Parental Compliance After Screening Social Development in Toddlers

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Objectives: To examine the prevalence of parents' compliance with follow-up measurements after their child tested positive at a screening to assess problems in social development, as well as to find demographic, screening-related, and child-specific factors associated with parental compliance.

Design: Two-stage screening design.

Setting: Utrecht, the Netherlands.

Participants: A random population of 31,724 children were screened at well-baby clinics at age 14 to 15 months (screen 1). Three hundred sixty-four children underwent screen 2 (255 children who scored positive at screen 1 [population screening] and 109 children younger than 36 months who were identified by surveillance because of suspected problems in their social development).

Main Exposure: A 2-stage screening was applied.

Main Outcome Measures: Compliance with recommendations of having either a second screening (after screen 1) or clinical evaluation (after screen 2).

Results: Of 370 children who tested positive at screen 1, parents of 255 children (69%) complied with screen 2. Three groups were distinguished after screen 2 (n=173): early compliance (clinical evaluation within 6 months) (68%), late compliance (clinical evaluation after 6 months) (14%), and noncompliance (no clinical evaluation) (18%). Late compliance and noncompliance were more common in parents of younger children and children who were identified via population screening. Parents of children with either relatively high cognitive skills and/or low scores on screening measures were less inclined to comply.

Conclusions: Study results suggest higher effectiveness of surveillance over population screening. Screening may well be applied as a second step after surveillance to identify children who need further clinical evaluation.

Arch Pediatr Adolesc Med. 2007;161:363-368

IT IS GENERALLY AGREED THAT early identification of child health problems is desirable. Autistic spectrum disorders (ASDs) are chronic disabling disorders that manifest in early childhood and are characterized by impairments in social interaction and verbal and nonverbal communication as well as by a restricted repertoire of interests, activities, and behaviors. Because ASDs currently cannot be cured, aims of early detection lay in reducing impairments and disabilities (tertiary prevention). However, positive effects of particularly early intervention have been demonstrated in terms of IQ gains and improvements in social and communicative skills. Also, earlier identification of children with the disorder may help to reduce family stress (because parents can be educated about the disorder), speed up referral to specialized education, and promote discussion of family planning issues to inform parents about the genetic risks of having another child with ASD. Siblings of a child with autism have an increased risk of developing ASD. The risk of developing ASD in siblings is estimated at 3% to 7%. Parents of children with ASD consider early detection of the disorder to be important. Parental satisfaction was demonstrated to be related to the length of time between the first concerns and final diagnosis of ASD. Early detection can be accomplished by screening. The effectiveness of screening in terms of the willingness of parents to follow the professional advice if the screening result is positive is ignored in literature. In the case of screening children, the compliance of parents is important because they act as gatekeepers to referral. About 25% of parents do not follow the advice of having their child...
examined after a developmental screening.\textsuperscript{8,9} Similar percentages of parents do not follow treatment recommendations for their developmentally handicapped child\textsuperscript{10} or their child with language delay.\textsuperscript{11} Little is known about the factors that influence compliance with professional advice after screening.

As part of a research program studying the effects and benefits of early screening for ASD, the purpose of the current study was to examine demographic, screening-related, and child-specific factors associated with the attitude of parents after their child had been screened to assess problems in social development and the willingness of parents to accept the advice of follow-up evaluation. Screening was performed in 2 stages. Screen 1 was done at population level by physicians at well-baby clinics (population screening). Children at screen 2 had first failed the population screening (screen 1) or were identified through surveillance by physicians at well-baby clinics because of suspected problems in the children’s social development.

**METHODS**

**SCREEN 1 (POPULATION SCREENING)**

Screen 1 refers to the screening of a total population of 31,724 children aged 14 to 15 months (population screening) in a geographically defined area, the province of Utrecht, the Netherlands. This area is situated in the middle of the country and mainly has an urban character. Physicians at all well-baby clinics within the area used the 4-item Early Screening of Autistic Traits (ESAT) scale.\textsuperscript{12} Screening items refer to social development and play behavior and were asked of the parents. Although attendance of well-baby clinics is not compulsory, the attendance in the first year is as high as 98%, with an average of 6 visits in the first year.\textsuperscript{13} Parents were informed in advance of the screening program by means of a brochure and a poster in the waiting rooms of the well-baby clinics. The brochure and the screening test were also available in English, Arabic, and Turkish languages. As screen 1 would detect general problems in social development instead of ASD specifically, we communicated to parents that the screening was aimed to identify problems in social development. In our pilot study, 138 (90%) of 153 children with autism were identified with the 4-item ESAT scale. No typically developing children were identified (n=478); however, the instrument detected 14 (19%) of 76 children diagnosed with attention-deficit/hyperactivity disorder.\textsuperscript{12}

The parent had to give verbal consent to participate in the screening procedure. The completed 4-item ESAT scale list was sent to our department with minimal information regarding personal identification (first letter of the surname of the child, sex, birth date, and date of screening) to safeguard patient privacy. Parents were advised by the physician to continue with the screening procedure if they answered at least 1 of 4 items negatively. By signing a response card, the parents of screen-positive children gave their informed consent to be contacted by our research team. During a telephone call, we explained the procedure of the second screening to the parents. Further information on the population screening study can be found elsewhere.\textsuperscript{14}

**SCREEN 2**

Children who scored positive at screen 1 (population screening) and whose parents did comply (n=255) (Figure) and children aged up to 36 months identified by surveillance (n=109) underwent screen 2. Physicians of the same well-baby clinics who cooperated with the population screening study could identify children who in their opinion would need further assessment. The identification of these children is quite different from the formal referral to our clinic when problems are generally more evident. The average (SD) age at screen 2 was 16 (2) months for children recruited by the population screening and 27 (6) months for the referred group.

Screen 2 consisted of the 14-item ESAT scale\textsuperscript{13} and was done at a home visit by an experienced psychologist (C.D.) of our research team. Items refer to social development and play behavior. Also, the cognitive development of the child was examined by the Mullen Scales of Early Learning.\textsuperscript{15} Information on the 14-item ESAT scale was provided by the parents as well as by observation of the child. Children who failed at least 3 items of the 14-item ESAT scale were considered screen positive. Parents of screen-positive children were advised to allow their children to undergo a clinical evaluation at the Department of Child Psychiatry, University Medical Center Utrecht, because of the problems that were identified in social development. The results of screen 2 were discussed with the parent immediately after the screening or by a telephone call within 2 weeks after the home visit. Also, a written report was sent to parents. If parents refused to participate in further clinical evaluations, we asked whether we could contact them again for follow-up measurements.

Early-compliant parents were defined as those following the advice to have a clinical evaluation performed within 6 months after screen 2. As there were no waiting lists when the project was performed, a delay between screen 2 and the clinical evaluation could be attributed to the attitude of parents or the practical obstacles they experienced. Late-compliant parents were defined as parents who sought clinical evaluation for their child at our clinic at least 6 months after screen 2. Noncompliant parents were defined as those who did not comply with our follow-up evaluations.

**CLINICAL EVALUATION**

The parents of screen-positive children at screen 2 were advised to have diagnostic assessments for their children at the University Medical Center Utrecht. Assessments included developmental history, a standardized parental interview with the Autism Diagnostic Interview–Revised,\textsuperscript{16} the Vineland Social-Emotional

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Figure. Flowchart of the screening procedure.
Early Childhood Scales, standardized behavior observation using the Autism Diagnostic Observation Schedule–Generic, pediatric examination, and medical workup. On the basis of all available information, an experienced child psychiatrist (E.D., H.E., or J.K.B.) made a clinical judgment on whether the child was likely to meet Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, Revised criteria of specific categories by about the age of 3.5 years. Children were reevaluated at the average (SD) age of 45 (7) months, and final diagnoses were established according to Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, Revised criteria. Six children could not be traced for reevaluation, and they received preliminary diagnoses made at the first psychiatric assessment when they were an average (SD) age of 23 (6) months.

Results of the clinical evaluation were discussed with the parents by a child psychiatrist (E.D., H.E., or J.K.B). Whenever ASD or any other developmental problems were preliminarily diagnosed, this was communicated with the parents. Parents were offered care as usual, eg, parental education, mediation therapy, and/or day care for the child at a specialized psychiatric or educational unit. The interrater reliability of the clinical diagnosis made by 3 psychiatrists (E.D., H.E., and J.K.B.) was calculated for 2 diagnostic categories, ASD or other than ASD for a subgroup of 38 children. Agreement was reached in 35 (92%) of 38 cases. Agreement corrected for chance was 0.74 (Cohen κ). Diagnostic discrepancies were resolved at a consensus meeting.

The average (SD) age at first clinical evaluation was 24 (9) months for children recruited by the population screening and 31 (5) months for the surveillance group. In total, 141 children (66 children identified through the population screening and 75 identified through surveillance) were clinically evaluated. As a result, 58 children received an ASD diagnosis (19 children from the population screening and 39 from surveillance).

MEASUREMENTS

The following background and descriptive information was collected. Parental education level was measured on an 8-point scale, from level 1 (having finished elementary school only) to level 8 (having at least a university degree). Foreign cultural background was defined as having at least 1 parent who was not born in the Netherlands. For defining living environment, we made a rough division between living in a (small) apartment or living in a house. We further coded whether the child went to professional day care facilities for at least 1 day per week.

The Mullen Scales of Early Learning were used to evaluate the cognitive function of most children. Seventeen children were examined with the Psychoeducational Profile–Revised because of their disruptive and/or noncooperative behavior, as this instrument requires fewer task-taking skills. Six children who were examined in the pilot phase of our study were evaluated with the Dutch version of the Bayley scales.

The cognitive levels of 3 children could not be measured because of too-disruptive behavior.

The Social Communication Questionnaire (SCQ), a parent-completed questionnaire that measures symptoms of autism, was used at reevaluation. The items of the SCQ are very similar to those of the Autism Diagnostic Interview–Revised. A higher score on the SCQ reflects the presence of more autistic symptoms. Finally, we asked parents for their reasons to not (yet) comply with follow-up evaluations.

The study design and screening procedure were approved by the medical ethics review board of the University Medical Center Utrecht.

RESULTS

NONCOMPLIANCE AFTER SCREEN 1

Of 31,724 children screened, 370 (1%) were screen positive. Although most parents agreed to the home visit that was offered when the child scored positive at screen 1, 115 (31%) did not. Of these noncompliant parents, we had no information on the identity of 73 of them or on the reasons for their noncompliance, as they immediately mentioned that they were not interested in further screening; the remaining 42 noncompliant parents had signed informed consent to be called and were no longer anonymous. These parents gave the following reasons for noncompliance during a telephone conversation: the child was already being seen by a specialist because of medical problems (n=17 [15%]); the parents revised their first answer (n=12 [10%]); the parents were not worried about the positive screening result and found a home visit too much hassle (n=10 [9%]); they lacked time (2 children); or found their child too young to participate in a screening procedure (1 child).

Sixty-four (56%) of 115 children of noncompliant parents were boys and 169 (66%) of 255 children of compliant parents were boys. Children of noncompliant parents failed fewer items of the 4-item ESAT scale (mean [SD] score, 1.07 [0.56]) than children of compliant parents (mean [SD] score, 1.22 [0.55]) (t358=−2.32; P=.02).

NONCOMPLIANCE AFTER SCREEN 2

Three groups of parents were distinguished: an early-compliant group (117 [68%] of 173 parents), a non-compliant group (32 [18%] of 173 parents), and a late-compliant group (24 [14%] of 173 parents). These groups are compared here. The data are shown in the table.

Comparison of Early-Compliant and Noncompliant Groups in Screen 2

Fathers were more highly educated in the noncompliant group than in the early-compliant group (t175=−2.44; P=.02). Also, more children in the noncompliant group were recruited by the population screening (χ2=10.85; P=.001). These children were younger than those of the compliant parents (t175=4.14; P<.001). Children of noncompliant parents appeared to have a lower risk of ASD, reflected by lower scores on the 14-item ESAT scale (t175=2.91; P=.004), lower scores on the SCQ at follow-up measurement (t175=2.82; P=.007), and higher cognitive levels (t175=−6.08; P<.001). The percentage of items on the 14-item ESAT scale that the parent and expert agreed on was calculated for each child. Stronger agreement on the 14-item ESAT scale between the parent and expert was found in noncompliant parents (t175=2.69; P=.009). There were no group differences for other variables.
Children of late-compliant parents were clinically evaluated 16 months after screen 2 (mean [SD] referral time, 15.8 [9.4] months), in contrast to 3 months for early-compliant parents (mean [SD] referral time, 2.6 [1.4] months). The percentage of children diagnosed with ASD was similar in the 2 groups (48 [41%] of 117 children for early compliance and 56 [42%] of 24 children for late compliance). Children of late-compliant parents were more often recruited by the population screening (χ² = 17.20; P = .001) and were younger at screen 2 (t₁₁₀ = 5.07; P = .001).

Comparison of Early-Compliant and Late-Compliant Groups in Screen 2

Parents wanted to await further development (14 [58%] for late compliance and 10 [31%] for noncompliance), they had personal and/or practical reasons (5 [21%] for late compliance and 7 [22%] for noncompliance), or the child was already seen by a medical specialist (5 [21%] for late compliance and 6 [19%] for noncompliance). Parents of 2 children in the noncompliant group were afraid of the stigmatizing effects of psychiatric assessments. We do not know the reasons for noncompliance that 4 parents in the noncompliant group had.

Comparison of Late-Compliant and Noncompliant Groups in Screen 2

Girls were overrepresented in the noncompliant group as compared with the late-compliant group (χ² = 4.37; P = .04). Lower scores on the SCQ (t₁₁₅ = 2.59; P = .014) and higher cognitive levels were found in the noncompliant group (t₁₁₅ = 3.55; P = .001). For other variables, no group differences were found.

The following reasons for late compliance and noncompliance were given. Parents wanted to await further development (14 [58%] for late compliance and 10 [31%] for noncompliance), they had personal and/or practical reasons (5 [21%] for late compliance and 7 [22%] for noncompliance), or the child was already seen by a medical specialist (5 [21%] for late compliance and 6 [19%] for noncompliance). Parents of 2 children in the noncompliant group were afraid of the stigmatizing effects of psychiatric assessments. We do not know the reasons for noncompliance that 4 parents in the noncompliant group had.

Parental compliance with professional advice was examined after screening toddlers to assess problems in social development. Screening was done in 2 stages. Screen 1 was performed at population level at age 14 to 15 months. Screen 2 refers to a more extensive screening with the 14-item ESAT scale aimed for children who scored positive at screen 1 (population screening) or for children identified through surveillance by physicians at well-baby clinics.

Nearly a third of the parents did not comply with the advice given at both screening moments (115 parents [31%] at screen 1 and 56 [32%] at screen 2). These percentages are similar to those reported in other studies of parental compliance after developmental screening.⁸,⁹

Children of noncompliant parents after screen 1 had lower screening scores and therefore may have been at lower risk for problems in social development and ASD specifically. Owing to the study design, we have no information on most of these noncompliant families and are unable to draw conclusions as to whether the parental decision not to comply was justified, ie, because the child had a false-positive screen. As 17 (15%) of the parents who did disclose their identity indicated that their child was already being seen by a medical professional, at least part of the noncompliance after the population screening seems to be well motivated and understandable.
After screen 2, we found not only noncompliant parents but also late-compliant parents, each having different characteristics. We found fathers to be more highly educated in the noncompliant group than in the late-compliant group. This finding is difficult to interpret and seems to contradict the literature. Haines et al. found parents of low-income households and manual social class households to be less inclined to seek help for their child with psychological problems. Possibly, more highly educated fathers are more skeptical about assessments after screening when in their view no (serious) problems exist, whereas they are relatively quick to seek help when they identify problems in their children themselves. It can also be hypothesized that the more subtle problems found in the noncompliant group, together with the children’s higher cognitive skills, may have masked deficits and misled parents into thinking nothing is wrong. The high agreement between the parent and expert on the 14-item ESAT scale score that we found in the noncompliant group is interesting. It seems that parents do agree with the expert on specific aspects of social development of their children who are not (yet) well developed. Disagreement appears with the evaluation of these identified problems and the need to act on them with a clinical evaluation for their children. The overrepresentation of girls that we found in the noncompliant group seems in line with findings by Haines et al., who report that 63.5% of parents seek help for psychological problems for their boys in contrast to 48.4% of parents of girls.

Children in the late-compliant and noncompliant groups were younger (about 17 and 18 months of age) than children of early-compliant parents (about 23 months of age). Compliance may be compromised by the early age of screening because ASD-related problems are still difficult to recognize. By definition, some symptoms of ASD should be present before age 3 years. Indeed, symptoms have been documented in children as young as 8 to 12 months based on analysis of family home movies. However, symptoms and developmental problems progress with age, which means that most children with ASD will have more and clearer clinical manifestations after age 18 months than before.

Children of late-compliant parents had a risk of developing ASD similar to that of children of early-compliant parents, expressed by comparable percentages of ASD diagnoses at follow-up evaluation. The positive outcome of screening seems to have raised the awareness of late-compliant parents to potential problems of their children, perhaps prompting them to monitor the developmental progress of their children. These parents may have needed more time to face and accept the possibility that their children have a developmental disorder.

We have no way of knowing whether the noncompliant parents were right in their decision not to comply with clinical evaluation of their children. Follow-up of these children and case finding in the entire population originally screened are necessary to draw conclusions. Owing to privacy legislation, we do not have follow-up information on the children of parents who did not comply, and we do not know whether these parents had sought help elsewhere.

Our findings regarding compliance and noncompliance should be interpreted in the context of our study design. Although we told parents that we hoped to identify early problems in social development, we did not disclose that the aim of the study was to identify ASD. This was in accordance with the general policy of developmental surveillance in the Netherlands, which emphasizes that while many children may have transient and minor developmental problems, only a few will have persistent and serious problems. Our approach was consistent with the properties of our screening instrument, which was developed to have maximal sensitivity for ASD at the cost of identifying developmental problems other than ASD. We did not wish to burden parents with information on ASD and make them overly anxious or embarrassed. Nonetheless, it can be questioned whether the parents were able to make informed decisions in terms of having had sufficient knowledge about the advantages and disadvantages of all possible courses of action.

We found the route by which children came in for screening (via either population screening or surveillance) to be highly related to the compliance rate. Far more noncompliance and late compliance were found in the population screening group. It is evident that non-compliance lowers the effectiveness of screening. However, the same holds for late compliance, as it jeopardizes the intention of very early detection of problems. While children of our population screening were 11 months younger than the surveillance group at screen 2, the age difference was reduced to 7 months at the time of clinical evaluation. Although in individual cases it can still be very important that problems are identified as early as possible, the necessity of implementing a screening routine for ASD on the population level can be questioned. Our findings favor surveillance methods (case finding), defined as the ongoing process of monitoring the development of the child, with screening as a second step in identified children. This is all the more so as a higher percentage of children received an ASD diagnosis within the surveillance group (39 [59%] of 66 children evaluated) than within the population screening group (19 [25%] of 75 children evaluated). Evidently, the earlier age at population screening will have also contributed to the lower detection rate.

To conclude, a proportion of parents need time after screening for problems in social development in their toddlers. Study results suggest higher effectiveness of surveillance over population screening to identify children who need further clinical evaluation. Greater as well as faster compliance in parents of children who were first identified by surveillance were found. Screening may well be applied as a second step within identified children.

Accepted for Publication: December 4, 2006.

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Financial Disclosure: None reported.

Funding/Support: This study was supported by grants 940-38-045 and 940-38-014 from the Chronic Disease Program, grant 28.3000–2 from the Praeventiefonds-ZONMW, and grants from the Netherlands Organisation for Scientific Research, the Dutch Ministry of Health, Welfare, and Culture, Cure Autism Now, and the Korczak Foundation.

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