

University of Groningen

The Friesland study

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Document Version

Publisher's PDF, also known as Version of record

Publication date:

2003

[Link to publication in University of Groningen/UMCG research database](#)

Citation for published version (APA):

Bildt, A. A. D. (2003). *The Friesland study: pervasive developmental disorders in mental retardation*. [Thesis fully internal (DIV), University of Groningen]. [s.n.].

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Chapter 1

Introduction

1.1 Introduction

From clinical experiences and from research, it is well known that the co-occurrence of a pervasive developmental disorder in mental retardation causes many additional problems for children and their environment (Tsai, 1996; Kraijer, 1997, 1998). With respect to behavior, these additional problems are reflected in a higher amount of problem behavior, and a lower level of adaptive behavior, compared to children with mental retardation without a pervasive developmental disorder (Kraijer, 1997, 1998). Bryson (1996) mentioned significant behavior problems, or additional psychiatric problems in 50% of the individuals with mental retardation and a pervasive developmental disorder. The co-morbid, more or less non-specific problems known to occur, vary from separation anxiety, self-injurious behavior, general anxiety or fears, hyperactivity, compulsions/rituals, depression, irritability/agitation, sleep problems, tics, excessive masturbation and rumination (Kraijer, 1997). Besides, results of various studies on adaptive behavior, reveal both an overall impairment in adaptive behavior, as well as limitations in specific areas, particularly the social domain (Volkmar et al., 1987; Kraijer, 1987; Sparrow, 1997; Carter et al., 1998; Kraijer, 2000).

Due to the effects of pervasive developmental disorders on the (mal)adaptive behavior of children with mental retardation, it is of major importance that health or educational workers in the area of mental retardation are aware of the possibility of a co-morbid pervasive developmental disorder. On a community or organizational level, this awareness is needed for provision and planning of additional services for children and adolescents affected. Moreover, on the individual level, awareness of clinicians of pervasive developmental disorders and their effects, is the start for an early diagnostic process, to investigate whether a pervasive developmental disorder is indeed present. Early identification, leading to a timely provision of specific interventions and services, increases the opportunities for prevention of major behavior problems and stimulation of the development of skills needed to function in day-to-day life. This will improve the quality of life of the child or adolescent, and his/her environment.

To contribute to early identification and intervention, three factors are important. First, adequate tools or instruments are needed, to measure pervasive developmental disorders in children and adolescents with mental retardation. Second, information on the prevalence of pervasive developmental disorders in

children and adolescents with mental retardation, will increase the awareness of professionals that pervasive developmental disorders occur in this population. Third, insight into the additional and specific effects of pervasive developmental disorders on (mal)adaptive behavior of children and adolescents with mental retardation, will create a better basis for decisions concerning the most accurate and effective character, moment and duration of an intervention or other service for the individual child.

The present study was undertaken to contribute specifically to the issues of identification and prevalence of pervasive developmental disorders, and to their role in (mal)adaptive behavior. By examining several instruments to identify pervasive developmental disorders in children and adolescents with mental retardation, this study will provide information on their value for establishing a reliable estimate of the prevalence of pervasive developmental disorders in the population, or for diagnosing the disorder on an individual level. Through investigating the role of pervasive developmental disorders in (mal)adaptive behavior of children and adolescents with mental retardation, we hope the present study will lead to an increased awareness of the suppressing effect of pervasive developmental disorders on the abilities of these children, and the necessity of appropriate intervention.

In the paragraphs 1.2 and 1.3, a short description of mental retardation and pervasive developmental disorders will be given, to outline the terminology and definitions used in this study. Paragraph 1.4 considers the literature available on the combination of pervasive developmental disorders and mental retardation, its prevalence, and its effects on (mal)adaptive behavior. In this paragraph the objectives of the present study will also be specified. In paragraph 1.5 the method of the study will be explained, with respect to the participants and the sampling procedure.

1.2 Mental retardation

Terminology

Currently, the terminology concerning mental retardation is not unequivocal. Different countries and different disciplines use different terms, such as 'intellectual disabilities', 'developmental disabilities', 'learning disabilities', or even 'people with challenges', etc. The main reason for this, which is specifically reflected by the last

term, is to avoid the negative connotations that the term mental retardation has for some people. As described by the American Association on Mental Retardation (AAMR) in their last edition of *'Mental retardation, definition, classification and systems of supports'* (2002), mental retardation is still the official and scientific term at this moment, although changes may occur in the near future (see also the DSM-IV-TR (APA, 2000) and the ICD-10 (WHO, 1992)). In concordance with the AAMR we use the term mental retardation in the present study, since there is no consensus on an acceptable alternative yet.

Definition

The most recent definition of mental retardation is from the AAMR (2002): *'Mental retardation is a disability characterized by significant limitations both in intellectual functioning and in adaptive behavior as expressed in conceptual, social and practical adaptive skills. This disability originates before age 18'*. With respect to intelligence, the significant limitations should be approximately two standard deviations below the mean. The significant limitations in adaptive behavior are defined as two standard deviations below the mean on either conceptual, social or practical skills, or on the overall adaptive skills as measured with standardized instruments for adaptive behavior. The emphasis of the definition is on the combination of limitations in both intelligence and adaptive behavior.

Categories or levels of mental retardation

Classification of individuals based on the level of severity of mental retardation is mostly based on IQ-scores only. This is the reason why the AAMR dropped this classification in categories of level of mental retardation in 1992. The DSM-IV-TR and ICD-10 still discriminate four levels, with criteria based on intelligence only. In the care for people with mental retardation, at least in the Netherlands, but probably also in other countries, these levels or categories are still common and widely used. In the present study, we followed the classification of the DSM-IV-TR. Using the terms commonly known, enables us to communicate our results to researchers and clinical workers in this area more clearly.

The largest category (approximately 85% of the population with mental retardation) is the category of mild mental retardation, defined by an IQ range of 50-55 to approximately 70. Moderate mental retardation is defined by an IQ range of 35-

40 to 50-55, and includes approximately 10% of the people with mental retardation. Severe mental retardation occurs in only 3%-4% of the population with mental retardation. The level of intelligence is required to be between 20-25 and 35-40 for this category. The last category, profound mental retardation, is the smallest (1%-2% of the population with MR) and is defined by an IQ below 20-25 (DSM-IV-TR, APA, 2000).

Instead of a classification based on IQ-scores, the AAMR (1992, 2002) suggests an alternative classification, based on the intensity of needed supports. Four levels of intensity are distinguished: intermittent, limited, extensive, and pervasive. Several factors of the services needed are taken into account for this classification, e.g. time duration, time frequency, settings in which the support is needed, resources required for the service and the degree of intrusiveness in one's life; yet strict and reliable criteria for these levels of support are still lacking. In this system of classification, the level of adaptive behavior is more important than in the DSM-IV-TR, and therefore is more concordant with the current ideas about mental retardation. We agree with the AAMR that categories should be based on more than IQ only, and that adaptive behavior, skills and needs should be incorporated. However, at least in the Netherlands, this alternative system has not been widely implemented yet.

Prevalence

The prevalence rate of mental retardation is between 1 to 3 % in the total population. The percentage of the population considered having a level of intelligence of two standard deviations below the mean is 2 to 3%. However, with adaptive behavior incorporated, the prevalence decreases, especially in the highest level of intellectual functioning. Rutter et al. (1976) already illustrated this in the early seventies. They found a prevalence rate of 2.5% of children with mental retardation, defined by IQ score only. Yet, when they looked at the children who received special services for mental retardation, or who went to special schools etc., they found a decreased prevalence rate of 1.3%. Additionally, it is known that after the school period, the prevalence of mental retardation decreases, to 1% or even lower. This is caused by the fact that some children with an IQ below 70, who were identified as having mental retardation in school, function adaptively and adequately and live an independent life, when academic skills are less important. Especially with the

emphasis on limitations both in intelligence and adaptive behavior in the definition of mental retardation, the prevalence of 1% as given by the DSM-IV-TR, is considered to be the most reliable estimate. This prevalence appears to best exclude incorrectly classified individuals in the levels of intellectual functioning around the cut-off for mental retardation. The male:female ratio is 60%:40% for the total population with mental retardation (DSM-IV-TR, APA, 2000).

1.3 Pervasive Developmental Disorders

Terminology

The terminology on pervasive developmental disorders is at least as confusing as on mental retardation. The main terminological confusion exists, not surprisingly, in the area of the so-called pervasive developmental disorders-not otherwise specified. Again, different terms are used to describe the same concept: 'autism spectrum disorders', 'the lesser variant', 'the broader phenotype', etc. In the most recent version of the DSM (DSM-IV-TR, APA, 2000) and in the last version of the ICD (ICD-10, WHO, 1992), the term pervasive developmental disorders is still the official term. Therefore, this is the term used in the present study.

Generally, in the present study, the term pervasive developmental disorders is used to address 'Autistic Disorder' (AD) through 'Pervasive Developmental Disorder-Not Otherwise Specified' (PDD-NOS). With respect to instruments however, we follow the definitions that underlie the concept of the different instruments. Since some instruments are based on definitions from the DSM, and some are not, this means that we sometimes have to use slightly different terms. When this occurs, this will be specified in the text.

Definition

According to the DSM-IV-TR, *'pervasive developmental disorders are characterized by severe and pervasive impairment in several areas of development: reciprocal social interaction skills, communication skills, or the presence of stereotyped behavior, interests and activities. The qualitative impairments that define these conditions are distinctly deviant relative to the individual's developmental level or mental age'*. The disorder is usually evident in the first years of life.

Categories of pervasive developmental disorders

The pervasive developmental disorders considered in the present study are Autistic Disorder and Pervasive Developmental Disorder-Not Otherwise Specified. The specific criteria of these disorders, from the DSM-IV-TR, are shown in table 1.1. The criteria for Pervasive Developmental Disorder-Not Otherwise Specified are, in concordance with its name, clearly less straightforward than for Autistic Disorder.

The categories Rett's Disorder, Childhood Disintegrative Disorder, and Asperger's Disorder, were left out of consideration for the present study. Rett's Disorder and Childhood Disintegrative Disorder are very rare. A classification of Asperger's Disorder is officially not concordant with mental retardation, and was therefore not applicable in our population.

Prevalence

The prevalence of pervasive developmental disorders in the general population has been subject of study for many years. The DSM-IV-TR reports a prevalence of 5 per 10,000 for Autistic Disorder, referring to rates ranging from 2 to 20 per 10,000 in different studies. For the spectrum of pervasive developmental disorders, Yeargin-Allsopp et al. (2003) found a prevalence rate of 34 per 10,000 in children between 3 and 10 years of age. According to Fombonne (2003) this is likely to be an underestimate. Based on recent studies (Baird et al., 2000; Chakrabarti & Fombonne, 2001; Bertrand et al., 2001), Fombonne estimates the prevalence rate of pervasive developmental disorders at 60 per 10,000.

1.4 Pervasive developmental disorders in mental retardation

Over the years many studies have shown that mental retardation occurs very frequently in children with a pervasive developmental disorder, especially in autism or Autistic Disorder, with estimates between 70 and 90% (DeMyer et al., 1974; Wing & Gould, 1979; Wing, 1981; Rutter, 1983; Steffenburg & Gillberg, 1986; Wing, 1993; Bryson, 1996, 1997; APA, 2000). Due to the increasing attention for High Functioning Autism and Asperger's Syndrome, the percentage of individuals with a pervasive developmental disorder and mental retardation tends to decrease. Nevertheless, it is still substantial (Gillberg, 1999). Conversely, no unambiguous estimate is available of how many children with mental retardation have a co-morbid pervasive developmental disorder.

Table 1.1 Diagnostic criteria for Autistic Disorder and Pervasive Developmental Disorder-Not Otherwise Specified, DSM-IV-TR (APA, 2000)

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 gesture 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.

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 or 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. For example, this category includes 'atypical autism' - presentations that do not meet the criteria for Autistic Disorder because of late age at onset, atypical symptomatology, or subthreshold symptomatology, or all of these.

The overlap in the definitions of both disorders implicates difficulties in differentiating a pervasive developmental disorder from mental retardation. In both disorders, limitations in social and communicative behavior are present. Although not part of the definition, stereotyped behavior is known to occur frequently in children with mental retardation, especially in the lower levels of functioning (Kraijer, 1997, 2002), and also in children with a pervasive developmental disorder. Due to the behavioral overlap between children with mental retardation and children with a pervasive developmental disorder, the identification of pervasive developmental disorders in children with mental retardation is complicated (DiLavore, Lord, & Rutter, 1995). A pervasive developmental disorder should at least be considered when a child shows problems in social, communicative or stereotyped behavior. On an individual level, an extensive diagnostic process is then needed to identify whether or not a co-morbid pervasive developmental disorder affects the behavior of the child.

Identification of pervasive developmental disorders in children and adolescents with mental retardation

Standardized instruments developed for the assessment of pervasive developmental disorders can have great value for such a diagnostic process, although each instrument has its limitations (Lord, Rutter, & Le Couteur, 1994). One type of limitations does not specifically seem to be related to the instruments, but to the difficulties in applying the concept of pervasive developmental disorders in mental retardation, as mentioned above. However, to standardize the diagnostic process of children and adults, instruments are needed for clinical as well as for research purposes. The other type of limitations regards the psychometric qualities of an instrument. Instruments are only valuable, if they measure pervasive developmental disorders validly and reliably. Therefore it is important to study these psychometric qualities and the utility of the available instruments.

The *first* objective of the present study therefore was to investigate instruments developed for identifying pervasive developmental disorders (also) in children with mental retardation.

As a first step, we aimed to increase the insight into the performance of two screening instruments for pervasive developmental disorders, the Scale of Pervasive Developmental Disorder in Mentally Retarded persons, PDD-MRS (Kraijer, 1999) and the Autism Behavior Checklist, ABC (Krug, Arick, & Almond, 1980) in a total

population of children and adolescents with mental retardation. The aim was to investigate the interrelationship between the screening instruments, and their sensitivity and specificity in relation to the diagnostic instruments Autism Diagnostic Interview-Revised, ADI-R (Lord et al., 1994) and the Autism Diagnostic Observation Schedule, ADOS (Lord, Rutter, DiLavore, & Risi, 1998; Lord et al., 2000), and to the clinical DSM-IV-TR classification (APA, 2000). Chapter 2 describes the results and implications of this study.

Additionally, we investigated the ADI-R and ADOS in 184 children and adolescents with mental retardation, including the lowest functioning individuals. Since no specific information on the utility of these instruments in this population had been presented before, this study will contribute to understanding the value of these instruments in the diagnostic process in this population. The specific aim of this study was to examine the interrelationship between the instruments and the criterion related validity compared with a clinical DSM-IV-TR classification. The results of this study are presented in chapter 3.

Prevalence of pervasive developmental disorders in children and adolescents with mental retardation

Reviewing the literature reveals various estimates of the prevalence rate of pervasive developmental disorders in children and adolescents with mental retardation, ranging from 3% to 50% (Wing & Gould, 1979; Gillberg, 1983, 1984; Gillberg, Persson, Grufman, & Themner, 1986; Steffenburg, Gillberg, & Steffenburg, 1996; Benassi et al., 1990; Kraijer, 1991; King, DeAntonio, McCracken, Forness, & Ackerland, 1994; Deb & Prasad, 1994; Kraijer, 1997). Methodological issues explain the differences between the estimates. The studies differ from each other with respect to the concept under study and the sample. Although the concept is often defined with the DSM, the exact definitions of the DSM-III (APA, 1980) and DSM-III-R (APA, 1987) differ. Besides, Wing & Gould (1979), Wing (1981), Gillberg et al. (1986) and Steffenburg et al. (1996) also investigated broader definitions that go beyond the DSM. Additionally, the instruments from the studies are based on different theoretical bases and times, thereby also influencing the concept that is eventually measured. The samples in the studies differ with respect to age, level of mental retardation, and representativity.

Based on this literature, three issues are concluded to be important, to establish a reliable estimate of the prevalence: 1) a clear definition of the concept (which disorder is actually represented by the prevalence rate?), 2) standardized instruments (to increase the standardization, and thereby the comparability of results in different studies) and 3) a well described, representative sample (for the appropriate application of the prevalence rate to other groups).

The *second* objective of the present study was to establish a reliable estimate of the prevalence, taking these three issues into consideration. Therefore, in our prevalence study, we defined the concept as 'Autistic Disorder' and 'Pervasive Developmental Disorder-Not Otherwise Specified' according to the DSM-IV-TR criteria (APA, 2000). With respect to the importance of uniformity in research, we used standardized instruments, widely used both nationally and internationally. The representativity has been addressed by undertaking a population-based study. In chapter 4, the prevalence rates based on this study will be presented.

(Mal)adaptive behavior in children and adolescents with mental retardation and pervasive developmental disorders

Limitations in adaptive behavior in general or in specific areas, have become increasingly important in the definition of mental retardation (AAMR, 1992, 2002). Additionally, various studies show impairments in adaptive behavior in children with pervasive developmental disorders, also reflected in the general level and in specific areas. The limitations in the social area of adaptive behavior are specifically high, whereas the areas of motor skills and daily living skills seem relatively unaffected. Limitations in the area of communication are comparable to those of children with Down syndrome or language disorders, yet are clearly more limited when compared to other children with mental retardation without a pervasive developmental disorder (Volkmar et al., 1987; Freeman, Ritvo, Yokota, Childs, & Pollard, 1988; Volkmar, Carter, Sparrow, & Cicchetti, 1993; Vig & Jedrysek, 1995; Carpentieri & Morgan, 1996; Carter et al., 1998; Kraijer, 2000; Liss et al., 2001).

For children with both disorders, adaptive behavior is therefore dually affected. Understanding the specific effects of pervasive developmental disorders on adaptive and maladaptive behavior in children with mental retardation, will contribute to the diagnostic and the treatment process. Knowing what specific behavior of a child may be influenced by a pervasive developmental disorder, will ring alarm bells when that

behavior is present, leading to further examination of the child. For the treatment process, this understanding will contribute to a timely provision of a specific intervention or service to stimulate the development of skills and to prevent or decrease behavior problems.

The *third* objective of the present study therefore was to contribute to understanding the effect of a co-morbid pervasive developmental disorder on adaptive behavior, by investigation of three specific questions in three separate studies.

Before being able to judge limitations or skills in adaptive behavior, the level of adaptive behavior has to be measured or evaluated. In the first study, presented in chapter 5, a widely used and well-known instrument for measuring adaptive behavior, the Vineland Adaptive Behavior Scales, VABS (Sparrow, Balla, & Cicchetti, 1984) is thoroughly investigated. Although well developed and normed for the population of American children without mental retardation, no previous information was available on the application, reliability, validity or norms for children with mental retardation. This study contributes to the knowledge of the psychometric qualities, and thereby the value of the VABS, in measuring adaptive behavior in individuals with mental retardation.

The second study aimed to investigate social skills in children with mild and moderate mental retardation, with and without pervasive developmental disorders. Particularly subtle social skills were investigated, based on the Children's Social Behaviour Questionnaire, CSBQ (Luteijn, Minderaa, & Jackson, 2002), to see whether a measure of these subtle social skills would contribute to differentiating between children of different levels of intellectual functioning, with and without a pervasive developmental disorder. This study is described in chapter 6.

Chapter 7 presents the last study, in which the interrelationship between autistic and general behavior problems, the level of adaptive behavior and academic achievement was investigated, in a group of children within the highest, small range of IQ (60-70). The main objective was to increase the insight into the influence of (mal)adaptive behavior on academic achievement in children with mental retardation. Insight into this relationship will be of major clinical importance with respect to increasing the opportunities of children who have high enough IQ's to achieve certain skills and reach a higher level of education, but who are restricted by other factors.

The general conclusions of the separate studies, and their clinical and research implications, will be discussed in chapter 8.

1.5 Method

The overall design of this study involved investigation of a total population of children with mental retardation between 4 and 18 years of age. This paragraph gives an overview of the general sample of participants, their characteristics and how we approached them.

Procedure

The children and adolescents for this study were recruited from the province of Friesland, a province in the northern part of the Netherlands. At the time of the study, the total population consisted of approximately 618,000 inhabitants, including approximately 120,000 children between 4 and 18 years old (Provinsje Fryslân, 1998).

All 4 through 18 year-old children (suspected) with mental retardation in Friesland were asked to participate (N = 1436). All levels of mental retardation were included. No children were excluded based on etiology, sensory or motor impairments, psychiatric disorders or behavior problems. The only children excluded were from families who did not speak the Dutch or Frisian language. Children were approached through the facilities (schools, day-care facilities and institutions) they were known to (IDC-Friesland, 1998).

Schools

At the beginning of the study, two levels of special education for children with mental retardation existed: 'schools for children with mild learning problems' (in Dutch MLK), and 'schools for children with severe learning problems' (in Dutch ZML). The first type of school was the highest level of special education for children with mental retardation and provided education for children from 4 through approximately 12 years and for children from approximately 13 through 16-18 years (in Dutch VSO-MLK). The aim of these schools is that children achieve the objectives of general primary education, e.g. learn to read, write and arithmetic, by receiving additional support, adjusted to the specific needs of each child. Some objectives from general primary education have to be adjusted for these children, and some other objectives will not be achievable for them at all. During the study, this type of school has been

joined with another type of special education (i.e. in Dutch LOM), and does not longer exist as an independent type of education.

Children in schools for severe learning problems, have severely restricted abilities with respect to learning due to mental retardation, although they are able to participate in some form of education in a classroom setting. The schools provide education for children from 4 through 20 years. The objectives of general primary education are unachievable for these students, or can only partially be achieved. Therefore, aims in this type of school more often concern training of daily living skills and other basic skills. Children in this type of school receive many additional services and are often supported individually. During the study, an IQ of 60 or below was the criterion to attend a school for children with severe learning difficulties. Nevertheless, in practice some children with an IQ higher than 60 were admitted, due to the limiting effect on learning of other factors, such as behavior problems.

In total, all 30 schools for children with mental retardation in the province of Friesland participated. Some schools had more than one location, sometimes providing both types of education. Twenty schools or departments of schools were for children with mild learning problems, and 10 were for children with severe learning problems.

Day-care facilities

In day-care facilities for children with mental retardation, most children function at the lower levels of mental retardation. A day-care facility is accessible for children from 2 through 15-17 years old. However, many children leave the day-care facility around 6-8 years, to visit a school. Day-care facilities aim to stimulate adaptive behavior, and social, emotional, cognitive and motor development, all within the boundaries of the child. Special support is provided to meet the specific needs of the particular child, e.g. psychological, medical, or physical. In Friesland, there were 4 day-care facilities, and they all participated.

Institutions

At the beginning of the study, there were two institutions for mental retardation in Friesland that provided places for children and adolescents with IQ's of 70 and below. Both institutions participated in our study. Each of them had various locations throughout a wider area, including separate group homes that were sometimes located in the community. Depending on the level of mental retardation and co-morbid physical or behavior problems, the group homes differ in the level of support

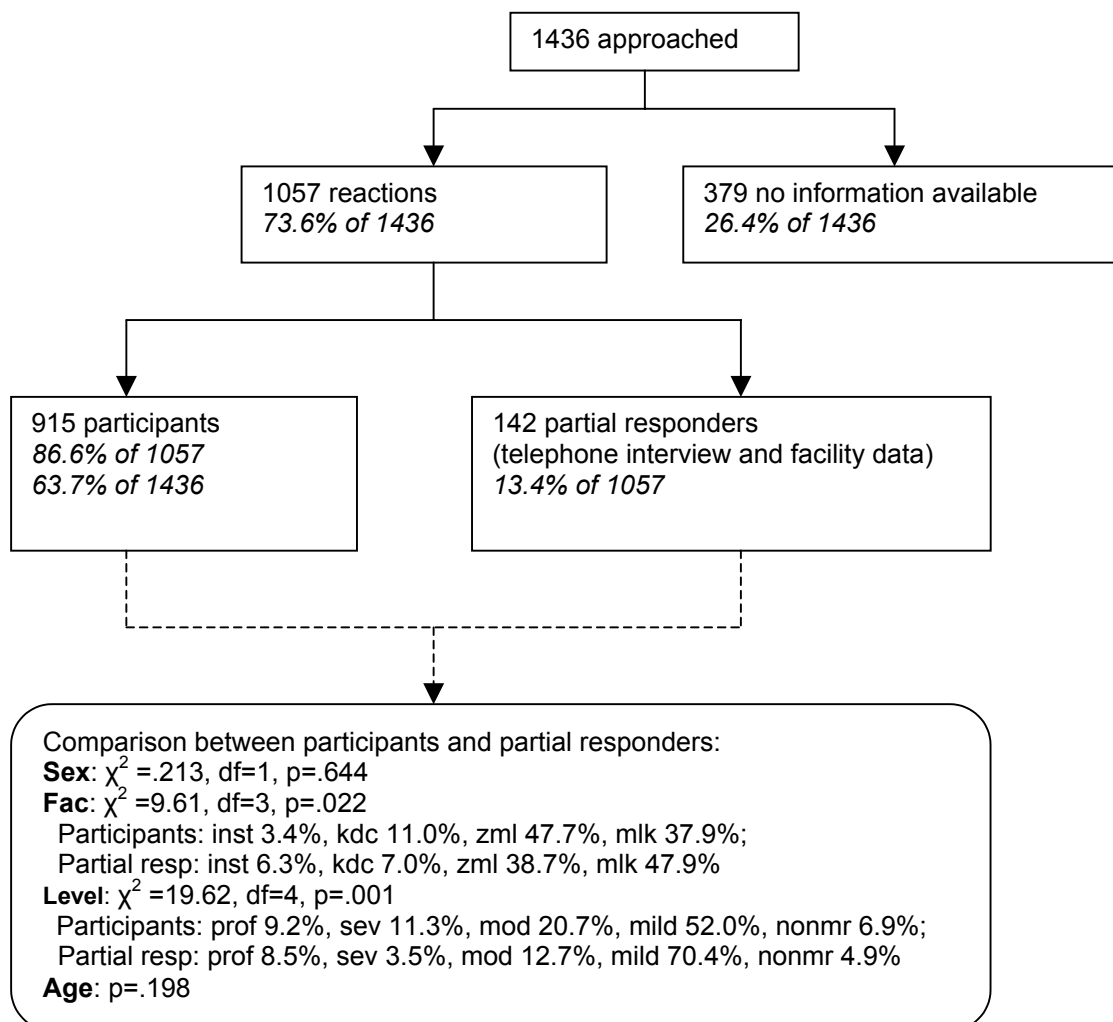
they provide. During the day most children and adolescents go to school, day-care facility or work. The children in our study, approached through the institutions mostly had low IQ's, falling into the severely and profoundly mentally retarded range. Some children had higher IQ's, however these children were also approached through their schools.

Due to privacy reasons, we were not able to approach parents directly. Therefore, schools, day-care facilities and institutions sent out a letter to all parents about the study. This letter included a letter of the school, day-care facility or institution, to recommend the study; a letter with information from the university; an application form and a stamped return-envelope. Parents were asked to return the application form to the University of Groningen, if they were interested in participation. Not all parents responded after the letter only. In cooperation with schools, day-care facilities and institutions, parents who did not respond received a new letter, asking if they would allow the university to call them. Only a very small proportion of parents objected to calling. The other parents received a telephone call, to explain the purpose of the study, how much time it would take to participate, and the exact procedure. Often parents said they had read the information, but either forgot to send the form back, or they had some unanswered questions that could be answered during the telephone conversation. When parents decided to participate, the usual procedure of the study was started. When parents decided not to participate, in most cases a non-response questionnaire was completed during the telephone call. In all cases we asked parents if we could send a questionnaire to school, day-care facility or institution.

The procedure of data collection started with setting a date with parents for an interview. Psychologists, social workers or last-year students in these areas administered the interview, after training in administering and scoring. The interviews were recorded on audiocassette and were scored double randomly. Regularly organized meetings during data collection served to maintain consensus on administration and scoring of the interview. Approximately two weeks before the interview, parents received a booklet of questionnaires on the behavior of their child that had to be completed before the interview. The parents returned the questionnaires to the interviewer during the interview. This enabled interviewers to check if all questions were completed, and to assist parents in questions that they

had difficulties with. Sometimes, parents needed help with all questions, and the interviewer visited the parents more often to complete the questionnaires together.

Figure 1.1 Sampling of children and adolescents with mental retardation in Friesland



Participants

In the following paragraphs, the participants of the study will be described. First, an overview will be given of the total population. Second, the children and adolescents who were reached after approach will be described. This sample will be compared to the total population, in order to examine the characteristics of the non-response population.

Total population

In figure 1.1, a schematic overview of the procedure is presented, with the numbers of children and adolescents in each step of sampling. The initial number of

1436 children, was the maximum estimate of children to include in our study. This number is probably too high, due to the fact that it is highly likely to include children who do not fulfill our inclusion criteria. In the group of children, whose parents provided all information, approximately 10% of the children did not fulfill all criteria, e.g. being 4 through 18 years old, having mental retardation, and being from a family that speaks the Dutch or Frisian language. These children were therefore excluded from the analyses.

Non-response analyses

Unfortunately, we were not able to analyze all inclusion criteria for the 379 children of whom we had no information, due to the privacy reasons already mentioned. However, we were able to compare the distribution of facilities between the 1057 responders and the initial 1436. This comparison showed that the responders were significantly deviant from the total population concerning this one aspect ($\chi^2=34.4$, $df=3$, $p<.001$). The response group contained more children from schools for severe learning problems, whereas the non-response group ($n=379$) largely consisted of children and adolescents from schools for mild learning problems. When interpreting the results of our study, this is something that should be taken into consideration.

Reached children and adolescents

With the procedure as described above, 1057 children and adolescents (666 males, 391 females) were reached (73.6%). In table 1.2 the characteristics of this sample are presented. In 86.6% ($n=915$) of the cases we received information from parents and from the school, day-care facility or institution. This group is referred to as 'participants'. In 13.4% ($n=142$) of the cases, we received information from the parents during a telephone interview, and information from school, day-care facility or institution. This group is referred to as 'partial responders', since we have facility information on their behavior, and some background information from parents.

In general, when only considering the 86.6% of the children of whom data were available from parents and school, day-care facility or institution, the response rate was highest in schools for severe learning problems (71.3%), compared to the initial 1436 children and adolescents who were approached. The lowest response rate was found in institutions (40.3%). However, it is important to mention that higher-functioning children who lived in an institutionalized setting were approached through

their school or day-care facility, and that they were not incorporated in this response rate, keeping it artificially low. The response rates in schools for mild learning problems (58.4%) and in day-care facilities (64.9%) were in between.

Table 1.2 Characteristics of the population of children and adolescents with MR in Friesland (n=1057)

		Institution		Day-care facility		School for severe learning problems		School for mild learning problems	
		n	%	n	%	n	%	n	%
Sex	Male	31	75.6	60	55.6	320	64.8	255	61.6
	Female	10	24.4	48	44.4	174	35.2	159	38.4
Level of mental retardation	Profound	29	70.7	62	57.4	4	0.8	1	0.2
	Severe	5	12.2	29	26.9	68	13.8	6	1.4
	Moderate	1	2.4	9	8.3	153	31.0	44	10.6
	Mild	6	14.6	5	4.6	237	48.0	328	79.2
	Non-MR	0	0.0	3	2.8	32	6.5	35	8.5
Age	< 12	9	22.0	87	80.6	211	42.7	190	45.9
	≥ 12	32	78.0	21	19.4	283	57.3	224	54.1
	Total	41		108		494		414	

Non-response analyses

Comparing the partial responders with the participants revealed no significant differences between the two groups with respect to sex or age. However, a significant difference existed with respect to the facility of the child ($\chi^2=9.61$, $df=3$, $p=.019$), and with respect to level of mental retardation ($\chi^2=19.62$ $df=4$, $p=.001$). In the response group, 47.7% of the children attended a school for severe learning problems, and 37.9% attended a school for mild learning problems. In the non-response group, the percentages were reversed, 38.7% attended a school for severe learning problems, and 47.9% attended a school for mild learning problems. The differences between the groups were much less explicit in institutions (resp. 3.4%, non-resp. 6.3%) and day-care facilities (resp. 11.0%, non-resp. 7.0%). Additionally, the group of participants contained relatively many children from the severe and moderate levels of mental retardation, as opposed to the group of partial responders, that for 70.4% consisted of children from the mild level of mental retardation. These factors are related to each other, since children from schools for mild learning

problems are more likely to have mild mental retardation than children from schools for severe learning problems (84.1% vs. 48.8%).

The differences between the two groups with respect to the distribution of facilities and the level of mental retardation, point into the same direction: children from lower levels of functioning are overrepresented in our sample, whereas higher functioning children are underrepresented, as compared to the total population. This probably leads to a bias in our findings, that we have to keep in mind when interpreting the results of the various studies.

Levels of mental retardation

Assignment of the participants to the four levels of mental retardation as mentioned in the DSM-IV-TR (APA, 2000) was based on IQ score, available from the facility, and/or a measure of adaptive behavior, as administered during the study. In 80% of the cases, this classification was based on information from intelligence tests or developmental tests obtained by the facility. In most cases these were Dutch versions of standardized tests, e.g. Wechsler Intelligence Scale for Children-Revised, WISC-R (Wechsler, 1974; Vander Steene et al., 1986) Wechsler Preschool and Primary Scale for Intelligence-Revised, WPPSI-R (Wechsler, 1989; Vander Steene & Bos, 1997), Snijders-Oomen Niet-verbale intelligentie test-Revisie, SON-R (Snijders, Tellegen, Winkel, & Laros, 1996), and the Bayley scales of Infant Development (Bayley, 1969; Van der Meulen & Smrkovsky, 1983). In the other 20% of the cases, participants were assigned to one of these categories based on the Vineland Adaptive Behavior Scales, VABS (Sparrow et al., 1984), administered as part of the study, and clinical review of functioning.

The cut-off criteria we followed for differentiation between the four levels were an IQ-score from 0 through 20 for profound, an IQ-score from 21 through 35 for severe, an IQ-score from 36 through 50 for moderate, and an IQ-score from 51 through 70 for mild mental retardation.

In the various studies, presented in the following chapters, specific (sub)samples of participants were included, depending on the objective of the specific study. Additionally, in each study specific instruments were selected, to measure the behavior or concept that was investigated. Each chapter will therefore give an overview of the specific sample and the instruments that were applied.

