reliability;
- inter-item Reliability;
- inter-scorer Reliability;

To determine the validity of the HBS, Neale & Liebert (1986) give the following types of validity:

- construct validity;
- content validity;
- criterion validity;
- convergent validity;
- discriminant validity.

In the following paragraphs we will discuss the results of the studies of both reliability and validity.

Test-Retest Reliability

Test-retest reliability is used to determine whether a test measures a constant phenomenon. The HBS, however, is a developmental scale. Especially for children, the score is expected to change, i.e. to improve over a period of time. So test-retest is only suitable to establish reliability when the two measurements are carried out within a short period of time.

Inter-item Reliability

Inter-item reliability is used to determine the internal consistency of a test, by correlating scores on two or more subtests (e.g. split-half reliability). The scores of the HBS, however, are not homogeneous. Every item measures a different aspect of development.

Van Berckelaer & Van Duijn (1993) measured the internal consistency by means of Cronbach's Alpha of the developmental subscales, i.e. motor, self-care, language, nonverbal communication, imagination/social and practical skills, and schoolwork. They recoded the developmental scores in order to obtain interval/ratio levels. Internal consistency was found to be satisfactory. The alphas varied from .74 to .92 for the entire group, and from .20 to .87 for the group consisting only of autistic subjects. Only one alpha was .20.

Inter-scorer Reliability

This form of reliability is given by a comparison of the scores obtained by two different raters. Wing & Gould (1978) reported on a small inter-scorer study (with highly trained interviewers), in which they obtained inter-scorer scores of close to 100%. The same authors (1978) also reviewed agreement between researchers. Agreement was 70% or more for 52 of the 62 sections rated and above 50% for all sections. In a study of the German version of the HBS (the IFFV), Ort & Liepmann (1981) reported an inter-scorer score of above 60%, with an average of 93%. Using the Kappa index, five items were noted as 'dubious or problematic'. Analysis of these items revealed that these problems were due mainly to misinterpretation of the questions.

Construct Validity

Construct (or theoretical) validity is used to determine whether a test is able to measure a certain theoretical construct. The HBS was designed by Lorna Wing as a tool to

determine impairments in social interaction, communication and imaginative skills. This 'triad of impairments' is a theoretical construct of autism (Wing & Gould, 1978; 1979 and Wing, 1996). In these and many other studies, the HBS has been shown to be able to distinguish between specific social and communicational impairments and other impairments.

Lund (1989) found, using a latent-trait model, that the two subscales for behaviour problems with and without social awareness are able to measure 'behaviour disorder'.

Criterion validity

This form of validity involves the question of whether test scores predict behaviour in all of those situations in which they are supposed to. The HBS was designed as an interview and gathers information from primary caretakers on daily life achievements. Criterion validity is not a big issue in this case.

Content validity

Content validity refers to the suitability of the sampled items to operationalize the construct, in this case development. Most items of the HBS were derived from previous scales, for example the Kushlick interview schedule and the VSMS (Wing & Gould, 1978). The items were picked by reviewing the results of early examples of the schedule and on the basis of practical experience. In the study by Van Berckelaer & Van Duijn (1993) described above, high correlations and equal mean age equivalents were found between the PsychoEducational Profile (Schopler & Reichler, 1979) and the HBS. This suggests that the content validity is satisfactory.

Convergent validity

This form of validity is used to determine whether two measures of the same construct have a high correlation. In other words: do two instruments that presumably measure the same thing indeed do so. Rapin's study (1996) of preschool children with inadequate communication showed that the responses of teachers to the HBS largely confirmed the picture created by the parents' reports and also the rating given by the child psychiatrists (p. 163).

Van Berckelaer & Van Duijn (1993) found a high correlation (0.81) between the scores of the HBS and the PsychoEducational Profile (Schopler & Reichler, 1979).

All of these findings suggest good convergent validity.

Discriminant validity

This form of validity is used to determine whether the scale is able to distinguish between the different concepts, as it is supposed to. In the case of the HBS this means is the scale able to distinguish between the autistic spectrum disorders and other disorders?

In several unpublished studies, the HBS was found to be able to discriminate between specific groups (e.g. differences in developmental and behavioural profiles between people with autism and people with Down's syndrome, with developmental ages between eight and ten). Dijkxhoorn & Martens (1992) reported differences in developmental and behavioural profiles between mentally handicapped children with and without autism. Both groups of children had developmental ages between three and four. A preliminary study by Liepmann & Ort (1980) reported promising results of

profile analysis. In the study of Rapin et al. (1996) differences were noted between children with developmental language disorders, children with high-functioning and low-functioning autism and children with developmental delay.

The HBS was designed to be used for populations with severe developmental disorders. The research data gathered over the years have shown satisfactory reliability and validity. Although the instrument has not been extensively researched, it has proven its value in both theory and practice.

3.3 The second phase of the study

Starting with the information collected in the first phase, we designed the second phase to explore the behaviour problems of children with a Pervasive Developmental Disorder and Mental Retardation in more detail. In the second phase the data was collected by means of individual assessments of the children, interviews with and questionnaires for caretakers and file analysis. In this phase information was collected at the four different contexts, namely the interpersonal, the intrapersonal, the super-ordinate and the organic contexts across time.

3.3.1 Sample selection

The sample of children with an ASD and MR was selected with the help of the regional advisory teams for people with MR and extreme challenging behaviour. We asked them to refer those children who matched the following criteria:

- 1) chronological age below 13 years;
- 2) mild to severe mental retardation (mental age 6-96 months), according to DSM-IV criteria (APA, 1994);
- 3) presence of a Pervasive Developmental Disorder, according to DSM-IV criteria (APA, 1994);
- 4) no serious additional sensory or motor handicaps (e.g. blindness, deafness, cerebral palsy);
- 5) a CEP score of 3 or higher.

The Consensus Protocol Severe Challenging behaviour (CEP; Schuring et al., 1990) had already been filled out, because this is a requirement when a person is referred to an advisory team and is eligible for financial support. So the information was already available for the children with challenging behaviour.

Schuring et al. (1990) developed the Consensus Protocol Severe Behaviour problems (CEP) to be able to distinguish between people with MR with challenging behaviour and those without, because the standard classification systems fall short in this respect. In the CEP the behaviours, the treatment and expected future perspectives must be indicated. The items about challenging behaviour separated into five prototypical categories, as proposed by Van Gemert (1985). These categories are:

- 1) the type with mainly self-injurious behaviours;
- 2) the type with mainly episodic outbursts;
- 3) the type with mainly reactive behaviour problems;

- 4) the type with mainly obsessive behaviours;
- 5) the type with mainly self-involved behaviours.

A 'consensus method' was used to assess the severity of the behaviour. At least three people involved in the care of the mentally retarded person with challenging behaviour had to fill out a questionnaire. In the final questionnaire the conclusion reached by consensus was described. Following a specific instruction, the level of severity of the behaviours was determined. These levels are:

1. least severe problems
2. moderate problems
3. very severe problems
4. extreme problems.

Kramer (1992) found that the validity of the level of severity is high ($\kappa = .91$). They measured this by comparing the CEP score with a ranking of the behaviours by caretakers familiar with the mentally retarded person.

We decided to include only children who live in a family setting, because the majority of the children referred did live at home and this gave us an opportunity to specify the research group further. The goal was a group of 20 children. All of the children who fitted the criteria have been included in the study. None of the parents refused to cooperate. After we had reached our goal of 20 cases, we stopped collecting data.

A control group was formed by means of coupled matching. The psychologist or pedagogue of the day-care centre or school that the child with ASD and MR and challenging behaviour attended was asked to find a match. Some children of the control group were referred for diagnosis to the clinic at Leiden University. Matching took place on the basis of the factors established in the preliminary research project, namely: level of functioning and chronological age.

The level of functioning was determined by means of the total score of the PEP-R (Schopler et al., 1990). For a further explanation of this scale see 3.6.1.

All of the children who fitted the criteria were included in the study. None of the parents refused to cooperate.

3.4 Description of the sample

The sample consisted of 20 children ranging in age from 4 years 7 months to 12 years. Seventeen children attended day-care centres for children with Mental Retardation (KDC); three visited a school for children with mild to moderate MR (ZMLK). All of the children were recruited to participate in the research project through the regional advisory teams and lived with their family.

The control group consisted of 20 children, ranging in age from 4 years 5 months to 12 years 10 months. Twelve children attended day-care centres for children with MR, seven visited a school for children with mild to moderate MR and one child was home-schooled. The children were recruited from the same schools and day-care

centres as the first sample; others were clients of the Ambulatorium at Leiden University. All lived with their family.

Table 3-2: Mean age, developmental age, male: female ratio and speaking: non speaking ratio of the two groups

| | Sample | Control Group |
|------------------------------------|-----------------|------------------|
| Mean Chronological Age (in months) | 92.35 (SD=26.7) | 94.30 (SD=26.45) |
| Mean Developmental age (in months) | 22.95 (SD=9.09) | 24.40 (SD=9.64) |
| Male: female ratio | 17: 3 | 17: 3 |
| Speaking: non-speaking ratio | 11: 9 | 11: 9 |
| Total | N=20 | N=20 |

The control group was slightly older and the developmental age was also slightly higher. The differences between the two groups were not significant ($\text{Sig} = .609$ and $.605$, respectively).

All children in the sample had a CEP score of three or higher, the children in the control group had a CEP score of two or lower.

3.5 Operationalization of the variables

The developmental framework of Wenar & Kerig (1999) contains of five dimensions. All variables that were investigated can be fitted into this framework. The instruments listed in this table will be discussed in paragraphs 3.6 and 3.7.

Table 3-3: The variables that were the focus of the research project, fitted into the developmental framework

| Time | Intrapersonal | Interpersonal | Super-ordinate | Organic |
|----------------------|--|---------------------------------------|--|--|
| Pre-natal | | | <i>Nationality</i> <i>Parents</i> <i>Siblings</i> | <i>Family history</i> (DISCO I) <i>Prenatal history</i> (DISCO I & file analysis) |
| Perinatal | | | | <i>Perinatal history</i> (DISCO I & file analysis) |
| Postnatal/first year | <i>First year of life</i> (DISCO I & file analysis) | | | <i>Postnatal history</i> (DISCO I & File analysis) |
| History | <i>Developmental History</i> (DISCO I & file analysis) <i>Psychological assessments</i> (file analysis) | <i>Changes as mentioned in files.</i> | <i>Family Structure</i> <i>Changes as mentioned in files.</i> | <i>Medical history</i> (DISCO I & file analysis) <i>Medical assessments</i> (DISCO I & file analysis) |
| Present age | <i>Development (PEP-R)</i> <i>Adaptive functioning</i> (VABS) <i>Communicative functioning</i> (COMVOOR/ | <i>Pedagogical situation</i> (NVOS) | <i>Family Structure</i> <i>Present caretakers</i> <i>SES</i> <i>Parental Stress</i> | <i>Cause of the disorder</i> <i>Present medication</i> <i>Present state of health</i> |

| Time | Intrapersonal | Interpersonal | Super-ordinate | Organic |
|------|--|---------------|----------------|---------|
| | VABS comm.) <i>Social functioning</i> (VABS soc./ Wing cat.) <i>Play</i> (SPT/VABS play) <i>Motor Skills</i> (VABS mot/PEP-R mot) <i>Challenging behaviour</i> (DBC, DISCO II) | | (NOSIK) | |

3.6 Description of the instruments: Intrapersonal level

In this paragraph we will describe the instruments used for our research. An overview of these instruments can be found in table 3.1

3.6.1 PsychoEducational Profile, revised (PEP-R; Schopler, Reichler, Bashford, Lansing & Marcus, 1990)

In 1972 the Division for the Treatment and Education of Autistic and related Communication handicapped CHildren (TEACCH) was founded. In their attempts to assess the children who were referred to them, Schopler and Reichler developed an instrument that could be administered in a more flexible fashion (Schopler & Reichler, 1979). This resulted in the first version of the PsychoEducational Profile (PEP). The Dutch version was published in 1982. In 1990 a revised version was published: the PEP-R (Schopler et al., 1990). The Dutch version of the PEP-R was translated and published by the Researchgroup Severe Developmental Disorders at Leiden University (1998).

The materials of the PEP-R have been adapted to the needs of the child with relational and communication problems; therefore very little language comprehension is necessary (Schopler et al., 1979). According to the manual the PEP-R can be used for children between 2 and 12 years old; the highest score however has an age equivalent of 78 months (Schopler et al., 1990). Research at Leiden University into the Dutch version of the PEP-R has established a range from 6 months to 5 years (Van Deyl, 1997). These ages are the developmental ages. Experience has shown that the PEP-R is very useful for older children (and even adults) with MR.

The PEP-R consists of two subscales, namely a behavioural scale and a developmental scale. The behavioural scale contains four subscales to identify the specific behavioural patterns of children with autism. These are:

- relating and effect;
- play and interest in materials;
- sensory responses;
- language.

The developmental scale consists of nine subscales:

- imitation;

- perception;
- fine motor skills;
- gross motor skills;
- eye-hand integration;
- cognitive performance;
- cognitive verbal.

These subscales were not established on the basis of factor analysis but on clinical experience.

Literature on the reliability and validity of the PEP-R is scarce. Schopler et al. (1990) described satisfactory inter-scoring reliability. The intraclass correlation was found to be .92 across different raters. The internal consistency of the PEP-items was established for a group of 72 children with different developmental disorders. The results showed correlations ranging between .79 and .96 (Van Berckelaer-Onnes & van Duijn, 1993). One would not expect a significant change in these figures for the PEP-R, because the construct has not changed. For the content validity of the developmental scale, significant correlations were found between the PEP-R and the Merrill-Palmer Scale (.85), the VSMS (.84), the Bayley Scales of Infant Development (.77) and the Peabody Picture Vocabulary Test (.71). The correlations with formal intelligence tests (e.g. WISC and WPPSI) were however much lower (.47).

Investigation of the Dutch version of the PEP-R has shown that the correlations between the scores of the American reference group and the Dutch group are high, except for the domain perception. In the Dutch group the maximum score for this item reflected a lower developmental age (van Deijl, 1997).

Kraijer & Plas (2002) in particular have expressed fierce criticism of the PEP-R. The research underlying the pathology scale, the profile for the subscales and the use of the 'emerging' score are too weak to validate use. We have been using the developmental scale to assess a general level of functioning and to compare the scores of our two groups.

3.6.2 The Vineland Adaptive Behavior Scales (VABS; Sparrow, Balla & Cicchetti, 1984)

Since the publication of the latest version of the AAMR definition of Mental Retardation (Luckasson et al., 1992), awareness of the importance of adaptive behaviour scales has grown considerably. Since then assessing adaptive behaviour has received preference above assessing intelligence, especially for subjects with severe to profound MR. To determine adaptive functioning, active co-operation of the client is not necessary because the information can be gathered through an informant who knows the client very well. Another advantage is that adaptive behaviour assessment covers actual behaviour over a longer period of time (Kraijer, 2000).

The Vineland Adaptive Behavior Scales (Sparrow et al., 1984) are probably the most widely used. The VABS are administered through a semi-structured interview with the primary caretaker, in our study the parents. The interview can only be carried out

by trained interviewers. In this study we used the expanded version of the scales, which collects information on four domains and 11 sub-domains:

- communication (receptive, expressive and written language);
- daily living skills (personal, domestic and community);
- socialization (interpersonal, play and leisure and social skills);
- motor skills (gross and fine).

Dutch standardization of the scales is still in progress at the Leiden and Groningen Universities (Kraijer, 2002). In the Friesland study, norms for the Dutch MR population have been established (De Bildt, 2003). Preliminary cross-cultural studies show small differences for the domain daily living skills in France (Fombonne & Achard, 1993) and differences in both daily living skills and communication in the Netherlands.

People with an ASD showed a specific scatter across the four domains. People with an ASD exhibited relative weaknesses in the socialization domain and relative strengths in the daily living skills domain. The scores for the communication domain were found to be intermediate (Carter, et al., 1998). These findings have resulted in a supplementary set of norms for people with an ASD. Four new norm groups were formed. The norms are formulated for two different age levels and there are separate norms for speaking and non-speaking subjects.

The American standardization procedure was performed on the Survey version with 3000 subjects. The internal consistency ranged from .70 to .98 for the domains and from .24 to .95 for the sub-domains. Test-retest reliability was determined for the domains, ranging from .76 to .93, and the inter-scorer agreement ranged from .62 to .78 for the different domains (Sparrow et al., 1984).

The construct validity was good; there were significant relationships between the scores and age and developmental age. Factor analyses yielded four factors which do not completely overlap with the domains. The factors are: Communication, Socialization, Daily Living Skills and Motor skills; the fourth factor contains the sub-domains Community Skills and Written Language. A study of the criterion validity revealed low correlations with the IQ-tests, ranging from .14 to .50. The correlations with the developmental scales were higher, around .55 (Sparrow et al., 1984). We performed some studies of the correlation between the PEP-R (Schopler et al., 1990) and the VABS and found a correlation of .74 for children.

3.6.3 The ComVoor (Verpoorten, Noens & Van Berckelaer-Onnes, 1999)

The name ComVoor is derived from the Dutch words for precursors in communication; in English the instrument is called ComFor, for Forerunners in Communication. The ComVoor is still an experimental instrument, designed to determine indicators for the use of augmentative communication for people with an ASD and none or limited verbal skills (Noens, Berckelaer-Onnes & Verpoorten, 2000).

The ComVoor is based on the following communication model of Verpoorten (1996):

Table 3-4: The communication model of Verpoorten (1996)

| Level | |
|---------------------|---|
| Sensation | Primary senses: tasting, smelling, feeling, looking, and listening. |
| Presentation | The presence of an object is the cue for acting. |
| Representation | A representation of an image or object is the cue for acting. |
| Meta-representation | Imagination; attribution of a new function to an object or picture. |

The test consists of five series with a total of 40 items and collects data on two levels of pre-verbal communication, the level of presentation (the first three series) and the level of representation (the last two series). The first level consists of matching tasks; the second contains sorting tasks above the level of literal association (e.g. sorting different balls or different photographs of cars). The test has been designed specifically to meet the needs of people with an ASD (Noens et al., 2000). The items of the ComVoor were designed to determine out whether the child needs augmentative communication on the presentational or representational level. The level of the sensations is not covered in the ComVoor. Children who obtain no score on the ComVoor are considered to function at the level of sensation (Noens & Van Berckelaer, 2002).

3.6.4 The Symbolic Play Test (SPT; Lowe & Costello, 1976)

The SPT was designed to assess the language potential in children who have failed to develop receptive and/or expressive language. The goal was a test that would explore comprehension of semantic relationships, early concepts and the ability to deal with concepts in young children (Lowe & Costello, 1976 p. 5). The test consists of four sets of miniature toys designed to evoke spontaneous non-verbal play activities in a structured situation. It takes about 15 minutes to administer the test. The total score can be converted to an age-equivalent.

The authors determined the reliability of the SPT. Both the split half and test-retest reliability are good ($r > .70$). The validity was measured by means of correlations with tests of language ability; those correlations were low ($\leq .30$). The scores increase over time, the authors call that 'later language ability'. After one year the correlations exceed .60.

For some of the children in our sample, the materials were too fragile and even dangerous to handle. The children for whom this applied were given 0-scores.

3.6.5 The Developmental Behaviour Checklist (DBC; Einfeld & Tonge, 1995; Dekker, Nunn & Koot, 2002)

Stewart Einfeld and Bruce Tonge, two psychiatrists from Australia, designed the DBC to be able to gather information on the relationship between behavioural and emotional disturbance in children and adolescents with MR. With this instrument

they are able to obtain prevalence data about these problems (Einfeld & Tonge, 1995). They describe the DBC as a ‘standardised instrument completed by lay informants to assess behavioural and emotional disturbance in children and adolescents with mental retardation’ (Einfeld & Tonge, 1995 p. 81). The construction of the instrument was based on the widely acclaimed format of the Child Behavior Checklist (CBCL; Achenbach & Edelbrock, 1983).

Item selection was based on the following definition of behavioural and emotional disorder: qualitative and quantitative deviance that cannot be explained on the basis of developmental delay alone and causes significant distress to the child, caretakers or the community, and causes significant added impairment. In total 664 files were researched for these criteria; after this process they ended up with 135 descriptions. After eliminating procedures based merely on inter-scorer reliability, 96 descriptions of behaviour remained.

The DBC is a questionnaire that can be filled out by parents (DBC-P/VOG-P), caretakers and/or teachers (DBC-T/VOG-L). The list contains 6 subscales namely:

- disruptive;
- self-absorbed;
- language deviance;
- anxiety relating;
- autistic relating;
- antisocial.

A factor analysis carried out by Hastings, Brown, Mount & Cormack (2001) resulted in 5 factors; they are:

- stereotype and self-stimulatory behaviour;
- disruptive and antisocial behaviour;
- internalising behaviour;
- inhibited behaviour lacking social sensitivity;
- autistic-like behaviour.

In the Dutch version (the VOG) five factors have been established, namely:

Communication Disturbance, Social Relating, Disruptive/Antisocial, Self-absorbed and Anxiety. These five factors explain 43.7% of the variance (Dekker, Nunn, Einfeld, Tonge & Koot, 2002). In this version norms for the Total Problem Behaviour Score (TBPS) have also been established (Koot & Dekker, 2001).

The items have been translated into language suitable for non-professionals. In the Teacher version the questions about disturbed sleep were deleted. Every item can be scored between 0 (‘not true as far as you know’) and 2 (‘often true or very true’). The sum of all positive items results in a Total Behavior Problem Score (TBPS). The six factors that were calculated on the basis of Principal Component Analysis provide six subscale scores.

A Receiver Operating Characteristics (ROC) analysis was conducted to obtain a ‘cut-off’ score. The area below the ROC curve visualizes the true positive rate against the false positive rate. The point where ROC equals 45 degrees is the optimum cut-off.

This equals a TBPS of 46 (Einfeld & Tonge, 1995). Using the ROC curve a valid prediction of psychiatric caseness can be made. This measure has been carried out along the same lines as the procedure used to design the CBCL (Achenbach & Edelbrock, 1983). Similar findings have been published for the Dutch version (Dekker et al., 2002a).

The developers of the DBC have published several papers on the reliability (or consistency) of the DBC (Einfeld & Tonge, 1996a, b & c). In total 63 pairs of caretakers were asked to fill out the DBC at two-week intervals to assess test-retest reliability. The resulting coefficient was .83. The internal consistency (Cronbach's alpha) varied from .67 (antisocial) to .91 (disruptive). The mothers and fathers of 42 children were asked to fill out the questionnaire independently to obtain inter-scorer scores. Teachers carried out the same procedure. The resulting inter-scorer scores varied from .56 to .90 for parents and .28 to .81 for teachers. Parent-Psychiatrist agreement has been used as a measure of convergent validity. The Pearson product-moment correlation between the DBC total score and rating by the professionals was .81. Hastings et al. (2001) found very similar factor structures and internal consistency. They have however reservations about use of the DBC for epidemiological purposes, because of the noted differences between raters.

The total score on the DBC has also been compared with that for other instruments used to assess psychopathology in children with MR. A correlation of .86 was found between the DBC and the Adaptive Behavior Scale of Lambert & Windmiller (1981) and a correlation of .72 was noted between the DBC and the Problem Behavior Section of the Scales of Independent Behaviors of Bruininks et al. (1984). Dekker c.s. reported reasonable relationships between the scores for the VOG and the CBCL. They also found correlations between the score for the VOG-O and the DSM-IV diagnoses anxiety disorders, behaviour disorders and mood disorders (Dekker et al. 2002a).

3.6.6 The Diagnostic Interview for Social and Communication Disorder (DISCO; Wing, 1993a)

The DISCO was designed by Lorna Wing to collect information on development and behaviour in a structured manner from the parents of the client. The DISCO is actually a new and improved version of the HBS (Wing, 1979), which we used in the preliminary phase. The DISCO gathers information on (Wing, Leekam, Libby, Gould & Lacombe, 2002):

- infancy;
- age of recognition of deviant development and setbacks in development;
- developmental skills;
 - current level of functioning in each domain;
 - delay in reaching milestones relevant to the domain;
 - rated untypical behaviour in relation to the domain;
- untypical behaviour not directly linked to a specific developmental domain.

Inter-scorer reliability was found to be high, kappa .75 or higher, for over 80% of the items (Wing et al., 2002). Ongoing research is focussing on developing algorithms for ICD-10, DSM-IV and the Wing and Gould autistic spectrum disorder criteria (Leekam, Libby, Wing, Gould & Taylor, 2002).

For the Dutch version the research group Severe Developmental Disorders at Leiden University translated the behavioural section, but left out the items on current development. Information on current development is, according to the research group, better assessed by the VABS (Sparrow et. al., 1984); whereas in the English version the VSMS (Doll, 1965) was used. The remainder of the items have been divided into two parts.

The first part, DISCO I, collects information about the history of the client. It was designed especially for the purpose of obtaining key background information needed to classify the ASD's.

With the second part, DISCO II, the interviewer can collect information on the behaviours of the client. Both the behaviours specific to the ASD's (e.g. responses to stimuli, routines, resistance to change) and non-specific behaviours (e.g. maladaptive behaviours with and without social awareness) are mapped. We have arranged the items according to the sub-domains of the VABS. The specific behaviours are summarized in an extra domain, called 'specific behavioural patterns'.

Some preliminary research has been carried out to assess the inter-scorer reliability of the Dutch versions. A study in which 14 cases were double-scored by two raters found an inter-scorer agreement of 100% for 83% of the items; 16% of the items exhibited an agreement of 93% and only 1% of the items had an inter-scorer agreement of 86%.

For the English version the inter-scorer agreement varied across the domains from 69% for problems with daily living skills to 91% for problems with motor skills (Wing et al., 2002).

3.7 Instruments for the Interpersonal context

3.7.1 The Nijmegen questionnaire for the pedagogical situation (NVOS; Wels & Robbroeckx, 1996)

The NVOS was constructed by Wels and Robbroeckx (1996) to assess family stress. It has been designed from a pedagogical viewpoint, for use in families with a demanding child up to the age of 14 years old. The scale is based upon the stress-model of Lazarus and the attribution theory of Weiner (in Wels & Robbroeckx, 1996). The NVOS consists of four subscales namely:

- A. subjective family stress (46 items);
- B. judgement of the pedagogical situation (1 item);
- C. attributions (34 items);
- D. help expectancy (36 items).

In our study the questionnaire is filled out mainly by parents, except for those who (according to the estimation of the researcher) would find the language too difficult. In such cases the questionnaire was filled out together with at least one of the parents.

The validity and the reliability of the questionnaire are good. Investigation of the test-retest reliability yielded coefficients between .60 and .94 for the clinical group. Section 2 has proven to be the weakest. There are differences between the scores of fathers and mothers. The mothers perceive more problems than the fathers (Wels & Robbroeckx, 1996).

The internal consistency lies between .68 and .92, with the exception of the subcategory 'Perspective' that has proven to have a weak internal consistency. Factor analysis has shown that the theoretically constructed subcategories of the factors A and C are very similar to the categorization obtained through factor analysis. Factor D has to be interpreted more prudently, mainly because of the small sample size.

3.7.2 The Nijmegen parental stress index, short version (NOSIK; Brock, Vermulst, Gerris & Abidin, 1992)

The NOSI is the Dutch adaptation of the Parenting Stress Index (PSI), which was designed by Abidin (1983). The scale is based on the assumption that stress is a multidimensional concept with three actors, namely the child, the parent and life events (Brock et al., 1992).

In our research project we used the short version of the scale (NOSIK), which consists of the 25 items with the highest scores in a factor analysis of the NOSI. Ten of the original subscales are represented in the NOSIK, namely:

- competence (6 items);
- attachment (1 item);
- depression (3 items);
- health (1 item);
- adaptation (1 item);
- mood (2 items);
- distractibility (2 items);
- over demanding (5 items);
- positive reinforcement (1 item);
- acceptance (3 items).

The NOSIK has been constructed for parents of children between the ages of two and 13 years. A clinical and a non-clinical norm group have been established, so that the stress level of the parents can be compared with that of parents of normal children and with that of parents of children with some problems. The levels have been divided into seven classes from the 5% parents with the lowest level of stress to the 5% parents with the highest level of stress (Brock et al., 1992).

The reliability of the NOSIK can be rated as 'very good', with alpha's ranging from .92 to .95. The criterion validity has been rated as 'good': the NOSIK is able to distinguish between clinical and non-clinical groups.

3.7.3 File analysis

The files that are kept in the day-care centres or schools that the children attend were analysed by the researcher, using the above-mentioned model by Wenar & Kerig (1999).

The file that is kept of every child contains relevant documents about the medical and psychological history, together with treatment plans and relevant administrative information.

All the files were analysed by one researcher. The analysis was carried out on the basis of semi-structured qualitative research. The model was used as a means to structure the amount of information.

Files are not kept for the purpose of research, so the information found in the files differs immensely from one file to the next. Whether the files deliver reliable information remains to be seen. We found that the files, however, were very useful to obtain the information from the past because they helped to trigger the memory of the parents. Most of the information about the history was collected by means of the DISCO I (Wing, 1993a).

3.7.4 Observations

During all of the individual assessments and the interviews with parents, observations were written down by the researcher. These observations, which often provide extra information about the children, were processed qualitatively and reported separately from the quantitative analyses.

4 Results of Phase I

4.1 Introduction

As mentioned in the previous chapters we found surprisingly little information in the literature on the causes of challenging behaviour of children with an ASD. Therefore we decided to start with a preliminary study. The goal of this preliminary study was to determine the general factors that contribute to challenging behaviour of children with an ASD. The factors that were looked at are: presence of mental retardation, chronological age, social age, sex, level of functioning, level of communication and level of social interaction.

For the purpose of the preliminary research we used data that had already been collected with the Dutch version of the Handicaps, Behaviour and Skills Schedule (Wing & Gould, 1982; Dutch version, 1983).

In this part we will describe the results of the preliminary research project. We will start with a description of the sample and then we will show the results of the analyses across the different behavioural clusters.

4.2 Description of the total sample

The data consisted of data collected by the research group ‘Severe Developmental Disorders’ of the University of Leiden and by staff of the University Hospital in Utrecht, since the HBS was used in both clinics for diagnostic and research purposes in the period 1986-1996. The total sample consisted of 374 children with an ASD of 12 years and younger.

The composition of the sample was as follows:

Table 4-1: Sample composition

| N=374 | Social Age (SA) in months (years) | Chronological Age (CA) in months (years) | Social Quotient (SA/CA x100) |
|---------------------|--------------------------------------|---|---------------------------------|
| Mean | 40.56 (3.38) | 64.72 (5.39) | 62 |
| Standard. Deviation | 25.32 (2.11) | 27.37 (2.28) | 24 |

4.2.1 Description of the two samples: ASD MR and ASD non-MR

As mentioned in chapter 3, people with an ASD and MR display challenging behaviour more frequently than people with an ASD without MR. In most research a division in level of functioning is made: for example, Rapin (1996), in her research project on communication problems, divided the ASD group into low-functioning autism (LAD) en high-functioning autism (HAD). We consider the LAD group to be the group with MR.

In order to establish a similar division, we needed to find a method to separate the two groups, because not all cases in our sample had a clear clinical diagnosis of MR. This was mainly because some children were too young to determine the level of MR

at the time of their assessment. This is especially true for those with milder levels of MR. We therefore defined Mental Retardation in this sample by the social quotient (SQ) calculated with the HBS ($SQ = \text{Social Age} / \text{Chronological Age}$). Sparrow et al. (1984, p. 52) stated that a VSMS-SQ of 88 equals a VABS-index of 70. However, by definition children with an ASD exhibit a delay in adaptive functioning, due to problems with socialization and communication (Carter et al., 1998). Therefore, we decided to divide the group with an SQ higher than 70 into a group with an SQ between 70 and 88, this is probably the group of children with mild MR combined with the group with 'borderline-intelligence', and a group with an SQ higher than 88, this is the group with an ASD without Mental Retardation. The specifics of the three samples are shown in table 4-2.

Table 4-2: Sex, Mean Social Age (SA), Mean Chronological Age (CA) and Social Quotient (SA/CA) of the sample

| | ASD (total sample) | ASD with MR ($SQ \leq 70$) | ASD with mild MR ($70 < SQ \leq 88$) | ASD without MR ($SQ > 88$) |
|--------------------------|-----------------------|---------------------------------|---|---------------------------------|
| N | 374 | 240 | 75 | 59 |
| Sex (percentage male) | 74.70 | 70.80 | 82.7 | 79.7 |
| Mean SA (in years) | 3.38 (SD=2.11) | 2.35 (SD=1.14) | 4.26 (SD=1.73) | 6.38 (SD=2.24) |
| Mean CA (in months) | 64.72 (SD=27.37) | 61.91 (SD=27.51) | 65.52 (SD=26.28) | 76.24 (SD=24.35) |
| SA/CA | .62 (SD=.24) | .48 (SD=.14) | .78 (SD=.05) | .99 (SD=.09) |

By definition SA/CA and SA are higher in the groups with mild and no MR. Even though the chronological age of the non-MR sample is slightly higher, the samples are comparable, considering the standard deviation. The percentage males in the total sample are similar to the percentage found in the literature on the ASD's (Van Berckelaer-Onnes & Van Engeland, 1997; Kraijer, 1998). The percentage of males in the non-MR sample is slightly higher; this finding has also been reported in literature (Kraijer, 1998; Volkmar, Szatmari & Sparrow, 1993).

More than 60% of the total sample has an $SQ \leq .70$, these children are therefore considered to be mentally retarded. In literature a percentage between 70 and 80% of the population with the Autistic Disorder is usually mentioned (Howlin, 1997; Van Berckelaer-Onnes & Van Engeland, 1997). When we include the sample with mild MR, 84% of our sample has MR. This is a slight overrepresentation. The group ASD with mild MR probably includes the low end of the HFA-curve and the high end of the LFA-curve.

4.3 HBS clusters

As mentioned in paragraph 3.2.1., the HBS consists of a developmental scale and a behavioural scale. We have used the following questions of the HBS for our analyses:

Table 4-3: HBS clusters

| Item cluster: | Code |
|---|--------|
| 11. Abnormalities of Speech or Sign Language (7 items) | HSPR |
| 23. Abnormal Response to Sounds (2 items) | HAUD |
| 24. Abnormal Response to Visual Stimuli (4 items) | HVIS |
| 25. Abnormal Proximal Sensory Stimulation (6 items) | HNAB |
| 26. Abnormal Bodily Movements (7 items) | HBEW |
| 27. Routines and Resistance to Change (5 items) | HWST |
| 28. Behaviour problems with Limited or no Social Awareness (11 items) | HGZS |
| 29. Behaviour problems with Social Awareness (4 items) | HGMS |
| Specific Behaviour problems (HNAB +HWST + HGZS) | HG |
| Cluster Lund (HGZS + HGMS) | HCLUND |

The question ‘other behaviours associated with’ has been excluded from this list, because of the broad range of answers possible. In the HBS the presence of the behaviour of an item is graded 0, the absence 2. To facilitate interpretation of the results we have changed the scores. A 0 for an item in this study means the child does not express this behaviour, a 2 means that the child frequently exhibits this behaviour. The scores for the separate items have been added and divided by the number of items in the cluster. This implies that the maximum score for each cluster is 2, the minimum score is 0. This way all of the clusters are comparable.

4.4 Results of the comparisons between the MR and Non-MR groups of the behavioural clusters of the HBS

To compare the three groups One-way ANOVA was performed to see whether the total scores for the different clusters differ between the three groups. The results are shown in table 4-4.

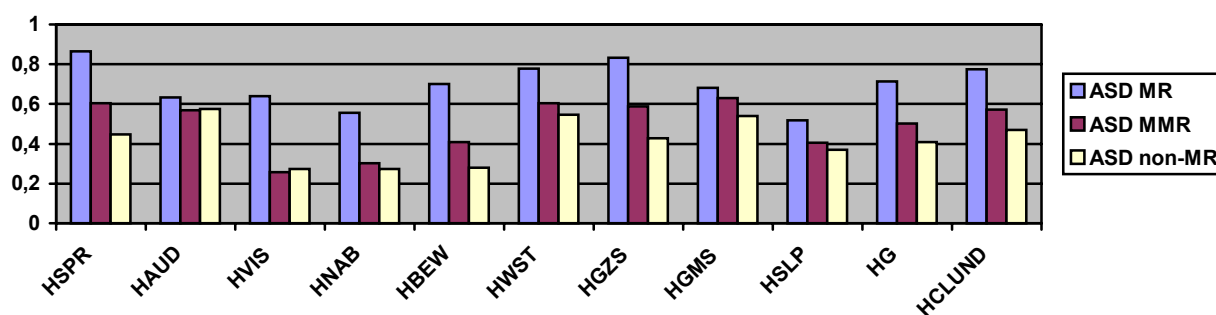
Table 4-4: One-way ANOVA of the different behavioural clusters of the HBS for the groups without MR (Non-MR) and the group with an SQ between 70 and 88 (Mild Mental Retardation; MMR) and with MR (MR)

| Item cluster | | N | Mean | SD | F | Sig |
|---------------------------------------|--------|-----|-------|-------|--------|--------|
| Abnormalities of Speech | Non-MR | 47 | .4468 | .3003 | 13.446 | .000** |
| | MR | 89 | .8636 | .5314 | | |
| | MMR | 48 | .6042 | .4643 | | |
| Abnormal Responses to Sounds | Non-MR | 59 | .5763 | .6748 | .420 | .657 |
| | MR | 230 | .6348 | .6032 | | |
| | MMR | 71 | .5704 | .5994 | | |
| Abnormal Responses to Visual Stimuli | Non-MR | 56 | .2723 | .4629 | 22.015 | .000** |
| | MR | 237 | .6403 | .5727 | | |
| | MMR | 73 | .2568 | .3460 | | |
| Abnormal Proximal Sensory Stimulation | Non-MR | 58 | .2730 | .3742 | 17.822 | .000** |
| | MR | 234 | .5556 | .4370 | | |
| | MMR | 67 | .3010 | .3018 | | |
| Abnormal Bodily Movements | Non-MR | 55 | .2805 | .2564 | 30.753 | .000** |
| | MR | 225 | .7010 | .4630 | | |
| | MMR | 68 | .4076 | .2965 | | |
| Routines and Resistance to | Non-MR | 55 | .5455 | .4981 | | |

| Item cluster | | N | Mean | SD | F | Sig |
|--|--------|-----|-------|-------|--------|--------|
| Change | MR | 232 | .7767 | .5062 | 6.665 | .001** |
| | MMR | 68 | .6059 | .4478 | | |
| Behaviour problems with limited or no Social Awareness | Non-MR | 54 | .4276 | .3609 | 25.062 | .000** |
| | MR | 211 | .8341 | .4435 | | |
| | MMR | 61 | .5872 | .3229 | | |
| Behaviour problems with Social Awareness | Non-MR | 55 | .5409 | .4684 | 1.569 | .210 |
| | MR | 183 | .6817 | .5385 | | |
| | MMR | 62 | .6290 | .5216 | | |
| Sleep Problems | Non-MR | 57 | .3684 | .6717 | 1.254 | .287 |
| | MR | 230 | .5174 | .7690 | | |
| | MMR | 69 | .4058 | .7137 | | |
| Specific Behaviour problems | Non-MR | 51 | .4079 | .3179 | 24.163 | .000** |
| | MR | 204 | .7139 | .3320 | | |
| | MMR | 59 | .5007 | .2629 | | |
| Cluster Lund | Non-MR | 53 | .4704 | .3589 | 14.479 | .000** |
| | MR | 174 | .7734 | .4223 | | |
| | MMR | 57 | .5739 | .3268 | | |

The same data is shown as a bar chart in figure 4-1, to make comparisons easier.

Figure 4-1 Mean scores for the behavioural clusters Abnormalities of Speech (HSPR), Abnormal Responses to Sounds (HAUD), Abnormal Responses to Visual Stimuli (HVIS), Abnormal Proximal Sensory Stimulation (HNAB), Abnormal Bodily Movements (HBEW), Routines and Resistance to Change (HWST), Behaviour Problems with Limited or no Social Awareness (HGZS), Behaviour Problems with Social Awareness (HGMS), Sleep Problems (HSLP), Specific Behaviour Problems (HG) and Cluster Lund (HCLUND) for the groups ASD with Mental Retardation (MR), ASD with mild Mental Retardation (MMR) and ASD without Mental Retardation (Non-MR)



As can be seen in figure 4-1, the group ASD-MR has the highest score for all of the behavioural clusters.

We performed t-tests to establish the significant differences between the ASD-MR group and the ASD-MMR group, on the one hand, and the ASD-MMR group and the ASD group without MR, on the other hand. The full description of the t-tests can be found in Appendix B.

When comparing the ASD-MR and ASD-MMR groups we found significant differences for exactly the same clusters as listed in table 4-4. The groups differ in every cluster except for the clusters 'abnormal responses to sounds' ($p=.432$), 'behaviour problems with social awareness' ($p=.503$) and 'sleep problems' ($p=.284$).

When comparing the ASD-MMR with the ASD non-MR group, we found significant differences for the clusters ‘abnormalities of speech or sign language’ ($p=.053$), ‘abnormal bodily movements’ ($p=.013$) and ‘behaviour problems with limited or no social awareness’ ($p=.014$). So the ASD-MMR group is similar to the ASD non-MR group in eight out of the eleven clusters and the ASD-MMR group differs in eight out of the eleven clusters from the ASD-MR group.

Summarizing, we note that the groups with MR and MMR and the group without MR differ in eight of the eleven clusters. These results verify the assumption already found in literature that children with an ASD and MR show more challenging behaviour than children with ASD without MR. In other words: MR is a contributing factor to the presence of challenging behaviour of children with ASD. The two groups, MR and non-MR, have totally different profiles and are considered to be different subgroups. The group with MMR has a different pattern; therefore we decided to use a very strict cut-off point, when $SQ \leq .70$, for further analyses.

4.5 Results of the MR group across the different behavioural clusters of the HBS

In this section we will look at the following six factors namely: chronological age, social age (HBS composite score), sex, level of functioning (social age/chronological age), level of communication and level of social interaction. These factors may contribute to challenging behaviour of children with an ASD and MR. We have calculated Pearson Correlations and will only report on the significant correlations ($p \leq .05$).

We performed forward regression analyses to determine the explained variance of the factors. For each factor the results are described in the following sections. Full descriptions of the analyses can be found in Appendix B.

4.5.1 The relationship between chronological age and the behavioural clusters of the HBS

The significant correlations between chronological age and the different behavioural clusters of the HBS are shown in table 4-5.

Table 4-5: The relationship between chronological age and the behavioural clusters ($p \leq .05$)

| | Abnormalities of Speech | Behaviour problems with Social Awareness | Sleeping Problems | Cluster Lund |
|---------------------|-------------------------|--|-------------------|--------------|
| Pearson Correlation | -.426 | .182 | .168 | .175 |
| p-value | .000 | .014 | .011 | .022 |
| N | 88 | 181 | 228 | 172 |

As the chronological age of children with an ASD and MR increases, the abnormalities of speech diminish. Sleeping problems, behaviour problems with social awareness and the behaviours clustered according to Lund increase with the age of the child.

Using forward regression analysis with the behavioural clusters as dependent variables and the six factors as independent variables, we found that chronological age explains 2.9% of the variance in the cluster abnormal proximal sensory stimulation and 3.8 % of the variance in the cluster sleeping problems ($F \leq .50$ to enter).

4.5.2 The relationship between social age and the behavioural clusters of the HBS

As we can see in table 4-6 abnormalities of speech, abnormal responses to sounds, abnormal response to visual stimuli, abnormal proximal sensory stimulation and abnormal bodily movements decrease, when the social age increases. Behaviour problems with social awareness increase as the social age increases.

Table 4-6: The relationship between social age and the behavioural clusters ($\leq .05$)

| | Abnormalities of Speech | Abnormal Responses to Sound | Abnormal Response to Visual Stimuli | Abnormal Proximal Sensory Stimulation | Abnormal Bodily Movements | Behaviour problems with Social Awareness |
|---------------------|-------------------------|-----------------------------|-------------------------------------|---------------------------------------|---------------------------|--|
| Pearson Correlation | -.550 | -.134 | -.271 | -.279 | -.213 | .196 |
| p-value | .000 | .043 | .000 | .000 | .001 | .008 |
| N | 88 | 228 | 235 | 232 | 223 | 181 |

4.5.3 The relationship between sex and the behavioural clusters of the HBS

As sex is a dichotomous variable the difference between boys and girls is calculated with a t-test. Only one significant difference was found: the girls score higher in the cluster abnormalities of speech ($p=.007$).

4.5.4 The relationship between level of functioning and the behavioural clusters of the HBS

Level of functioning (SQ) is determined by calculating CA/SA. Level of functioning exhibits a significant negative correlation with nine out of the 11 clusters. If the SQ rises, the behaviour problems decrease. Only the clusters routines and resistance to change and behaviour problems with social awareness do not show a significant correlation with level of functioning.

Table 4-7: The relationship between level of functioning and the behavioural clusters Abnormalities of Speech (HSPR), Abnormal Response to Sounds (HAUD), Abnormal Response to Visual Stimuli (HVIS), Abnormal Proximal Sensory Stimulation (HNAB), Abnormal Bodily Movements (HBEW), Behaviour problems with limited or no Social Awareness (HGZS), Sleeping Problems (HSLP), Specific Behaviour problems (HG) and Cluster Lund (HCLund) of the HBS (p≤ .05)

| | HSPR | HAUD | HVIS | HNAB | HBEW | HGZS | HSLP | HG | HCLund |
|---------------------|-------|-------|-------|-------|-------|-------|-------|-------|--------|
| Pearson Correlation | -.305 | -.176 | -.249 | -.414 | -.349 | -.191 | -.154 | -.281 | -.151 |
| p-value | .004 | .008 | .000 | .000 | .000 | -.006 | .020 | .000 | .048 |
| N | 88 | 228 | 235 | 232 | 223 | 209 | 228 | 202 | 172 |

Using forward regression analysis with the behavioural clusters as dependent variables and the six factors as independent variables we found that level of functioning (SQ) explains 3.3% of the variance in the cluster abnormal response to sounds, 11.9% of the variance in the cluster abnormal response to visual stimuli, 20.5% of the variance in the cluster abnormal proximal sensory stimulation, 14.8% of the variance in the cluster abnormal bodily movements, 3.8% of the variance in the cluster behaviour problems with limited or no social awareness, 4.4% of the variance in the cluster behaviour problems with social awareness, 7.5 % of the variance in the cluster specific behaviour problems and 4% of the variance in the cluster according to Lund ($F \leq .50$ to enter).

4.5.5 The relationship between level of communication and the behavioural clusters of the HBS

Table 4-8: The relationship between level of communication and the behavioural clusters (p≤ .05)

| | Abnormalities of Speech | Abnormal Response to Visual Stimuli | Abnormal Proximal Sensory Stimulation | Abnormal Bodily Movements | Behaviour problems with Social Awareness |
|---------------------|-------------------------|-------------------------------------|---------------------------------------|---------------------------|--|
| Pearson Correlation | -.681 | -.254 | -.270 | -.193 | .288 |
| p-value | .000 | .000 | .000 | .005 | .000 |
| N | 88 | 221 | 219 | 210 | 169 |

The increase in the level of communication, in particular speech, coincides with a decrease of abnormalities in speech, abnormal response to visual stimuli, abnormal proximal sensory stimulation and abnormal bodily movements. Behaviour problems with social awareness increase when the level of communication increases.

Using forward regression analysis with the behavioural clusters as dependent variables and the six factors as independent variables we found that the level of communication explains 55.3% of the variance in the cluster abnormalities of speech, 4.1% of the variance in the cluster routines and resistance to change and 4.8 % of the variance in the cluster behaviour problems with social awareness ($F \leq .50$ to enter).

4.5.6 The relationship between level of socialization and the behavioural clusters of the HBS

Table 4-9: The relationship between level of socialization and the behavioural clusters (p ≤ .05)

| | Abnormalities of Speech | Abnormal Response to Visual Stimuli | Abnormal Proximal Sensory Stimulation | Abnormal Bodily Movements |
|---------------------|-------------------------|-------------------------------------|---------------------------------------|---------------------------|
| Pearson Correlation | -.433 | -.174 | -.202 | -.248 |
| P-value | .004 | .044 | .020 | .005 |
| N | 42 | 134 | 132 | 130 |

When the level of socialization increases, abnormalities of speech, abnormal responses to visual stimuli, abnormal proximal sensory stimulation and abnormal bodily movements decrease.

Using forward regression analysis with the behavioural clusters as dependent variables and the six factors as independent variables we found that the level of socialization explains 2.4% of the variance in the cluster routines and resistance to change ($F \leq .50$ to enter).

4.6 Conclusion

The primary analyses showed that the group with MMR shows a very different pattern of challenging behaviour. The analyses of variance were therefore performed on the group with an $SQ \leq .70$.

The results are summarized in table 4-10.

Table 4-10: Explained variance of chronological age, social age, SQ, sex, level of communication, level of socialization and total explained variance of the behavioural clusters¹

| | Chronological Age | Social Age | Social age eEquivalent | Sex | Level of Communication | Level of Socialization |
|------|-------------------|------------|------------------------|------------|------------------------|------------------------|
| HSPR | - 55.3% | - | - | Girls>Boys | - | - |
| HAUD | | - | - 3.3% | | | |
| HVIS | | - | - 11.9% | | - | - |
| HNAB | 2.9% | - | - 20.5% | | - | - |
| HBEW | ¹ | - | - 14.8% | | - | - 2.5% |
| HWST | | | | | 4.1% | 2.4% |
| HGZS | | | - 3.8% | | | |

¹ Significant negative correlations are marked (-), positive correlations are marked (+)

| | Chronological Age | Social Age | Social age eQuivalent | Sex | Level of Communication | Level of Socialization |
|--------|-------------------|------------|-----------------------|-----|------------------------|------------------------|
| HGMS | - 4.5% | + | | | + | 5.1% |
| HSLP | + | | - | | | |
| HG | | | - 7.5% | | | |
| HCLund | + | | - 4.0% | | | |

¹ Significant negative correlations are marked (-), positive correlations are marked (+)

The behaviour problems directly associated with the ASD's, namely specific reactions to sensory stimuli (HAUD, HVIS, HNAB, HBEW), diminish when the child develops positively, thus as the social age increases. The other group of specific behaviour problems (HWST and HG) seems to be more or less independent of the factors we have examined. The non-specific behaviour problems (HGMS, HSLP, and HCLund) yielded mixed results. They decrease when the level of functioning (SQ) increases, but increase with chronological and social age.

In conclusion, we can explain a reasonable amount of variance of the behavioural clusters involving specific behaviour. However, we can only explain a fraction of the variance across the behavioural clusters of the HBS with regard to the non-specific behaviour problems. The non-specific behaviour problems include the most severe problems, e.g. self-injurious behaviours, aggression, destructive behaviours, etc. The factor SQ explains the highest proportion of variance across several behavioural domains.

These results only strengthen the need for further research.

In phase II of the study we included these factors in the selection criteria for the sample. Level of functioning and and chronological age are the main matching criteria.

5 Results of Phase II

5.1 Introduction

In this chapter we will summarize the results of phase II of the study. The chapter is divided into three parts. In the first part we will describe the scores across the different instruments for the group with ASD and MR and extreme challenging behaviour (ASD/MR+) and the control group of children with ASD and MR without extreme challenging behaviour (ASD/MR). We will also look into the distribution of the data, more precisely whether the distributions are normal. We will also describe the information obtained from file analysis and some qualitative information. In the second part we will look at the differences between the two groups and establish whether the differences between the groups are of significance. In the third part we will use some analyses to explore the influences of the different variables on the presence of challenging behaviour.

5.2 Description of the variables

In this part we will start with a description of the history and family background, followed by present information on behaviour, level of functioning and medical issues. We will describe both groups to obtain an overall picture of the two groups.

5.2.1 History

Information on the history of the subjects was gathered by means of file analyses (see 3.7.3) and a standardized interview with the parents, using the DISCO I (Wing, 1993a, see 3.6.6.).

5.2.1.1 Prenatal History

The heading prenatal history covers the family history and information about the pregnancy.

Family history

The DISCO I (Wing, 1993a) includes questions about relevant conditions in any blood relations. The interviewer can fill in the diagnosis, whether there is a known aetiological cause and what the relationship with the child with an Autistic Spectrum Disorder is. We collected data on two family members (I and II). If the parents mentioned more family members, we have presented the data on the two closest family relations.

Table 5-1 Family Background: Diagnosis of family members I and II (N=20)^a

| Diagnosis | ASD/MR+ | ASD/MR+ | ASD/MR | ASD/MR | Total | Total |
|-----------|---------|---------|--------|--------|-------|-------|
| | I | II | I | II | I | II |
| None | 9 | 16 | 11 | 17 | 20 | 33 |

| Diagnosis | ASD/MR+ | ASD/MR+ | ASD/MR | ASD/MR | Total | Total |
|----------------------------|---------|---------|--------|--------|-------|-------|
| | I | II | I | II | I | II |
| PDD | 3 | 1 | 2 | 0 | 5 | 1 |
| Specific Learning Disorder | 1 | 1 | 1 | 0 | 2 | 1 |
| Mental Retardation | 4 | 0 | 3 | 1 | 7 | 1 |
| Psychiatric disorder | 1 | 2 | 1 | 1 | 2 | 3 |
| Other | 2 | 0 | 2 | 1 | 4 | 1 |
| Total | 20 | 20 | 20 | 20 | 40 | 40 |

^a For all 20 cases in each group the data was gathered on 2 family members, or 40 people in total

For eleven children from the group with extreme challenging behaviour at least one family member had a psychiatric diagnosis, for the group without challenging behaviour nine members of the immediate family had some kind of psychiatric diagnosis. For nine cases of the group with challenging behaviour and three of the group without, two family members had a psychiatric disorder.

From the file analysis we learn that ‘autism in the family’ occurred three times in the group with challenging behaviour and twice in the group without. ‘MR in the family’ occurred four times in the group with challenging behaviour and three times in the group without.

Table 5-2 Family Background: Cause of problems for family members I and II (N=20) ^a

| Cause | ASD/MR+ | ASD/MR+ | ASD/MR | ASD/MR | Total | Total |
|---------------------|---------|---------|--------|--------|-------|-------|
| | I | II | I | II | I | II |
| No cause known | 10 | 17 | 14 | 18 | 24 | 35 |
| Tuberosis sclerosis | 0 | 0 | 1 | 0 | 1 | 0 |
| Family history | 2 | 1 | 0 | 0 | 2 | 1 |
| Autism | 1 | 0 | 0 | 0 | 1 | 0 |
| Down’s Syndrome | 1 | 1 | 0 | 0 | 1 | 1 |
| Other Genetic Cause | 1 | 0 | 1 | 0 | 2 | 0 |
| Infections | 0 | 0 | 1 | 1 | 1 | 1 |
| Other | 5 | 1 | 3 | 1 | 8 | 2 |
| Unknown | | | | | | |
| Total | 20 | 20 | 20 | 20 | 40 | 40 |

^a For all 20 cases in each group the data was gathered on 2 family members, or 40 people in total

For ten family members of the group with challenging behaviour and six members of the group without, known causes were given for the first family member. In five cases for the group with challenging behaviour and three cases of the group without, an underlying problem is presumed, but the specific cause is not yet known. For the second family member the cause was known in only three cases from the group with challenging behaviour and two cases from the group without.

Table 5-3 Family Background: Relationship to Object^a

| Relationship | ASD/MR+ | | ASD/MR | | Total | |
|------------------------------|---------|----|--------|----|-------|----|
| | I | II | I | II | I | II |
| No diagnosis | 9 | 16 | 10 | 17 | 19 | 33 |
| Sibling | 2 | 0 | 5 | 0 | 7 | 0 |
| Mother | 1 | 0 | 1 | 0 | 2 | 0 |
| Father | 0 | 1 | 0 | 0 | 0 | 1 |
| Grandparent maternal side | 0 | 0 | 1 | 0 | 1 | 0 |
| Grandparent paternal side | 1 | 1 | 1 | 0 | 2 | 1 |
| Other maternal | 2 | 1 | 1 | 2 | 3 | 3 |
| Other paternal | 5 | 1 | 1 | 1 | 6 | 2 |
| Total | 20 | 20 | 20 | 20 | 40 | 40 |

^a For all 20 cases in each group the data was gathered on 2 family members, or 40 people in total

For the group with challenging behaviour two siblings had a psychiatric diagnosis, in the group without, five siblings had a psychiatric diagnosis. The parents were mentioned twice in the group with challenging behaviour and once in the group without.

These questions often triggered a discussion between the parents about all kinds of odd family members within the respective families. Only family members with official diagnoses, who sought help for their problems, have been included.

Family

The DISCO I (Wing, 1993a) included questions about the composition of the family. Seventeen children of the group of children with an ASD and MR with challenging behaviour lived with both their biological father and biological mother. For the group without challenging behaviour that number was 18. In each of the groups one child was cared for by two parents, one of whom was the biological mother; in each group one child also lived with only the biological mother. In the group with challenging behaviour, one child lived with her grandparents.

In the files we found that in each group one couple got divorced in the first year of the life of the child; in each group one couple got divorced when the child was five years old or older. The parents of the child who lives with the grandparents were also divorced.

Table 5-4 Number of Biological Parents' Children

| Number of children | ASD/MR+ | ASD/MR | Total |
|--------------------|---------|--------|-------|
| 1 | 3 | 3 | 6 |
| 2 | 12 | 13 | 25 |
| 3 | 5 | 3 | 8 |
| 5 | 0 | 1 | 1 |
| Total | 20 | 20 | 40 |

In each of the two groups three children were the only child of both of their biological parents. Most children had one sibling, 12 in the group with challenging

behaviour and 13 in the group without. In the ASD/MR+ group five children had two siblings, in the group without three. In the group without challenging behaviour one child had four siblings.

When we look at the data for the biological mothers' children, we see a slightly different picture. In each of the groups four children are the only child of their mother. In the group with challenging behaviour 11 children have one sibling, in the group without 12. Five mothers of the group with challenging behaviour have three children and two mothers of the group without have the same number. In the group children without challenging behaviour there is one mother with four children and one mother with five children.

In the group children with challenging behaviour, nine children are the oldest children, nine are the second and two are the third child of their respective mothers. In the group of children without challenging behaviour six are the oldest child, 13 the second and one child is the fifth child of its mother.

All the siblings lived within the family at the time of the interview with the parents.

Pregnancy

The DISCO I (Wing, 1993a) contains questions about concerns during pregnancy. In this case therefore the question was not whether a doctor had diagnosed problems, but whether the mother perceived problems.

Table 5-5 Pre-natal Concerns

| Problems | ASD/MR+ | ASD/MR | Total |
|---------------|---------|--------|-------|
| No Problems | 17 | 15 | 32 |
| Some Problems | 3 | 5 | 8 |
| Total | 20 | 20 | 40 |

Three mothers of the group of children with challenging behaviour mentioned problems during pregnancy. In all of these cases the pregnancy was called 'unplanned' by the mother. In one case hyperbilirubinaemia had been diagnosed. In the group without challenging behaviour five mothers perceived problems during pregnancy. Hyperbilirubinaemia was mentioned three times in this group.

Table 5-6 Gestation, Birth weight and Mother's age at birth

| | Group | N | Mean | SD | SEM |
|-------------------------------|---------|----|-------|------|------|
| Gestation in months | ASD/MR+ | 17 | 39.47 | .80 | .19 |
| | ASD/MR | 18 | 37.11 | 7.70 | 1.81 |
| Birth weight in kg. | ASD/MR+ | 13 | 3.46 | .58 | .16 |
| | ASD/MR | 6 | 3.52 | .65 | .27 |
| Mothers age at birth in years | ASD/MR+ | 14 | 28.43 | 4.13 | 2.06 |
| | ASD/MR | 17 | 30.88 | 3.72 | .95 |

The information about birth weight is missing in several cases, mainly because the parents did not remember the exact birth weight and it could not be found in the files. The standard deviations for 'gestation' differ immensely. In the group without challenging behaviour three children were born (very) early compared to none in the ASD/MR+ group. In both groups four children were born 'late', but gestation never exceeded 43 weeks.

5.2.1.2 Perinatal History

The DISCO I (Wing, 1993a) includes a question about problems during birth.

Table 5-7 Perinatal Problems

| Problems | ASD/MR+ | ASD/MR | Total |
|---------------|---------|--------|-------|
| No Problems | 13 | 11 | 24 |
| Some Problems | 7 | 9 | 16 |
| Total | 20 | 20 | 40 |

Seven mothers of the group of children with challenging behaviour reported problems during birth. From the files we learn that two children were delivered by a C-section and two by vacuum extraction. One child needed oxygen immediately after birth. Of the group of children without challenging behaviour two children were delivered by C-section and three by vacuum extraction. Four children needed oxygen. In each group meconium was seen in the amniotic fluid of one child.

Parents often do not remember the Apgar scores. One parent of the group of children with challenging behaviour remembered that it was very low.

5.2.1.3 Postnatal History

The DISCO I (Wing, 1993a) includes questions about the period immediately after birth.

Table 5-8 Postnatal Problems

| Problems | ASD/MR+ | ASD/MR | Total |
|---------------|---------|--------|-------|
| No Problems | 10 | 12 | 22 |
| Some Problems | 10 | 7 | 17 |
| Total | 20 | 19 | 39 |

Ten parents of the group of children with challenging behaviour mentioned problems shortly after birth. In the files we find: 'feeding problems' (3x), 'child is blue' (3x), and 'child is yellow' (2x). In addition cardiac problems, constipation and an immediate operation were reported. One child was placed in an incubator. Seven parents of the group of children without challenging behaviour encountered problems shortly after birth. In the files we find: 'feeding problems' (2x), 'child was placed in incubator' (4x), 'child is yellow' (2x) and 'motor delay' (2x). Also

mentioned were ‘child is blue’, ‘an infection’, ‘child is very big’ and ‘problems in brain’. For one child Down’s syndrome was recognized immediately after birth.

5.2.1.4 Present Medical Status

The DISCO I (Wing, 1993a) contains several items on medical problems during childhood.

Additional problems

Four separate questions concerned the presence of epilepsy, hearing impairment, visual impairment and other additional problems during development.

Table 5-9 Presence of Epilepsy, Hearing Impairment, Visual Impairment and Other Additional Problems

| | ASD/MR+ | ASD/MR | Total |
|---------------------------|---------|--------|-------|
| Epilepsy | 4 | 7 | 11 |
| Hearing Impairment | 6 | 5 | 11 |
| Visual Impairment | 9 | 9 | 18 |
| Other Additional Problems | 17 | 15 | 32 |
| Total | 36 | 36 | 72 |

Four children from the group with challenging behaviour and seven children from the group without challenging behaviour have suffered epileptic seizures at one point or another. Six children from the group with challenging behaviour and five children of the group without have (had) auditory problems. From the files we learn that the most common problem was frequent ear infections, often leading to the placement of tubes. Nine children from each group had visual problems, the most common problem being that they needed glasses.

Seventeen children of the group with challenging behaviour and 16 children of the group without have had additional problems at one point or another. The most frequently named in the files were bowel problems (5x in group with challenging behaviour and 1x in group without), infections (7x in group with challenging behaviour and 4x in group without) and motor problems (2x in group with challenging behaviour and 4x in group without).

Medical examinations

The children in both groups have undergone numerous examinations, mainly carried out to discover the cause of their problems. The DISCO I (Wing, 1993a) includes questions about whether certain examinations have been carried out and whether the results were positive or not.

Table 5-10 Medical Examinations

| | ASD/MR+ | ASD/MR | Total |
|-------------------|---------|--------|-------|
| EEG | 16 | 16 | 32 |
| Cat-scan | 13 | 13 | 26 |
| Other brain | 13 | 14 | 27 |
| Fra-X | 16 | 13 | 29 |
| Other chromosomes | 16 | 15 | 31 |
| Blood | 14 | 13 | 27 |

| | | | |
|---------|----|----|----|
| Urine | 15 | 13 | 28 |
| Hearing | 19 | 16 | 35 |
| Vision | 19 | 15 | 34 |
| Other | 10 | 8 | 18 |

Numerous medical examinations have been carried out in both groups. For most children brain function, the presence of chromosomal disorders, the presence of metabolic disorders and the presence of hearing and visual problems were examined. For the group with challenging behaviour a mean of 7.00 (SD=2.87) medical examinations per child were performed, for the group without 6.35 (SD=2.87). The results of the examinations are presented in the next table.

Table 5-11 Positive Results Medical Examinations

| | ASD/MR+ | ASD/MR | Total |
|-------------------|---------|--------|-------|
| EEG | 4 | 9 | 13 |
| Cat-scan | 2 | 7 | 9 |
| Other brain | 3 | 7 | 10 |
| Fra-X | 0 | 0 | 0 |
| Other chromosomal | 1 | 3 | 4 |
| Blood | 1 | 2 | 3 |
| Urine | 0 | 2 | 2 |
| Hearing | 3 | 5 | 8 |
| Vision | 7 | 4 | 11 |
| Other | 2 | 2 | 4 |

For the group with challenging behaviour more examinations were carried out, but the group without challenging behaviour exhibited more positive results. In total a mean of 1.15 (SD=1.14) positive results were obtained for the group with challenging behaviour, for the group without the mean was 2.25 (SD=.52). Nine children of the group without challenging behaviour and four children of the group with challenging behaviour had an abnormal EEG. For seven children of the group without challenging behaviour and two children in the group without, malformations of the brain were diagnosed. For one child of the group with challenging behaviour and three children of the group without, a chromosomal disorder was diagnosed. Each group contained one child with Down's syndrome; two children in the group with challenging behaviour were suspected of having Williams Beuren syndrome. One child in the group with challenging behaviour was suspected of having Angelman's syndrome. One child in the group without challenging behaviour had tuberous sclerosis. For the other children the parents were told that there was a chromosomal disorder, but the name was not known by the parents and could not be found in the files.

Use of Medication

In the following table the medications prescribed for the two groups are described.

Table 5-12 Use of Medication

| | ASD/MR+ | ASD/MR | Total |
|---------------------|---------|--------|-------|
| Major tranquillizer | 7 | 2 | 9 |

| | ASD/MR+ | ASD/MR | Total |
|---------------------|---------|--------|-------|
| Minor tranquillizer | 1 | 1 | 2 |
| Antidepressant | 1 | 0 | 1 |
| Anticonvulsant | 0 | 4 | 4 |
| Other | 5 | 5 | 10 |
| Total | 14 | 12 | 25 |

In total 11 children from the group with challenging behaviour and eight children from the group without took one or more forms of medication. For the group with challenging behaviour tranquillizers were prescribed the most, for the group without challenging behaviour anticonvulsants.

5.2.1.5 Developmental History

In this part we will report on the developmental history of the two groups. DISCO I included questions about the developmental milestones in the first year of life. First we will consider behaviour in the first two years of life. Parents were asked whether they were concerned about various aspects of behaviour. The results are shown in table 5-13.

Table 5-13 Behaviour during Infancy

| Behaviour | Worries | ASD/MR+ | ASD/MR | Total |
|-------------------------------|------------------------------------|---------|--------|-------|
| Feeding | Problems up to 6 weeks | 1 | 2 | 3 |
| | Problems continued after 6 weeks | 4 | 4 | 8 |
| Vomiting | Minor problem | 0 | 1 | 1 |
| | Major problem | 1 | 1 | 2 |
| Excessive crying/screaming | Minor problem | 1 | 1 | 2 |
| | Major problem | 5 | 4 | 9 |
| Woke up screaming | Minor problem | 1 | 0 | 1 |
| | Major problem | 4 | 3 | 7 |
| Sleeping pattern | Minor problem | 2 | 2 | 4 |
| | Major problem | 9 | 7 | 16 |
| Unusually good/quiet/passive | Minor problem | 0 | 3 | 3 |
| | Marked problem | 11 | 11 | 22 |
| Demand for social attention | Sometimes | 2 | 4 | 6 |
| | Did not ask | 14 | 14 | 28 |
| Response to cuddling | Passively accepted | 2 | 4 | 6 |
| | Stiff and non-responsive | 12 | 8 | 20 |
| Eye contact | Some eye contact | 5 | 3 | 8 |
| | No eye contact | 10 | 13 | 23 |
| Getting ready to be picked up | Some anticipation | 4 | 4 | 8 |
| | No anticipation | 13 | 13 | 26 |
| Reciprocation in baby games | Enjoyed, no reciprocation | 4 | 5 | 9 |
| | No interest or enjoyment | 14 | 13 | 27 |
| Waving bye-bye | Had to be prompted up to age 3 yrs | 0 | 0 | 0 |
| | Did not wave bye-bye | 16 | 18 | 34 |
| Babbling | Less than expected | 8 | 7 | 15 |
| | Little or no babbling | 9 | 10 | 19 |
| Pre-speech 'conversation' | Occasional | 5 | 4 | 9 |
| | Did not take turns | 14 | 16 | 30 |

| Behaviour | Worries | ASD/MR+ | ASD/MR | Total |
|----------------------------|------------------------------|---------|--------|-------|
| Response to speech | Variable response | 2 | 4 | 6 |
| | Marked lack of response | 14 | 14 | 28 |
| Intoned pre-speech sounds | Some intonation | 4 | 6 | 10 |
| | No intoned pre-speech sounds | 13 | 13 | 26 |
| Interest in environment | Some interest | 2 | 2 | 4 |
| | Little or no interest | 16 | 15 | 31 |
| Looked when others pointed | Occasional | 1 | 0 | 1 |
| | Did not look | 17 | 18 | 35 |
| Joint referencing | Occasional sharing | 0 | 1 | 1 |
| | Did not share interests | 18 | 18 | 36 |
| Special Interests | Minor | 3 | 4 | 7 |
| | Marked | 15 | 12 | 27 |
| Attachment to one parent | Minor | 3 | 3 | 6 |
| | Marked | 8 | 3 | 11 |
| Dislike of care procedures | Minor | 3 | 0 | 3 |
| | Marked | 6 | 2 | 8 |
| Oversensitive to noise | Minor | 1 | 6 | 7 |
| | Marked | 12 | 5 | 17 |
| Odd movements | Minor | 0 | 2 | 2 |
| | Marked | 13 | 5 | 18 |
| Other problems | Minor | 0 | 0 | 0 |
| | Marked | 2 | 0 | 0 |

The children in both groups showed marked problems in the first year of life as far as the items for specific autistic features are concerned, e.g. joint attention, speech, and responsiveness.

Table 5-14 Developmental Milestones

| Developmental domain | Time of achievement | ASD/MR+ | ASD/MR | Total |
|------------------------|---------------------|---------|--------|-------|
| Independent Sitting | Normal | 11 | 8 | 19 |
| | Late | 9 | 12 | 21 |
| Crawling | Normal | 11 | 8 | 19 |
| | Late | 8 | 11 | 19 |
| | Still not able | 1 | 1 | 2 |
| Walking | Normal | 10 | 9 | 19 |
| | Late | 10 | 10 | 20 |
| | Still not able | 0 | 1 | 1 |
| Riding tricycle | Normal | 8 | 6 | 14 |
| | Late | 6 | 11 | 17 |
| | Still not able | 6 | 3 | 9 |
| Riding bicycle | Normal | 6 | 5 | 11 |
| | Late | 4 | 8 | 12 |
| | Still not able | 10 | 7 | 17 |
| Clean and dry in day | Normal | 1 | 2 | 3 |
| | Late | 6 | 10 | 16 |
| | Still not able | 13 | 8 | 21 |
| Clean and dry at night | Normal | 1 | 1 | 2 |
| | Late | 7 | 8 | 15 |
| | Still not able | 12 | 11 | 23 |

The children in both groups were very delayed in achieving some basic self-help skills. The motor skills were developed within the normal range for about half of the children in both groups.

The parents could answer the questions about whether they were worried very easily, but for the developmental milestones specific ages in months were requested. A majority of the parents were not able to answer such questions, so the answers were clustered into the categories: normal/late and still not able.

5.2.1.6 *Age of Recognition of Problems*

In this paragraph the questions about age of onset, from the point of view of the researcher, and the age at which the parents first became worried are described. Another question of the DISCO I deals with the onset of untypical behaviour. Finally the age at which an Autistic Spectrum Disorder was first diagnosed is considered.

Table 5-15: Age of Recognition of Problems

| | Group | N | Mean | SD | SEM |
|--|---------|----|-------|-------|------|
| Age of first worries in months | ASD/MR+ | 20 | 9.85 | 10.61 | 2.37 |
| | ASD/MR | 20 | 10.20 | 10.42 | 2.33 |
| Age of first problems in months | ASD/MR+ | 20 | 2.35 | 6.83 | 1.52 |
| | ASD/MR | 20 | 4.40 | 8.60 | 1.92 |
| Age of onset in months | ASD/MR+ | 20 | 16.90 | 16.51 | 3.69 |
| | ASD/MR | 20 | 11.45 | 8.51 | 1.90 |
| Age of onset untypical behaviour (rater) in months | ASD/MR+ | 20 | 4.45 | 9.70 | 2.17 |
| | ASD/MR | 20 | 3.45 | 7.42 | 1.66 |
| Age at diagnosis in months | ASD/MR+ | 20 | 30.80 | 21.63 | 4.84 |
| | ASD/MR | 20 | 40.50 | 26.02 | 5.82 |

Most parents started worrying about the development of their child before the first birthday. In retrospect they give an even earlier start of the first problems. The age of onset according to the parents was older than the age presumed by the researcher. The age of a diagnosis within the autistic spectrum was lower for the group with challenging behaviour than for the group without. Most children had been diagnosed before the age of four years.

The standard deviation for age of onset in the ASD/MR+ group is very high. Four children in that group are suspected of 'late onset' (after 36 months) compared to one in the ASD/MR group.

Parents often mentioned a setback in development as the starting point for the problems. This will be discussed in the next paragraph.

5.2.1.7 *Setback in development of skills*

Parents were asked whether or not there was a setback in skills, and at what age. In the columns of table 5-16 the number of children is listed who suffered a setback in a particular area. The data was available for all children.

Table 5-16: Setback in development of skills

| Developmental area | ASD/MR+ | ASD/MR | Total |
|-----------------------------|---------|--------|-------|
| Motor Development | 4 | 6 | 10 |
| Toilet Training | 4 | 0 | 4 |
| Other self care | 2 | 0 | 2 |
| Language/Communication | 9 | 4 | 13 |
| Play | 5 | 2 | 7 |
| Social Interaction | 6 | 6 | 12 |
| Feeding | 5 | 0 | 5 |
| Other adaptive Skills | 8 | 0 | 8 |
| Total (Setback in any area) | 15 | 10 | 25 |

A setback has been described more often for children with challenging behaviour than for those without. A setback in language and/or communication skills was reported most often. The category ‘other adaptive skills’ has only positive answers for the group with challenging behaviour. In all cases it was a setback in behaviour, especially obeying rules.

In total a setback in one or more developmental areas was found for 15 children with challenging behaviour and ten children without. Especially in the group with challenging behaviour the setback occurred in more than one domain.

For two children of the ASD/MR+ group West syndrome (early-onset epilepsy) was mentioned as the cause of the setback. In both groups one child had severe problems in the brain, resulting in brain surgery that was named as the cause of the setback. For all of the other children no cause has been found.

5.2.2 Socio-economic Status of the family

The DBC (Einfeld & Tonge, 1995) contained questions about the country of origin of the biological parents, the educational level and the level of the current job.

Table 5-17 Ethnicity of the biological parents

| | ASD/MR+ | | ASD/MR | | Total |
|-----------|---------|--------|--------|--------|-------|
| | Mother | father | mother | father | |
| Dutch | 19 | 17 | 16 | 14 | 66 |
| Non-Dutch | 1 | 3 | 4 | 6 | 14 |
| Total | 20 | 20 | 20 | 20 | 80 |

When we look at the group of children with challenging behaviour, we see that one mother is not of Dutch origin, both parents of this particular child are from Turkey. The other non-Dutch fathers are of Romanian and Indonesian descent. In the group of children without challenging behaviour, three couples are from Turkey, Morocco and Pakistan, respectively. One mother is from Indonesia. Three fathers are from Italy, Japan and Ghana, respectively.

All of the children in the group with challenging behaviour were born in the Netherlands. Of the group without challenging behaviour one child was born in

Malaysia and one in Italy. Both of these children have a Dutch mother and the Dutch nationality.

Both groups are mixed, as far as ethnicity is concerned.

In table 5-18 the level of education of the biological parents is given. We have arranged the information in the following categories: 'low' means no education or only primary school, 'medium' means secondary education and 'high' means at least a bachelor's degree.

Table 5-18 Level of Education of the biological parents^a

| | ASD/MR+ | | ASD/MR | | Total |
|---------|---------|--------|--------|--------|-------|
| | mother | father | mother | father | |
| High | 4 | 4 | 4 | 4 | 16 |
| Medium | 10 | 9 | 8 | 6 | 33 |
| Low | 6 | 5 | 8 | 8 | 27 |
| Unknown | 0 | 2 | 0 | 2 | 4 |
| Total | 20 | 20 | 20 | 20 | 80 |

^a We have clustered the level of education as follows:

High: at least a bachelor's degree

Medium: at least High School or vocational training

Low: Primary Education or less

This information is unknown for two fathers in each group. These fathers do not live with their children. In both groups four fathers and four mothers have at least a bachelor's degree. In the group of children with challenging behaviour six mothers and five fathers followed only primary school or less. In the group without challenging behaviour eight mothers and eight fathers attended only primary school or less.

The level of education of the parents is mixed for both groups.

Table 5-19 Current job level of the biological parents^a

| | ASD/MR+ | | ASD/MR | | Total |
|---------|---------|--------|--------|--------|-------|
| | mother | father | Mother | father | |
| High | 4 | 5 | 5 | 6 | 20 |
| Medium | 9 | 9 | 7 | 8 | 33 |
| Low | 0 | 3 | 2 | 2 | 7 |
| No job | 7 | 1 | 6 | 2 | 16 |
| Unknown | 0 | 2 | 0 | 2 | 4 |
| Total | 20 | 20 | 20 | 20 | 80 |

^a We have clustered the level of education as follows:

High: at least a bachelor's degree

Medium: at least High School or vocational training

Low: Primary Education or less

The data are missing for the four fathers who do not live with their children. The three fathers who do not have a job are all unable to work due to chronic illnesses.

The mothers who do not have a job are housewives. Almost all of the fathers work fulltime, whereas most of the mothers work part-time for at least two days a week. Most of the parents hold jobs according to their level of education.

5.2.3 Current Functioning

In the following paragraphs we will discuss the information gathered about current behaviour.

We will describe the scores of the children for the various instruments in the following areas: level of functioning, challenging behaviour, child-rearing situation and parental stress. We will also describe some qualitative information, gathered through observations.

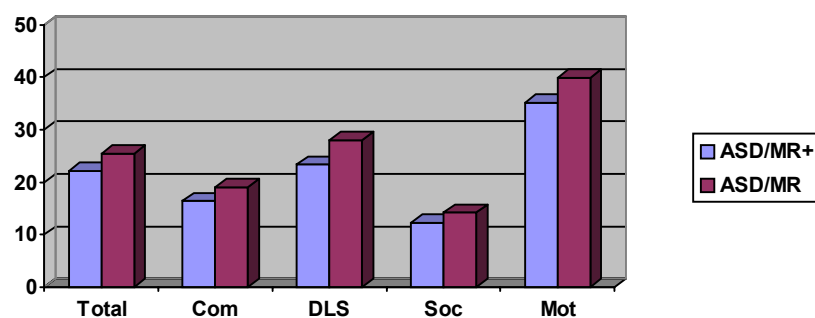
5.2.3.1 Level of Functioning

The level of functioning was measured with several instruments that cover different domains. Cognitive functioning was measured by the PsychoEducational Profile Revised (Schopler et al., 1990), adaptive functioning by the Vineland Adaptive Behavior Scales (Sparrow et al., 1984), the level of symbolic play by the Symbolic Play Test (Lowe et al., 1976), and the precursors of communication by the ComVoor (Verpoorten et al., 1999).

Adaptive Functioning

The VABS (Sparrow et al., 1984) describes four domains of adaptive functioning. Three domains were divided into three sub-domains, one into two. To make comparisons between the different domains possible, the scores were converted into developmental ages. Data were available for all subjects.

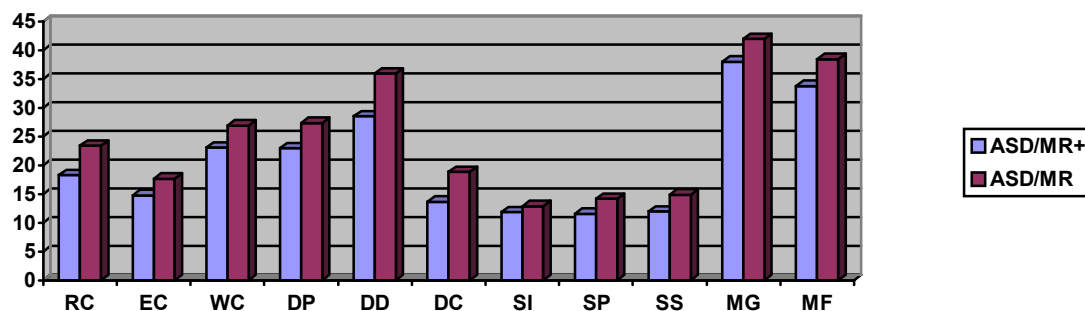
Figure 5-1 Developmental age in months for the four domains (Communication (Com); Daily Living Skills (DLS); Socialization (Soc); Motor Skills (Mot) of the VABS and the composite score (Total) for the two groups (ASD/MR+ and ASD/MR)



As can be seen in figure 5-1 the trend is that the scores of the group with challenging behaviour were lower in all four domains as was the total score. We see the characteristic profile of people with an ASD (Carter et al., 1998), namely relatively high scores, compared to the total score, for Daily Living Skills (DLS) and Motor Skills (Mot) and low scores for Communication (Com) and Socialization (Soc).

The distributions are normal for both groups, with the exception of the domain Communication for the group without challenging behaviour. The full descriptive statistics can be found in Appendix C.

Figure 5-2 Developmental ages in months for the 11 sub-domains (Receptive Communication (RC); Expressive Communication (EC); Written Language (WC); Personal Skills (DP); Domestic Skills (DD); Community Skills (DC); Interpersonal Relations (SI); Play and Leisure Time (SP); Social Skill (SS); Gross Motor Skills (GM); Fine Motor Skills (MF)) of the VABS for the two groups (ASD/MR+ and ASD/MR)



The scores of the group without challenging behaviour were slightly higher for all sub-domains. The distributions were not even in almost all of the sub-domains. Normal distributions were noted for both groups for the sub-domains: Receptive Communication (RC), Personal Skills (DP), Play and Leisure Time (SP) and Fine Motor Skills (MF) (see Appendix C).

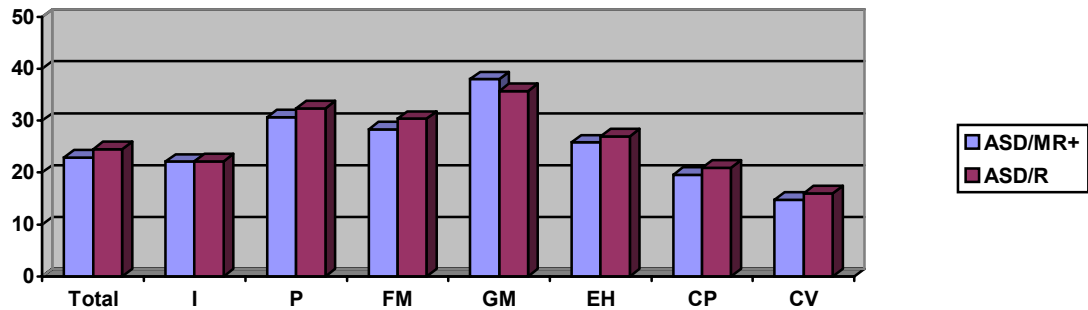
When we look more closely at the scores we find that the parents of the ASD/MR+ group reply more often that their children are not allowed to engage in, or are restricted from, certain activities: e.g. ‘not left unattended because they make a mess’ or ‘put themselves in dangerous situations’, ‘they can’t play with toys because they break them’, etc. These children are more restricted in their independence because of the use of restraints in certain situations. Restraints were reported for 17 children in the ASD/MR+ group compared to seven children in the ASD/MR group. Eleven children of the ASD/MR+ group were restrained most of the time in a ‘highchair’, wheelchair or (play)pen. These restrictions of their freedom to move and act independently could have diminished the scores, especially for the domains Daily Living Skills and Socialization.

Cognitive Functioning

We used the PEP-R (Schopler et al., 1990) to describe cognitive functioning in seven domains and a total score. The PEP-R is by no means a test that provides a full description of the cognitive functioning of a child, but we wanted a test that could be administered to all of the children in the project.

Again, to make comparisons possible the scores were converted to developmental ages.

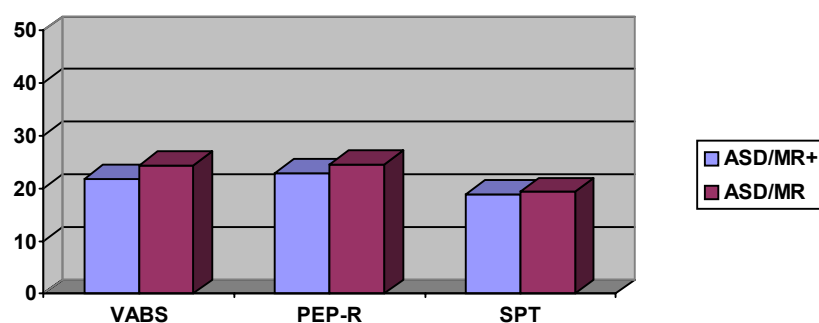
Figure 5-3 Developmental ages in months for the total score (Total) and the subscales Imitation (I), Perception (P), Fine Motor Skills (FM), Gross Motor Skills (GM) Eye-Hand Integration (EH), Cognition Performat (CP) and Cognition Verbal (CV) according to the PEP-R for the two groups (ASD/MR+ and ASD/MR)



Again we see slightly higher scores for the group without challenging behaviour, with the exception of the sub-domain Gross Motor Skills (GM). For this sub-domain the score was higher for the group with challenging behaviour. The profile is scattered, the highest scores being obtained for the motor scales and the lowest scores for cognition (CP and CV). This is as expected for children with an ASD and MR. The sub-domains of the PEP-R are very weak, as far as the internal consistency is concerned (Kraijer & Plas, 2002), so no comments can be made about the profile.

The distributions were not normal for the sub-domains Cognition Performat (CP) and Cognition Verbal (CV) (see Appendix C for the full description of the variables). A lot of children scored 0 for these sub-domains.

Figure 5-4 Mean developmental age in months for the Vineland Adaptive Behavior Scales (VABS), PsychoEducational Profile Revised (PEP-R) and Symbolic Play Test (SPT) for the ASD/MR+ and ASD/MR groups



In figure 5-4 we see the combined graph for the three instruments that depict cognitive and adaptive functioning and the score on the Symbolic Play Test (SPT; Lowe, 1976). We see the same tendency of slightly higher scores for the group without challenging behaviour.

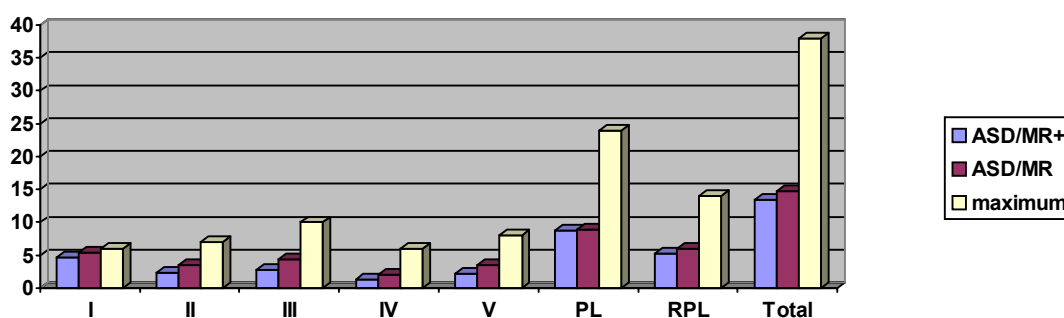
The developmental age for the Symbolic Play Test was lower for both groups. Because of the impairment in imagination, the play of children with an ASD is weak (see 2.3.3.4.). The scores for the SPT were not evenly distributed (see Appendix C). Eleven children from the group with challenging behaviour and nine children from the group without scored no points at all on the SPT. Two children from the group with challenging behaviour and one child from the group without obtained the maximum score (25).

There is a qualitative difference between the children from the ASD/MR+ group and those from the ASD/MR group. The children who scored no points at all only show the simplest forms of manipulative play: mouthing and banging. The children of the ASD/MR+ group throw the materials away, bang them and almost break the (very fragile!) materials. The children of the ASD/MR group often do not even touch the materials.

ComVoor

The ComVoor (Verpoorten et al., 1999) is a very specific test, dealing with perception as a precursor for communication. In the next graph the mean scores for the five series (I till V) and the composite scores for presentational (PL) and representational (RPL) are given, together with the total score.

Figure 5-5 Mean maximum scores and the scores of the ASD/MR and ASD/MR+ groups for the ComVoor for the 5 series (I till V), the score for presentational level (PL) and representational level (RPL), and the total score (Total).



The scores of the group with challenging behaviour are slightly lower for the five series and the three composite scores (PL, RPL and total). The distributions are not normal (see Appendix C). Five children of the group with challenging behaviour obtained a 0 score (total) compared to two children of the ASD/MR group.

Again we see a difference in the way the children handled the materials. The design of the ComVoor is adapted to the needs of people with an ASD. If the children understood what was expected of them, they performed the tasks. If they did not understand what was expected or the tasks became too difficult, the children of the ASD/MR+ group started to throw the materials away or broke them, whereas the children of the ASD/MR stopped handling the materials.

5.2.3.2 Challenging Behaviour

Data on the challenging behaviour of the groups were collected by means of two instruments, namely the DISCO II (Wing, 1993a) and the DBC (Einfeld et al., 1995). The two instruments gather different information. The DISCO was designed mainly for people with an ASD and is administered in the form of an interview. The DBC was designed for children with MR and is a questionnaire.

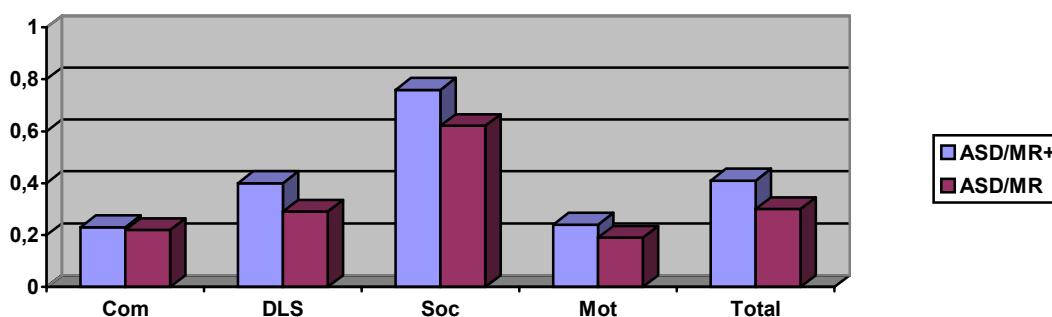
The DISCO data are available for all cases; the DBC data are available for 18 cases from the group with challenging behaviour and 19 cases from the group without. The mean age and developmental age of the groups did not differ significantly, when we exclude these three cases (see Appendix C).

DISCO II (Wing, 1993a)

With DISCO II the interviewer collected information about non-adaptive behaviours in the eleven domains of the VABS. Furthermore information about repetitive and stereotyped activities and challenging behaviour with and without social awareness was gathered.

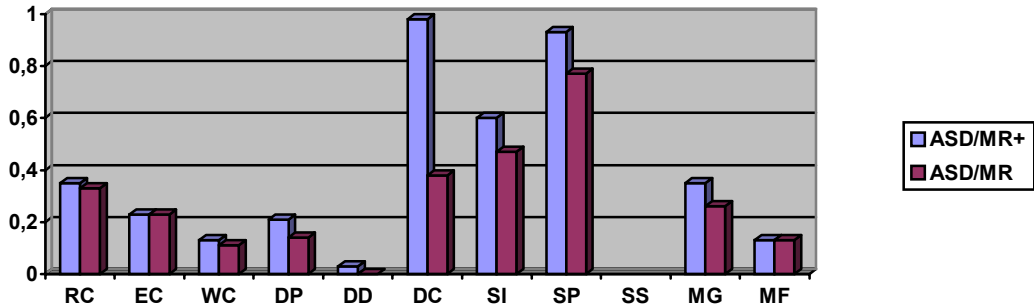
All of the domains consisted of different numbers of items. To make comparisons between the domains possible, the sum of the scores was divided by the number in that domain to obtain a score between one and zero.

Figure 5-6 Scores of the DISCO for the VABS-domains Communication (Com), Daily Living Skills (DLS), Socialization (Soc), Motor Skills (Mot) and Total Non-Adaptive Behaviour (Total) for the ASD/MR+ and ASD/MR groups



The group with challenging behaviour had higher scores for all the domains. Both groups obtained the highest score for the domain problems with socialization. The distribution was normal for both groups in this domain. For a full description of the variables see Appendix C.

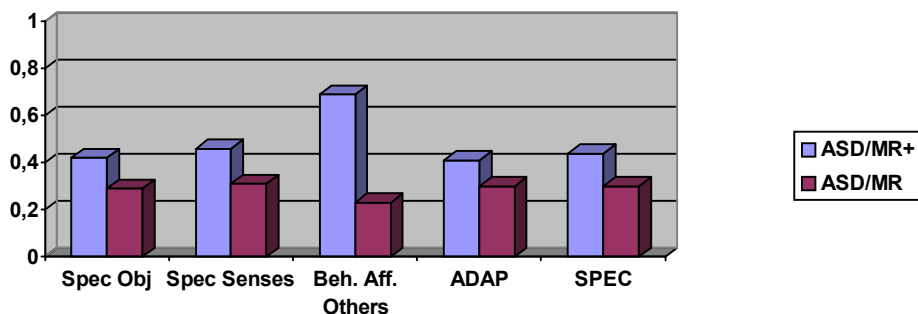
Figure 5-7 Scores of the DISCO II for the VABS-sub-domains Receptive Communication (RC), Expressive Communication (EC), Written Language (WC), Personal Skills (DP), Domestic Skills (DD), Community Skills (DC), Interpersonal Relations (SI), Play and Leisure Time (SP), Social Skills (SS), Gross Motor Skills (MG) and Fine Motor Skills (MF) for the ASD/MR+ and ASD/MR groups



The main pattern was higher scores for the group with challenging behaviour. The scores for the sub-domains Community, Interpersonal Relations and Play were very high, especially for the group with challenging behaviour. We see very low scores for the sub-domains Domestic Skills and Social Skills. The children from both groups did not really have any skills in that area, so no problems either. The distributions were not normal, with the exception of the sub-domain Interpersonal Relationship (SI). For a full description of the variables, see Appendix C.

As mentioned in 3.6.6., DISCO II has an extra domain, specific behavioural patterns. In figure 5-8 we have presented the scores for the sub-domains ‘Specific Behaviour towards Objects’ (Spec Obj), ‘Specific Behaviour concerning the Senses’ (Spec Senses), ‘Behaviour Affecting Others’ (Beh. Aff. others), the composite of these three (SPEC), the total score Problems with Adaptive Behaviour (ADAP), and the total score Specific Behaviours (SPEC).

Figure 5-8 Total scores for Behaviours Affecting Others (Beh. Aff. Others) and total Maladaptive Behaviour (ADAP) and Specific Behaviour with Objects (Spec. Obj.), Specific Behaviour Senses (Spec. Senses) and total Specific behaviours (SPEC) on the DISCO II for the ASD/MR+ and ASD/MR groups



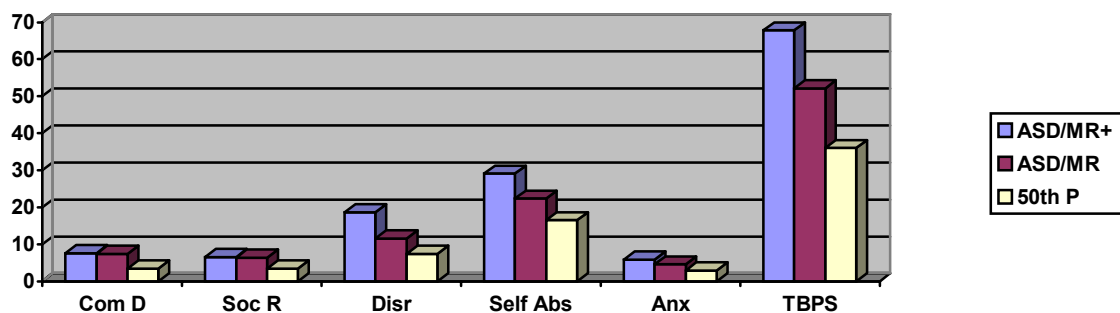
For the group with challenging behaviour the scores were higher for all five domains of maladaptive and specific behaviour of the DISCO II. For the group without challenging behaviour we see an even profile. For the group with challenging behaviour we see an especially high score for the sub-domain Behaviour Affecting Others. This domain contains the items: wandering off, aggressive behaviours, destructive behaviours, lack of cooperation, attention seeking and need for supervision.

We only find normal distributions for Specific Behaviour towards Objects and Specific Behaviour concerning the Senses (see Appendix C).

Developmental Behaviour Checklist scales (Einfeld & Tonge, 1995; Dekker, Nunn & Koot, 2002)

The DBC collects information by means of a questionnaire. In the Dutch version (Dekker et al., 2002) five factors were established: namely, Communication Disorder (Com D), Social Relations (Soc R), Disruptive/Antisocial Behaviours (Disr), Selfabsorbed (Self Abs) and Anxiety (Anx). The total score is called TBPS. For these factors the 50th percentile was calculated, according to the preliminary Dutch norms (Koot & Dekker, 2001). We calculated the mean of the norms for boys and girls.

Figure 5-9 Scores for the factors Communication Disorder (Com D), Social Relations (Soc R), Disruptive/Antisocial Behaviours (Disr), Selfabsorbed (Self Abs) and Anxiety (Anx) of the DBC and total score (TBPS) for the ASD/MR+ and ASD/MR groups and the 50th Percentile



The DBC is a normative instrument (see 3.6.5.), so we have included the scores for the 50th percentile in figure 5-9. Our groups score higher than the 50th percentile for all factors. The scores for the factors Disruptive/Antisocial and Self-Absorbed and the TBPS were higher for the group with challenging behaviour. For the other factors there are no differences.

The distributions of the scores are normal (see Appendix C).

5.2.3.3 The Pedagogic Situation and Parental Stress

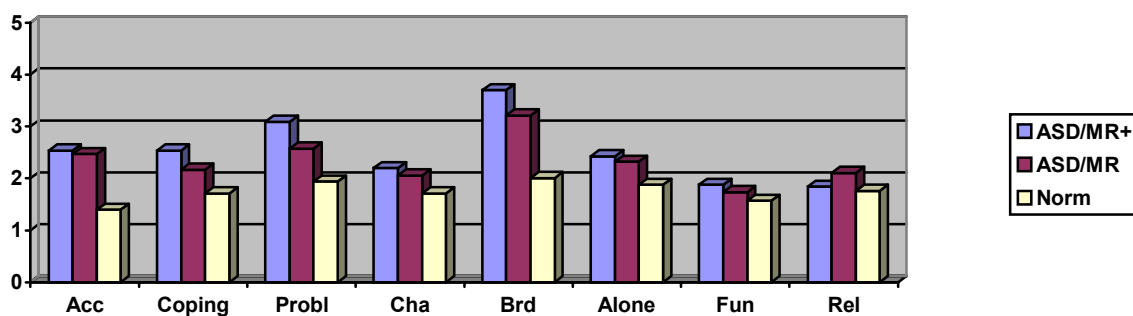
The Pedagogic Situation and Parental Stress were measured, using two instruments.

NVOS (Wels et al., 1996) and NOSIK (de Brock et al., 1992)

The scores on the NVOS were converted by calculating the sum and dividing that by the maximum. In the graphs you can also see the score for the norm group of mothers. The data was collected for 17 cases in both groups. The mean chronological age and mean developmental age of the groups do not differ significantly if we exclude the missing cases (see Appendix D). Missing data is due to the fact that some parents found the questions too confronting and personal, they did not return the questionnaires.

The NVOS is a normative instrument, so we have also included the scores for the norm group in the graphs 5-10, 5-11 en 5-12.

Figure 5-10 Scores for the NVOS for the factors Acceptation (Acc), Coping, Having Problems (Probl), Willing to Change (Cha), Child is Burden (Brd), Being Alone (Alone), Having Fun with the Child (Fun) and Relationship (Rel) of the domain Subjective Stress for the ASD/MR+ and ASD/MR groups and the norm group of the mothers



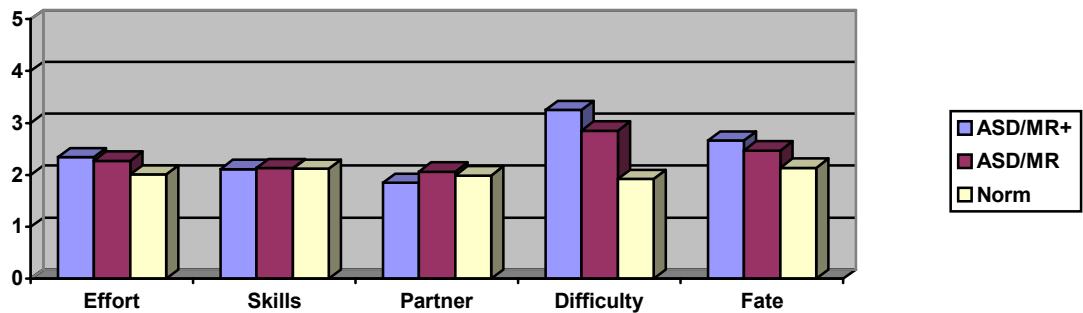
The scores of the norm group were lower than the scores of our groups for all of the domains. The scores for Coping, Having Problems (Probl), Willing to Change (Cha), Child is Burden (Brd), Being alone (Alone) and Having Fun with the child (Fun) were higher for the group with challenging behaviour. The score for 'Having a Good Relationship with the Child' (Rel) was higher for the group without challenging behaviour.

The distributions were not even for at least one of the groups for Child is burden (Brd), Being Alone (Alone), Having Fun with the child (Fun) and Having a Good Relationship with the Child (Rel).

The parents of the children with challenging behaviour judged the pedagogic situation more negatively (\underline{M} =4.76) than the parents of children without challenging behaviour (\underline{M} =3.65). The score of the norm group was much lower (\underline{M} =2.38).

This part contains only one item and the scores were not distributed evenly.

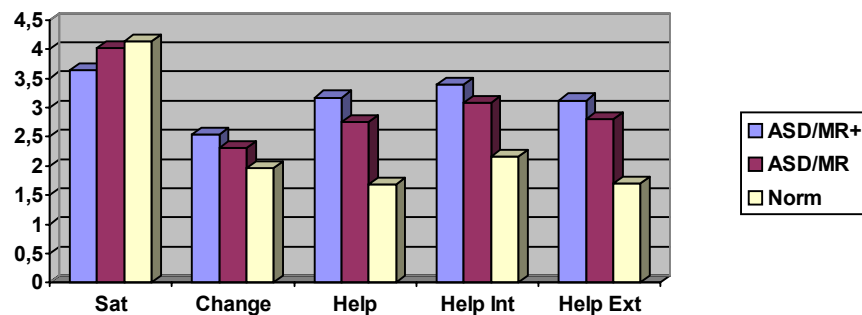
Figure 5-11 Scores for the NVOS for Attributions for the ASD/MR+ and ASD/MR groups and the mothers of the norm group



When we look at Attributions, the mothers of our groups did not rate their efforts and skills and the help of the partner any lower than the mothers of the norm group. The mothers of our groups rated their children as more difficult, especially the mothers of the children with challenging behaviour. They also perceived the influence of fate as being slightly higher.

The distributions for Effort, Skills and Difficulty of the child were normal; the distributions for the other domains were not (see Appendix C).

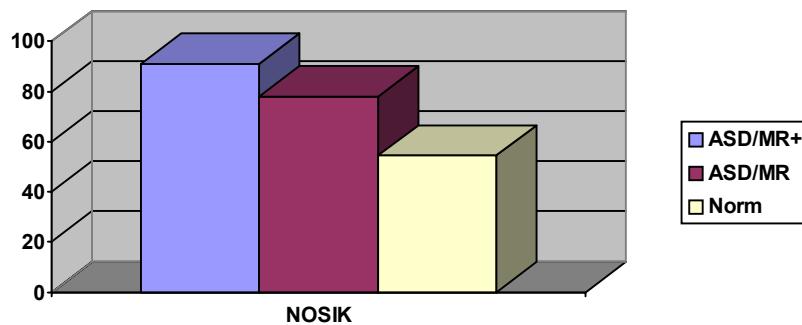
Figure 5-12 Scores for the NVOS for the sub-domains Satisfaction (Sat), Wanting Change (Change), Wanting Help (Help), Internal Help Expectancy (Help Int), External Help Expectancy (Help Ext) of the domain Help Expectancy for the ASD/MR+ and ASD/MR groups and the norm group of the mothers



In Figure 5.12 we see that both of our groups were less satisfied (Sat) with their current child-rearing situation than the norm group. We see higher scores for both groups for Wanting Change (Change), Need for Help (Help) and Internal (Help Int) and External Help Expectancy (Help Ext). The scores for the group with challenging behaviour were higher than the scores for the group without.

The distributions were normal, except for Satisfaction (see Appendix C).

Figure 5-13 Scores on the NOSIK, parental stress for the ASD/MR+ and ASD/MR groups and the mothers of the norm group



The NOSIK is a normative scale, so we have included the graph for the norm group. The parents of the group with challenging behaviour experience more stress than the parents of the group without. Compared to the non-clinical norm group, the score for the ASD/MR+ group can be rated as ‘very high’, whereas the ASD/MR group scores within the ‘high’ range.

The distributions are normal (see Appendix C).

5.3 Comparisons of the ASD/MR+ and the ASD/MR groups

5.3.1 Introduction

In this chapter we will discuss the differences between the group of children with an ASD and MR and challenging behaviour (ASD/MR+) and the group of children with an ASD and MR without challenging behaviour (ASD/MR). We will describe the results according to the model as proposed in 3.1. We will start with the Organic context, followed by the Super-ordinate, Interpersonal and Intrapersonal contexts. In each paragraph the history and current information will be reported (the time-dimension).

Different statistical tests were used to compare means, adapted to the level of the measurements and whether or not the distributions were normal. This will be explained for every test.

The sample is relatively small (N=20); consequently differences have to be relatively large to reach a level of significance (Meerling, 1989). If significant differences are found however, they are often clear (Lamberbon, 1991). Therefore the results are also described at a significance level of .1.

5.3.2 Differences in the Organic Context

In this paragraph we will describe the differences in the history of the children and the present medical status.

5.3.2.1 History

Prenatal History

The description of the variables of the prenatal history of the two groups can be found in 5.2.1.1. The variables Family Background and Pre-natal History were tested using the Chi-square test, because the variables are measured at the categorical level. The chi-statistic tests whether the two variables are independent. If $\text{Sig} \leq .05$ we reject the hypothesis that the variables are independent, hence they are in some way related (Field, 2000). Using the information from 5.2.1.1, we can determine the direction of the relationship.

Table 5-20 Chi-square tests for Family Background: Diagnosis, Cause and Relationship and Presence of Pre-natal problems

| | N | Value | Df | Asymp. Sig. (2-sided) |
|--------------------|----|--------|----|-----------------------|
| Family Background: | | | | |
| Diagnosis | 40 | 7.553 | 9 | .582 |
| Cause | 40 | 7.7167 | 7 | .412 |
| Relationship | 40 | 8.338 | 10 | .596 |
| Pre-natal Problems | 40 | .625 | 1 | .695 |

All of these variables are independent.

For the variables Gestation (in weeks), Birth weight (in grams) and Mothers Age at Birth (in years), t-tests for independent samples were used. If the significance level of Levene's test for Equality of Variance $\leq .05$, equal variances are not assumed (Field, 2000). The corresponding Sig-value can be found in the last column. In table 5-21 the results of these t-tests are presented.

Table 5-21 T-tests for Gestation, Birth weight and mother's age at birth

| | N MR/ASD+ | N MR/ASD | F | Levene's Test for Equality of Variances | | t-test for Equality of Means | |
|-------------------------|--------------|-------------|-------|--|--------|---------------------------------|---------------------|
| | | | | Sig. | T | Df | Sig. (2- tailed) |
| Gestation | 17 | 18 | 7.689 | .009* | 1.293 | 17.388 | .213 |
| Birth weight | 13 | 6 | .005 | .946 | -.186 | 17 | .855 |
| Mothers age at birth | 14 | 17 | .107 | .746 | -1.739 | 29 | .093* |

Gestation and Birth weight do not differ between the two groups. The age of the mothers is lower in the group with challenging behaviour than in the group without. The difference is significant at the .1 level, so we can speak of a trend. This result should be interpreted prudently, because data are only available for 31 cases.

Perinatal History

The description of the variable for Perinatal History can be found in 5.2.1.2. The variable Perinatal Problems was measured at the categorical level, so the Chi-square test was applied. The variables are independent: chi square (1, $N=40$) = .417, $\text{Sig}=.748$.

Postnatal History

The description of the variables for Postnatal History can be found in 5.2.1.3. The variable Postnatal Problems was measured at the categorical level, so the Chi-square test was applied. The variables are independent: chi square (1, $N=39$) = .408, $Sig=.523$.

Present Medical Status

The description of the variables for the Present Medical Status can be found in 5.2.1.4. The variables Additional Problems, Medical Examinations and Use of Medication were all measured at the categorical level, so Chi-square tests were used.

Table 5-22 Chi-square test for Presence of Epilepsy, Hearing Impairment, Visual Impairment and Other Additional Problems.

| | N | Value | Df | Asymp. Sig. (2-sided) |
|---------------------------|----|-------|----|-----------------------|
| Epilepsy | 40 | 1.129 | 1 | .480 |
| Hearing Impairment | 40 | .125 | 1 | .500 |
| Visual Impairment | 40 | 0.000 | 1 | 1.000 |
| Other Additional problems | 40 | .625 | 1 | .695 |

There are no differences between the two groups as far as these additional problems are concerned.

Table 5-23 Chi-square test for Medical Examinations performed

| | N | Value | Df | Asymp. Sig. (2-sided) |
|-------------------|----|-------|----|-----------------------|
| EEG | 40 | 0.000 | 1 | 1.000 |
| Cat-scan | 40 | 0.000 | 1 | 1.000 |
| Other brain | 40 | .114 | 1 | .736 |
| Fra-X | 40 | 1.129 | 1 | .288 |
| Other Chromosomal | 40 | .143 | 1 | .705 |
| Blood | 40 | .114 | 1 | .736 |
| Urine | 40 | .476 | 1 | .490 |
| Hearing | 40 | 2.057 | 1 | .151 |
| Vision | 40 | 3.137 | 1 | .077* |
| Other | 40 | .404 | 1 | .525 |

The total number of examinations performed does not differ significantly ($t=7.17$, $p=.478$). There is a significant difference at the $Sig=.1$ level for examinations of the eyes. In the group ASD/MR+ eye examinations were performed more often.

Table 5-24 Chi-square test for Positive Results Medical Examinations

| | N | Value | Df | Asymp. Sig. (2-sided) |
|-------------------|----|-------|----|-----------------------|
| EEG | 40 | 2.849 | 1 | .091* |
| Cat-scan | 40 | 3.584 | 1 | .058* |
| Other brain | 40 | 2.133 | 1 | .144 |
| Fra-X | 40 | - | - | - |
| Other Chromosomal | 40 | 1.111 | 1 | .292 |
| Blood | 40 | .360 | 1 | .548 |
| Urine | 40 | 2.105 | 1 | .147 |

| | N | Value | Df | Asymp. Sig. (2-sided) |
|---------|----|-------|----|-----------------------|
| Hearing | 40 | .625 | 1 | .429 |
| Vision | 40 | 1.129 | 1 | .288 |
| Other | 40 | 0.000 | 1 | 1.000 |

For the EEG and CAT scan these results are significant at the .1 level.

In total more positive results of Medical Examinations were found for the group ASD/MR ($t=-1.908$, $p=.067$). Nine children of the ASD/MR group had detectable problems on the EEG and for seven children abnormalities were detected on the Cat-scan. For the ASD/MR group, the cause of the problems is known more often.

Table 5-25 Chi-square test for use of Medication

| | N | Value | Df | Asymp. Sig. (2-sided) |
|--------------------|----|-------|----|-----------------------|
| Major tranquilizer | 40 | 8.063 | 2 | .018** |
| Minor tranquilizer | 40 | 1.027 | 2 | .598 |
| Antidepressant | 40 | 1.026 | 1 | 1.000 |
| Hypnotics | 40 | 0.000 | 1 | 1.000 |
| Anti-convulsant | 40 | 5.125 | 2 | .077* |
| Other | 40 | 1.027 | 2 | .598 |

As a trend the use of medication was higher for the group with challenging behaviour, except for anticonvulsants. They were used more in the group without challenging behaviour. In particular the use of major tranquilizers was significantly higher for the group ASD/MR+ ($p<.05$).

5.3.3 Differences in the Super-ordinate Context

In this paragraph we will discuss the differences in the composition of the family and the social and economical status of the families between the two groups. The data was gathered at the categorical level, so chi-square tests were performed. The chi-square tests whether the two variables are independent. If $\text{Sig} \leq .05$ we reject the hypothesis that the variables are independent, hence they are in some way related (Field, 2000). Using the information from 5.2, we can see the direction of the relationship.

Composition of the family

The variable presence of the biological mother is independent, chi-square (1, $N=40$) = .1026, $\text{Sig} = .500$, the same applies for the variable presence of the biological father, chi-square (1, $N=40$) = .229, $\text{Sig} = .100$.

None of the children have siblings who live outside the family. The number of live in brothers and sisters is also independent, chi-square (4, $N = 40$) = 3.326, $\text{Sig} = .505$.

Therefore we found no differences in the composition of the family between the two groups.

Social and economical status of the family

In this paragraph differences between the variables Ethnicity (whether or not the parents are Dutch), Level of Education of the Parents and Current Job Level of the Parents were examined.

Table 5-26 Chi-square test for Ethnicity of the parents

| | N | Value | Df | Asymp. Sig. (2-sided) |
|--------|----|-------|----|-----------------------|
| Father | 20 | 1.290 | 1 | .451 |
| Mother | 20 | 1.026 | 1 | 1.000 |

The variable ethnicity is independent. There are no differences in the ethnicity of the parents between the two groups.

Table 5-27 Chi-square test for level of Education of the Parents

| | N | Value | Df | Asymp. Sig. (2-sided) |
|--------|----|-------|----|-----------------------|
| Father | 20 | 1.292 | 2 | .731 |
| Mother | 20 | .508 | 2 | .776 |

Table 5-28 Chi-square test for current Job Level of the Parents

| | N | Value | Df | Asymp. Sig. (2-sided) |
|--------|----|-------|----|-----------------------|
| Father | 20 | .683 | 3 | .953 |
| Mother | 20 | 2.438 | 3 | .487 |

Both the level of education and the current job level are independent. We found no differences in the social and economical status of the families.

The backgrounds of the families are identical for the variables we have investigated. We have found no differences in the super-ordinate context.

5.3.4 Differences in the Interpersonal Context

In this paragraph we will discuss the information we have collected at the interpersonal context. We will start with the history, current information on interpersonal functioning and the current pedagogical situation.

Finally we will discuss the variables for the pedagogical situation as measured with the NVOS (Wels et al., 1996). The description of these data can be found in 5.2.3.3.

The differences between the groups were tested using t-tests for independent samples. If the significance level of Levene's test for Equality of Variance $\leq .05$, equal variances are not assumed (Field, 2000). The corresponding Sig-value can be found in the last column. In the following tables the results of these t-tests are described.

5.3.4.1 History

Firstly we will describe the Age of Recognition of the problems. We have no data on early attachment, but presume that early onset of problems could influence attachment. The description of this variable can be found in paragraph 5.2.1.6. The differences between the groups were tested using t-tests for independent samples. If

the significance level of Levene's test for Equality of Variance $\leq .05$, equal variances are not assumed (Field, 2000). The corresponding Sig-value can be found in the last column. In the following tables the results of these t-tests are described.

Age of Recognition of Problems

Table 5-29 T-tests for Age of First Worries

| Age in months | MR/ASD+ | MR/ASD | F | Levene's Test for Equality of Variances | | t-test for Equality of Means | |
|--|---------|--------|-------|---|--------|------------------------------|-----------------|
| | | | | Sig. | T | Df | Sig. (2-tailed) |
| First Worries | 9.85 | 10.20 | .136 | .714 | -.105 | 38 | .917 |
| First Problems | 2.35 | 4.40 | 1.384 | .247 | -.105 | 38 | .409 |
| Age of onset | 16.90 | 11.45 | 3.392 | .073 | 1.312 | 38 | .197 |
| Age of onset untypical behaviour (rater) | 4.45 | 3.45 | .839 | .365 | .365 | 38 | .716 |
| Age at diagnosis | 30.80 | 40.50 | .724 | .400 | -1.282 | 38 | .208 |

We found no differences between the two groups as far as the age of first worries; age of onset of the problems and age of diagnosis are concerned.

5.3.4.2 Current Pedagogical Situation

Interpersonal Relationships

In this paragraph we will discuss the variable development of 'Interpersonal Relationships' as measured for the sub-domain of the same name by the VABS (Sparrow et al., 1984) and the problems in that sub-domain as measured by the DISCO II (Wing, 1993a).

Development

The group ASD/MR+ had slightly higher scores for the sub-domain Interpersonal Relationships of the VABS, in contrast to the other sub-domains of the VABS, but the differences are not significant ($t=-.312$, $p=.757$).

In table 5-30 Problems with Interpersonal Relationships as measured by the DISCO II (Wing, 1993a) are shown.

Table 5-30 T-tests for Problems with Interpersonal Relationships

| N=40 | Mean MR/ASD+ | Mean MR/ASD | F | Levene's Test for Equality of Variances | | t-test for Equality of Means | |
|-------------------------|-----------------|----------------|--------|---|--------|------------------------------|-----------------|
| | | | | Sig. | T | Df | Sig. (2-tailed) |
| Total interpersonal | .601 | .470 | .615 | .438 | 2.833 | 38 | .007** |
| Staring at people | .700 | .200 | 19.092 | .000 | 2.107 | 37.696 | .044** |
| Lack of emotions | .005 | .300 | 16.403 | .000 | -1.823 | 30.058 | .081* |
| Unhappy | .850 | .350 | 12.798 | .000 | 2.028 | 32.003 | .051* |
| Confused | 1.600 | .500 | .005 | .942 | 4.395 | 38 | .000** |
| Fears | 1.100 | .300 | 26.079 | .000 | 2.947 | 32.433 | .006** |
| Non verbal soc. Int. | 1.550 | .950 | 1.996 | .166 | 2.139 | 38 | .039** |

| N=40 | Mean MR/ASD+ | Mean MR/ASD | F | Levene's Test for Equality of Variances | | t-test for Equality of Means | |
|-----------------------------|-----------------|----------------|-------|--|-------|---------------------------------|---------------------|
| | | | | Sig. | T | Df | Sig. (2- tailed) |
| Reaction to visitors | 1.500 | .900 | .237 | .629 | 2.698 | 38 | .010** |
| Seeking physical comfort | 1.450 | 1.050 | 4.729 | .036 | 1.843 | 36.193 | .074* |

The group MR/ASD+ had significantly more problems with interpersonal relationships than the MR/ASD group. We found significantly more problems in the ASD/MR+ group for the items 'staring at people', 'feeling unhappy', 'being confused', 'fears', 'problems with non verbal social interaction', 'reaction to visitors' and 'seeking physical comfort'. The group ASD/MR, however, had significantly more problems with the item 'lack of emotions'.

Problems in Relationship with Peers

Table 5-31 Chi-square tests for Problems with Relationship with Peers

| N=40 | Value | Df | Asymp. Sig. (2-sided) |
|------------------------|-------|----|-----------------------|
| Attitude towards Peers | 9.750 | 5 | .083* |
| Play with Peers | 7.914 | 4 | .095* |

We found significant differences at the Sig = .1 level for the two questions concerning the relationship with peers. The group ASD/MR+ exhibited more negative behaviour towards peers. Seven children showed aggressive behaviour towards peers, nine children ignored their peers. In the ASD/MR group seven children ignored other children, but 11 children played next to other children. The variables are dependent (Sig=.1) and differ between the two groups. Nineteen children of the ASD/MR+ group did not play with other children, compared to 12 children of the ASD/MR group.

There are noticeable differences between the children of the two groups. In both groups a proportion of the children did not seek interaction with other children, but of the remaining children who did seek to play with other children, the children of the ASD/MR+ group got into trouble when playing with others. The children of the ASD/MR group tended to react more passively towards their peers, whereas the ASD/MR+ group had a more bizarre reaction.

Subjective Stress

Table 5-32 T-tests for NVOS Subjective Stress

| | Mean (N=17) MR/ASD+ | Mean (N=17) MR/ASD | F | Levene's Test for Equality of Variances | | t-test for Equality of Means | |
|--------------------|---------------------------|--------------------------|-------|--|-------|---------------------------------|---------------------|
| | | | | Sig. | t | Df | Sig. (2- tailed) |
| Acceptation | 2.5441 | 2.476 | 2.198 | .148 | .255 | 32 | .800 |
| Coping | 2.5394 | 2.1665 | .489 | .489 | 1.243 | 32 | .223 |
| Having Problems | 3.0918 | 2.5688 | .478 | .495 | 2.079 | 32 | .044** |

| | Mean (N=17) MR/ASD+ | Mean (N=17) MR/ASD | F | Levene's Test for Equality of Variances | | t-test for Equality of Means | |
|----------------------|---------------------------|--------------------------|-------|--|-------|---------------------------------|---------------------|
| | | | | Sig. | t | Df | Sig. (2- tailed) |
| Wanting to Change | 2.2047 | 2.0600 | 1.269 | .268 | 2.097 | 32 | .620 |
| Child is Burden | 3.7047 | 3.2100 | .349 | .559 | .501 | 32 | .159 |
| Being Alone | 2.4265 | 2.3235 | .110 | .743 | 1.442 | 32 | .766 |
| Having Fun | 1.8824 | 1.7412 | .400 | .532 | .541 | 32 | .592 |
| Good Relationship | 1.8471 | 2.1059 | .515 | .478 | -.963 | 32 | .343 |

The mothers of the group of children with challenging behaviour perceived significantly more problems with their child than the mothers of the other group. For all the other variables the differences are not significant.

Judgement of Pedagogical Situation

Table 5-33 T-test for NVOS Judgement of Pedagogical Situation

| | N | N | Levene's Test for Equality of Variances | | | t-test for Equality of Means | |
|---|---------|--------|---|------|-------|------------------------------|-----------------|
| | | | F | Sig. | t | Df | Sig. (2-tailed) |
| | MR/ASD+ | MR/ASD | | | | | |
| B | 17 | 17 | .300 | .587 | 2.049 | 32 | .049** |

The mothers of the group with ASD/MR+ had a significantly worse perception of their pedagogical situation than the mothers of the other group.

There were a lot of comments about this question written down on the forms. The mothers of the ASD/MR+ group often expressed their worries about the future and their need for respite care, whereas the mothers of the ASD/MR group tended to comment about the questions that were suggestive of behaviour problems. They often wrote down: 'my child does not have a behaviour disorder, he/she has autism!'

Attributions

Table 5-34 T-tests for NVOS Attributions

| | Mean (N=17) MR/ASD+ | Mean (N=17) MR/ASD | Levene's Test for Equality of Variances | | | t-test for Equality of Means | |
|---------------------|------------------------|-----------------------|--|------|-------|---------------------------------|---------------------|
| | | | F | Sig. | t | Df | Sig. (2- tailed) |
| Effort | 2.341 | 2.274 | .007 | .932 | .376 | 32 | .710 |
| Skills | 2.106 | 2.139 | .027 | .871 | -.203 | 32 | .886 |
| Proportion partner | 1.855 | 2.057 | .511 | .480 | -.772 | 32 | .446 |
| Level of difficulty | 3.251 | 2.849 | .925 | .343 | 1.449 | 32 | .157 |
| Coincidence/Fate | 2.662 | 2.471 | 2.096 | .157 | .520 | 32 | .607 |

The parents of both groups rate their situations similarly. We found no significant differences in the above-mentioned variables.

Help Expectancy

Table 5-35 T-tests for NVOS Help Expectancy

| | Mean (N=17) MR/ASD+ | Mean (N=17) MR/ASD | Levene's Test for Equality of Variances | | t-test for Equality of Means | | |
|-----------------------------|---------------------------|--------------------------|--|------|---------------------------------|----|---------------------|
| | | | F | Sig. | t | Df | Sig. (2- tailed) |
| Satisfaction | 3.646 | 4.020 | .551 | .463 | -1.331 | 32 | .192 |
| Will to change | 2.539 | 2.314 | .013 | .909 | 1.202 | 32 | .238 |
| Wants help | 3.167 | 2.745 | 1.873 | .181 | 1.513 | 32 | .140 |
| Help expectancy internal | 3.388 | 3.075 | 1.839 | .185 | 1.531 | 32 | .135 |
| Help expectancy external | 3.118 | 2.802 | 2.084 | .159 | 1.161 | 32 | .254 |

We found no differences between the groups as far as help expectancy goes. The parents of both groups expect the same help and are equally satisfied.

Parental Stress

Table 5-36 T-test for NOSIK Parental Stress

| | Mean (N=17) MR/ASD+ | Mean (N=17) MR/ASD | Levene's Test for Equality of Variances | | t-test for Equality of Means | | |
|--------------------|------------------------|-----------------------|--|------|---------------------------------|-------|---------------------|
| | | | F | Sig. | T | Df | Sig. (2- tailed) |
| Parental Stress | 91.12 | 78.06 | 3.26 | .08 | 1.50 | 27.33 | .15 |

The parents of both groups perceive high stress levels. The differences between the two groups were not significant.

As far as the pedagogical situation is concerned the parents and the skills of the parents do not differ between the two groups. We do however find differences in the perception of their children. The parents of the ASD/MR+ group find their children more difficult and view their situation more negatively. They do not think that they lack the pedagogical skills.

5.3.5 Differences in the Intrapersonal Context

In this paragraph we will discuss the differences in the Intrapersonal Context. We will start with the developmental history and then describe the differences in the cognitive and behavioural levels.

5.3.5.1 Developmental History

In this paragraph we will discuss the differences between the two groups in behaviour during infancy, the reaching of developmental milestones and setbacks in the development of skills.

The data were gathered at the categorical level, so chi-square tests were used. The chi-square tests whether the two variables are independent. If $\text{Sig} \leq .05$ we reject the hypothesis that the variables are independent, hence they are in some way related (Field, 2000). Using the information from 5.2, we can see the direction of the relationship.

Behaviour during Infancy

Table 5-37 Chi-square tests for Behaviour during Infancy

| | N | Value | Df | Asymp. Sig. (2-sided) |
|-------------------------------|----|-------|----|-----------------------|
| Feeding | 40 | .451 | 2 | .798 |
| Vomiting | 40 | 1.086 | 2 | .581 |
| Excessive Crying/Screaming | 40 | .086 | 2 | .958 |
| Woke up Screaming | 40 | 1.268 | 2 | .531 |
| Sleeping Pattern | 40 | .450 | 2 | .799 |
| Unusually Good/Quiet/Passive | 40 | 3.600 | 2 | .165 |
| Demand for social attention | 40 | 1.333 | 2 | .513 |
| Response to cuddling | 40 | 1.752 | 2 | .416 |
| Eye contact | 40 | 1.002 | 2 | .606 |
| Getting ready to be picked up | 40 | .000 | 2 | 1.000 |
| Reciprocation in baby games | 40 | .148 | 2 | .929 |
| Waving bye-bye | 40 | .784 | 1 | .661 |
| Babbling | 40 | .119 | 2 | .942 |
| Pre-speech conversation | 40 | 1.244 | 2 | .537 |
| Response to speech | 40 | 1.333 | 2 | .513 |
| Intoned pre-speech sounds | 40 | 1.400 | 2 | .497 |
| Interest in environment | 40 | .232 | 2 | .890 |
| Looked when others pointed | 40 | 1.029 | 2 | .598 |
| Joint referencing | 40 | 1.333 | 2 | .513 |
| Special Interests | 40 | 1.143 | 2 | .565 |
| Attachments to one parent | 40 | 3.360 | 2 | .186 |
| Dislike of care procedures | 40 | 6.690 | 2 | .035** |
| Oversensitive to noise | 40 | 6.704 | 2 | .035** |
| Odd movements | 40 | 7.356 | 2 | .025** |
| Other Problems | 40 | 2.105 | 1 | .487 |

Most of the variables are independent. However, we do find significant differences for the items 'dislike of care procedures', 'oversensitive to noise' and 'odd movements'. For all of these items the ASD/MR+group had significantly higher scores ($\text{Sig} \leq .05$). All of these problems were detected at a very early age and could be an indication for oversensitivity to certain stimuli.

Developmental Milestones

The data has been collected at the categorical level, so chi-square tests were used. These tests establish whether the two variables are independent. If $\text{Sig} \leq .05$ we reject the hypothesis that the variables are independent, hence they are in some way related (Field, 2000). The description of the variables can be found in 5.2.1.5.

Table 5-38 Chi-square tests Developmental Milestones

| Developmental milestones | N | Value | Df | Asymp. Sig. (2-sided) |
|--------------------------|----|-------|----|-----------------------|
| Independent Sitting | 40 | .902 | 1 | .527 |
| Crawling | 40 | .947 | 2 | .623 |
| Walking | 40 | 1.053 | 2 | .591 |
| Riding Tricycle | 40 | 2.756 | 2 | .252 |
| Riding Bicycle | 40 | 1.954 | 2 | .377 |
| Clean and Dry during day | 40 | 2.524 | 2 | .283 |
| Clean and Dry at night | 40 | .110 | 2 | .946 |

All of these variables are independent. We do not find differences between the two groups in the age at which the developmental milestones were reached.

Setback in the development of skills

Table 5-39 Chi-square tests setback in development of skills

| Developmental area | N | Value | Df | Asymp. Sig. (1-sided) |
|------------------------|----|--------|----|-----------------------|
| Motor Development | 40 | .533 | 1 | .358 |
| Toilet Training | 40 | 4.444 | 1 | .053* |
| Other Self Care | 40 | 2.105 | 1 | .244 |
| Language/Communication | 40 | 2.849 | 1 | .088* |
| Play | 40 | 1.558 | 1 | .204 |
| Social Interaction | 40 | .000 | 1 | .634 |
| Feeding | 40 | 5.714 | 1 | .024** |
| Other Adaptive Skills | 40 | 10.000 | 1 | .002** |

The variables Setback in Feeding and Other Adaptive Skills are not independent ($\text{Sig} \leq .05$). The group ASD/MR+ had a setback significantly more often than the ASD/MR group. For the variables Toilet Training and Language/Communication $\text{Sig} \leq .1$, there is a trend that the group ASD/MR+ also had setbacks in these variables more often.

When we calculate a sum-score for all of the items that map a setback, we find that the group ASD/MR+ had Setbacks in Infancy in any of the above-mentioned areas more often than the ASD/MR group ($t=2.320$, $p=.027$).

5.3.5.2 Current Functioning

In this paragraph we will discuss the differences between the two groups in the variables for Cognition, the Triad of Impairments, Motor Skills, Practical Skills and Challenging Behaviour. The description of the data can be found in 5.2.3. The differences between the two groups were tested using t-tests for independent samples. If the significance level of Levene's test for Equality of Variance $\leq .05$, equal variances are not assumed (Field, 2000). The corresponding Sig-value can be found in the last column.

Cognition

Under the heading of Cognition we gathered information on general intellectual functioning, attention, splinter abilities and memory. The PEP-R was used as a

measure of general development. Specific instruments to map, for instance, Theory of Mind or Executive Functions are very rare and are not adapted to the level of functioning of our group. The DISCO II included several questions about the above-mentioned subjects; we will discuss those results here.

Level of Functioning

The total score on the PEP-R (Schopler et al., 1990) was used as a matching criterion, so the total scores do not differ. In the following table the t-tests of the sub-domains Imitation, Perception, Cognition Performal and Cognition Verbal are presented. The differences between the two groups were tested using t-tests for independent samples.

Table 5-40 T-tests for PEP-R

| | Mean DA MR/ASD+ | Mean DA MR/ASD | Levene's Test for Equality of Variances | | | t-test for Equality of Means | |
|------------------------|--------------------|-------------------|--|------|------|---------------------------------|---------------------|
| | | | F | Sig. | t | Df | Sig. (2- tailed) |
| Imitation | 22.10 | 22.15 | .842 | .37 | -.01 | 38 | .99 |
| Perception | 30.70 | 32.40 | .146 | .70 | -.20 | 38 | .77 |
| Cognition Performal | 19.60 | 20.45 | .106 | .75 | -.48 | 38 | .63 |
| Cognition Verbal | 14.75 | 16.05 | .802 | .38 | -.42 | 38 | .68 |
| Total | 22.95 | 24.50 | .199 | .66 | -.52 | 38 | .61 |

We found no differences in the above-mentioned sub-domains of the PEP-R. All Sig's were above .1.

Attention

In the DISCO II (Wing, 1993a) two questions reflect Attention. These questions gather categorical data, so Chi-square-tests were performed. The chi-statistic tests whether the two variables are independent.

One question deals with Concentration for Self-chosen activities, the second question deals with concentration for activities that are proposed by others.

Table 5-41 Chi-square test for attention

| Developmental milestones | N | Value | Df | Asymp. Sig. (2-sided) |
|---|----|-------|----|-----------------------|
| Concentration Self-Chosen Activities | 40 | 7.140 | 4 | .129 |
| Concentration Activities Proposed by Others | 40 | 9.905 | 4 | .042** |

The variable Attention for Activities Proposed by Others is not independent. When we look at the results we see that the ASD/MR+ group has a lower score for this variable. About half of the children in this group cannot perform a task without supervision.

For the DBC (Einfeld et al., 1995) we find significant differences for the items: lack of concentration ($t=1.705$, $\underline{\text{Sig}}=.097$), easily distracted ($t=2.660$, $\underline{\text{Sig}}=.012$), inpatient ($t=1.981$, $\underline{\text{Sig}}=.056$), impulsive ($t=2.121$, $\underline{\text{Sig}}=.041$), overactive/restless ($t=1.687$,

Sig=.100), asks too much attention ($t=3.761$, Sig=.001). All of these items are indicative of hyperactivity and attention problems.

The information we gathered on the attention skills of the two groups, although only based on item scores, shows that the ASD/MR+ group has more problems with attention and is much more often considered to be hyperactive. This finding can be confirmed with observations. The constant need for supervision and the inability to do anything independently is the main complaint of the parents of the ASD/MR+ group. In the structured situation of the individual assessments we did not find this difference between the two groups.

Splinter Abilities

In the DISCO II one question concerns Special Skills. The groups do not differ in this variable ($t=1.125$, Sig=.267). Abilities on a level higher than expected for their age are very rare in these children. The severity of the level of Mental Retardation in both groups is probably the explanation.

Memory

In the DISCO II one question was about Memory. The groups do not differ in this respect ($t=1.050$, Sig=.300).

Socialization

Socialization was measured by the domain Socialization of the VABS (Sparrow et al., 1984). Part of this domain, namely interpersonal relationships, has been described under the heading of interpersonal context (see 5.3.4.2). Because of the need for a complete overview, the entire domain socialization will be described here. Problems with socialization were measured by the DISCO II (Wing, 1993a). The differences between the two groups were tested using t-tests for independent samples. If the significance level of Levene's test for Equality of Variance $\leq .05$, equal variances are not assumed (Field, 2000). The corresponding Sig-value can be found in the last column.

Table 5-42 T-tests for domain Socialization VABS

| | Mean MR/ASD+ | Mean MR/ASD | Levene's Test for Equality of Variances | | t | t-test for Equality of Means | |
|--------------------------------|-----------------|----------------|--|------|--------|---------------------------------|---------------------|
| | | | F | Sig. | | Df | Sig. (2- tailed) |
| Total Socialization | 12.25 | 14.20 | .841 | .365 | -1.030 | 38 | .310 |
| Interpersonal Relationships | 11.85 | 12.90 | .005 | .944 | -.413 | 38 | .682 |
| Play and Leisure Time | 11.55 | 14.20 | .697 | .409 | -1.440 | 38 | .158 |
| Social Skills | 11.95 | 14.90 | 2.022 | .163 | -1.490 | 38 | .144 |

We found no differences between the two groups in the socialization domain of the VABS.

Table 5-43 T-tests for domain Problems with Socialization DISCO II

| | Mean MR/ASD+ | Mean MR/ASD | Levene's Test for Equality of Variances | | t | t-test for Equality of Means | |
|--------------------------------|-----------------|----------------|--|------|-------|---------------------------------|---------------------|
| | | | F | Sig. | | Df | Sig. (2- tailed) |
| Total problems | .5874 | .4414 | .033 | .856 | 4.747 | 38 | .000** |
| Socialization | .5874 | .4414 | .033 | .856 | 4.747 | 38 | .000** |
| Problems | | | | | | | |
| Interpersonal Relationships | .6006 | .4700 | .615 | .438 | 2.833 | 38 | .007** |
| Play and Leisure Time | .9271 | .7729 | 3.963 | .054 | 3.003 | 38 | .005** |
| Problems Social Skills | .2344 | .0813 | 4.725 | .036 | 1.865 | 37.449 | .070** |

The group ASD/MR+ had significantly more problems in the domain socialization and its sub-domains.

Communication

As described in 3.5, in both groups eleven children could speak and nine were non-speaking. Speaking or non-speaking was not a matching criteria. The development of communication was measured by the ComVoor, which maps the Precursors of Communication and the VABS with the domain Communication containing the sub-domains Receptive and Expressive Communication and Written Language. The differences between the two groups were tested using t-tests for independent samples.

Table 5-44 T-tests for ComVoor

| | Mean MR/ASD+ | Mean MR/ASD | Levene's Test for Equality of Variances | | t | t-test for Equality of Means | |
|----------------|-----------------|----------------|--|------|--------|---------------------------------|-----------------|
| | | | F | Sig. | | Df | Sig. (2-tailed) |
| Serie I | 4.70 | 5.45 | 3.997 | .053 | -.912 | 38 | .368 |
| Serie II | 2.40 | 3.55 | .264 | .611 | -1.202 | 38 | .237 |
| Serie III | 2.75 | 4.45 | 1.432 | .239 | -1.354 | 38 | .184 |
| Serie IV | 1.25 | 2.00 | 1.357 | .251 | -1.079 | 38 | .288 |
| Serie V | 2.25 | 3.50 | 2.530 | .120 | -1.111 | 38 | .273 |
| Presentation | 9.80 | 13.45 | .015 | .902 | -1.306 | 38 | .200 |
| Representation | 3.55 | 5.50 | 2.299 | .138 | -1.107 | 38 | .275 |
| Total | 13.35 | 19.45 | .788 | .380 | -1.368 | 38 | .179 |

Although the scores of the ASD/MR+ group were overall lower, we did not find significant differences. However, we did find significant differences for two items: namely, item 12, sorting pictograms ($t=-1.798$, $\text{Sig}=.081$) and item 13, sorting letters ($t=-2.135$, $\text{Sig}=.040$).

The group ASD/MR had higher scores for these items.

Table 5-45 T-tests for VABS communication

| | Mean MR/ASD+ | Mean MR/ASD | Levene's Test for Equality of Variances | | t-test for Equality of Means | | |
|-----------------------------|-----------------|----------------|--|------|------------------------------------|----|---------------------|
| | | | F | Sig. | t | Df | Sig. (2- tailed) |
| Total Communication | 16.40 | 19.00 | .445 | .509 | -.752 | 38 | .457 |
| Communication Receptive | 18.30 | 23.40 | 3.909 | .055 | - 1.481 | 38 | .147 |
| Communication Expressive | 14.80 | 17.65 | .798 | .377 | -.691 | 38 | .494 |
| Written Language | 23.05 | 26.90 | 2.283 | .139 | -.766 | 38 | .448 |

We found no differences between the two groups for the domain Communication of the VABS.

Table 5-46 T-tests for DISCO Problems with Communication

| | Mean MR/ASD+ | Mean MR/ASD | Levene's Test for Equality of Variances | | t-test for Equality of Means | | |
|--|-----------------|----------------|--|------|---------------------------------|----|---------------------|
| | | | F | Sig. | t | Df | Sig. (2- tailed) |
| Total Communication | .2345 | .2206 | .728 | .399 | .311 | 38 | .758 |
| Communication Receptive | .3500 | .3250 | .008 | .930 | .503 | 38 | .618 |
| Communication Expressive Speaking | .2286 | .2286 | .019 | .891 | 0.000 | 38 | 1.000 |
| Communication Expressive Non-speaking | .4000 | .2938 | 1.911 | .175 | 1.345 | 38 | .187 |
| Written language | .1250 | .1083 | .343 | .561 | .293 | 38 | .771 |

We have found no differences between the two groups on Problems with Communication of the DISCO II.

The communication skills of the two groups do not differ. The scores of the two groups were very low.

Imagination

The development of imagination was measured by the SPT (Lowe & Costello, 1976) and the sub-domains Play and Leisure Time of the VABS (Sparrow et al., 1984). The problems with Play and Leisure time were measured by the DISCO II (Wing, 1993a). The differences between the two groups were tested by means of t-tests for independent samples. If the significance level of Levene's test for Equality of Variance $\leq .05$, equal variances cannot be assumed (Field, 2000). The corresponding Sig-values are shown in the last column.

Table 5-47 T-tests for Imagination as assessed by VABS and SPT

| | | | Levene's Test for Equality of Variances | | | t-test for Equality of Means | |
|--------------------------------|--------------|-------------|---|------|--------|------------------------------|-----------------|
| | N MR/ASD+ | N MR/ASD | F | Sig. | T | Df | Sig. (2-tailed) |
| Play & Leisure Time (V) (N=20) | 11.55 | 14.20 | .605 | .441 | -1.440 | 38 | .158 |
| SPT (N=18) | 18.950 | 19.367 | .007 | .934 | -.130 | 33.852 | .897 |

We found no differences between the two groups for the Symbolic Play Test and the sub-domain Play and Leisure Time assessed by the VABS.

Table 5-48 T-test for problems with Play and Leisure Time DISCO

| | | | Levene's Test for Equality of Variances | | | t-test for Equality of Means | |
|------------------------------|-----------------|----------------|---|------|-------|------------------------------|-----------------|
| | Mean MR/ASD+ | Mean MR/ASD | F | Sig. | T | Df | Sig. (2-tailed) |
| Problems Play & Leisure Time | .9271 | .7729 | 3.963 | .054 | 3.003 | 38 | .005** |

The group ASD/MR+ exhibited significantly more problems in the sub-domain Play and Leisure Time of the DISCO II.

Therefore, the level of play does not differ between the two groups, but the ASD/MR+ group experiences much more problems with play.

Motor Skills

The development of motor skills was measured by means of the VABS (Sparrow et al., 1984), namely Fine and Gross Motor Skills, and the PEP-R (Schopler et al., 1990), namely Eye-Hand-Integration, Fine and Gross Motor Skills. With the DISCO II (Wing, 1993a) problems in the VABS domains Fine and Gross Motor Skills were measured. The differences between the two groups were tested using t-tests for independent samples. If the significance level of Levene's test for Equality of Variance $\leq .05$, equal variances cannot be assumed (Field, 2000). The corresponding Sig-value can be found in the last column. The results of the two groups can be found in the following tables.

Table 5-49 T-tests for Motor Skills on VABS (V) and PEP-R (P)

| | | | Levene's Test for Equality of Variances | | | t-test for Equality of Means | |
|------------------------|--------------------|-------------------|---|------|-------|------------------------------|-----------------|
| | Mean DA MR/ASD+ | Mean DA MR/ASD | F | Sig. | t | Df | Sig. (2-tailed) |
| Total Motor V | 35.10 | 39.85 | 3.509 | .069 | -.814 | 38 | .421 |
| Gross Motor V | 38.00 | 41.90 | 1.489 | .230 | -.616 | 38 | .545 |
| Fine Motor V | 33.75 | 38.40 | 1.288 | .234 | -.717 | 38 | .478 |
| Eye-Hand-Integration P | 25.80 | 27.00 | .016 | .901 | -.280 | 38 | .781 |
| Gross Motor P | 38.00 | 35.70 | 1.411 | .242 | .382 | 38 | .705 |
| Fine Motor P | 28.30 | 30.35 | .548 | .464 | -.419 | 38 | .678 |

We found no differences between the ASD/MR+ and the ASD/MR groups in the variables of the development of motor skills.

Table 5-50 T-tests for Problems with Gross and Fine Motor Skills on the DISCO II

| | N MR/ASD+ | N MR/ASD | Levene's Test for Equality of Variances | | t-test for Equality of Means | | |
|-------------------------|--------------|-------------|--|------|---------------------------------|----|---------------------|
| | | | F | Sig. | t | Df | Sig. (2- tailed) |
| Total Problems Motor | .2415 | .1925 | .324 | .573 | 1.303 | 38 | .201 |
| Problems Gross Motor | .3517 | .2600 | .311 | .580 | 2.153 | 38 | .038** |
| Problems Fine Motor | .1313 | .1250 | .287 | .595 | .119 | 38 | .906 |

The group ASD/MR+ had significantly more problems with Gross Motor Skills than the group ASD/MR. In particular there was one item of the sub-domain Problems with Gross Motor Skills with a significant difference, namely 'spinning around own axis' ($t=2.107$, $\underline{\text{Sig}}=.047$).

Practical Skills

The development of Practical Skills was measured with the VABS (Personal, Domestic Skills and Community Skills). With the DISCO II problems in the VABS domains Fine and Gross Motor Skills were assessed. The differences between the two groups were tested with t-tests for independent samples. The results of the t-tests can be found in the following tables.

Table 5-51 T-tests for VABS Daily Living Skills

| | Mean DA MR/ASD+ | Mean DA MR/ASD | Levene's Test for Equality of Variances | | t-test for Equality of Means | | |
|---------------------------|--------------------|-------------------|--|------|---------------------------------|----|---------------------|
| | | | F | Sig. | t | Df | Sig. (2- tailed) |
| Total Daily Living Skills | 23.35 | 27.95 | .440 | .511 | -1.378 | 38 | .176 |
| Personal | 18.48 | 22.52 | 2.385 | .131 | -1.270 | 38 | .212 |
| Domestic | 17.98 | 23.02 | .523 | .474 | -1.623 | 38 | .113 |
| Community | 16.73 | 24.27 | .123 | .728 | -1.159 | 38 | .253 |

We found no significant differences between the ASD/MR+ and the ASD/MR groups in the variables for the development of Daily Living Skills.

Table 5-52 T-tests for DISCO II Daily Living Skills

| | Mean MR/ASD+ | Mean MR/ASD | Levene's Test for Equality of Variances | | t-test for Equality of Means | | |
|--------------------|-----------------|----------------|--|--------|---------------------------------|--------|---------------------|
| | | | F | Sig. | t | Df | Sig. (2- tailed) |
| Total Problems DLS | .4029 | .1721 | 10.318 | .003** | 6.559 | 30.093 | .000** |
| Problems Personal | .2088 | .1412 | 1.607 | .213 | 1.563 | 38 | .127 |

| | | | Levene's Test for Equality of Variances | | | t-test for Equality of Means | |
|--------------------|--------------|-------------|---|--------|-------|------------------------------|-----------------|
| | Mean MR/ASD+ | Mean MR/ASD | F | Sig. | t | Df | Sig. (2-tailed) |
| Problems Domestic | .0025 | 0.000 | 4.457 | .041** | 1.000 | 19.00 | .330 |
| Problems Community | .9750 | .3750 | 37.520 | .000** | 6.564 | 22.052 | .000** |

The group ASD/MR+ exhibited significantly more problems with the Total Problems Daily Living Skills and Problems with Community Skills.

When we look at the items we find significant differences in the items 'smearing faeces' ($t=1.742$, $Sig=.096$) and 'cooperation with dressing' ($t=2.478$, $Sig=.021$) of the problems of the Personal Skills domain and both items of the problems of the Community Skills domain, namely 'need for supervision' ($t=6.564$, $Sig=.000$) and 'lack of common sense' ($t=2.163$, $Sig=.042$). For all of these variables the scores were higher for the ASD/MR+ group.

Challenging behaviour

The presence of challenging behaviour was measured by the DISCO II (Wing, 1993a) and the DBC (Einfeld et al., 1995). The information about problems with adaptive behaviours was described above. In this section we will discuss the differences between the two groups in the specific behaviour domains of the DISCO II, Behaviour Affecting Others and Total Problems with Adaptive Behaviour and Total Specific Behaviour. The five factors of the DBC, namely Communication Disturbance, Social Relating, Disruptive/Antisocial, Self-absorbed, Anxiety and the Total Problem Behaviour Score will also be described. The description of the variables can be found in 5.2.3.2.

The differences between the groups were tested using t-tests for independent samples. If the significance level of Levene's test for Equality of Variance $\leq .05$, equal variances cannot be assumed (Field, 2000). The corresponding Sig-value can be found in the last column.

Table 5-53 T-tests for challenging behaviour in DISCO II and DBC

| N=36 | | | Levene's Test for Equality of Variances | | | t-test for Equality of Means | |
|----------------------------|--------------|-------------|---|--------|--------|------------------------------|-----------------|
| | Mean MR/ASD+ | Mean MR/ASD | F | Sig. | t | Df | Sig. (2-tailed) |
| Specific Objects | .4245 | .2875 | 5.337 | .026** | 4.195 | 34.004 | .000** |
| Specific Senses | .4638 | .3063 | 1.027 | .317 | 3.725 | 38 | .001** |
| Total Specific | .4441 | .2969 | .094 | .760 | 5.093 | 38 | .000** |
| Behaviour Affecting Others | .6885 | .2250 | .115 | .736 | 11.711 | 38 | .000** |
| Total Non-Adaptive | .4107 | .3017 | 1.065 | .309 | 4.463 | 38 | .000** |
| Communication | | | | | | | |
| Disturbance | 7.6667 | 7.4211 | .708 | .406 | .179 | 35 | .859 |
| Social Relating | 6.6111 | 6.4737 | .051 | .822 | .135 | 35 | .893 |
| Disruptive/Antisocial | 18.67 | 11.63 | .482 | .492 | 2.696 | 35 | .011** |
| Self-absorbed | 29.1667 | 22.3684 | 1.016 | .320 | 1.923 | 35 | .063* |
| Anxiety | 5.9444 | 4.5789 | 4.116 | .050 | 1.088 | 30.940 | .289 |
| TBPS | 67.8333 | 52.1579 | 2.057 | .160 | 2.166 | 35 | .037** |

We found significant differences ($Sig \leq .05$) between the two groups in the domains for specific behaviour problems (Specific Behaviour with Objects, Specific Behaviour Senses and Total Specific Behaviour); for Self-absorbed we found a score that is significant at the .1 level. We also found significant differences for most of the domains for non-specific behaviours (Behaviour Affecting Others, Disruptive/Antisocial behaviour and the Total Behaviour Problem score).

When we look at the items of the DISCO II, we find significant differences in the sub-domain Specific Sensory Behaviours for the items ‘smearing’ ($t=4.277$, $Sig=.000$), ‘self-injurious behaviour’ ($t=3.339$, $Sig=.002$), ‘self-stimulatory behaviour’ ($t=2.552$, $Sig=.015$), ‘repetitive destructive behaviour’ ($t=4.625$, $Sig=.000$), ‘insensitivity to pain’ ($t=2.135$, $Sig=.039$) and ‘oversensitivity for sounds’ ($t=1.966$, $Sig=.057$).

In the sub-domain Specific Behaviours with Objects we found significant differences in the items ‘repetitive behaviour with objects’ ($t=2.107$, $Sig=.042$) and ‘limited pattern of activities’ ($t=1.838$, $Sig=.075$).

For all of these items the group ASD/MR+ had a higher score than the ASD/MR group.

With the DBC we found significant differences for the items: ‘rude, swears at others’ ($t=2.062$, $Sig=.055$), ‘bangs head’ ($t=2.929$, $Sig=.008$), ‘lack of concentration’ ($t=1.705$, $Sig=.097$), ‘cries without reason’ ($t=2.436$, $Sig=.020$), ‘easily distracted’ ($t=2.660$, $Sig=.012$), ‘impatient’ ($t=1.981$, $Sig=.056$), ‘inappropriate sexual activities’ ($t=2.209$, $Sig=.042$), ‘impulsive’ ($t=2.121$, $Sig=.041$), ‘kicks or hits others’ ($t=2.412$, $Sig=.021$), ‘loud’ ($t=2.767$, $Sig=.009$), ‘overactive, restless’ ($t=1.687$, $Sig=.100$), ‘demands too much attention’ ($t=3.761$, $Sig=.001$), ‘sleeps too little/disturbed sleep’ ($t=2.057$, $Sig=.049$), ‘throws clothes’ ($t=2.8200$, $Sig=.008$), ‘throws objects’ ($t=3.086$, $Sig=.004$) and ‘manipulates or provokes others’ ($t=1.961$, $Sig=.058$). For all these items the group ASD/MR+ had higher scores.

The group ASD/MR had higher scores for the items: ‘does not react to feelings of others’ ($t=-1.773$, $Sig=.085$) and ‘shy’ ($t=-2.931$, $Sig=.006$).

Sleeping Problems

The DISCO II contains four items on sleeping problems. The t-tests for these items can be found in the following table.

Table 5-54 T-tests for DISCO-items for Sleeping Problems

| | Mean MR/ASD+ | Mean MR/ASD | Levene’s Test for Equality of Variances | | t-test for Equality of Means | | |
|-------------------------------|-----------------|----------------|--|--------|---------------------------------|--------|---------------------|
| | | | F | Sig. | t | Df | Sig. (2- tailed) |
| Sleep Medication | .4029 | .1721 | 23.834 | .000** | 2.354 | 28.892 | .026** |
| Falling Asleep | .2088 | .1412 | 7.811 | .008** | 1.584 | 33.204 | .123 |
| Sleeping Through the Night | .0025 | 0.000 | 38.223 | .000** | 3.532 | 24.590 | .002* |
| Nightmares | .9750 | .3750 | 38.000 | .000** | 2.373 | 19.000 | .028** |

The group ASD/MR+ had significantly more problems with sleeping through the night and had nightmares more often. They also used sleep medication more often.

5.4 Regression Analysis

In this paragraph we will report on a regression analysis performed to determine whether the variables that differ between the groups ASD/MR+ and ASD/MR have predictive value. We would like to know to which extent these variables can predict whether a child belongs in the group with challenging behaviour or the group without. Because of the number of variables, we made clusters of variables where possible. Since some of the variables and the variable 'group' are categorical, we need to apply logistic regression analysis (Field, 2000). The analyses will be described separately for the Biological, the Interpersonal and the Intrapersonal Contexts. We found no differences between the groups in the Superordinate context (see 5.3.3); therefore no analysis was performed on these variables.

5.4.1 Results of logistic regression analyses of the variables of the Biological Context

We used forward logistic regression analysis with the variable 'belonging to the group ASD/MR+ or ASD/MR' as the dependent variable and the following independent variables that were found to be significant in 5.3.2: mother's age at birth, examination of the eyes, result of EEG, Result of Cat-scan, use of major tranquillizer and use of anticonvulsant. In table 5-55 the results of this analysis are presented.

Table 5-55 Summary of Forward Logistic Regression Analysis of the variables of the Biological context (N=40)

| Variable | B | SE B | Wald | Sig | Percent Correct |
|---------------------------------|----------|----------|-------|-------|-----------------|
| Group (constant) | 8.7076 | 77.7791 | .0125 | .9109 | 54.84 |
| Step I | | | | | |
| Use of major tranquillizer | | | .0373 | .9815 | 74.19 |
| Use of major tranquillizer (1) | 10.3334 | 69.9346 | .0218 | .8825 | |
| Use of major tranquillizer (2) | -18.3789 | 148.0134 | .0154 | .9012 | |
| Step II | | | | | |
| Positive Result of CAT-scan (1) | -18.5890 | 104.5878 | .0316 | .8589 | 77.42 |

In total two variables from the biological context contributed significantly to prediction of the outcome. Whether a client belongs to group 2 (ASD/MR) has proven to be the most valid factor. All together 77.42% of cases can be predicted correctly on the basis of the model.

5.4.2 Results of logistic regression analysis of the variables of the Interpersonal Context

We applied a forward logistic regression analysis with the variable 'belonging to the group ASD/MR+ or ASD/MR' as the dependent variable and the following

independent variables that were found to be significant in 5.3.4: total problems with interpersonal relationships, having problems with the child and judgement of pedagogical situation. In table 5-56 the results of this analysis are described.

Table 5-56 Summary of Forward Logistic Regression Analysis of the variables of the Interpersonal context (N=40)

| Variable | <u>B</u> | <u>SE B</u> | Wald | <u>Sig</u> | Percent Correct |
|---|----------|-------------|--------|------------|-----------------|
| Group (constant) | 2.8625 | 1.4873 | 3.7045 | .0543 | 50 |
| Step I | | | | | |
| Total Problems with interpersonal relationships | -5.2108 | 2.6052 | 4.0007 | .0455 | 67.65 |

The variable Total Problems with interpersonal relationships predicts a significant part of the outcome. Whether a client belongs to group 1 (ASD/MR+) has proven to be the most valid. All together 67.65% of the cases can be predicted correctly on the basis of the model.

5.4.3 Results of logistic regression analysis of the variables of the Intrapersonal Context

We used forward logistic regression analysis with the variable 'belonging to the group ASD/MR+ or ASD/MR' as the dependent variable and two groups of independent variables, namely the intrapersonal factors of the development and the intrapersonal factors of the challenging behaviour.

5.4.3.1 Results of logistic regression analysis of the variables of Development

The following independent variables that were found to be significant in 5.3.5, were included in the analysis: dislike of care procedures, oversensitive to noise, odd movements, total setback in development of skills, total problems with socialization, problems with social interactions, problems with social skills, problems with play and leisure time, total problems with daily living skills, problems with community skills and problems with gross motor skills. In table 5-57 the results of this analysis are described.

Table 5-57 Summary of Forward Logistic Regression Analysis of the variables of the Intrapersonal context Development (N=40)

| Variable | <u>B</u> | <u>SE B</u> | Wald | <u>Sig</u> | Percent Correct |
|--------------------------------|----------|-------------|--------|------------|-----------------|
| Group (constant) | 44.4841 | 227.2569 | .0383 | .8448 | 50 |
| Step I | | | | | |
| Problems with community skills | -32.0034 | 227.1644 | .0198 | .8880 | 87.50 |
| Step II | | | | | |
| Dislike of care procedures | -7.8435 | 56.7684 | .0191 | .8901 | 87.50 |
| Step III | | | | | |
| Oversensitive to noise | -2.2218 | 1.2408 | 3.2063 | .0734 | 90.00 |
| Step IV | | | | | |
| Total Problems Socialization | -22.3615 | 13.9876 | 2.5557 | .1099 | 92.50 |

Four variables predict, together with the constant, a significant part of the outcome. All of the variables predict whether a client belongs to group 1, the group with challenging behaviour. In total the two variables for Problems with Adaptive Behaviour (Problems with Community and Total Problems Socialization) and the two variables for problems in the history (dislike of care procedures and oversensitive to noise) predict 92.5% of the cases correctly.

5.4.3.2 *Results of logistic regression analysis of the variables for Challenging Behaviour*

The following independent variables that proved to be significant in 5.3.5 were included in the analysis: Total Specific Problems, Specific Problems with Objects, Specific Problems with Senses, Behaviour Affecting Others, Total Non Adaptive Problems, Total Problem Score, Disruptive/Antisocial Behaviour, and Self-absorbed Behaviour. In table 5-58 the results of this analysis are described.

Table 5-58 Summary of Forward Logistic Regression Analysis of the variables in the Intrapersonal context Challenging Behaviour (N=40)

| Variable | B | SE B | Wald | Sig | Percent Correct |
|----------------------------|----------|--------|--------|-------|-----------------|
| Group (constant) | 7.6942 | 2.7042 | 8.0958 | .0044 | 51.35 |
| Step I | | | | | |
| Behaviour affecting others | -16.4267 | 5.5257 | 8.8376 | .0030 | 91.89 |

The variable 'behaviour affecting others' is a significant predictor of the outcome. It predicts whether a client belongs to the group ASD/MR+. Together with the constant, the predictive value is 91.89%. So more than nine out of ten cases will be assigned correctly to the group with challenging behaviour.

6 Summary

The aim of this project was to study challenging behaviour of children with an Autistic Spectrum Disorder. In the first phase we established that the group of children with an Autistic Spectrum Disorder and Mental Retardation show far more challenging behaviour than the group without Mental Retardation. Not only the prevalence of specific behaviours differs between the two groups, but especially the prevalence of non-specific behaviours. In fact, the more severe forms of challenging behaviour (e.g. aggression, destruction and self-injurious behaviour) seem to be the most distinguishing factor between the two groups, obviously next to the level of functioning. So challenging behaviour is much more common in the group with MR. The two groups can even be distinguished on the basis of the prevalence of challenging behaviour. The group with mild Mental Retardation seems to exhibit a different pattern. On the basis of this result we decided to perform the second phase of our study with the group of children with an ASD and MR, leaving out the group with mild Mental Retardation. The other factors that were of influence, namely chronological age, sex, and level of functioning, were used as matching criteria for the second phase.

The second phase was designed to investigate both the qualitative and the quantitative aspects of the development of children with an ASD and MR and extreme challenging behaviour. We designed a framework, using the bio-ecological model of Bronfenbrenner & Ceci (1994) and the developmental framework of Wenar & Kerig (1999), to gather the information in a more structured way. This framework has proven to be a useful tool in structuring the information. The group of children with an ASD and MR and extreme challenging behaviour was small (N=20) although we came in contact with many of the children who fitted our criteria and were referred by the advisory teams at that time; we must take the small sample size into consideration. Finding significant differences in small samples is much more difficult. Because of the lack of standardized instruments we drew up a control group of children with an ASD and MR, without extreme challenging behaviour. The matching procedure was based on coupled matching; for every child with challenging behaviour we searched for a child of the same chronological age, sex and developmental age. The matched group was by no means a-select.

In the following the results of the two phases in the intrapersonal, interpersonal, super-ordinate and organic contexts are summarized.

The Intrapersonal context

In the first phase we investigated mainly the intrapersonal factors. We found that the presence of Mental Retardation is a very important factor. The groups with and without mental retardation differed significantly in the following clusters: abnormalities of speech, abnormal responses to visual stimuli, abnormal proximal sensory stimulation, abnormal bodily movements, routines and resistance to change,

behaviour problems with limited or no social awareness, specific behaviour problems and the cluster according to Lund (1989).

Therefore the presence of Mental Retardation must be considered a significant risk factor.

In the second phase we focussed on the differences between the group with extreme challenging behaviour and the group without. When analysing the developmental history of the two groups we found significant differences at the Sig= .05 level for the items 'dislike of care procedures' and 'odd movements', the group with challenging behaviour had higher scores for these items. These items could be an indication of oversensitivity to certain stimuli. We also found that the group with challenging behaviour experienced a setback in skills more often ($p=.027$). The setback was particularly evident for Toilet Training Skills (Sig=.053), Language/Communication Skills (Sig=.088), Independent Feeding (Sig=.024) and Behaviour (Sig=.002).

We found no differences in the factor Intellectual Functioning between the two groups in the second phase. We had already established that this is an important factor and used it as a matching criterion. We found however differences in the cognitive function 'attention', namely the item 'concentration on activities proposed by others' ($p=.042$) and several items of the DBC for problems with hyperactivity and attention.

When we look at the behavioural level, we did not find differences at the level of development of communication, socialization and imagination. We did however find that the group of children with challenging behaviour experienced more problems in the Socialization domain (Sig=.000). Children with challenging behaviour also have more problems in the domains Gross Motor Skills (Sig=.038) and Daily Living Skills (Sig=.000).

The largest differences involved challenging behaviour. Both groups showed challenging behaviour, but the group ASD/MR+ exhibited more challenging behaviour, especially in DISCO II domains. With DISCO II we found differences in both the specific and non-specific behaviour domains. With the DBC we found significant differences in the Total Score and the domains Disruptive/Antisocial and Self-absorbed. The group ASD/MR+ also suffered significantly more Sleeping Problems.

From the regression analysis we learn that the problems with Community Skills, Dislike of Care Procedures, Oversensitivity to Noise and Total Problems with Socialization are the most predictive factors for the assignment to the group with challenging behaviour.

The Interpersonal context

As mentioned in the last paragraph, we found no differences in the development of social behaviour, but the group with challenging behaviour encounters more problems in their social interaction with both adults and peers. The only item of both

DISCO II and the DBC with a higher score for the group ASD/MR was 'lack of emotions' (Sig=. 081).

The parents of the group ASD/MR+ perceive their children as more problematic (Sig=. 044) and judge their pedagogical situation as more negative (Sig=. 049). This is not surprising, because their children are more difficult.

Regression analysis shows that 'problems with interpersonal relationships' is the best predictor for assignment to the group with challenging behaviour.

We found no differences in the super-ordinate context.

The Organic context

As far as the history is concerned, there was a difference in the age of the mothers. The mothers of the group with challenging behaviour were older (Sig=. 093). Almost all of the children in the group with challenging behaviour had eye examinations; this is more than in the group without (Sig=. 077). The group with challenging behaviour had many more positive results of medical examinations than the group without challenging behaviour, especially examinations of the brain. For the group without challenging behaviour the cause of abnormalities was known more often. This involves mainly disfunctioning of the brain. In this group more anticonvulsants were administered. In the group with challenging behaviour, more tranquillizers were used.

The use of tranquillizers and a positive result of a CAT-scan have proven to be the best predictors for assignment to the group without challenging behaviour.

7 Discussion and Recommendations

In this chapter we will discuss the results, give recommendations for future research and consider the consequences of our results for clinical practice.

7.1 Limitations of the study

Before discussing the results, we have to look at the limitations of this study. When designing a study in two phases, as we did in this project, it is desirable to draw independent samples from the subjects used in the first phase. So the selection of the groups ASD/MR with and ASD/MR without challenging behaviour should have been drawn at random from the sample of the first phase. This was however not possible. First the amount of time that passed between the two phases was too long, so a lot of the children would have been too old to be included in the sample. Another problem was that the subjects of this study could not be traced, because of the anonymity of the data.

The data included very limited information about the children, for example no information about the family and bio-medical data was available, and so no further factors could be examined using this particular set of data. Hence the choice to use the first phase only as a means to identify the factors necessary for sample selection for the second phase.

In the second phase, it was not difficult to draw up a group with challenging behaviour. We designed a study for 20 cases and the advisory teams Noord-Holland/Utrecht and Zuid-Holland/Zeeland initially came up with enough cases to fulfil the required 20 cases. None of the parents refused to cooperate, but some cases were not included because they did not meet the criteria: the level of functioning of the children was below 12 months, they did not live with their families at the time we contacted them, their additional handicaps were too severe or they could not be reached. So we included in our sample all children recommended by the two advisory teams who fitted our criteria. This procedure accounts for 15 cases. The others were referred to the Ambulatorium by the advisory teams Noord-Brabant/Limburg (four cases) and Gelderland/Overijssel/Flevoland (one case). We stopped collecting data at 20 cases. So the sample is not representative for the Netherlands, but almost all of the children with an ASD and MR and extreme challenging behaviour who were referred to the advisory teams in 1999 and fitted our criteria have been included in the sample. So it is safe to say that the sample is a fair representation of the group of children with an ASD and MR whose challenging behaviour has become so severe, that their caretakers have turned for help to the regional advisory teams.

Initially we did not plan to form a control group. However, when selecting the instruments we could use for our research, we found very few instruments that were

reliable and valid enough to justify any conclusions. Therefore we composed a control group. This was not an easy task. We asked the psychologist/pedagogue of the day-care centre or school the children attended to select a child on the basis of sex, age and developmental level and without extreme challenging behaviour. However, these professionals were often not able to provide a valid estimation of the level of functioning. The children they selected, often scored much higher on the test we used to establish the developmental age (PEP-R). And in addition, others also had very high scores on the Consensus Protocol, the scale we had selected as a criterion for the presence of challenging behaviour. All children with an ASD exhibited behaviour that was problematic for their surroundings, and by definition no child with an ASD and MR is without challenging behaviour. So we found 13 children by the above-mentioned method and matched the seven remaining children with clients who were referred to the clinic at Leiden University and fitted our criteria. Therefore the control group cannot be considered a-select. However, the two groups do not differ in background variables investigated (see 3.4).

A final point of discussion concerns the variables we have been investigating. We have studied an immense number of variables but have by no means been exhaustive. The information from the files and the available instruments for this specific group have their limitations. Especially for cognitive functioning, very few to no instruments are available to assess, for example, neuropsychological functions in these children. As far as treatment variables are concerned, we found considerable 'pollution' due to the fact that not only children with an ASD and MR get extra help but also all children who show extreme challenging behaviour. At the time of the assessments, 14 children of the group with challenging behaviour had a 1:1 child/caretaker ratio, compared to none of the control group. All 20 children with challenging behaviour were in a specific group for children with autism. In contrast, only eight of the children from the group without challenging behaviour were. So we could not look into the influence of treatment on the challenging behaviour, because of sample bias.

Another limitation concerns the biological factors; research was carried out from a behavioural point of view, not a bio-medical one. We obtained information from the files and we interviewed parents. We did not set out to gather medical data ourselves.

7.2 Discussion of the results

In this paragraph we will discuss the results, following the categorization as proposed by O'Brien (2000).

Predisposing factors

Children with an Autistic Spectrum Disorder show by definition behaviours that are not 'normal' and are difficult to understand by others. Since the groundbreaking study by Rutter et al. (1970) several researchers have shown that people with MR are at an increased risk for additional psychopathology (e.g. Benson, 1985; Došen, 1990; Emerson, 2001; O'Brien, 2000). This finding has recently been replicated in the Netherlands (Dekker, Koot, Van der Ende, & Verhulst, 2002). Fewer studies, however, have been carried out on children with the dual diagnosis Autistic

Spectrum Disorder and Mental Retardation. We have found that children with the dual diagnosis exhibit more specific behaviour problems than their age-matched peers without Mental Retardation; but more importantly, they also show more non-specific behaviour problems, such as self-injurious behaviour, aggression, destructive behaviour, etc. This finding is very similar to that of Kraijer (1998). So we must conclude that the presence of Mental Retardation is the main predisposing factor for the development of challenging behaviour of children with an Autistic Spectrum Disorder.

Dykens & Hodapp (2001, see 2.5) drew up a very long list of possible risk factors. We have not investigated all of these factors, mainly because of the low level of functioning of our group, especially social functioning. Therefore factors like less differentiated self-concepts, social strain and stigma have not been included in our study. However, we did find some of their biological risk factors, such as high rates of sensory impairments and biochemical or neurological anomalies. The children of the ASD/MR+ group have more medical problems. We found that much more specific result. We have also found some indications for sensory problems, such as oversensitivity to touch and sounds. Another indication of biomedical problems is the increased use of medication.

The level of functioning is clearly a significant risk factor for the development of problems with social awareness and problems with communication. Although poor communication and socialization skills are considered to be risk factors, a person needs at least some basic level to be able to show these kinds of problems. So both high and very low communication and socialization skills must be considered a protective factor. We have found that the quality of the social interaction of the ASD/MR+ group differs from that of the group without challenging behaviour. Whereas the group without challenging behaviour tends to be aloof or passive as far as the social interaction is concerned, the ASD/MR+ group shows a more bizarre form of interaction. The interaction cannot be fully characterized as 'active but odd' (Wing, 1996), because their functioning is too poor to really fit that category. These children do seek active contact, especially with adults, but they tend to cling to them. They seek attention actively, but seem unable to sustain a positive interaction. When they try to hug, they strangle the other person and when another child seeks contact, they often react aggressively. Their way of social interacting may best be categorized as 'disorganized'. They seem to have a difficult temperament and are very difficult to soothe. As a result, these children are often removed from social situations and are literally restrained in their seats. We see a similar pattern as far as communication is concerned. Whereas the group without challenging behaviour tends not to communicate, the group with challenging behaviour communicates to a major extent through their challenging behaviour. Although poor communication and social skills are risk factors, a child should at least show some intention to communicate before problems in this area can occur. All of the communication functions, as formulated by Durand & Carr (1991), can be found in this group, whereas the group without challenging behaviour tends not to ask for help and protest against unwanted events and activities. When escaping a stressful situation the group without challenging behaviour tends to escape in a more passive way.

Although we included the oversensitivity to sounds and touch under medical problems, the adverse reactions of children with challenging behaviour, could also be the result of more problems with sense making. We have found some scattered evidence that children with challenging behaviour have more problems with sustaining attention and are more easily distracted.

Janssen et al. (2002) proposed another theory to explain the emergence of challenging behaviour, namely as a result of stress and the lack of coping mechanisms. We have found some child factors that could result in more stress, especially the strong reaction to certain stimuli. The more aloof attitude of the children without challenging behaviour could also be viewed as a useful coping mechanism to deal with stressful stimuli.

Perpetuating factors

All children with an ASD and MR show at least some challenging behaviour. The forms of behaviour of the group with extreme challenging behaviour cannot, however, be ignored. The supervising adult has to react to the extremes of behaviour the child is showing, especially because so often other children are at risk. 'Adult attention' as a reinforcer is a very important perpetuating factor. As mentioned before, the behaviours per se define the amount of supervision. This finding together with the finding of Taylor & Carr (1992 a&b) that aloof children show more challenging behaviour when adult attention is increased and attention-seeking children show less challenging behaviour when adult attention is increased poses an interesting insight into how caregivers deal with these children. This mechanism could easily function as a self-fulfilling prophecy.

The problems with socialization and the resulting social isolation, for sure, lessen the chance of positive social interactions and learning experiences.

An important interpersonal factor is the perceived stress of the parents. The parents of children with challenging behaviour perceive their situation as difficult and they are often exhausted. Especially problems with the adaptive skills (e.g. feeding, sleeping, washing, etc.) are extremely stressful everyday routines for the parents. Other factors that could influence parental stress are the fact that in the group with challenging behaviour the cause of the problems is often not known. Parents and doctors are still searching for the reason and as a result more visits to the doctor are required due to further investigations and use of medication. Another factor that could play a role in enhancing parental stress is the fact that the children with challenging behaviour more often exhibit a setback in development.

We totally agree with the statement of Dykens & Hodapp (2001) that we have to consider these families as 'families under stress'. In this research project we found that the distinguishing factors between the two groups are the main factors that other researchers have found to influence parental stress, namely: extrovert behaviour problems, low adaptive skills and a poor physical condition (Van Berkum, 1993) and the challenging behaviour itself (Quine & Paul, 1985)

We have not found any indications that the parents or the parental skills differ between the two groups. The children of the group with challenging behaviour were more often described as difficult babies, with many problems in dealing with outside stimuli, including resistance to care procedures. This, together with the forms of behaviour these children show, especially their social behaviour, does raise the question of the quality of attachment between the parents and the afflicted child. We do not have specific data on this bond, but from observations we find that especially the bond between the mothers and the children is very close. The mother often functions as a buffer and interpreter between the child and the outside world.

Precipitating factors

We have not found any differences between the two groups as far as environmental and super-ordinate factors are concerned. The starting point of the challenging behaviour is rarely known, with the exception of the two children with West syndrome. The children with challenging behaviour were difficult almost from birth. We have found no indications that more environmental changes or stressful life-events occurred in the lives of the children with extreme challenging behaviour. It seems that they react more extremely to similar events.

7.3 Clinical implications

This research project has given us at least some insight into the risk and protective factors of the emergence of extreme challenging behaviour in children with an ASD and MR. These factors are summarized in table 7.1. We found factors within different contexts and emerging at different moments in the child's life. This finding calls for in-depth assessments. File analysis, amnestic interviews and a thorough assessment of current functioning are absolutely necessary to have any idea about the cause and nature of the problems.

Table 7-1 Risk factors for the emergence of challenging behaviour

| Intrapersonal Context | Interpersonal Context | Super-ordinate Context | Organic Context |
|---|---|------------------------|---|
| <u>History</u> | | | |
| * Odd Movements * Dislike of care procedures | * Difficult to soothe * More problems caring for | - | * Older mothers * Oversensitivity to certain stimuli |
| <u>Setback in skills</u> | | | |
| * Setback in being toilet-trained * Setback in development of communication * Setback in feeding-skills * Worsening of behaviour | - | - | - |

| Intrapersonal Context | Interpersonal Context | Super-ordinate Context | Organic Context |
|--|--|------------------------|---|
| <u>Current functioning</u> | | | |
| <ul style="list-style-type: none"> * Low level of general cognitive functioning * Attention problems * Hyperactive behaviour * Problems associated with gross motor skills * Problems associated with Daily Living Skills * Autism Specific behaviours * Disruptive/antisocial behaviours * Self-absorbed behaviours | <ul style="list-style-type: none"> * Problems with interpersonal relationships with both peers and parents * Children perceived as problematic by their parents * Negative judgement of pedagogical situation | - | <ul style="list-style-type: none"> * Cause of the problems unknown * Not clear medical problems * Use of tranquilizers |

The above-mentioned factors can be used to detect children at risk. Recognizing ‘at risk children’ is a very important means to prevent further escalation of the problems. These necessary assessments take time, time that should be spent on young children when it is still not clear what the cause of the problems is. Too often the occurrence of challenging behaviour is the cue to start further investigations, instead of assessing possibilities and problems when, for instance, they start at a day-care centre. The current system of indicating children for financial support should be based on the assessment of risk factors, instead of the occurrence of challenging behaviour.

Although we cannot classify the problems of the group with challenging behaviour according to DSM-IV (APA, 1994) criteria, most of the children do seem to have co-morbid conditions. The problems with the classification systems have already been noted (e.g. Došen, 1990; Dykens, 2000; Emerson, 2001), but that does, however, not mean that diagnosticians can neglect their duty to describe the symptoms as clearly as possible and take them into account when developing a treatment program. In this study we noted mainly problems with hyperactivity, psychotic tendencies and conduct problems in the ASD/MR+ group.

Exhaustive clinical assessments and observations, for the purpose of both diagnosis and evaluation, should be performed regularly. The variety of problems these children and their families face demand a multi-disciplinary approach. Too often the different disciplines perform their own diagnostics and treatment, without consulting other disciplines. The finding of Hodapp and Dykens (1994) that research into Mental Retardation had been split up into ‘two cultures’, namely the biomedical research and the behavioural research, can in our opinion also apply for clinical workers. Challenging behaviour is often viewed from either a behavioural or a medical point of view and if the treatment does not have the desired effect, the other discipline can be consulted. In our opinion challenging behaviour should be viewed

as a developmental problem and specialists from different ‘cultures’ should assess and treat the problems together.

Because of the immense problems these parents face, both parental guidance and parental support are indispensable. Van Berkum (1993) has named extrovert behaviour problems, low adaptive skills and poor physical condition of the child as the main inducers of parental stress. All of these factors are clearly visible in children with an ASD and MR and extreme challenging behaviour.

7.4 Implications for future research

The differences in the forms of challenging behaviour between the groups with and without MR and in the group with and without challenging behaviour again raise the issue of co-morbidity. The first phase clearly established that as far as challenging behaviour goes, the groups with and without MR are clearly distinct. But the groups from the second phase, namely ASD/MR with challenging behaviour and ASD/MR without, can also be distinguished on the basis of the challenging behaviour. Volkmar & Woolston (1995) have stated that two disorders should be viewed as a distinctive category, when the combination of the two differs from the two single disorders (see 2.3.5). This seems to be a correct assumption for both ASD with MR and ASD/MR with extreme challenging behaviour. Research into these separate groups is very crucial, because we have already established that research involving only the group ASD without MR does not provide us with sufficient information to deal with the challenges of the other groups.

As far as the cognitive development of the children with an ASD and MR and extreme challenging behaviour is concerned we found differences in attention. The early problems with sensory stimuli could be an indication of extra problems with central coherence (Frith, 2003), but there are also indications for more problems with the executive functions (Ozonoff, 1994) of this group. There are not only more problems with attention but the inhibition skills also seem much worse. The group without challenging behaviour tends to explore much less and therefore does not get into trouble and needs much less supervision, whereas the group with challenging behaviour seems to react to every stimulus. Whether this is on a cognitive or a biomedical level remains to be seen. It is even doubtful whether we should use terms like ‘attention’, ‘inhibition’, ‘concentration’, etcetera. The problems seem to occur at a very basic, organic level. The trouble with exploring this further is the total lack of instruments to assess the cognitive problems in low functioning children. Further research into these areas is crucial for insight in these problems. This research should take place at the three levels; clinical practice is not helped by independent biological, psychological or behavioural research. What we need are studies of the overall biological, psychological and behavioural level and its coherence. The group with mild Mental Retardation was excluded from this research project. It is very possible that within the group with mild MR both the low end of the high functioning population and the high end of the low functioning population are represented. This group calls for further study in itself.

The setback in skills exhibited by so many children of the group with challenging behaviour also needs further research. This result has been reported before (e.g. Hoshino et al., 1987; Davidovitch, et al., 2000). For some children one could argue that the children fit the category 'Childhood Disintegrative Disorder' (APA, 1994) better, but usually the setback occurred earlier, often around two years of age. Moreover the setback often occurred after the development had already been described as delayed. Further research on the influence of early development on the emergence of challenging behaviour is crucial, especially because the problems seem to be so persistent. The newest version of the DISCO (10th edition; Wing, 1999) provides us with the opportunity to gather information about current and past behaviour and could therefore become an important research tool.

Our research can only be qualified as 'exploratory' and much more research is needed. The proposed framework has however proven to be a useful tool for gathering information on the different dimensions. The results indicate that it is necessary to obtain information on all of these dimensions. The occurrence of challenging behaviour is a very complex phenomenon that needs extensive multi-disciplinary research, both for clinicians faced with a child with these problems and for researchers to gain further insight into these problems. Recently two large epidemiological studies were carried out in the Netherlands (Dekker, 2003; De Bildt, 2003), so the focus should now shift towards more individually based studies. The instruments described in table 3-3 have all proven to be useful tools for individual assessments that should form the basis for these studies.

Samenvatting

Onderhavig onderzoeksproject startte met de idee het probleemgedrag van kinderen met een autistische spectrumstoornis te onderzoeken. Daartoe is een opzet in twee fasen ontworpen. In de eerste fase, waarin gebruik werd gemaakt van een bestaand databestand van de Handicaps Behaviour and Skills Schedule (HBS; Wing, 1980), hebben we vastgesteld dat kinderen met een autistische spectrumstoornis én een verstandelijke beperking veel meer probleemgedrag vertonen dan kinderen met een ASD zonder een verstandelijke beperking. Dit geldt niet alleen voor het specifieke probleemgedrag, maar juist ook voor het voorkomen van niet-specifiek probleemgedrag. Het gaat hierbij om de meest ernstige vormen van probleemgedrag, zoals agressie, destructie en zelfverwondend gedrag. Sterker nog, de groepen onderscheiden zich, naast vanzelfsprekend door het niveau van functioneren, voornamelijk door deze vormen van probleemgedrag. Probleemgedrag blijkt één van de belangrijkste voorspellers voor de co-morbiditeit van een autistische spectrumstoornis en een verstandelijke beperking. Het probleemgedrag neemt af, naarmate de ontwikkeling toeneemt. De groep met een lichte verstandelijke beperking laat echter een ander patroon zien.

Op basis van deze resultaten hebben we besloten ons in de tweede fase specifiek te richten op de groep kinderen met een autistische spectrumstoornis én een verstandelijke beperking. De andere factoren waarvan was vastgesteld dat ze van invloed waren op het voorkomen van probleemgedrag, te weten chronologische leeftijd, sekse en niveau van functioneren, hebben we gebruikt als criteria om de controlegroep te matchen aan de onderzoeksgroep. Ook is bewust gekozen om alleen thuiswonende kinderen met een matige tot ernstige verstandelijke beperking bij het onderzoek te betrekken.

In de tweede fase van het onderzoek hebben we ons gericht op de kwalitatieve en kwantitatieve aspecten van de ontwikkeling van kinderen met een autistische spectrumstoornis en zeer ernstig probleemgedrag. We hebben een onderzoeksmodel ontworpen, gebruik makend van het bio-ecologisch model van Bronfenbrenner & Ceci (1994) en het 'developmental framework' van Wenar & Kerig (1999), aan de hand waarvan op een gestructureerde wijze informatie kon worden verzameld. We hebben zeer veel data verzameld over een relatief kleine groep. De groep kinderen met een autistische spectrumstoornis en zeer ernstig probleemgedrag omvatte slechts 20 kinderen. Deze 20 omvatten echter het leeuwendeel van de kinderen die aan de selectiecriteria voldeden en door het Consulenteenteam waren doorverwezen. Vanwege het gebrek aan gestandaardiseerde instrumenten juist voor deze doelgroep, hebben we middels gepaarde matching een controlegroep samengesteld van kinderen met een autistische spectrumstoornis en een verstandelijke beperking zonder probleemgedrag. Het ontwikkelingsniveau, de sekse en de chronologische leeftijd waren de belangrijkste matchingscriteria. De controlegroep is niet op aselechte wijze tot stand gekomen.

De intrapersoonlijke context

In de eerste fase hebben we alleen intrapersoonlijke factoren onderzocht. De aanwezigheid van een verstandelijke beperking bleek de belangrijkste factor. Concluderend kunnen we stellen dat de co-morbiditeit van een autistische spectrumstoornis met een verstandelijke beperking de belangrijkste risicofactor voor het ontstaan van probleemgedrag is.

In de tweede fase hebben we ons gericht op de verschillen tussen de groep met en de groep zonder zeer ernstig probleemgedrag. Als we kijken naar de ontwikkelingsgeschiedenis van de twee groepen, vinden we significant hogere scores voor de groep met zeer ernstig probleemgedrag op de items: hekel aan verzorging en vreemde bewegingen. Deze items lijken een indicatie voor een overgevoeligheid voor bepaalde stimuli. We hebben ook vastgesteld dat de groep met ernstig probleemgedrag vaker een achteruitgang in vaardigheden laat zien, vooral met betrekking tot zindelijkheid, taal/communicatie, zelf eten en gedrag.

We hebben geen verschillen met betrekking tot de intelligentie tussen de groepen gevonden. We hebben echter wel een aantal verschillen met betrekking tot de cognitieve functies gevonden, namelijk met betrekking tot de aandacht. Bij de groep met ernstig probleemgedrag worden meer problemen met de concentratie voor activiteiten die door anderen zijn voorgesteld, gesignaleerd en deze groep scoort ook hoger op een aantal items van de VOG (Koot & Dekker, 2002), vooral de items die betrekking hebben op hyperactiviteit en problemen met de aandacht.

Wanneer we naar het gedragsniveau kijken, vinden we geen verschillen met betrekking tot de ontwikkeling van communicatie, socialisatie en verbeelding. Er wordt echter wel veel meer probleemgedrag op het socialisatiedomein geconstateerd. Ook op de domeinen grove motoriek en dagelijkse vaardigheden worden meer problemen geconstateerd.

Het grootste verschil heeft echter betrekking op het probleemgedrag zelf. De groep met ernstig probleemgedrag scoort veel hoger op zowel de specifieke als de niet-specifieke gedragsdomeinen. Op de VOG behaalt de groep met ernstig probleemgedrag een veel hogere totaalscore en hogere scores op de domeinen antisociaal gedrag en in zichzelf gekeerd gedrag. Ook vertoont de groep met ernstig probleemgedrag veel meer slaapproblemen.

Uit de regressie-analyse blijkt dat problemen met maatschappelijke vaardigheden, hekel aan verzorging, overgevoeligheid voor geluid en problemen m.b.t. socialisatie de hoogste voorspellende waarde voor toewijzing aan de groep met ernstig probleemgedrag hebben.

De interpersoonlijke context

De ontwikkeling van het sociale gedrag verschilt niet tussen de twee groepen, maar de groep met ernstig probleemgedrag laat wel veel meer problemen zien in de relaties met zowel volwassenen als met kinderen. Er is slechts één item waarop de groep zonder probleemgedrag hoger scoort en dat is 'gebrek aan emoties'. Problemen met interpersoonlijke relaties blijken de beste voorspeller voor toewijzing tot de groep met ernstig probleemgedrag.

De ouders van de groep met ernstig probleemgedrag ervaren hun kinderen als meer problematisch en waarderen de opvoedingssituatie negatiever.

We hebben geen verschillen met betrekking tot de superordinatie context gevonden.

De organische context

De moeders van de groep met ernstig probleemgedrag zijn ouder dan de moeders uit de controlegroep.

In beide groepen zijn frequent medische onderzoeken uitgevoerd. Er worden vaker medische problemen geconstateerd in de groep met ernstig probleemgedrag, bijvoorbeeld visusproblemen en onduidelijke hersenafwijkingen. Van de groep zonder probleemgedrag is echter de etiologische diagnose bekend, dit zijn veelal specifieke hersendisfuncties. In deze groep worden ook vaker anti-epileptica voorgeschreven, terwijl in de groep met ernstig probleemgedrag meer frequent kalmerende middelen gebruikt worden.

Het niet gebruiken van kalmerende middelen en een positief resultaat op een CAT-scan blijken de beste voorspellers van toewijzing tot de groep zonder ernstig probleemgedrag.

Beperkingen van het onderzoek

Bij een studie in twee fasen is het voor de hand liggend om in de tweede fase een onafhankelijke steekproef uit de eerste fase te trekken. Dit was in dit geval echter niet meer mogelijk. Ten eerste was er te veel tijd verstreken, waardoor veel kinderen al ouder dan 12 jaar waren en ten tweede waren veel kinderen niet meer te traceren. De dataset uit de eerste fase bevatte zeer weinig informatie; gegevens over familieomstandigheden, biomedische informatie enzovoort waren niet beschikbaar voor analyse. Dit heeft ertoe geleid dat de resultaten van de eerste fase vooral zijn gebruikt ten behoeve van het vaststellen van de selectiecriteria voor de onderzoeksgroep en de controlegroep van de tweede fase.

Het bleek geen probleem om kinderen met een ASS en een verstandelijke beperking en ernstig probleemgedrag te vinden. De consulenteams Noord-Holland/Utrecht en Zuid-Holland/Zeeland leverden 20 cases. Alle ouders wilden meewerken, maar een aantal kinderen zijn uitgesloten, omdat ze niet aan de gestelde criteria voldeden; het niveau van functioneren bleek lager dan 12 maanden, ze woonden ten tijde van het onderzoek niet meer thuis, er waren te ernstige bijkomende problemen of het lukte niet hen te traceren. Uiteindelijk hebben 15 van de 20 aangebrachte kinderen deelgenomen aan het onderzoek. De andere vijf kinderen zijn naar het Ambulatorium verwezen door de consulenteams Noord-Brabant/Limburg (vier) en Gelderland/Overijssel/Flevoland (één). De steekproef is niet aselekt tot stand gekomen, maar bijna alle Nederlandse kinderen met een ASS en een verstandelijke beperking en zeer ernstig probleemgedrag waarvan de ouders/begeleiders in 1999 contact met de consulenteams hebben opgenomen en die aan de criteria voldeden, zijn in de steekproef opgenomen. We mogen ervan uitgaan dat de steekproef representatief is voor de Nederlandse populatie thuiswonende kinderen met een ASS en een matige tot ernstige verstandelijke beperking en ernstig probleemgedrag.

In de eerste opzet was geen controlegroep opgenomen. Echter bij het selecteren van een onderzoeksbatterij bleken er zeer weinig betrouwbare en valide instrumenten met normen voor de geselecteerde groep. Om die reden moest een controlegroep worden samengesteld. De gedragswetenschapper van het dagcentrum of de school waar het kind uit de proefgroep overdag heen ging, is gevraagd een kind als controle te selecteren op basis van sekse, leeftijd en ontwikkelingsleeftijd. Snel bleek echter dat vooral de ontwikkelingsleeftijd vaak fout werd ingeschat. Ook bleken de kinderen die voor de controlegroep aangedragen werden toch vaak veel probleemgedrag te vertonen. Vanwege de aard van hun problematiek hebben veel kinderen met een ASS gedragsproblemen. Uiteindelijk zijn via de dagverblijven en scholen 13 kinderen geselecteerd. De overige zeven waren cliënten van het Ambulatorium en voldeden aan de gestelde criteria. De controlegroep is niet op aselechte wijze tot stand gekomen. De groepen verschillen echter niet wat betreft de achtergrondvariabelen (zie 3.4).

Een laatste discussiepunt betreft de onderzochte variabelen. Er is een zeer groot aantal factoren bekeken, maar deze kunnen geenszins als uitputtend worden beschouwd. De informatie uit de dossiers en de voor de doelgroep beschikbare instrumenten hebben hun beperkingen, zeker met betrekking tot het cognitieve niveau. Er zijn bijvoorbeeld geen instrumenten beschikbaar die neuropsychologische functies bij deze groep kinderen kunnen vaststellen. Het bleek ook onmogelijk naar de behandeling/begeleiding te kijken, vanwege vervuilende factoren. Alle kinderen met ernstig probleemgedrag bleken extra hulp te krijgen, 14 hadden zelfs voortdurend 1:1-begeleiding. Ook bleken alle kinderen met probleemgedrag in een anti-groep te zitten, tegenover slechts acht kinderen uit de controlegroep. Onderhavig onderzoek is uitgevoerd door een orthopedagoog en niet door een medicus. Alle medische informatie is verkregen uit de tweede hand, namelijk uit de dossiers of via de interviews met ouders.

Discussie van de resultaten

De resultaten worden bediscussieerd aan de hand van de categorieën van O'Brien (2000).

Risicofactoren voor het ontstaan van probleemgedrag

Kinderen met een autistische spectrumstoornis vertonen per definitie gedrag dat door anderen als abnormaal wordt gezien. Sinds de eerste studies van Rutter et al. (1970) is meermalen vastgesteld dat mensen met een verstandelijke beperking een verhoogd risico voor psychopathologie lopen (zie bijvoorbeeld: Benson, 1985; Došen, 1990; Emerson, 2001; O'Brien, 2000). Recent is ook in Nederland onderzoek uitgevoerd dat deze bevindingen bevestigt (Dekker, Koot, Van der Ende & Verhulst, 2002). Er is echter veel minder onderzoek uitgevoerd bij kinderen met de dubbele diagnose autistische spectrumstoornis en verstandelijke beperking. Wij hebben vastgesteld dat kinderen met deze dubbele diagnose meer specifiek probleemgedrag dan hun leeftijdgenoten zonder een verstandelijke beperking vertonen; veel belangrijker is echter dat ze ook meer niet-specifiek probleemgedrag als zelfverwonding, agressie en destructie vertonen. Dit is in 1998 door Kraijer ook bij volwassenen vastgesteld. De aanwezigheid van een verstandelijke beperking is de belangrijkste risicofactor

voor het ontstaan van probleemgedrag bij kinderen met een autistische spectrumstoornis.

Dykens & Hodapp (2001, see 2.5) hebben een lange lijst risicofactoren geformuleerd. Wij hebben niet al deze factoren bekeken vanwege het lage functioneringsniveau van onze onderzoeksgroep, vooral m.b.t. de sociale ontwikkeling. Dit impliceert dat factoren als zelfbewustzijn en sociale stigmata niet onderzocht zijn. We hebben echter wel gelijkaardige biologische factoren als een hoog percentage zintuiglijke problemen en biochemische en neurologische afwijkingen gevonden. De kinderen met een ASS en een verstandelijke beperking en ernstig probleemgedrag hebben vaker medische problemen, veelal zonder duidelijke oorzaak. In deze groep worden ook vaker indicaties voor zintuiglijke problemen als overgevoeligheid voor bepaalde prikkels en geluiden gevonden. Ook wordt in deze groep meer medicatie gebruikt.

Met betrekking tot de sociale interactie valt op dat er kwalitatieve verschillen tussen de twee groepen zijn. De groep zonder probleemgedrag kan eerder gekarakteriseerd worden als afzijdig of passief, terwijl de groep met probleemgedrag de interactie wel aangaat, maar dan snel in de problemen geraakt. Ze klampen zich vast en vragen voortdurend en uitsluitend de aandacht. Een knuffel wordt een wurggreep en ze reageren vaak agressief op pogingen tot contact van anderen. Dit geldt zowel voor de interactie met volwassenen als met kinderen. De interactie kan het best gekarakteriseerd worden als bizar, maar past niet helemaal in de ‘actieve maar bizarre’ categorie van Wing (1996), vanwege het zwakke niveau. Deze kinderen lijken een moeilijk temperament te hebben en zijn moeilijk te troosten. De sociale omgeving van deze kinderen wordt ernstig ingeperkt en deze kinderen worden vaak zelfs letterlijk in hun stoeltje vastgezet. Ook met betrekking tot de communicatie zien we een dergelijk verschil; de groep zonder probleemgedrag doet nauwelijks pogingen tot communicatie, terwijl de groep met probleemgedrag voornamelijk communiceert middels probleemgedrag.

Hoewel slechte communicatieve en sociale vaardigheden risicofactoren zijn, moet een kind in staat zijn tot intentionaliteit om problemen op deze gebieden te laten zien. Dus zowel relatief goede, als slechte sociale en communicatieve vaardigheden moeten als protectieve factoren worden beschouwd.

De overgevoeligheid voor geluid en aanraking zijn beschreven onder de medische problemen; deze afweer kan echter ook een gevolg zijn van meer problemen met de betekenisverlening. We hebben tevens een aantal aanwijzingen gevonden dat de kinderen met ernstig probleemgedrag meer problemen hebben om de aandacht vast te houden en dat ze sneller afgeleid zijn.

Janssen et al. (2002) beweren dat probleemgedrag het resultaat van stress en een gebrek aan ‘coping’-strategieën is. We hebben een aantal kindfactoren gevonden die zouden kunnen resulteren in een hoger stressniveau, vooral de afwijkende reactie op bepaalde stimuli. De meer teruggetrokken houding van de kinderen zonder probleemgedrag zou ook gezien kunnen worden als een zinvolle coping-strategie om met deze prikkels om te gaan.

Factoren die probleemgedrag in stand houden

Alle kinderen met een ASS en een verstandelijke beperking laten enige vorm van probleemgedrag zien. De vorm en de ernst van het probleemgedrag van de onderzoeksgroep kunnen echter niet worden genegeerd. De verantwoordelijke volwassene moet wel reageren, zeker als andere kinderen in gevaar komen. De aandacht van volwassenen als gedragsversterker is een gekende factor die gedrag in stand kan houden. Echter, juist het probleemgedrag is een zeer bepalende factor bij de bepaling van de begeleider: kind-ratio. Dit gegeven, samen met de bevindingen van Taylor & Carr (1992 a&b) dat de gedragsproblemen bij kinderen die in zichzelf gekeerd zijn toenemen als de aandacht toeneemt en bij kinderen die veel aandacht vragen het probleemgedrag juist afneemt met toenemende aandacht, werpt een nieuw licht op de gewenste wijze van omgang met deze kinderen. Er zou hier wel eens sprake kunnen zijn van een ‘self-fulfilling prophecy’.

De stress die ouders ervaren is een belangrijke interpersoonlijke factor. De ouders van de kinderen met ernstig probleemgedrag verkeren voortdurend in een moeilijke situatie en zijn vaak uitgeput. Vooral de dagelijks terugkerende problemen rondom eten en slapen zijn zeer belastend. Een andere factor die van invloed lijkt op de stress die ouders ervaren is de onduidelijkheid over de oorzaak van de problematiek. Ouders zijn vaak nog op zoek naar duidelijkheid, met regelmatig doktersbezoek als gevolg. Ook het verlies van reeds verworven vaardigheden is voor ouders moeilijk te accepteren.

Dykens en Hodapp (2001) beschrijven families met een kind met een verstandelijke beperking en probleemgedrag als ‘families under stress’. Externaliserend probleemgedrag, zwakke adaptieve vaardigheden en problemen met de gezondheid (zie ook Van Berkum, 1993) en het probleemgedrag zelf (zie ook Quine & Paul, 1985) blijken ook in dit onderzoek de belangrijkste stressoren.

De opvoedingsvaardigheden van de ouders lijken niet te verschillen. De kinderen met ernstig probleemgedrag worden van baby af aan beschreven als moeilijk en de problemen in de verwerking van prikkels worden ook vanaf zeer jong beschreven. Dit gegeven samen met vooral de problemen in de sociale interactie, vormen een bedreiging voor het vormen van veilige hechting. Hoewel hechting geen onderwerp van dit onderzoek was, viel wel op dat er vaak sprake is van een zeer hechte band tussen de moeders en de kinderen. De moeders lijken als buffer tussen en vertaler voor de buitenwereld en het kind te fungeren.

Factoren die een directe aanleiding voor probleemgedrag kunnen zijn

We hebben geen verschillen in de omgevingsfactoren (bijvoorbeeld veranderingen in de omgeving, significante life-events) gevonden. Het lijkt alsof de kinderen uit de groep met ernstig probleemgedrag sterker reageren op gelijkaardige gebeurtenissen.

Implicaties voor de klinische praktijk

Onderhavig onderzoeksproject heeft ons in ieder geval enig inzicht in de risico en protectieve factoren voor het ontstaan van zeer ernstig probleemgedrag bij kinderen met een autistische spectrumstoornis en een verstandelijke beperking gegeven. Deze

factoren zijn samengevat in onderstaande tabel. We hebben factoren in de verschillende contexten beginnend op verschillende momenten in het leven van een kind, gevonden. Dit vraagt om het uitgebreid in kaart brengen van de kinderen. Dossieronderzoek, anamnestiche onderzoeken en het uitgebreid in kaart brengen van het huidige functioneren zijn voorwaarden om de oorzaken en het verloop van de problemen in kaart te kunnen brengen.

Risicofactoren voor het ontstaan van ernstig probleemgedrag

| Intrapersoonlijke context | Interpersoonlijke context | Superordinatie | Organische context |
|--|---|----------------|---|
| <u>Voorgeschiedenis</u> | | | |
| * Vreemde bewegingen * Weerstand tegen verzorgd worden | * Moeilijk te troosten * Veel problemen in de dagelijkse verzorging | - | * Oudere moeders * Overgevoeligheid voor bepaalde stimuli |
| <u>Achteruitgang in vaardigheden</u> | | | |
| * Achteruitgang zindelijkheid * Achteruitgang m.b.t. de communicatie * Achteruitgang m.b.t. het eten * Gedragsverslechtering | - | - | - |
| <u>Huidig functioneren</u> | | | |
| * Laag gemiddeld functioneringsniveau * Aandachtsproblemen * Hyperactief gedrag * Problemen m.b.t. de grove motoriek * Problemen m.b.t. de dagelijkse vaardigheden * Specifiek autistisch gedrag * Storend en antisociaal gedrag * In zichzelf gekeerd gedrag | * Problemen m.b.t. de interpersoonlijke relaties zowel met leeftijdsgenoten als volwassenen * Kinderen worden door de ouders moeilijk gevonden * Negatieve waardering van de opvoedingssituatie | - | * Oorzaak van de problemen onbekend * Geen duidelijke medische problemen * Gebruik van kalmerende |

De bovengenoemde factoren kunnen gebruikt worden om risicokinderen op te sporen. Dit is noodzakelijk om verdere escalatie van de problemen te voorkomen. De daartoe benodigde onderzoeken kosten tijd, deze tijd moet worden besteed aan jonge kinderen waarbij nog zo veel onduidelijk is. Nog te vaak is het ontstaan van probleemgedrag de aanleiding om verder onderzoek in te zetten in plaats van het in kaart brengen van mogelijkheden en problemen op het moment van opname in bijvoorbeeld een kinderdagcentrum. De risicotaxatie zou financiële ondersteuning moeten genereren, in plaats van al bestaand probleemgedrag.

Hoewel we de problemen van de groep met ernstig probleemgedrag niet kunnen classificeren m.b.v de DSM-IV (APA, 1994), is er bij de meeste kinderen sprake van comorbide stoornissen. In dit onderzoek hebben we vooral hyperactiviteitsproblemen, psychotische verschijnselen en gedragsproblemen gesignaleerd. De problemen met de classificatiesystemen zijn ook al door andere auteurs beschreven (zie ook Došen, 1990; Dykens, 2000; Emerson, 2001), maar dat ontslaat diagnostici niet van hun plicht de symptomen zo duidelijk mogelijk te omschrijven en te betrekken bij het behandelingsplan.

Uitgebreide klinische diagnostiek en observaties, zowel ten behoeve van de diagnostiek als de evaluatie zijn noodzakelijk. De vele soorten probleemgedrag waar deze kinderen en hun families mee te maken hebben, vereisen een multidisciplinaire benadering. Te vaak doet elke discipline zijn eigen ding, zonder andere deskundigen daarbij te betrekken. Hodapp en Dykens (1994) hebben reeds aangegeven dat onderzoek bij mensen met een verstandelijke beperking is verdeeld in twee culturen, namelijk het biomedische onderzoek en het gedragsonderzoek. Wat ons betreft geldt dit ook voor het werk van klinici. Probleemgedrag wordt vaak bekeken ofwel vanuit de gedragsmatige, ofwel vanuit de medische invalshoek; als de ene behandelingswijze te weinig effect sorteert, mag de andere discipline het proberen. Gedragsproblemen zijn ontwikkelingsproblemen en specialisten vanuit de verschillende ‘culturen’ moeten de problemen gezamenlijk in kaart brengen en behandelen.

Vanwege de gigantische problemen waar ouders mee te maken hebben is ouderbegeleiding en –ondersteuning een absolute noodzaak. Van Berkum (1993) heeft reeds aangegeven dat externaliserend probleemgedrag, zwakke adaptieve vaardigheden en een slechte gezondheid de meeste ouderlijke stress veroorzaken. Al deze factoren zien we ook in onze groep kinderen met een autistische spectrumstoornis en een verstandelijke beperking en zeer ernstig probleemgedrag.

Implicaties voor verder onderzoek

De verschillen in de vormen van probleemgedrag tussen de groepen met en zonder verstandelijke beperking en de groepen met en zonder ernstig probleemgedrag roepen opnieuw de vraag naar comorbiditeit op. In de eerste fase is vastgesteld dat de groepen met en zonder verstandelijke beperking duidelijk verschillen. Maar de groepen uit de tweede fase met en zonder ernstig probleemgedrag kunnen ook op basis van het probleemgedrag worden onderscheiden. Volkmar en Woolston (1995) geven aan dat twee stoornissen als apart moeten worden beschouwd als de combinatie van de twee verschilt van de twee aparte stoornissen (zie 2.3.5). Dit lijkt ook op te gaan voor de groepen met en zonder ernstig probleemgedrag. Onderzoek naar deze twee groepen is van wezenlijk belang, omdat we al hebben vastgesteld dat onderzoek naar alleen de groep zonder een verstandelijke beperking te weinig informatie oplevert om de andere groepen adequaat te kunnen behandelen.

Met betrekking tot de cognitie hebben we verschillen in de aandacht gevonden. De vroege problemen m.b.t. de sensorische stimuli zouden een indicatie voor grotere

problemen met de centrale coherentie kunnen zijn (Frith, 2003). Maar we vinden ook aanwijzingen voor meer problemen met de executieve functies (Ozonoff, 1994) in de groep met ernstig probleemgedrag. Niet alleen zijn er meer aandachtsproblemen, ook de inhibitie lijkt veel zwakker. De groep zonder ernstig probleemgedrag exploreert veel minder en geraakt niet in de problemen en heeft daardoor minder toezicht nodig, terwijl de groep met probleemgedrag lijkt te reageren op elke stimulus. Of dit een probleem op het cognitieve of biomedische niveau is, verdient verder onderzoek. Het is zelfs twijfelachtig of we kunnen spreken in termen als ‘aandacht’, ‘inhibitie’, ‘concentratie’ enz. De problemen lijken zich voor te doen op een basaal, organisch niveau. Het is moeilijk om dit verder te onderzoeken, vanwege het volledig ontbreken van instrumentarium om de cognitieve problemen van laagfunctionerende kinderen in kaart te brengen. Toekomstig onderzoek op dit gebied zou op drie niveaus moeten plaatsvinden. De klinische praktijk is niet geholpen bij op zichzelf staand biologisch, psychologisch of gedragsonderzoek. Er is een grote behoefte aan onderzoek dat zowel de biologische, psychologische alsook de gedragsfactoren en de onderlinge samenhang bekijkt.

De groep met een lichte verstandelijke beperking is uitgesloten van dit onderzoek. Het is goed mogelijk dat in deze groep zowel de zwakste groep hoogfunctionerende kinderen, alsook de sterkste groep laagfunctionerende kinderen zijn vertegenwoordigd. Deze groep moet als aparte groep verder worden bekeken.

Ook de achteruitgang in vaardigheden die de groep kinderen met ernstig probleemgedrag laat zien, moet verder bezien worden. Dit resultaat is eerder gerapporteerd (bijv. Hoshino et al., 1987; Davidovitch et al., 2000). Sommige kinderen lijken eerder te voldoen aan de criteria van de categorie ‘Desintegratieve Psychose van de Kinderleeftijd’ (APA, 1994), maar veelal doet de terugval zich rond het tweede jaar voor. Sterker nog, de achteruitgang begon een aantal keer als de ontwikkeling al als (licht) vertraagd was aangemerkt. Onderzoek naar de invloed van de vroege ontwikkeling op het ontstaan van ernstig probleemgedrag is cruciaal, vooral omdat de problemen zo persistent blijken. De laatste versie van de DISCO (10^e versie; Wing, 1999) biedt de mogelijkheid om zowel huidig als vroeger gedrag in kaart te brengen en is daarmee een belangrijk onderzoeksinstrument.

Wij hebben een exploratief onderzoek uitgevoerd en er is nog veel meer onderzoek noodzakelijk. De gepresenteerde modellen zijn bruikbaar gebleken bij het verzamelen en ordenen van informatie op de verschillende dimensies. Het vóórkomen van ernstig probleemgedrag is een zeer gecompliceerd fenomeen dat vraagt om intensief multidisciplinair onderzoek, zowel ten behoeve van de praktijk als ten behoeve van de wetenschap. Recentelijk zijn twee grote Nederlandse epidemiologische onderzoeken uitgevoerd (Dekker, 2003; De Bildt, 2003), dus het wordt tijd om de aandacht te verleggen naar meer op het individu gerichte studies.

References

- Abidin, R.R. (1983). *Parenting Stress Index: Manual*. Charlottesville, VA: Pediatric Psychology Press.
- Achenbach, T.M. (1997). *Empirically based assessment of child and adolescent psychopathology: Practical implications*. Thousand Oaks, CA: Sage.
- Achenbach, T.M., & Edelbrock, C. (1983). *Manual for the Child Behavior Checklist and revised Child Behavior Profile*. Burlington, VT: Vermont University Association in Psychiatry.
- American Psychiatric Association (1994). *Diagnostic and statistical manual* (4th ed.). Washington, DC: APA.
- American Psychiatric Association (1998). *Publication manual of the American psychological association* (10th ed.). Washington, DC: APA
- American Psychiatric Association (2000). *Diagnostic and statistical manual*, (4th ed.), text revision. Washington, DC: APA.
- Asperger, H. (1944). Die 'Autistische Psychopathen' im Kindesalter. *Archives Psychiatrie Nervenkreis*, 117, 76-136.
- Bailey, A., Philips, W., & Rutter, M. (1996). Autism: Towards an integration of clinical, genetic, neuropsychological and neurobiological perspectives. *Journal of Child Psychology, Psychiatry and allied disciplines*, 37, 89-126.
- Baron-Cohen S. (1989). Perceptual role-taking and protodeclarative pointing in autism. *British Journal of Developmental Psychology*, 7, 113-127.
- Baron-Cohen, S., Leslie, A.M., & Frith, U. (1985). Does the autistic child have a 'theory of mind'? *Cognition*, 21, 37-46.
- Benson, B.A. (1985). Behaviour disorders and mental retardation: Associations with age, sex and level of functioning in an outpatient clinic sample. *Applied Research in Mental Retardation*, 6, 79-85.
- Berkson, G., Tupa, M., & Sherman, L. (2001). Early development of stereotyped and self-injurious behaviors: I. Incidence. *American Journal on Mental Retardation*, 106, 539-547.
- Bernsen, A.H. (1980). The children's Handicap, Behaviour and Skills (HBS) schedule. In E. Strömgen, A. Dupont & J.A. Nielsen (Eds.). *Epidemiological research as a basis for the organization of extramural psychiatry* (pp. 133-139). Copenhagen: Munksgaard.
- Bettelheim, B. (1967). *The empty fortress. Infantile autism and the birth of the self*. New York/London: Free Press.
- Bleuler, E. (1911). Dementia Praecox oder Gruppe der Schizophrenien. In *Handbuch der Psychiatrie*, spezieller Teil, 4. Abt. 1. Hälfte. Leipzig-Wien.
- Bodfish, J.W., Symons, F.J., Parker, D.W., & Lewis, M.H. (2000). Varieties of repetitive behavior in autism: Comparisons to mental retardation. *Journal of Autism and Developmental Disorders*, 30, 237-243.
- Borthwick-Duffy, S.A. (1994). Prevalence of destructive behaviors. In T. Thompson & D.B. Gray (Eds.), *Destructive behavior in developmental disabilities: Diagnosis and treatment* (pp. 3-23). Thousand Oaks, CA: Sage.

- Bronfenbrenner, U., & Ceci, S.J. (1994). Nature-nurture reconceptualized in developmental perspective: A bioecological model. *Psychological Review*, *101*, 568-586.
- Bruininks, R.H., Hill, B.K., & Morreau, L.E. (1988). Prevalence and implications of maladaptive behaviors and dual diagnosis in residential and other service programs. In J.A.F.J. Menolascino, M.H. Albarelli & V.C. Gray (Eds.), *Mental retardation and mental health*. New York, NY: Springer-Verlag.
- Brylewski, J., & Wiggs, L. (1999). Sleep and challenging behaviour. *Journal of Intellectual Disability Research*, *43*, 504-512.
- Burack, A.J. (1997). The study of atypical and typical populations in developmental psychopathology: The quest for a common science. In S.S. Luthar, J.A. Burack, D. Cicchetti & J.R. Weisz (Eds.), *Developmental psychopathology; Perspectives on adjustment, risk and disorder* (pp. 139-165). Cambridge, MA: University Press.
- Carr, E.G., & Durand, V.M. (1985). Reducing behavior problems through functional communication training. *Journal of Applied Behavior Analysis*, *18*, 111-126.
- Carter, A.S., Volkmar, F.R., Sparrow, S.S., Wang, J., Lord, C., Dawson, G., Fombonne, E., Loveland, K., Messio, G., & Schopler, E. (1998). The Vineland Adaptive Behavior Scales: Supplementary norms for individuals with autism. *Journal of Autism and Developmental Disorders*, *28*, 287-299.
- Chakrabarti, S., & Fombonne, E. (2001). Pervasive developmental disorders in preschool children. *The Journal of the American Medical Association*, *285*, 3093-3099.
- Creak, E.M. et al. (1961). Schizophrenic syndrome in childhood: Progress report of working party. *British Medical Journal*, *107*, 889.
- Davidovitch, M., Glick, L., Holtzmann, G., Tirosh, E., & Safir, M.P. (2000). Developmental regression in autism: Maternal perception. *Journal of Autism and Developmental Disorders*, *30*, 113-118.
- De Bildt, A.A. (2003). *The Friesland Study; Pervasive Developmental Disorders in Mental Retardation*. Groningen, NL.: Proefschrift.
- De Brock, A.J.L.L., Vermulst, A.A., Gerris, J.R.M., & Abidin, R.R. (1992). *NOSI; Nijmeegse Ouderlijke Stress Index*. Lisse, NL: Swets & Zeitlinger.
- De Vos, M.P.A., Buysse, W.H., & Van Berckelaer-Onnes, I.A. (1996). *Het consulententeam Zuid-Holland en Zeeland geëvalueerd; de effecten van interventies bij de aanmeldingen in 1993 en 1994*. Leiden, NL: vakgroep Orthopedagogiek.
- Dekker, E. (1999). *Niet alleen achter, maar ook anders*. Leiden, NL: thesis.
- Dekker, M.C. (2003). *Psychopathology in children with intellectual disability*. Rotterdam, NL: PhD thesis.
- Dekker, M.C. , Koot, H.M., Van de Ende, J., & Verhulst, F.C. (2002). Emotional and behavioral problems in children and adolescents with and without intellectual disability. *Journal of Child and Adolescent Psychology and Psychiatry*, *43*, 1087-1098.
- Dekker, M.C., Nunn, R.J., & Koot, H.M. (2002a). Psychometric properties of the revised Developmental Behaviour Checklist scales in Dutch children with intellectual disability. *Journal of Intellectual Disability Research*, *46*, 61-75.

- Dekker, M.C., Nunn, R.J., Einfeld, S.E., Tonge, B.J., & Koot, H.M. (2002b). Assessing psychopathology in children and adolescents with intellectual disability. Revisiting the factor structure of the Developmental Behaviour Checklist. *Journal of Autism and Developmental Disorders* (accepted).
- Didden, R., Duker, P., & Korzilius, H. (1997). A meta-analytic study on treatment effectiveness for problem behaviours with individuals who have a mental handicap. *American Journal on Mental Retardation*, *101*, 387-399.
- Dijkxhoorn, Y.M., & Martens, S. (1992). *De PEP-R en de HBS*. Leiden, NL: Thesis.
- Doll, E. (1965). *Vineland social maturity scale*. Circle Pines, MN: American Guidance Service.
- Došen, A. (1990). *Psychische en gedragsstoornissen bij zwakzinnigen*. Meppel: Boom.
- Duker, P., & Didden, R. (1996). Functionele analyse van probleemgedrag: Een kritisch overzicht van observatie-instrumenten. In J.H.L. Oud & B.T.M. Van Waesberghe. *Gedragsproblemen bij jonge gehandicapte kinderen*. Arnhem, NL: Groot Klimmendaal.
- Dunlap, G., Robbins, F.R., & Darrow, M.A. (1994). Parents' reports of their childrens challenging behaviors: Results of a statewide survey. *Mental Retardation*, *32*, 206-212.
- Durand, V.M., & Carr, E.G. (1991). Functional communication training to reduce challenging behaviour: Maintenance and application in new settings. *Journal of Applied Behavior Analysis*, *24*, 251-254.
- Durand, V.M., & Crimmins, D.B. (1988). Identifying the variables maintaining self-injurious behavior. *Journal of Autism and Developmental Disorders*, *18*, 99-117.
- Dykens, E.M. (2000). Annotation: Psychopathology in children with intellectual disability. *Journal of Child Psychology, Psychiatry and Allied Disciplines*, *41*, 407-417.
- Dykens, E.M., & Hodapp, R.M. (2001). Research in mental retardation: Towards an etiologic approach. *Journal of Child Psychology, Psychiatry and Allied Disciplines*, *42*, 49-71.
- Eaton, W.W., Mortensen, P.B., Thomson, P.H., & Frydenberg, M. (2001). Obstetric complications and risk for severe psychopathology in childhood. *Journal of Autism and Developmental Disorders*, *31*, 279-285.
- Einfeld, S.W., & Tonge, B. J. (1995). The Developmental Behavior Checklist: The development and validation of an instrument to assess behavioral and emotional disturbance in children and adolescents with mental retardation. *Journal of Autism and Developmental Disorders*, *25*, 81-104.
- Einfeld, S. W., & Tonge, B.J. (1996a). Population prevalence of psychopathology in children and adolescents with intellectual disability: I Rationale and methods. *Journal of Intellectual Disability Research*, *40*, 91-98.
- Einfeld, S. W., & Tonge, B.J. (1996b). Population prevalence of psychopathology in children and adolescents with intellectual disability: II Epidemiological findings. *Journal of Intellectual Disability Research*, *40*, 99-109.
- Einfeld, S. W., & Tonge, B.J. (1996c). The use of factor analysis for ascertaining patterns of psychopathology in children with intellectual disability. *Journal of Intellectual Disability Research*, *40*, 198-207.

- Eisenmajer, R., & Prior, M. (1991). Cognitive linguistic correlates of 'theory of mind' ability in autistic children. *British Journal of Developmental Psychology*, 9, 351-364.
- Emerson, E. (2001). *Challenging behaviour; Analysis and intervention in people with severe intellectual disabilities*. Cambridge, MA: University Press.
- Emerson, E., & Bromley, J. (1995). The form and function of challenging behaviours. *Journal of Intellectual Disability Research*, 39, 388-398.
- Evans, I.M., & Meyer, L.H. (1985). *An educative approach to behavior problems: A practical decision model for interventions with severely handicapped learners*. Baltimore, MD: Paul H. Brookes Publishing Co.
- Fay, W.H., & Schuler, A.L. (1980). *Emerging language in autistic children*. In R.L. Schiefelbusch (Series Ed.). *Language intervention series* (vol. 5). Baltimore, MD: University Park Press.
- Feldman, M.A., & Griffiths, D. (1997). Comprehensive assessment of severe behavior problems. In N.N. Singh (Ed.). *Prevention & treatment of severe behavior problems; Models and methods in developmental disabilities* pp. (23-48). Pacific Grove, CA: Brooks/Cole Publishing Company.
- Field, A. (2000). *Discovering statistics using SPSS for windows. Advanced techniques for the beginner*. London, UK: SAGE publications.
- Folstein, S., & Rutter, M. (1977). Infantile autism: A genetic study of 21 twin pairs. *Journal of Child Psychology and Psychiatry*, 18, 297-321.
- Fombonne, E. (1998). Epidemiological surveys of autism. In F.R. Volkmar (Ed.) *Autism and pervasive developmental disorders* (pp. 32-63). Cambridge, MA: University Press.
- Fombonne, E., & Achard, S. (1993). The Vineland Adaptive Behavior Scale in a sample of normal French children: A research note. *Journal of Child Psychology and Psychiatry*, 34, 1051-1058.
- Freeman, B.J., Guthrie, D., Ritvo, E., Schroth, P., & Ball, J. (1978). The behavior observation scale for autism. Initial methodology, data analysis and preliminary findings on 89 children. *Journal of the American Academy of Child Psychiatry*, 17, 576-588.
- Frith, U. (2003). *Autism: Explaining the enigma*. Oxford: Basil Blackwell.
- Frith, U., & Happé, F. (1998). Why specific developmental disorders are not specific: Online and developmental effects in autism and dyslexia. *Developmental Science*, 1, 267-272.
- Gardner, W.I., & Whalen, J.P. (1996). Discussion: A multimodal behavior analytic model for evaluating the effects of medical problems on non-specific behavior symptoms in people with developmental disabilities. *Behavioral Interventions*, 11, 147-161.
- Gillberg, C. (1991). The treatment of epilepsy in autism. *Journal of Autism and Developmental Disorders*, 21, 61-77.
- Gillberg, C. (1992). Autism and autistic-like conditions: Subscales among disorders of empathy. *Journal of Psychology and Psychiatry*, 33, 813-842.
- Gillberg, C. (1999). Neurodevelopmental processes and psychological functioning in autism. *Development and Psychopathology*, 11, 567-587.

- Gillberg, C. (2001). Asperger syndrome and high functioning autism; Shared differences or different disorders? *The Journal of Developmental and Learning Disorders*, 5, 81-95.
- Gillberg, C., & Coleman, M. (1996). The biology of the autistic syndromes. A review of the literature. *Developmental Medicine and Child Neurology*, 38, 191-202.
- Gillham, J.E., Carter, A.S., Volkmar, F.R., & Sparrow, S.S. (2000). Towards a developmental operational definition of autism. *Journal of Autism and Developmental Disorders*, 30, 269-278.
- Happé, F., Briskman, J., & Frith, U. (2001). Exploring the cognitive phenotype of autism: Weak 'central coherence' in parents and siblings of children with autism: I. Experimental tests. *Journal of Child Psychology and Psychiatry*, 42, 299-307.
- Happé, F. (1999). Autism: Cognitive deficit or cognitive style. *Internetcongress Autism99*.
- Harris, J.C. (1992). Neurobiological factors in self-injurious behavior. In J.K. Luiselli, J.L. Matson & N.N. Singh (Eds.). *Self-injurious behavior: Analysis, assessment and treatment* (pp. 59-92). New York, NY: Springer-Verlag.
- Hastings, R.P., Brown, T., Mount, R.H., & Magnus Cormack, K.F. (2001). Exploration of psychometric properties of the Developmental Behavior Checklist. *Journal of Autism and Developmental Disorders*, 31, 423-431.
- Hermelin, B., & O'Connor, N. (1970). *Psychological experiments with autistic children*. London, UK: Pergamon Press.
- Hobson, R.P. (1986). The autistic child's appraisal of expressions of emotion. *Journal of child Psychology, Psychiatry and Allied Disciplines*, 27, 321-342.
- Hobson, R.P. (1987). The autistic child's recognition of age- and sex-related characteristics of people. *Journal of Autism and Developmental Disorders*, 17, 63-79.
- Hobson, R.P. (1989). Beyond Cognition: A theory of autism. In G. Dawson (Ed.), *Autism: Nature, diagnosis, and treatment* (pp. 22-48). New York, NY: Guilford Press.
- Hobson, R.P. (1990). On acquiring knowledge about people and the capacity to pretend: Response to Leslie (1987). *Psychological Review*, 97, 114-121.
- Hobson, R.P., Ouston, J., & Lee, A. (1988). What's in a face? The case of autism. *British Journal of Developmental Psychology*, 79, 441-453.
- Hodapp, R.M. (1997). Children with disabilities. In S.S. Luthar, J.A. Burack, D. Cicchetti & J. Weisz (Eds.). *Developmental psychopathology; perspectives on adjustment, risk, and disorder* (pp. 198-207). Cambridge, MA: University Press.
- Hodapp, R.M., & Dykens, E.M. (1996). Mental Retardation. In E.J. Mash & R.A. Barkley (Eds.), *Child Psychopathology* (pp. 362-389). New York, NY: Guilford.
- Hodapp, R.M., & Zigler, E. (1995). Past, present, and future issues in the developmental approach to Mental Retardation and developmental disabilities. In D. Cicchetti & C.J. Cohen (Eds.), *Developmental psychopathology: vol. 2. Risk, disorder and adaptation* (pp. 299-331). New York, NY: Wiley.

- Hoshino, Y., Kaneko, M., Yashino, Y., Kumashiro, H., Volkmar, F.R., & Cohen, D.J. (1987). Clinical features of autistic children with setback course in their infancy. *Japanese Journal of Psychiatry*, *41*, 237-246.
- Howlin, P. (1997). *Autism; Preparing for adulthood*. London, UK: Routledge.
- Howlin, P. (1998). Practitioner review: Psychological and educational treatments for autism. *Journal of Child Psychology and Psychiatry and Allied Disciplines*, *39*, 307-322.
- Howlin, P., & Goode, S. (1998). Outcome in adult life for people with autism and asperger's syndrome. In F.R. Volkmar (Ed.). *Autism and pervasive developmental disorders* (pp.209-236). Cambridge, MA: University Press.
- Howlin, P., Mawhood, L., & Rutter, M. (2000). Autism and developmental receptive language disorder – a follow-up comparison in early adult life. II: Social, behavioral, and psychiatric outcomes. *Journal of Child Psychology and Psychiatry*, *41*, 561-578.
- Janssen, C.G.C., Schuengel, C., & Stolk, J. (2002). Gedragsproblemen bij mensen met een ernstige verstandelijke beperking, gehechtheidsproblemen en psychologische stress. Een verklaringsmodel met implicaties voor onderzoek en praktijk. *Nederlands Tijdschrift voor de Zorg aan verstandelijk gehandicapten*, *28*, 3-20.
- Kanner, L. (1943). Autistic disturbances of affective contact. *Nervous Child*, *2*, 217-250.
- Kanner, L. (1954). Childhood schizophrenia, round table. *American Journal of Orthopsychiatry*, *24*, 526-528.
- Kars, H. (1995). *Ernstig probleemgedrag bij zwakzinnige mensen: Een systematische benadering van ernstig probleemgedrag in de zwakzinnigenzorg*. Houten, NL: Bohn Stafleu van Loghum.
- Kars, H., & Swets, J.H.J. (1998). *Problemen met het gedrag van mensen met een verstandelijke handicap*. Houten, NL: Bohn Stafleu Van Loghum.
- Kiernan, C., & Kiernan, D. (1994). Challenging behaviour in schools for pupils with severe learning difficulties. *Mental Handicap Research*, *7*, 117-201.
- Klin, A. (1991). Young autistic children's listening preferences in regard to speech: a possible characterization of the symptom of social withdrawal. *Journal of Autism and Developmental Disorders*, *23*, 15-35.
- Klin, A., Volkmar, F.R., & Sparrow, S.S. (1992). Autistic social dysfunction: Some limitations of the theory of mind hypothesis. *Journal of Child Psychology and Psychiatry*, *36*, 1127-1140.
- Klin, A., Jones, W., Schultz, R., & Volkmar, F. (2003). The enactive mind, or from action to cognition: Lessons from autism. *Philosophical Transactions Royal Society London Biology*, *358*, 345-360.
- Konstantareas, M.M., & Homatitidis, S. (1987). Ear infections in autistic and normal children. *Journal of autism and Developmental Disorders*, *20*, 591-593.
- Koot, J.M., & Dekker, M.C. (2001). *Voorlopige normgegevens bij de Developmental Behavior Checklist (Vragenlijst over ontwikkeling en gedrag van kinderen)*. Rotterdam, NL: Erasmus Universiteit.
- Kraijer, D.W. (1994). *Autisme- en verwante stoornissenschaal-Revisie*. Lisse, NL: Swets & Zeitlinger.

- Kraijer, D.W. (1998). *Autistische stoornissen en verstandelijke beperking; Ontwikkelingsstoornis en Ontwikkelingstekort*. Lisse, NL: Swets & Zeitlinger.
- Kraijer, D.W. (2000). Review of adaptive behavior studies in mentally retarded people with autism/pervasive developmental disorders. *Journal of Autism and Developmental Disorders*, 30, 39-47.
- Kraijer, D.W., & Kema, G.N. (1994). *SGZ, Storend Gedragsschaal voor zwakzinningen. Handleiding*. Lisse, NL: Swets & Zeitlinger.
- Kraijer, D.W., & Plas, J.J. (1997). *Psychodiagnostiek in de zorg voor verstandelijk gehandicapte mensen; Classificatie, test- en schaalgebruik*. Lisse, NL: Swets & Zeitlinger.
- Kraijer, D.W., & Plas, J.J. (2002). *Handboek psychodiagnostiek en verstandelijke beperking*. Lisse, NL: Swets & Zeitlinger.
- Kramer, G.J.A. (1992). *Consensusprotocol Ernstig Probleemgedrag. Handleiding voor het beschrijven en beoordelen van probleemsituaties rond bewoners van intramurale instellingen voor verstandelijk gehandicapten*. Utrecht, NL: Nzi.
- Krevelen, D.A. van (1963). Zur Ätiologie des Kannerschen Autismus under der Aspergeschen autistischen Psychopathie. In *Bericht der 5. Psychiatertagen des Landschaftsverbanden Rheinland Düsseldorf* (pp. 28-39).
- Lambermon, M.W.E. (1991). *Video of folder? Korte- en langetermijneffecten van voorlichting over vroegkinderlijke opvoeding*. Leiden, NL: Universiteit Leiden.
- Lambert, N., & Windmiller, M. (1981). *AAMD Adaptive Behavior Scale school edition*, Monterey, CA: McGraw-Hill.
- Leekam, S.R., Libby, S.J., Wing, L., Gould, J., & Taylor, C. The Diagnostic Interview for Social and Communication Disorders: Algorithms for ICD-10 childhood autism and Wing and Gould autistic spectrum disorder. *Child Psychology and Psychiatry*, 43, 327-342.
- Liepmann, M.C., & Ort, M. (1980). Assessment of handicaps in mentally retarded children: Preliminary findings from a study using the schedule of children's Handicaps, Behaviour and Skills. In E. Strömgen, A. Dupont & J.A. Nielsen (Eds.), *Epidemiological research as a basis for the organization of extramural psychiatry* (pp. 187-195). Copenhagen, DK: Munsgaard.
- Lord, C., & Magill, J. (1989). Methodological and theoretical issues in studying peer-directed behaviour and autism. In G. Dawson, (Ed.), *Autism: Nature, diagnosis, and treatment*. New York, NY: Academic Press.
- Lovaas, O.I., & Buch, G. (1997). Intensive behavioral intervention with young children with autism. In N.N. Singh (Ed.), *Prevention & treatment of severe behavior problems; Models and methods in developmental disabilities* (pp. 61-86). Pacific Grove, CA: Brooks/Cole Publishing Company.
- Lowe, M., & Costello, A.J. (1976). *Manual for the Symbolic Play Test*. England: NFER-NELSON.
- Lucangeli, D. (1997). *Connecting a fragmented world; Cognitive and meta-cognitive capabilities of high-functioning Autistic Children*. Leiden, NL: PhD thesis.
- Luckasson, R., Coulter, D.L., Polloway, E.A., Reiss, S., Shalock, R.L., Snell, M.E., Spitalnik, D.M., & Stark, J.A. (1992). *Mental Retardation: Definition, classification and systems of supports*. Washington, DC: AAMR.
- Lund, J. (1989). Measuring behaviour disorder in mental handicap. *British Journal of Psychiatry*, 155, 379-383.

- Mahler, M. (1965). On early infantile psychosis. *Journal of the American Academy of child Psychiatry*, 554-568.
- Matson, J.L., Bamburg, J.W., Mayville, E.A., & Khan, I. (1999). Seizure disorders in people with intellectual disability: An analysis of differences in social functioning, adaptive functioning and maladaptive behaviours. *Journal of Intellectual Disability Research*, 43, 531-539.
- McCune Nicolich (1981). Toward symbolic functioning: Structure of early pretend games and potential parallels with language. *Child Development*, 52, 785-797.
- Meerling (1989). *Methoden en technieken van psychologisch onderzoek. Deel I: Model, observatie en beslissing*. Amsterdam, NL: Boom Meppel.
- Minshew, N.J. (1991). Indices of neural functioning in autism: Clinical and biological implications. *Pediatrics*, 31, 774-780.
- Mundy, P., Sigman, M., Ungerer, J., & Sherman, T. (1987). Non-verbal communication and play correlates of language development in autistic children. *Journal of Autism and Developmental Disorders*, 17, 349-364.
- Murphy, G., Hall, S., Oliver, C., & Kissi-Debra, R. (1999). Identification of early self-injurious behaviour in young children with intellectual disability. *Journal of Intellectual Disability Research*, 43, 149-163.
- National Research Council (2001). *Educating Children with Autism*. Committee on educational interventions for children with autism. Division of behavioral and social sciences and education. Washington, DC: National Academy Press.
- Neale, J.M., & Liebert, R.M. (1986). *Science and Behaviour: An introduction to methods of research*, 3rd ed., New Jersey, NY: Prentice Hall.
- Noens, I.L.J., & Van Berckelaer-Onnes, I.A. (2002). Communicatie bij mensen met autisme en verstandelijke beperking: Van inzicht naar interventie. *Nederlands Tijdschrift voor Zorg aan verstandelijk gehandicapten*, 4, 212-226.
- Noens, I.L.J., Van Berckelaer-Onnes, I.A., & Verpoorten, R. (2000). Indicatiestelling van ondersteunende communicatie bij mensen met een autistische spectrumstoornis. *Logopedie en Foniatrie*, 72, 144-247.
- O'Brien, G. (2000). Learning Disability. In C. Gillberg & G. O'Brien (Eds.), *Developmental disability and behaviour* (pp. 12-26). London, UK: Mac Keith Press.
- Ort, M., & Liepmann, M.C. (1981). The schedule of children's handicaps behaviours and skills: Reliability and discrimination. In P. Mittler (Ed.). *Frontiers of knowledge in mental retardation. Proceedings of the 5th congress of the international association for the scientific study of mental deficiency*. Baltimore, MD: University Park Press.
- Ozonoff, S. (1995). Executive functions in autism. In E. Schopler & G. Mesibov (Eds.), *Learning and cognition in autism* (pp. 199-220). New York, NY: Plenum Press.
- Prior, M., & Ozonoff, S. (1998). Psychological factors in autism. In Volkmar, F.R. (Ed.), *Autism and pervasive developmental disorders* (pp. 64-105). Cambridge, MA: University Press.
- Quine, L., & Paul, J. (1985). Examining the causes of stress in families with mentally handicapped children. *British Journal of Social Work*, 15, 501-517.

- Quereshi, H. (1994). The size of the problem. In E. Emerson, P. McGill & J. Mansell (Eds.). *Severe learning disabilities and challenging behaviours: Designing high quality services and therapy*. London, UK: Chapman & Hall.
- Rapin, E. (ed.) (1996), *Preschool children with inadequate communication.*: Cambridge, UK: MacKeith Press.
- Rogers, S.J., & Bennetto, L. (2000). Intersubjectivity in autism: The Roles of imitation and executive functions. In A.M. Wetherby & B.M. Prizant (Eds.). *Autism spectrum disorders; A transactional developmental perspective* (pp. 79-108). Baltimore, MD: Paul Brooks Publishers.
- Romanczyk, R.G., Weiner, T., Lockshin, S., & Ekdahl, M. (1999). Research in autism: Myths, controversies and perspectives. In D. Berkell Zager (Ed.). *Autism; Identification, education and treatment* (pp. 23-62). London, UK: Lawrence Erlbaum Associates.
- Rutgers, A.H., Bakermans-Kranenburg, M.J., Van IJzendoorn, M.H., & Van Berckelaer-Onnes, I.A. (submitted). Autism and Attachment: A Review and Meta-Analysis.
- Rutter, M. (1978). Diagnosis and definition. In M. Rutter & E. Schopler (Eds.). *Autism: A reappraisal of concepts and treatment* (pp. 1-25). New York, NY: Plenum Press.
- Rutter, M. (1979). Language, cognition and autism. In R. Katzman (Ed.), *Congenital and acquired cognitive disorders* (pp. 247-264). New York, NY: Raven Press.
- Rutter, M. (1985). Treatment of autistic children. *Journal of Child Psychology and Psychiatry*, 26, 193-214.
- Rutter, M. (2000). Genetic studies of autism: From the 1970's into the millenium. *Journal of Abnormal Psychology*, 28, 3-14.
- Rutter, M., Bailey, A., Bolton, P., & Le Couteur, A. (1993). Autism: Syndrome definition and possible genetic mechanisms. In R. Plomin & G.E. McClearn (Eds.), *Nature, nurture and psychology* (pp. 269-284). Washington, DC: APA Books.
- Rutter, M. Graham, P., & Yule, W. (1970). *A neuropsychiatric study in childhood*. London, UK: SIMP/Heineman.
- Schopler, E. (1989). Principles for directing both educational treatment and research. In Gillberg, C. (Ed.). *Diagnosis and treatment of autism* (pp. 167-183). Göteborg, S: University of Göteborg.
- Schopler, E. (1995) (Ed). *Parent survival manual; A guide to crisis resolution in autism and related developmental disorders*. New York, NY: Plenum Press.
- Schopler, E., & Reichler, R.J. (1979). *Individualized assessment and treatment for autistic and developmentally disabled children, vol. 1*. Psycho Educational Profile. Austin, TX: PRO-ED.
- Schopler, E., Reichler, R.J., Bashford, A., Lansing, M.D., & Marcus, C.M. (1990). *Individualized assessment and treatment for autistic and developmentally disabled children, vol. 1*. Psycho Educational Profile revised. Austin, TX: PRO-ED.
- Schuler, A.L., Prizant, B.M., & Wetherby, A. (1997). Enhancing language and communication development: Prelinguistic approaches. In D. Cohen & F. Volkmar (Eds.), *Handbook of autism and pervasive developmental disorders* (2nd ed., pp. 539-571). New York, NY: John Wiley & Sons.

- Schuring, G., Barnhard, M.C., Kardaun, H.H., Kramer, G.J.A., & Schoep, H.F. (1990). *Ernstig probleemgedrag. Een inventarisatie van probleemsituaties rond bewoners in de intramurale zwakzinnigenzorg*. Utrecht, NL: NZI.
- Shah, A., & Frith, U. (1983). An islet of ability in autistic children: A research note. *Journal of Child Psychology and Psychiatry*, 24, 613-620.
- Shattock, P., & Whiteley, P. (2002). Biochemical aspects in autism spectrum disorders: Updating the opioid-excess theory and presenting new opportunities for biomedical intervention. *Expert Opinion of Therapeutic Targets*, 6, 175.
- Sigman, M., & Ungerer, J.A. (1984). Attachment behaviors in autistic children. *Journal of Autism and Developmental Disorders*, 14, 231-244.
- Simonoff, E. (2000). Extracting meaning from co morbidity: Genetic analyses that make sense. *Journal of Child Psychology and Psychiatry and Allied Disciplines*, 40, 19-56.
- Sparrow, S.S., Balla, D.A., & Cicchetti, D.V. (1984). *The Vineland Adaptive Behavior Scales; a revision of the VSMS by E.A. Doll: Expanded Form*. Minnesota, MI: AGS.
- Stenfert Kroese, B., & Fleming, K. (1993). Prevalence and persistency of challenging behaviour of children. In Fleming & Stenfert Kroeze (Eds.) *People with learning disability and severe challenging behaviour; new developments in services and therapy*. Great Britain: B. Biddles Ltd. Guildford and King's Lynn.
- Strømme, P., & Diseth, T.H. (2001). Prevalence of psychiatric diagnoses in children with Mental Retardation: Data from a population-based study. *Developmental Medicine and Child Neurology*, 42, 266-270.
- Tager-Flusberg, H. (1989). A psycholinguistic perspective on language development in the autistic child. In G. Dawson (Ed.), *Autism: Nature, diagnosis and treatment* (pp. 92-115). New York, NY: Guilford Press.
- Taylor, J.C., & Carr, E.G. (1992a). Severe problem behaviors related to social interaction 1: Attention seeking and social avoidance. *Behavior Modification*, 16, 305-355.
- Taylor, J.C., & Carr, E.G. (1992b). Severe problem behaviors related to social interaction 2: A system Analysis. *Behavior Modification*, 16, 356-371.
- Taylor, J.C., Ekdahl, M.M., Romanczyk, R.G., & Miller, M.L. (1994). Escape behavior in task situations: Task versus social antecedents. *Journal of Autism and Developmental Disorders*, 24, 331-344.
- Taylor, J.C., Romanczyk, R. G. (1994). Generating hypotheses about the function of student behavior problems by observing teacher behavior. *Journal of Applied Behavior Analysis*, 27, 252-265.
- Tsai, L.Y. (1999). Medical treatment in autism. In D. Berkel Zager (Ed.), *Autism; Identification, education and treatment* (pp. 199-258). London, UK: Lawrence Erlbaum Associates.
- Van Berckelaer-Onnes, I.A. (1979). *Early childhood autism: A child-rearing problem*. Lisse, NL: Swets & Zeitlinger.
- Van Berckelaer-Onnes, I.A. (1994). Playtraining for autistic children. In Hellendoorn, v.d. Kooy & Sutton-Smith (Eds.), *Play and intervention* (pp. 173-18). Albany, NY: State University of New York Press.

- Van Berckelaer-Onnes, I.A. (1997). Communicatie en autisme. *Logopedie*, 10, 29-39.
- Van Berckelaer-Onnes, I.A., & Van Duijn, G. (1993). A comparison between the Handicaps Behaviour and Skills schedule and the PsychoEducational Profile. *Journal of Autism and Developmental Disorders*, 23, 263-272.
- Van Berckelaer-Onnes, I.A., Dijkxhoorn, Y.M., Jacobs, M.E., & Van der Ploeg, D.A. (1996). *Punter 6, Merwebolder. Een woongroep voor kinderen met een autistische stoornis en een verstandelijke handicap*. Universiteit Leiden, NL: Orthopedagogiek.
- Van Berckelaer-Onnes, I.A., & Van Engeland H. (1997). *Kinderen en autisme. Onderkenning, behandeling en begeleiding*. Meppel, NL: Boom.
- Van Berckelaer-Onnes, I.A., & Kwakkel-Scheffer, J.J.C. (1996). Spelinterventies voor kinderen met een autistische stoornis. *Kinder- en Jeugd Psychotherapie*, 2/3, 77-92.
- Van Berkum, H.W. (1993). Determinanten van stress bij ouders van een verstandelijk gehandicapt kind: Kenmerken van het kind. *Nederlands Tijdschrift voor Zorg aan verstandelijk gehandicapten*, 1, 3-20.
- Van Deyl, B. (1997). *De Nederlandse normen van de PEP-R*. Leiden, NL: thesis Universiteit Leiden.
- Van Gemert, G.H. (1985). *'Gedragsgestoordheid bij zwakzinnigen'*. Lisse, NL: Swets & Zeitlinger.
- Van IJzendoorn, M.H., Goldberg, S., Kroonenberg, P.M., & Frenkl, O.J. (1992). The relative effects of maternal and child problems on the quality of attachment: A meta-analysis of attachment in clinical samples. *Child Development*, 63, 840-858.
- Van IJzendoorn, M.H., Schuengel, C., & Bakermans-Kranenburg, M.J. (1999). Disorganized attachment in early childhood: Meta-analysis of precursors, concomitants, and sequelae. *Development and Psychopathology*, 11, 225-249.
- Van Strien, P.J. (1986). *Praktijk als wetenschap; Methodologie van het sociaal-wetenschappelijk handelen*. Assen/Maastricht: Van Gorcum.
- Verpoorten, R.A.W. (1996). Communicatie met verstandelijk gehandicapte autisten: Een multi-dimensioneel communicatiemodel. *Nederlands Tijdschrift voor Zorg aan verstandelijk gehandicapten*, 22, 106-120.
- Verpoorten, R.A.W., Noens, I.L.J., & Berckelaer-Onnes, I. A. van (1999). *De ComVoor – Voorlopers in communicatie, experimentele versie*. Leiden, NL: Universiteit Leiden, NL.
- Vig, S., & Jedrysek, E. (1999). Autistic features in young children with significant cognitive impairment: Autism or mental retardation? *Journal of Autism and Developmental Disorders*, 29, 235-248.
- Volkmar, F.R., Hoder, E.L., & Cohen, D.J. (1985). Compliance, negativism, and the effects of treatment structure in autism: A naturalistic, behavioral study. *Journal of Child Psychiatry*, 26, 865-877.
- Volkmar, F.R., & Lord, C. (1998). Diagnosis and definition of autism and other pervasive developmental disorders. In F.R. Volkmar (Ed), *Autism and pervasive developmental disorders* (pp. 1-31). Cambridge, MA: University Press.

- Volkmar, F.R., Sparrow, S.S., Goudreau, D., Cicchetti, D.V., Paul, R., & Cohen, D.J. et al. (1987). Social deficits in autism: An operational using the Vineland Adaptive Behavior Scales. *Journal of the American Academy of Child and Adolescent Psychiatry*, 26, 156-161.
- Volkmar, F.R., Szatmari, P., & Sparrow, S.S. (1993). *Sex differences in pervasive developmental disorders*, 23, 579-591.
- Volkmar, F.R., Woolston, J.L. (1995). Co-morbidity of psychiatric disorders in children and adolescents (unpublished).
- Wels, P.M.A., & Robbroeckx, L.M.H. (1996). *Nijmeegse Vragenlijst voor de Opvoedingssituatie*. Lisse, NL: Swets Test Services.
- Wenar, C., & Kerig, P. (1999). *Developmental psychopathology: from infancy through adolescence* (4th ed.) Boston, MA: McGraw Hill.
- Wetherby, A.M., & Prizant, R.M. (1998). Enhancing language and communication development in autism: Assessment and intervention guidelines. In D.E. Berkell-Zager (Ed.), *Autism; Identification, education and treatment* (pp. 141-174). Hillsday, NJ: Lawrence Erlbaum Associates, publishers.
- Wetherby, A.M., Prizant, R.M., & Hutchinson, T. (1998). Communicative, social affective, and symbolic profiles of young children with autism and pervasive developmental disorders. *American Journal of Speech-Language Pathology*, 7, 79-91.
- Wetherby, A.M., Prizant, B.M., & Schuler, L. (2000). Understanding the nature of communication and language impairments. In A.M. Wetherby & B.M. Prizant (Eds.). *Autism spectrum disorders; A transactional developmental perspective* (pp. 109-142). Baltimore, CA: Paul Brooks Publishers.
- Williams, E., Reddy, V., & Costali, A. (2001). Taking a closer look at functional play in children with autism. *Journal of Autism and Developmental Disorders*, 31, 67-77.
- Wing, L. (1980). The MRC Handicaps, Behaviour & Skills (HBS) Schedule. In A. Strömgen, A. Dupont & J.A. Nielsen (Eds.) *Epidemiological Research as a Basis for the Organization of Extramural Psychiatry* (pp. 187-195). Copenhagen, DK: Munksgaard.
- Wing, L. (1981). Language, social and cognitive impairments in autism and severe mental retardation. *Journal of Autism and Developmental Disorders*, 11, 31-44.
- Wing, L. (1993a). *Diagnostic Interview for Social and Communication Disorders* (3rd ed.). Bromley, UK: Center for Social and Communication Disorders, Elliott House.
- Wing, L. (1993b). The definition and prevalence of autism: A review of the literature. *Adolescent Psychiatry*, 2, 61-74.
- Wing, L. (1996). *The autistic spectrum*. A guide for parents and professionals. London, UK: Constable.
- Wing, L. (1999). *Diagnostic Interview for Social and Communication Disorders*, (10th rev.) Bromley, UK: NAS.
- Wing, L. (2001). *The Autistic Spectrum. A parent's Guide to Understanding and Helping Your Child*. Berkeley, CA: Ulysses Press.
- Wing, L., & Gould, J. (1978). Systematic recording of behaviours and skills of retarded and psychotic children. *Journal of Autism and Childhood Schizophrenia*, 8, 79-97.

- Wing, L., & Gould, J. (1979). Severe impairments of social interaction and associated abnormalities in children: Epidemiology and classification. *Journal of Autism and Developmental Disorders*, 9, 11-30.
- Wing, L., & Gould, J. (1982). *Schaal voor Handicaps en Vaardigheden*. London, UK: Medical Research Council. Nederlandse vertaling (1983). Leiden, NL: Universiteit Leiden.
- Wing, L., Leekam, S.R., Libby, S.J., Gould, J., & Larcombe, M. (2002). Diagnostic Interview for Social and Communication Disorders: Background, inter-rater reliability and clinical use. *Journal of Child Psychology and Psychiatry*, 43, 307-325.
- Wolery, M. (2000). Commentary: The environment as a source of variability: Implications for research with individuals who have autism. *Journal of Autism and Developmental Disorders*, 30, 379-381.
- World Health Organization (1992). *The ICD-10 classification of mental and behavioural disorders. Clinical descriptions and diagnostic guidelines*. Geneva, CH: WHO.
- Zigler, E. (1984). A developmental theory on mental retardation. In B. Blatt & R. Morris (Eds.), *Perspectives in special education: Personal Orientations* (pp. 173-209). Santa Monica, CA: Scott, Foresman.

Appendix A

DSM-IV-TR criteria Pervasive Developmental Disorders (APA, 1994).

Diagnostic criteria for 299.00 Autistic Disorder

A. A total of six (or more) items from (1), (2) and (3), with at least two from (1), and one each from (2) and (3):

- (1) qualitative impairment in social interaction, as manifested by at least two of the following:
 - (a) marked impairment in the use of multiple nonverbal behaviors such as eye-to-eye gaze, facial expression, body postures, and gestures to regulate social interaction
 - (b) failure to develop peer relationships appropriate to developmental level
 - (c) a lack of spontaneous seeking to share enjoyment, interests, or achievements with other people (e.g., by a lack of showing, bringing, or pointing out objects of interest)
 - (d) lack of social or emotional reciprocity
- (2) qualitative impairments in communication as manifested by at least one of the following:
 - (a) delay in, or total lack of, the development of spoken language (not accompanied by an attempt to compensate through alternative modes of communication such as gestures or mime)
 - (b) in individuals with adequate speech, marked impairment in the ability to initiate or sustain a conversation with others
 - (c) stereotyped and repetitive use of language or idiosyncratic language
 - (d) lack of varied, spontaneous make-believe play or social imitative play appropriate to developmental level
- (3) restricted repetitive and stereotyped patterns of behavior, interests, and activities, as manifested by at least one of the following:
 - (a) encompassing preoccupation with one or more stereotyped and restricted patterns of interest that is abnormal either in intensity or focus
 - (b) apparently inflexible adherence to specific, nonfunctional routines or rituals
 - (c) stereotyped and repetitive motor mannerisms (e.g. hand or finger flapping or twisting, or complex whole-body movements)
 - (d) persistent preoccupation with parts of objects

B. Delays or abnormal functioning in at least one of the following areas, with onset prior to age 3 years: (1) social interaction, (2) language as used in social communication, or (3) symbolic or imaginative play.

- C. The disturbance is not better accounted for by Rett's Disorder or Childhood Disintegrative Disorder.

Diagnostic criteria for 299.80 Rett's Disorder

- A. All of the following:
- (1) apparently normal prenatal and perinatal development
 - (2) apparently normal psychomotor development through the first 5 months after birth
 - (3) normal head circumference at birth
- B. Onset of all of the following after a period of normal development:
- (1) deceleration of head growth between ages 5 and 48 months
 - (2) loss of previously acquired purposeful hand skills between ages 5 and 30 months with the subsequent development of stereotyped hand movements (e.g., hand wringing or hand washing)
 - (3) loss of social engagement early in the course (although often social interaction develops later)
 - (4) appearance of poorly co-ordinated gait or trunk movements
 - (5) severely impaired expressive and receptive language development with severe psychomotor retardation

Diagnostic criteria for 299.10 Childhood Disintegrative Disorder

- A. Apparently normal development for at least the first 2 years after birth as manifested by the presence of age-appropriate verbal and nonverbal communication, social relationships, play and adaptive behaviour.
- B. Clinically significant loss of previously acquired skills (before age 10 years) in at least two of the following areas:
- (1) expressive or receptive language
 - (2) social skills or adaptive behavior
 - (3) bowel or bladder control
 - (4) play
 - (5) motor skills
- C. Abnormalities of functioning in at least two of the following areas:
- (1) qualitative impairment in social interaction (e.g. impairment in non-verbal behaviors, failure to develop peer relationships, lack of social or emotional reciprocity)
 - (2) qualitative impairments in communication (e.g. delay or lack of spoken language, inability to initiate or sustain a conversation, stereotyped and repetitive use of language, lack of varied make-believe play)
 - (3) restricted, repetitive, and stereotyped patterns of behavior, interests, and activities, including motor stereotypies and mannerisms
- D. The disturbance is not better accounted for by another specific Pervasive Developmental Disorder or by Schizophrenia.

Diagnostic criteria for 299.80 Asperger's Disorder

- (A) Qualitative impairment in social interaction, as manifested by at least two of the following:
 - (1) marked impairment in the use of multiple nonverbal behaviors such as eye-to-eye gaze, facial expression, body postures, and gestures to regulate social interaction
 - (2) failure to develop peer relationships appropriate to developmental level
 - (3) a lack of spontaneous seeking to share enjoyment, interests, or achievements with other people (e.g., by a lack of showing, bringing, or pointing out objects of interest to other people)
 - (4) lack of social or emotional reciprocity

- (B) restricted repetitive and stereotyped patterns of behavior, interests, and activities, as manifested by at least one of the following:
 - (1) encompassing preoccupation with one or more stereotyped and restricted patterns of interest that is abnormal either in intensity or focus
 - (2) apparently inflexible adherence to specific, non-functional routines or rituals
 - (3) stereotyped and repetitive motor mannerisms (e.g. hand or finger flapping or twisting, or complex whole-body movements)
 - (d) persistent preoccupation with parts of objects

- C. The disturbance causes clinically significant impairment in social, occupational, or other important areas of functioning.

- D. There is no clinically significant general delay in language (e.g., single words used by age 2 years, communicative phrases by age 3 years).

- E. There is no clinically significant delay in cognitive development or in the development of age-appropriate self-help skills, adaptive behavior (other than in social interaction), and curiosity about the environment in childhood.

- F. Criteria are not met for another specific Pervasive Developmental Disorder or Schizophrenia.

Diagnostic criteria for 299.80 Pervasive Developmental Disorder Not Otherwise Specified (Including Atypical Autism)

This category should be used when there is a severe and pervasive impairment in the development of reciprocal social interaction associated with impairment in either verbal and nonverbal communication skills, or with the presence of stereotyped behavior, interests, and activities, but the criteria are not met for a specific Pervasive Developmental Disorder, Schizophrenia, Schizotypal Personality Disorder, or Avoidant Personality Disorder.

Appendix B

Table 2: t-tests for the behavioural clusters of the group with mild Mental Retardation and an ASD (MMR/ASD) and the group with no MR and an ASD (non-MR/ASD)

| Item cluster | N MMR/ASD | N Non/MR ASD | Levene's Test for Equality of Variances | | | t-test for Equality of Means | |
|--|-----------|--------------|---|--------|-------|------------------------------|-----------------|
| | | | F | Sig. | t | Df | Sig. (2-tailed) |
| Abnormalities of Speech | 48 | 47 | 7.262 | .008** | 1.965 | 80.686 | .053 |
| Abnormal responses to sounds | 71 | 59 | .987 | .325 | -.052 | 128 | .985 |
| Abnormal responses to visual stimuli | 73 | 56 | 2.591 | .110 | -.217 | 127 | .828 |
| Abnormal Proximal Sensory Stimulation | 67 | 58 | 3.687 | .057 | .463 | 123 | .644 |
| Abnormal Bodily Movements | 68 | 55 | 2.443 | .121 | 2.508 | 121 | .013** |
| Routines and Resistance to Change | 68 | 55 | .175 | .677 | .708 | 121 | .481 |
| Behaviour problems with limited or no Social Awareness | 61 | 54 | .113 | .737 | 2.503 | 113 | .014** |
| Behaviour problems with Social Awareness | 62 | 55 | .978 | .325 | .957 | 115 | .341 |
| Sleep Problems | 69 | 57 | .411 | .523 | .300 | 124 | .764 |
| Specific Behaviour problems | 59 | 51 | .941 | .334 | 1.643 | 108 | .103 |
| Cluster Lund | 57 | 53 | .574 | .450 | 1.584 | 108 | .116 |

Table 2: t-tests for the behavioural clusters of the group with Mental Retardation and an ASD (MR/ASD) and the group with mild MR and an ASD (MMR/ASD)

| Item cluster | N MR/ASD | N MMR ASD | Levene's Test for Equality of Variances | | | t-test for Equality of Means | |
|--|----------|-----------|---|--------|-------|------------------------------|-----------------|
| | | | F | Sig. | t | Df | Sig. (2-tailed) |
| Abnormalities of Speech | 89 | 48 | 2.280 | .133 | 2.845 | 135 | .005** |
| Abnormal responses to sounds | 230 | 71 | .115 | .735 | .787 | 299 | .432 |
| Abnormal responses to visual stimuli | 237 | 73 | 30.234 | .000** | 6.973 | 201.064 | .000** |
| Abnormal Proximal Sensory Stimulation | 234 | 67 | 15.574 | .000** | 5.457 | 153.354 | .000** |
| Abnormal Bodily Movements | 225 | 68 | 11.767 | .001** | 6.192 | 173.926 | .000** |
| Routines and Resistance to Change | 232 | 68 | 1.142 | .286 | 2.509 | 298 | .013** |
| Behaviour problems with limited or no Social Awareness | 211 | 61 | 6.629 | .011** | 4.804 | 132.058 | .000** |
| Behaviour problems with Social Awareness | 183 | 62 | .130 | .719 | .671 | 243 | .503 |

| | | | Levene's Test for Equality of Variances | | | t-test for Equality of Means | |
|-----------------------------|-----|----|---|--------|-------|------------------------------|--------|
| Sleep Problems | 230 | 69 | 2.862 | .092 | 1.074 | 297 | .284 |
| Specific Behaviour problems | 204 | 59 | 1.589 | .209 | 4.535 | 261 | .000** |
| Cluster Lund | 174 | 57 | 5.795 | .017** | 3.705 | 122.201 | .000** |

Appendix C

VABS

Tests of Normality

| | Kolmogorov-Smirnov ASD/MR+ | | | Kolmogorov-Smirnov ASD/MR | | |
|------------------|-------------------------------|----|------|------------------------------|----|------|
| | Statistic | df | Sig. | Statistic | df | Sig. |
| Rec DA | .113 | 20 | .200 | .158 | 20 | .200 |
| Expr DA | .160 | 20 | .194 | .226 | 20 | .009 |
| WT DA | .438 | 20 | .000 | .396 | 20 | .000 |
| PS DA | .107 | 20 | .200 | .138 | 20 | .200 |
| DS DA | .217 | 20 | .014 | .130 | 20 | .200 |
| Com DA | .280 | 20 | .000 | .166 | 20 | .148 |
| Int. DA | .125 | 20 | .200 | .296 | 20 | .000 |
| SLT DA | .173 | 20 | .118 | .138 | 20 | .200 |
| SS DA | .343 | 20 | .000 | .267 | 20 | .001 |
| GM DA | .167 | 20 | .145 | .203 | 20 | .030 |
| FM DA | .182 | 20 | .082 | .182 | 20 | .081 |
| Com DA | .153 | 20 | .200 | .198 | 20 | .038 |
| DLS DA | .185 | 20 | .072 | .124 | 20 | .200 |
| Soc. DA | .135 | 20 | .200 | .158 | 20 | .200 |
| Mot DA | .100 | 20 | .200 | .159 | 20 | .200 |
| Total DA | .118 | 20 | .200 | .116 | 20 | .200 |
| Total norm DA | .123 | 20 | .200 | .117 | 20 | .200 |

PEP-R

Tests of Normality

| | Kolmogorov-Smirnov ASD/MR+ | | | Kolmogorov-Smirnov ASD/MR | | |
|----------|-------------------------------|----|------|------------------------------|----|------|
| | Statistic | df | Sig. | Statistic | Df | Sig. |
| Total DA | .148 | 20 | .200 | .121 | 20 | .200 |
| Age | .142 | 20 | .200 | .159 | 20 | .198 |
| ImS | .208 | 20 | .023 | .141 | 20 | .200 |
| ImDA | .180 | 20 | .089 | .190 | 20 | .056 |
| PS | .132 | 20 | .200 | .172 | 20 | .123 |
| PDA | .165 | 20 | .159 | .171 | 20 | .127 |
| FMS | .125 | 20 | .200 | .192 | 20 | .052 |
| FMDA | .126 | 20 | .200 | .159 | 20 | .197 |
| GMS | .212 | 20 | .019 | .183 | 20 | .077 |
| GMDA | .154 | 20 | .200 | .243 | 20 | .003 |
| EHS | .104 | 20 | .200 | .154 | 20 | .200 |
| EHDA | .089 | 20 | .200 | .158 | 20 | .200 |
| CPS | .184 | 20 | .076 | .132 | 20 | .200 |
| CPDA | .227 | 20 | .008 | .158 | 20 | .200 |
| CVS | .300 | 20 | .000 | .276 | 20 | .000 |
| CVDA | .331 | 20 | .000 | .322 | 20 | .000 |

SPT

Tests of Normality

| | Kolmogorov-Smirnov ASD/MR+ | | | Kolmogorov-Smirnov ASD/MR | | |
|------|-------------------------------|----|------|------------------------------|----|------|
| | Statistic | df | Sig. | Statistic | df | Sig. |
| SPTS | .378 | 18 | .000 | .262 | 18 | .002 |
| SPTO | .369 | 18 | .000 | .342 | 18 | .000 |

COMVOOR

Tests of Normality

| | Kolmogorov-Smirnov ASD/MR+ | | | Kolmogorov-Smirnov ASD/MR | | |
|--------------------------|-------------------------------|----|------|------------------------------|----|------|
| | Statistic | df | Sig. | Statistic | df | Sig. |
| Sum serie 1 | .308 | 20 | .000 | .308 | 20 | .000 |
| Sum serie 2 | .221 | 20 | .011 | .221 | 20 | .011 |
| Sum serie 3 | .255 | 20 | .001 | .255 | 20 | .001 |
| Sum serie 4 | .313 | 20 | .000 | .313 | 20 | .000 |
| Sum serie 5 | .255 | 20 | .001 | .255 | 20 | .001 |
| Sum presentational level | .178 | 20 | .095 | .178 | 20 | .095 |
| Sum representational | .273 | 20 | .000 | .273 | 20 | .000 |
| Total score | .214 | 20 | .017 | .214 | 20 | .017 |

DBC

Tests of Normality

| | Kolmogorov-Smirnov ASD/MR+ | | | Kolmogorov-Smirnov ASD/MR | | |
|----------------------------|-------------------------------|----|------|------------------------------|----|------|
| | Statistic | df | Sig. | Statistic | df | Sig. |
| Comm Dist | .114 | 18 | .200 | .195 | 19 | .055 |
| Social Relating | .159 | 18 | .200 | .167 | 19 | .175 |
| TBPS | .093 | 18 | .200 | .163 | 19 | .199 |
| Disruptive / Antisocial | .123 | 18 | .200 | .137 | 19 | .200 |
| Selfabsorbed | .101 | 18 | .200 | .138 | 19 | .200 |
| Anxiety | .193 | 18 | .073 | .111 | 19 | .200 |

DISCO II

Tests of Normality

| | Kolmogorov-Smirnov ASD/MR+ | | | Kolmogorov-Smirnov ASD/MR | | |
|----------|-------------------------------|----|------|------------------------------|----|------|
| | Statistic | df | Sig. | Statistic | df | Sig. |
| COMREC | .188 | 20 | .062 | .187 | 20 | .065 |
| COMEXPSN | .210 | 20 | .021 | .272 | 20 | .000 |
| COMEXPS | .158 | 20 | .200 | .204 | 20 | .028 |
| COMNSCH | .368 | 20 | .000 | .368 | 20 | .000 |
| COMSCH | .326 | 20 | .000 | .371 | 20 | .000 |
| COMSS | .190 | 20 | .057 | .199 | 20 | .036 |
| COMSSN | .174 | 20 | .114 | .194 | 20 | .046 |
| COMSNSN | .179 | 20 | .093 | .169 | 20 | .137 |
| COMSNS | .164 | 20 | .168 | .225 | 20 | .009 |
| DLS PERS | .257 | 20 | .001 | .156 | 20 | .200 |
| DLS DOM | .538 | 20 | .000 | a | a | a |
| DLS COM | .538 | 20 | .000 | .280 | 20 | .000 |
| DLS | .255 | 20 | .001 | .203 | 20 | .030 |
| SOCG26N | .257 | 20 | .001 | .336 | 20 | .000 |
| SOCG29N | .300 | 20 | .000 | .276 | 20 | .000 |
| SOCG34N | .283 | 20 | .000 | .228 | 20 | .008 |
| SOCG39N | .340 | 20 | .000 | .207 | 20 | .025 |
| SOCINT | .108 | 20 | .200 | .140 | 20 | .200 |
| SOCH3N | .345 | 20 | .000 | .327 | 20 | .000 |
| SOCH4N | .538 | 20 | .000 | .349 | 20 | .000 |
| SOCH6N | .481 | 20 | .000 | .275 | 20 | .000 |
| SOCPLAY | .364 | 20 | .000 | .142 | 20 | .200 |
| SOCPLAY4 | .188 | 20 | .061 | .128 | 20 | .200 |
| SS | .303 | 20 | .000 | .431 | 20 | .000 |
| SOC | .114 | 20 | .200 | .098 | 20 | .200 |
| MOTGR | .205 | 20 | .027 | .170 | 20 | .133 |
| MOTF | .261 | 20 | .001 | .305 | 20 | .000 |
| MOT | .170 | 20 | .130 | .193 | 20 | .048 |
| SPEC OBJ | .287 | 20 | .000 | .252 | 20 | .002 |
| SPEC | .150 | 20 | .200 | .133 | 20 | .200 |
| SENSES | | | | | | |
| SPECN22N | .327 | 20 | .000 | .243 | 20 | .003 |
| SPECN23N | .311 | 20 | .000 | .281 | 20 | .000 |
| SPECOBJ | .155 | 20 | .200 | .138 | 20 | .200 |
| SPECFMI | .216 | 20 | .016 | .144 | 20 | .200 |
| ADAPSS | .151 | 20 | .200 | .244 | 20 | .003 |
| ADAPSNSN | .118 | 20 | .200 | .127 | 20 | .200 |
| ADAPSSN | .121 | 20 | .200 | .139 | 20 | .200 |
| ADAPSNS | .134 | 20 | .200 | .196 | 20 | .044 |
| SPEC | .110 | 20 | .200 | .164 | 20 | .163 |

A daghuis is constant

NVOS/NOSIK

Tests of Normality

| | Kolmogorov-Smirnov ASD/MR+ | | | Kolmogorov-Smirnov ASD/MR | | |
|-----------------------------|-------------------------------|----|------|------------------------------|----|------|
| | Statistic | df | Sig. | Statistic | df | Sig. |
| Acceptation | .152 | 17 | .200 | .186 | 17 | .123 |
| Coping | .127 | 17 | .200 | .165 | 17 | .200 |
| Having problems | .171 | 17 | .197 | .135 | 17 | .200 |
| Wanting to change situation | .132 | 17 | .200 | .094 | 17 | .200 |
| Child is burden | .208 | 17 | .049 | .118 | 17 | .200 |
| Being alone | .206 | 17 | .053 | .267 | 17 | .002 |
| Having fun | .250 | 17 | .006 | .195 | 17 | .086 |
| Good relationship | .238 | 17 | .011 | .144 | 17 | .200 |
| B | .212 | 17 | .041 | .161 | 17 | .200 |
| Effort self and partner | .206 | 17 | .054 | .163 | 17 | .200 |
| Skills | .161 | 17 | .200 | .207 | 17 | .051 |
| Proportion partner | .233 | 17 | .015 | .291 | 17 | .000 |
| Level of difficulty | .154 | 17 | .200 | .131 | 17 | .200 |
| Coincidence/fate | .174 | 17 | .181 | .212 | 17 | .041 |
| Satisfaction | .196 | 17 | .081 | .275 | 17 | .001 |
| Will to change | .175 | 17 | .172 | .104 | 17 | .200 |
| Wants help | .183 | 17 | .134 | .189 | 17 | .107 |
| Help expectancy internal | .180 | 17 | .146 | .202 | 17 | .063 |
| Help expectancy external | .150 | 17 | .200 | .139 | 17 | .200 |
| Perspective | .169 | 17 | .200 | .181 | 17 | .140 |
| Nosik | .165 | 17 | .200 | .132 | 17 | .200 |

Curriculum Vitae

Yvette Marjo Dijkxhoorn is in 1966 te Rotterdam geboren. De lagere school heeft ze in Lekkerkerk doorlopen, waarna ze in 1979 naar de brugklas van de scholengemeenschap Crimpenerwaard te Krimpen a/d IJssel ging. In 1981 werd de schoolloopbaan voortgezet op de Gymnasium-afdeling van Rijksscholengemeenschap te Brielle. Na het eindexamen Gymnasium-B heeft ze een jaar Biologie gestudeerd aan de universiteit van Leiden waarna ze in 1987 overstapte naar Pedagogiek. In 1992 studeerde ze aan de Rijksuniversiteit Leiden af in de Orthopedagogiek met als specialisatie ontwikkelingsstoornissen.

Direct na haar afstuderen is ze in dienst getreden bij de afdeling Orthopedagogiek van de universiteit Leiden. Eerst als derde-geldstroomonderzoekster en sinds 1998 ook als docent. Haar onderzoekstaken richten zich met name op mensen met ernstige ontwikkelingsstoornissen, in het bijzonder probleemgedrag en specifieke zorg voor mensen met autisme. Daarnaast is zij sinds 1996 als consulent werkzaam voor de verschillende consulente teams. Sinds 1999 is ze geregistreerd als Gezondheidszorgpsycholoog.