J Autism Dev Disord (2006) 36:723–732
DOI 10.1007/s10803-006-0115-0

ORIGINAL PAPER

Screening for Autistic Spectrum in Children Aged 14 to 15 Months. I: The Development of the Early Screening of Autistic Traits Questionnaire (ESAT)

Sophie H. N. Swinkels · Claudine Dietz · Emma van Daalen · Ine H. G. M. Kerkhof · Herman van Engeland · Jan K. Buitelaar

Published online: 14 April 2006

Abstract This article describes the development of a screening instrument for young children. Screening items were tested first in a non-selected population of children aged 8–20 months (n = 478). Then, parents of children with clinically diagnosed ASD (n = 153, average age 87 months) or ADHD (n = 76, average age 112 months) were asked to score the items retrospectively for when their child was 14 months old. A 14-item screening instrument, Early Screening of Autistic Traits (ESAT) which had maximal sensitivity and specificity for ASD was developed. The sensitivity of the ESAT was checked in an independent sample of 34 children aged 16–48 months clinically diagnosed with ASD. A 4-item version appears to be a promising prescreening instrument.

Keywords Autistic spectrum disorder · Screening · Instrument · Early identification

Introduction

The importance of an early diagnosis of autistic spectrum disorders (ASD) is generally recognized (Bristol-Power & Spinella, 1999; Vig & Jedrysek, 1999) because it allows the early initiation of treatment, family support, and genetic counseling, and timely planning of educational objectives. Indeed, several studies suggested positive change in language, social, or cognitive outcomes of young children with ASD after early started interventions. Unfortunately, methodological limitations preclude the definitive attributions of this positive change, to specific intervention procedures (National Research Council, 2001). Further, early diagnosis makes it possible to increase our knowledge of the course of the clinical characteristics of ASD and to study the environmental, behavioral, cognitive, and biological factors that influence this course.

Identification of Abnormalities

Several attempts have been made to identify abnormalities specific to young preschool children and infants with autism. A study interviewing parents of children with and without autism, about their child's behavior in the first 2 years of life, identified abnormalities characteristic for autism (Wimpory, Hobson, Williams, & Nash, 2000). For example, children with autism did not show behaviors characteristic of person-to-person-object interactions, namely, to offer, give, show, or point to objects in relation to someone else. Nearly all of the infants in the control group but not a single child with autism followed another person's pointing. Items on pointing to objects, giving or showing objects, and following another person's pointing are also part of the Checklist for Autism in Toddlers (CHAT) (Baron-Cohen, Allen, & Gillberg, 1992; Baron-Cohen et al., 1996). ASD are characterized by impairments in three areas: reciprocal social interaction, communication, and repetitive and stereotyped patterns of interest and behavior. In accordance with the assumption that social impairments are central to the disorder, social deficits are

S. H. N. Swinkels · C. Dietz (✉) · E. van Daalen · I. H. G. M. Kerkhof · H. van Engeland
Department of Child and Adolescent Psychiatry, University Medical Center Utrecht, B01.201, Post Box 85500, 3508 GA, Utrecht, The Netherlands
e-mail: c.dietz@azu.nl

J. K. Buitelaar · S. H. N. Swinkels
Department of Psychiatry, University Medical Center Nijmegen, Nijmegen, The Netherlands

generally considered the most prominent characteristic of very young children with autism (Stone et al., 1999; Wimpory et al., 2000), and thus most studies on the early identification of ASD, including the ones mentioned above, have focused on aspects of social interactions. This exclusive focus on social deficits can be criticized. First of all, social impairments are not specific for children with ASD. Children with delayed language development, learning disabilities, mental retardation, attachment disorder, and environmental deprivation may also have problems with social interaction (Ghuman, Freund, Reiss, Serwint, & Folstein, 1998). Secondly, behaviors such as pointing to objects, giving, or showing objects and following another person's pointing all develop between age 9 and 18 months during normal development (Carpenter, Nagell, & Tomasello, 1998). A delay in the development of a behavioral skill can only become apparent at an age at which the vast majority of children have mastered the skill. This questions the usefulness of behaviors relating to person-to-person object interactions for the detection of ASD in children younger than 18 months.

The general approach of retrospective studies that have used interviews or questionnaires has been to ask about the child's behavior in the first 2 or 3 years of life. Few studies have focused exclusively on younger ages. An exception is the study of Vostanis et al. (1998), in which the parents of referred children with ASD completed a questionnaire on concerns regarding their child's development between 12 and 18 months. Play behavior items and lack of referential gestures were found to best predict the diagnosis of autism. Gillberg (1989) studied symptoms reported by the parents of children with autism younger than 2–3 years. The item ''Does not play like other children'' was among the three items with the strongest discriminatory power. Another study compared autistic children with blind children and found that, during the first year of life, autistic children were more fixated than blind children on certain very specific activities, such as whirling the wheels of toy cars, and were characterized as being completely absorbed by these activities, losing contact with outer reality (Janson, 1993). Sensorimotor functioning may also be a marker of ASD. Gillberg (1989) concluded that abnormal perceptual responses were possibly more important for the early identification of ASD than aspects of social and communication. Baranek (1999) also found several sensorimotor deficits to be subtle yet salient predictors of autism at 9–12 months in children with autism.

Screening activities are crucial to early diagnosis. The purpose of screening is to identify children at risk of autism as soon as possible so that they can be rapidly referred for full diagnostic assessment. It should be appreciated that decisions to introduce screening tests to the general population not only depend on the availability of adequate screening tests with sufficient psychometric properties. Additional considerations for such decisions were formulated by the National Screening Committee, UK (2003). They apply to the importance of the disorder for public health, the feasibility of the test (simple, safe, precise and validated), the availability of facilities for diagnosis and treatment, also the cost of screening should be economically balanced in relation to expenditure on the care and treatment of persons with the disorder as well as to medical care as a whole.

An example of an autism screening instrument is the Pervasive Developmental Disorders Screening Test (PDDST) (Siegel, 1996). However, the psychometric data for this instrument were not presented per age group and so its use for screening very young children is not clear. The only screening instrument that has been tested in a large unselected sample is the CHAT (Baron-Cohen et al., 1996). The CHAT was developed to screen children for autism at age 18 months and is based on three key psychological predictors, namely, a lack of pretend play, a lack of protodeclarative pointing, and a lack of gaze monitoring. The presence of two or more of these predictors at 18 months is predictive of a later diagnosis of autism. About 16,000 British 18-month-old children were screened with the CHAT by primary healthcare practitioners; children with a clear developmental delay were not screened. The population was monitored at 7 years of age to establish the sensitivity, specificity, and positive predictive value of the instrument (Baird et al., 2000). Specificity was close to 100% when using a high threshold. A two-stage procedure improved the Positive Predictive Value. The CHAT also identified some children with a broader defined ASD or language disorder or related developmental disorder by 42 months. However, the CHAT appeared to lack sensitivity at 18 months, i.e., it missed 62% of the children who were later found to have autistic disorder.

Robins, Fein, Barton, and Green (2001) made a modified version of the CHAT, the M-CHAT, and screened 1,122 children aged 18–30 months from a non-selected population and 171 children from a sample referred for early intervention services. The reported preliminary data are promising but a large-scale validation study is necessary to evaluate the usefulness of the M-CHAT as a screening instrument.

Goal of this Study

Primary healthcare practitioners would gain considerably from a simple screening instrument that alerts them to early signs of ASD and of the need for further specialist evaluation. No such instrument is currently available. The first step in the development of the screening instrument is to develop a provisional version that minimizes false-negative results and which selects the maximal number of children with ASD. The next step is to perform a large population study with the provisional version and to clinically assess all screen-positive children. The diagnostic classification of the screen-positive children provides important information on the screening properties of the provisional version, enabling the instrument to be fine-tuned to achieve an optimal balance between specificity and sensitivity. This article presents the results of the first step. In accordance with child screening routines in the Netherlands, emphasis was on the screening of children aged 14–15 months.

Study 1

Method

Development of ESAT

After a review of the literature of early symptoms of autism reported in retrospective studies, prospective studies, and family home movies (Adrien et al., 1992, 1993; Baron-Cohen et al., 1996; Duijnhoven & Hoekman, 1995; Gillberg, 1989; Gillberg et al., 1990; Gillberg, Nordin, & Ehlers, 1996; Janson, 1993; Lord, 1995; Osterling & Dawson, 1994), we selected 19 potential screening items that formed the preliminary version of the Early Screening of Autistic Traits (ESAT). This version of the ESAT included the three key items of the CHAT (Baron-Cohen et al., 1992, 1996) and covered the domains of pretend play, joint attention, interest in others, eye contact, verbal and nonverbal communication, stereotypes, preoccupations, reaction to sensory stimuli, emotional reaction, and social interaction. We rephrased the items to achieve optimal comprehensibility and to arrive at dichotomous scores, with a negative answer reflecting abnormality. The items of the ESAT are presented in Table 1.

Participants

The ESAT was completed by the parents/caregivers of 478 children aged 8–20 months with reference to the child's current behavior. In the Utrecht area, caregivers were approached at day-care units, shopping malls, swimming pools, and recreational areas. Locations were chosen in different neighborhoods to ensure that the sample consisted of a mixture of children from various socioeconomic backgrounds.

Of the 478 children, the mean age was 13 ± 3 months (*M* ± *SD*). Two hundred and forty-eight of these children were aged between 13 and 17 months (see Fig. 1). The total group consisted of 238 boys and 231 girls; nine caregivers forgot to tick the sex of the child. Most caregivers ($n = 442$) received the ESAT test in the form of a questionnaire that could be completed at home and returned by mail. Some caregivers ($n = 36$) were interviewed. The ESAT test was completed by the child's parents ($n = 375$) or by another caregiver who knew the child very well ($n = 103$).

We further asked parents ($n = 153$) who were members of the Dutch National Autistic Society and who had a child with a diagnosis of ASD (i.e., either autistic disorder, Asperger syndrome, or PDD-NOS) to complete the ESAT test retrospectively with reference to their child's behavior at 14 months of age. The mean age (± *SD*) of the 153 children was 87 (± 46) months (range 14–226 months); 86.4% of the children were boys. To check the test–retest reliability of retrospective reporting, we asked the parents of 50 children to complete the test a second time after a mean of 5.4 months ($SD = 1.7$, range 3–10 months). These 50 children were similar to the overall sample in terms of sex ratio and level of functioning but were older (*M* ± *SD* 115 ± 46 months) as to the overall group.

As a comparison group, the parents of 76 children with a diagnosis of ADHD (combined type) and without the diagnosis of ASD, completed the ESAT test retrospectively with reference to their child's behavior at 14 months of age. The mean age (± *SD*) of the 76 children was 112 (± 37) months (range 34–201 months); 90.6% of the children were boys.

Unfortunately, we were not able to match the children with ASD and ADHD, as children with ADHD were older, $t(241) = -3.77$, $P < .001$ (one-tailed). No information was available on the developmental level of the children within the ADHD comparison group.

Results

Non-selected Population

Caregivers other than parents were significantly more inclined than parents to give negative answers to the ESAT items (mean score 1.98 vs. 1.40, respectively; ANOVA with age as covariate $F(1, 472) = 11.0$, $P < .001$). It made no difference whether the ESAT was administered by interview or questionnaire: 442 caregivers gave an average of 1.52 negative answers to the questionnaire whereas 33 caregivers gave 1.50 negative answers in the interview.

Figure 1 shows the percentage of negative answers to each item by age for the children of the non-selected population. The percentage of negative answers for each item averaged over the age range of 13–17 months and the percentage of items left unanswered are presented in Table 1.

Table 1 Percentage of negative answers to the ESAT items in each group. The percentage of missing (unanswered) items in study 1 is given in parentheses

ESAT items	Study 1			Study 2
	Non-selected 13–17 months n = 248	ADHD at 14 months n = 76	ASD at 14 months n = 153	ASD at 14 months n = 34
1. Is your child interested in different sorts of objects and not for instance mainly in cars or buttons?	0 (0)	10.0 (2.5)	55.4 (1.2)	55.9
2. Can your child play with toys in varied ways (not just fiddling, mouthing or dropping them)?	1.6 (1.2)	20.0 (8.8)	65.1 (4.8)	55.9
3. When your child expresses his/her feelings, for instance by crying or smiling, is that mostly on expected and appropriate moments?	0 (0)	15.0 (6.3)	56.6 (12.0)	50
4. Does your child react in a normal way to sensory stimulation, such as coldness, warmth, light, sound, pain or ticking?	0 (.4)	7.5 (3.8)	61.4 (4.8)	52.9
5. Can you easily tell from the face of the child how he/she feels?	.5 (1.0)	13.8 (5.0)	59.6 (6.6)	47.0
6. Is it easy to make eye-contact with your child?	1.2 (.4)	32.5 (7.5)	65.1 (4.8)	73.5
7. When your child has been left alone for some time, does he/she try to attract your attention, for instance by crying or calling?	1.5 (1.5)	8.8 (6.3)	63.9 (2.4)	52.9
8. Is the behavior of your child free of stereotyped repetitive movements like banging his/her head or rocking his/her body?	4.0 (2.0)	18.8 (16.3)	59.6 (5.4)	50
9. Does your child, on his/her own accord, ever bring objects over to you or show you something?	1.2 (1.2)	5.0 (7.5)	65.7 (3.6)	35.3
10. Does your child show to be interested in other children or adults?	0 (0)	3.8 (3.8)	51.1 (12.8)	64.7
11. Does your child like to be cuddled?	1.2 (2.4)	23.8 (11.3)	34.0 (10.6)	23.5
12. Does your child ever smile at you or at other people?	.4 (0)	6.3 (2.5)	50.6 (6.0)	38.2
13. Does your child like playing games with others, such as peek-a-boo, ride on someone's knee, or to be swung?	0 (0)	3.9 (0)	42.6 (14.9)	14.7
14. Does your child react when spoken to, for instance, by looking, listening, smiling, speaking or babbling?	0 (.8)	5.0 (1.3)	33.7 (12.7)	47.1
15. Does your child speak a few words or utter various babbling sounds?	1.2 (1.2)	7.9 (4.8)	40.3 (3.4)	–
16. When you are pointing at something, does your child follow your gaze to see what you are pointing at?	2.8 (3.2)	0 (15.9)	55.5 (6.7)	–
17. Does your child ever use his/her index finger to point, to indicate interest in something?	5.2 (2.4)	1.6 (28.6)	63.9 (8.4)	–
18. Does your child ever use his/her index finger to point, to ask for something?	10.9 (2.8)	11.1 (19.0)	58.0 (5.9)	–
19. Does your child ever pretend, for example, to make a cup of tea using a toy cup and teapot, or pretend other things?	34.7 (12.1)	22.2 (22.2)	77.3 (5.9)	–

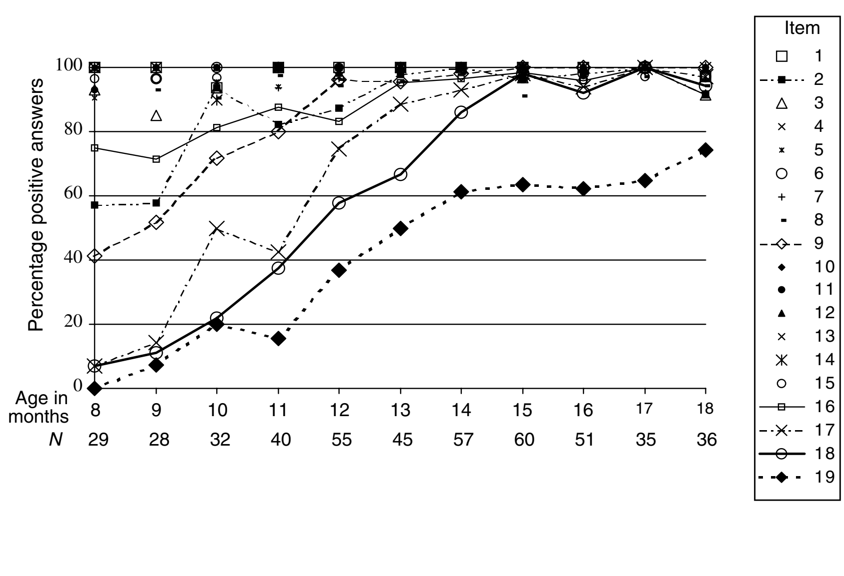

Fig. 1 The percentage of negative answers to each item by age (age in months) for the children from the non-selected population. The second row of the x-axis gives the number of children in each age block (n)

Most items were answered negatively in fewer than 2% of the children. Exceptions were the answers to the item on stereotyped repetitive movements, both items on pointing, and the item on pretend play. The rate of negative answers to each of these four items, averaged over the children aged 13–17 months, was higher than 4%. Further, the answers to most items were largely independent of age after 12 months. The answers to the three key items of the CHAT were most strongly related with age. At age 8 months, each of the CHAT behaviors was present in fewer than 10% of the children in the non-selected population. Over the months thereafter, these behaviors tended to be present more often. Protodeclarative pointing was present in more than 90% of the children at about 13 months and instrumental pointing at about 14–15 months. However, about 25% of the parents apparently did not observe pretend play by age 18 months (Table 2). The high percentage of missing data may reflect that parents found items difficult to understand or difficult to answer in a categorical yes/no format (Table 1). The items on pretend play (12.1%) and gaze following (3.2%) in particular had a relatively high percentage of missing data.

Children with Autistic Spectrum Disorder

The test–retest reliability (intraclass correlation coefficient) for the retrospective data for the children with clinically diagnosed ASD was .81 ($P < .001$), with sum scores for the 19 items of 10.0 ($SD = 4.1$, range 0–16) at the first rating and 11.0 ($SD = 4.8$, range 1–19) at the second rating. We then checked whether the number of items answered negatively was a function of the current age of the child and thus of the time since the child was 14 months old. Among this group of children with ASD, the Pearson correlation coefficient between the age of the child at the time that the parent completed the test retrospectively and the 19-item sum score was .20 ($P < .001$). Further, there was no significant relationship between the age of the child at the time the parent completed the test retrospectively and the number of missing values in the questionnaire ($r = -.11$, $P = .17$).

The percentage of negative answers to each of the ESAT items in the ASD group was between 33.7 and 77.3% (see Table 1). For each item, this percentage was significantly higher than for the children from the non-selected population (cross-table analysis, $P < .001$). The mean Phi for the ASD population versus the non-selected population for all 19 items was .64. The highest Phi values were found for item 2 (play in varied ways, .74), item 4 (react normal to stimulation, .74), and item 9 (bring or show, .74). With the exception of item 19 (pretend play, .40), all Phi values were higher than .5.

We also investigated the extent to which the percentage of negative answers to the ESAT items was dependent on the level of functioning of the child with ASD. To this end, the level of functioning was clustered into four categories on the basis of the school placement of the child (1 = non-specialized facilities for the normal population; 4 = specialized facilities for non-speaking children with very low level of functioning). Twenty-seven children were placed in category 1, 30 in category 2, 28 in category 3, and 54 in category 4. The remaining children did not go to school or a day-care facility, mostly because they were too young (see Table 3). The level of functioning (category number)

Table 2 The percentage of negative answers to ESAT items by age (age in months) from the children from the non-selected population

Age in months											
ESAT items (*n*)	8 (29)	9 (28)	10 (32)	11 (40)	12 (55)	13 (45)	14 (57)	15 (60)	16 (51)	17 (35)	18 (36)
1	100	100	94	100	100	100	100	100	100	100	97
2	57	58	94	82	87	98	100	97	98	100	92
3	93	85	94	100	98	100	100	97	100	100	91
4	100	100	100	100	100	100	100	100	100	100	100
5	91	100	96	94	98	100	100	100	97	100	100
6	100	96	100	100	96	100	100	97	98	100	97
7	100	100	100	94	98	100	100	96	97	100	96
8	100	93	91	97	94	96	98	91	98	97	94
9	41	52	72	80	96	96	98	100	100	100	100
10	100	100	100	100	100	100	100	100	100	100	100
11	93	100	100	100	100	98	100	100	100	100	100
12	100	100	100	100	100	100	100	97	100	100	100
13	100	100	100	100	100	100	100	100	100	100	100
14	100	100	90	100	97	100	100	100	100	100	100
15	97	96	97	100	100	100	100	100	96	97	97
16	75	71	81	88	83	95	96	98	96	100	91
17	7	14	50	43	75	87	93	98	94	100	97
18	7	11	22	38	58	67	86	98	92	100	94
19	0	7	20	15	37	50	61	64	62	65	74

Table 3 Percentage of negative answers to the ESAT items, for children diagnosed with ASD, and categorized by school placement

School placement	*n*	Mean number of negative answers	*SD*
1. Non-specialized facilities for the normal population	27	5.2	3.3
2. Specialized facilities for children with average cognitive functioning	30	10.1	3.2
3. Specialized facilities for children with mild retardation	28	10.0	4.2
4. Specialized facilities for non-speaking children with very low level of functioning	54	12.1	3.4

correlated significantly with the total number of negative answers on the ESAT test ($r = .56$, $P < .001$). Children in special schools for children with learning disabilities had higher scores than children visiting schools for children with about normal level of functioning. For each individual item, level of functioning was found to be related to percentage of negative scores, with the exception of item 13 (enjoys social play) and item 11 (likes cuddling). Differences were tested against $P < .05$, without correction for multiple comparisons. Items that were particularly sensitive to level of functioning were item 1 (interest different toys) (X^2 (3, $n = 124$) = 20.83, $P < .001$; item 2 (varied play) (X^2 (3, $n = 124$) = 32.42, $P < .001$; item 9 (bring or show) (X^2 (3, $n = 124$) = 24.93, $P < .001$; and item 17 (point to indicate interest) (X^2 (3, $n = 122$) = 27.10, $P < .001$).

To investigate the extent to which the items discriminated between children from a non-selected population and children with ASD with a high level of functioning, Phi values were calculated for ASD category 1 versus the non-selected population. The mean Phi value was .38, considerably lower than the mean Phi value for the comparison of all children with ASD versus the non-selected population. Relatively high Phi values (above .4) for children with ASD with a high level of functioning versus the non-selected population were found for items 1, 3, 4, 5, 6, 7, and 12.

For most ESAT items the percentage of negative answers for the children with ADHD was between that for the children from the non-selected population and the children with ASD (see Table 1). Overall, the Phi values for the ASD group of children versus the ADHD group of children were comparable, but slightly lower, than the Phi values for ASD versus the non-selected population. The exception was item 11 (like to be cuddled) with a Phi value for the ASD group versus the ADHD group of .28, considerably lower than the Phi value for the ASD group versus the non-selected group (.50). The percentage of missing items was significantly higher in the ASD and ADHD groups than in the non-selected group.

Cut-off values for Autistic Spectrum Disorder

We then derived a cut-off value for the detection of ASD. This value should satisfy two requirements: maximal sensitivity for ASD, and maximal specificity for ASD versus normal development. For these analyses, the questionnaires for two children in the non-selected population, 12 children in the ASD group, and 13 children in the ADHD group were excluded because there were more than three missing values.

Cross-tabulation analysis showed that by using all 19 items and a criterion of four (or more) negative answers, 89.5% of the children in the ASD group versus 2.4% of the children in the non-selected group would have screened positive for ASD. Omission of the two items that had a relatively high percentage of negative answers in the non-selected group diminished the sensitivity for ASD. However, omission of more items and changing the cut-off value to three or more negative answers improved the sensitivity without affecting the specificity for recognizing abnormal development. After testing different sets of items, we found that items 1–14 and a cut-off value of three (or more) negative answers detected 0% of the non-selected sample, 90.1% of the children with ASD, and 19.0% of the children with ADHD as screening positive for ASD.

Further analysis showed that 94.3% of the children who screened positive for ASD had one or more negative answer on items 1–4 whereas only 2.0% of the non-selected population had one or more negative answers on these items. For two other sets of four items (items 2, 4, 7, 8; and items 2, 4, 7, 12), 96.8 and 96.0% of children who screened positive for ASD had at least one negative answer, respectively. However, since 5.9 and 3.8% of the non-selected sample had at least one negative answer on these items, respectively, these two sets differentiated less well between children with ASD and children from the non-selected population than the set of the first four ESAT items.

Study 2

Method

The aim of our second study was to investigate the sensitivity of our provisional instrument and cut-off values in a new sample of relatively young and well-described children with ASD.

Subjects

Thirty-four young children with ASD were recruited from the preschool day treatment program of the Department of Child Psychiatry in Utrecht. The diagnoses of all children were made following extensive and protocol led evaluations including a developmental history, standardized psychiatric observation by means of the ADOS-G (Lord et al., 1989). Observations on the ward, testing of developmental level and language functions, pediatric examination, and full somatic work-up. Independent of the ADOS-G algorithm, the responsible child psychiatrist (EvD) reached a clinical diagnosis using all available information and a best estimate approach. Inclusion criteria were (1) diagnosis of autistic disorder by either clinical diagnosis or ADOS-G algorithm or both, and (2) if only one of the diagnoses (clinical diagnosis or ADOS-G algorithm) indicated the presence of autistic disorder, the other diagnosis should at least indicate the presence of ASD. We opted for this approach because we wanted to include only those children for whom it was highly likely that they would receive a diagnosis in the ASD spectrum at older age. It is difficult to distinguish between autistic disorder and ASD in young children, as it is to distinguish between milder variants of ASD and normal development (Cox et al., 1999). Furthermore, the concordance between clinical diagnosis and ADOS-G algorithm in very young children is lower than it is in older children (Lord, 1995).

The mean age of the children at the time of the interview was 37 months (SD = 8, range 16–48 months). The group consisted of 27 boys and 7 girls. Cognitive functioning was in the normal range in eight children (DQ $\geq$ 85), in the borderline range in seven children (DQ 70–85), and in the mentally retarded range in 19 children (DQ $<$ 70). Language development was appropriate for chronological age in four children, slightly delayed, but with five words spoken spontaneously, in 11 children, and seriously delayed (fewer than five words spoken) in 19 children.

Procedure

Parents were interviewed about when they first started to worry about their child's development, and were asked to describe their first concerns. After this interview and on a separate occasion, parents were asked to answer the first 14 items of the ESAT retrospectively with respect to their child's behavior at about 14 months of age. Both interviews took place when the child was visiting the Department of Child Psychiatry for a diagnostic evaluation and therefore before the parents were informed about the diagnosis of their child.

Results

Open Questions

Over a quarter of the parents (26.5%) indicated that their concerns had started almost immediately after birth or at least before their child was 3 months old. Another 47.1% noticed abnormalities in behavioral development before their child's first birthday, 17.6% started worrying in the period between 12 and 18 months, and only 8.8% started worrying after their child was 18 months old.

Parents most frequently mentioned problems of reciprocal social interactions when asked about their first concerns: 67.7% of the parents spontaneously mentioned concerns such as ''rarely took the initiative to interact'' and ''looked at a spot on the wall behind you, never at your face''. Other concerns were excessive crying or sleeping problems (64.7%), abnormalities of language (47.1%) or locomotion (26.5%); parents seldom mentioned other concerns, such as poor adaptation to change (11.8%), abnormal perceptual responses (5.9%), play development (5.9%).

Parents who had concerns before the first birthday of their child mentioned first worries about the development of language less frequently than parents who started worrying after the first birthday of their child (X^2 (1, n = 34) = 8.60, $P < .001$). No such difference was found for the other areas of development.

ESAT Items at about 14 months (retrospectively)

The retrospective data for the 14-item version of the ESAT, when the children were about 14 months, are summarized in Table 1. For most items the percentage of negative answers in this study was rather similar to the percentage found for the ASD group of study 1.

All but two children with ASD (32/34, 94%) tested positive for ASD when the cut-off value was three or more negative answers. The parents of these two screen-negative children were convinced that their child had developed normally until 15 months of age. Parents of one of these children mentioned their child had lost skills after a flu in the second year.

There were no significant associations between the number of negative answers to ESAT items and the child's level of cognitive functioning (r = .11) or language development (r = .14), nor was the answer to individual items significantly associated with either language development or cognitive functioning. All but three children (91%) had at least one negative answer to the first four ESAT items.

To explore the relationship between the answers to the ESAT and the age of first worry, the sample was divided into two subgroups: children whose parents had started worrying before the first birthday ($n = 25$) versus children whose parents had started worrying thereafter ($n = 9$). The two subgroups of children were similar in age at the time of the interview ($M = 36.4$ vs. 38.1 months, respectively) but differed significantly in the ESAT sum score ($M = 7.4$ vs. 4.6, respectively, t (32) = 2.52, $P = .02$). Two items, item 2 ''play in varied ways'' and item 3 ''express feelings on expected and appropriate moments'', were answered negatively significantly more often by parents who had started worrying before the first birthday than by parents who had started worrying thereafter (Fisher's exact test, $P = .02$ and $P < .01$, respectively).

Discussion

Our goal was to develop a provisional version of a screening instrument to detect ASD in very young children for use in the general population. We used data collected retrospectively from parents who had a child with ASD to construct an instrument that showed a higher than 90% sensitivity for ASD. This 14-item test also had a high specificity to differentiate normal from abnormal development but was less specific in distinguishing between ASD and other types of abnormal development. For example, 19% of children with ADHD scored above threshold. Furthermore, level of functioning was found to have a major influence, in addition to diagnosis, on the scores for the test, with lower level of functioning being related to higher scores. The results of study 1 must be further interpreted in the context of some limitations, such as the lack of a comparison group of children with mental retardation but without ASD. It is also unknown in which direction the results are biased given that ASD and ADHD groups were not matched on age and IQ. We evaluated the sensitivity of the ESAT test retrospectively in an independent and carefully documented sample of young children with ASD (on average 37 months), for when their child was about 14 months; sensitivity rate was higher than 90%. In this second study, the level of cognitive functioning was not found to affect test scores, which may have been due to the smaller sample size, the lower amount of variation in level of functioning, and the difference in categorizing level of functioning between this study and the first study. The young age of the children in the second study meant that cognitive functioning could only be divided into three broad categories, based on specific tests of cognitive functioning, whereas four categories of cognitive functioning were identified in the first study, based on school placement.

We started with a set of 19 ESAT items but found that the omission of five items improved the specificity of the test without affecting its sensitivity. None of the three key items of the Checklist for Autism in Toddlers (CHAT), (Baron-Cohen et al., 1992, 1996) were included in our preferred selection of items. This was mainly because these items, designed to be applied at 18 months, were less useful at an earlier age. Too many normally developing children lacked the skills implicated by the CHAT items before age 18 months (Willemsen-Swinkels, Buitelaar, & van Engeland, 2001). According to parent report, pretend play was absent in more than 25% of the unselected population even at age 18 months. Another item that was not included in our preferred selection of items concerned using words or babbling. Abnormalities in babbling were shown earlier not to distinguish children with autism from other children (Gillberg, 1989).

Our results suggested that over the age range studied (8–18 months), age affects some items but not others. For example, behaviors such as pretend playing, pointing, and bringing/showing objects are beginning to emerge in this age range and are therefore very sensitive to age. The items ''interest in other people'' and ''gaze following'' had a relatively high proportion of negative answers for children younger than 1 year, probably because these items concern aspects of behavior that are difficult to evaluate in very young children or refer to behavior that still need to be developed during the first year. The other items were found to have a high proportion of positive answers in the non-selected population from 8 months onward, indicating that these items involved aspects of behavior and skills that are present already in very young children.

A rater effect was found within the non-selected population with caregivers other than parents being more inclined to give negative answers. This might have been due to a slightly more objective (less biased) attitude toward the child or, since most of these caregivers were grandparents, it can be related to their more extensive experience with child behavior. Within the diagnostic groups all raters were parents. The difference in ratings between the non-selected population and the diagnostic groups would probably have been larger when all raters in the non-selected population had been parents too.

An issue to consider is whether to use the 14-item ESAT test or to use a much shorter version. Our analyses showed that a 4-item instrument, used as a quick screening instrument, identified almost all children with ASD. A disadvantage of the shorter version is the number of false-positive results. The 4-item version could thus be used as a quick prescreening instrument, to limit the number of children that need to be screened with the full 14-item version of the ESAT test.

Social impairments are considered to be central to ASD (Fein, Pennington, Markowitz, Braverman, & Waterhouse, 1986; Volkmar et al., 1987), and thus should form the focus of early detection (Gillberg et al., 1990). In accordance, we found good Phi values for items like ''Does your child, on his/her own accord, ever bring objects over to you or show you something?'' and ''Is it easy to make eye-contact with your child''. Our results suggest that, in addition to items on social impairments, items relating to lack of variability in play behavior and interests might also be of use for the very early detection of autism. The parents in our second study hardly ever spontaneously mentioned abnormalities in play development when asked about their first concerns. This might explain the relative unpopularity of play-related items in screening instruments. However, when specific questions were asked, most of the same parents reported that they had noticed a lack of variability in play or interests as early as 14 months. Gillberg et al. (1990) found that abnormalities of play were among the symptoms that seemed to be most typical of young children with autistic disorder. Libby, Powell, Messer, and Jordan (1998) reported unusual features in the early spontaneous play in children with autism. Stone et al. (1999) found that, in 2-year-old infants with autistic disorder, clinicians reported more impairments in the social and communication domains than impairments regarding restricted and repetitive activities. Even so, for 83% of the 2-year-old children at least one of the two clinicians reported restricted interests. A similar pattern was seen for items relating to abnormal perceptual responses. Parents hardly ever mentioned such abnormalities when asked about their first concerns but when questioned specifically parents had noticed these abnormalities very early on.

In these studies, parents were asked to report on the presence/absence of ESAT items when their child was about 14 months old. Not surprisingly, parents who had started worrying before this age had significantly more negative answers to the ESAT items than parents who started worrying later. The answers to item 2 ''play in varied ways'' and item 3 ''express feelings on expected and appropriate moments'' were particularly different between the ''early worriers'' and the ''late worriers''. It seems important to note that the age of concern is likely to influence the type of behavior that causes first concerns. For example at an age where language development or the formation of friendships is not yet expected to emerge, these aspects of development are not likely to be topics of concerns of parents.

At a very young age, children with ASD may show subtle abnormalities of social skills or early language development rather than striking impairments in these areas. These subtle abnormalities may be difficult for parents to recognize. In addition, behaviors that are perhaps less typical for ASD, such as lack of variability in play or abnormal perceptual responses, might be of use for early detection of ASD because they are relatively easy to observe and recognize. When children grow older and fail to develop more elaborate social skills, impairments in social development might become more specific indicators of the presence of ASD.

This journal volume includes a second paper on the topic of early detection of ASD discussing the results of a population screening study with the ESAT. This paper is entitled: Screening for Autistic Spectrum Disorder in children aged 14–15 months. II: Population Screening with the Early Screening of Autistic Traits. Design and General Findings.

Acknowledgments This study was supported by grants 940-38-045 and 940-38-014 (Chronic Disease Program), by grant 28.3000-2 of the Praeventiefonds-ZONMW, by the Netherlands Organization for Scientific Research (NWO), by a grant from the Dutch Ministry of Health, Welfare and Culture, and by grants from Cure Autism Now, and the Korczak Foundation.

References

Adrien, J. L., Barthelemy, C., Perrot, A., Roux, S., Lenoir, P., Hameury, L., & Sauvage, D. (1992). Validity and reliability of the infant behavioral summarized evaluation (IBSE) – a rating scale for the assessment of young children with autism and developmental disorders. *Journal of Autism and Developmental Disorders, 22,* 375–394.

Adrien, J. L., Lenoir, P., Martineau, J., Perrot, A., Hameury, L., Larmande, C., & Sauvage, D. (1993). Blind ratings of early symptoms of autism based upon family home movies. *Journal of the American Academy of Child and Adolescent Psychiatry, 32,* 617–626.

Baird, G., Charman, T., Baron-Cohen, S., Cox, A., Swettenham, J., Wheelwright, S., & Drew, A. (2000). A screening instrument for autism at 18 months of age: A 6-year follow-up study. *Journal of the American Academy of Child and Adolescent Psychiatry, 39,* 694–702.

Baranek, G. T. (1999). Autism during infancy: A retrospective video analysis of sensory-motor and social behaviors at 9–12 months of age. *Journal of Autism and Developmental Disorders, 29,* 213–224.

Baron-Cohen, S., Allen, J., & Gillberg, C. (1992). Can autism be detected at 18 months? The needle, the haystack and the CHAT. *British Journal of Psychiatry, 161,* 839–843.

Baron-Cohen, S., Cox, A., Baird, G., Swettenham, J., Nightingale, N., Morgan, K., Drew, A., & Charman, T. (1996). Psychological markers in the detection of autism in infancy in a large population. *British Journal of Psychiatry, 168,* 158–163.

Bristol-Power, M. M., & Spinella, G. (1999). Research on screening and diagnosis in autism: A work in progress. *Journal of Autism and Developmental Disorders, 29,* 435–438.

Carpenter, M., Nagell, K., & Tomasello, M. (1998). Social cognition, joint attention, and communicative competence from 9 to 15 months of age serial no 255.

Cox, A., Charman, T., Baron-Cohen, S., Drew, A., Klein, K., Baird, G., Swettenham, J., & Wheelwright, S. (1999). Autism spectrum disorders at 20 and 42 months of age: Stability of clinical and ADI-R diagnosis. *Journal of Child Psychology and Psychiatry, 40,* 719–732.

Duijnhoven, J., & Hoekman, J. (1995). Opsporing van kinderen met autisme in de jeugdgezondheidszorg. *Tijdschrift voor Jeugdgezondheidszorg, 27,* 8–11.

Fein, D., Pennington, B., Markowitz, P., Braverman, M., & Waterhouse, L. (1986). Toward a neuropsychological model of infantile autism: Are the social deficits primary? *Journal of the American Academy of Child Psychiatrists, 25,* 198–212.

Ghuman, J. K., Freund, L., Reiss, A., Serwint, J., & Folstein, S. (1998). Early detection of social interaction problems: Development of a social interaction instrument in young children. *Journal of Developmental and Physical Disabilities, 19,* 411–419.

Gillberg, C. (1989). Early symptoms in autism. In C. Gillberg (Ed.), *Diagnosis and Treatment of Autism* (pp. 23–32). New York: Plenum Press.

Gillberg, C., Ehlers, S., Schaumann, H., Jakobsson, G., Dahlgren, S. O., Lindblom, R., Bågenholm A., Tjuus, T., & Blidner, E. (1990). Autism under age 3 years: A clinical study of 28 cases referred for autistic symptoms. *Journal of Child Psychology and Psychiatry, 11,* 921–934.

Gillberg, C., Nordin, V., & Ehlers, S. (1996). Early detection of autism. Diagnostic instruments for clinicians. *European Child and Adolescent Psychiatry, 5,* 67–74.

Janson, U. (1993). Normal and deviant behavior in blind children with ROP. *Acta Ophthalmologica Supplement, 210,* 20–26.

Libby, S., Powell, S., Messer, D., & Jordan, R. (1998). Spontaneous play in children with autism: A reappraisal. *Journal of Autism and Childhood Schizophrenia, 28,* 487–497.

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

Lord, C., Rutter, M., Goode, S., Heemsbergen, J., Jordan, H., Mawhood, L., & Schopler, E. (1989). Autism diagnostic observation schedule: A standardized observation of communicative and social behavior. *Journal of Autism and Developmental Disorders, 19,* 185–212.

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.

National Screening Committee (2003). Criteria for appraising the viability, effectiveness and appropriateness of a screening programme. Retrieved December 2004, from http://www.nsc.uk.

Osterling, J., & Dawson, G. (1994). Early recognition of children with autism – A study of first birthday home videotapes. *Journal of Autism and Developmental Disorders, 24,* 247–257.

Robins, D. L., Fein, D., Barton, M. L., & Green, J. A. (2001). The modified checklist for autism in toddlers: An initial study investigating the early detection of autism and pervasive developmental disorders. *Journal of Autism and Developmental Disorders, 31,* 131–144.

Siegel, B. (1996). Pervasive Developmental Disorders Screening test. Unpublished work.

Stone, W. L., Lee, E. B., Ashford, L., Brissie, J., Hepburn, S. L., Coonrod, E. E., & Weiss, B. H. (1999). Can autism be diagnosed accurately in children under 3 years? *Journal of Child Psychology and Psychiatry, 40,* 219–226.

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., Sparrow, S., Goudreau, D., Cicchetti, D., Paul, R., & Cohen, D. J. (1987). Social deficits in autism: An operational approach using the Vineland Adaptive Behavior Scales. *Journal of the American Academy of Child and Adolescent Psychiatry, 26,* 156–161.

Vostanis, P., Smith, B., Corbett, J., Sungum-Paliwal, R., Edwards, A., Gingell, K., Golding, R., Moore, A., & Williams, J. (1998). Parental concerns of early development in children with autism and related disorders. *Autism, 2,* 229–242.

Willemsen-Swinkels, S. H. N., Buitelaar, J., & Engeland, van H. (2001). Is 18 months too early for the CHAT? *Journal of the American Academy of Child and Adolescent Psychiatry, 40,* 737–738.

Wimpory, D. C., Hobson, R. P., Williams, J. M. G., & Nash, S. (2000). Are infants with autism socially engaged? A study of recent retrospective parental reports. *Journal of Autism and Developmental Disorders, 30,* 525–536.