Advancing early detection of autism spectrum disorder by applying an integrated two-stage screening approach

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Background: Few field trials exist on the impact of implementing guidelines for the early detection of autism spectrum disorders (ASD). The aims of the present study were to develop and evaluate a clinically relevant integrated early detection programme based on the two-stage screening approach of Filipek et al. (1999), and to expand the evidence base for this approach. Methods: The integrated early detection programme encompassed: 1) training relevant professionals to recognise early signs of autism and to use the Early Screening of Autistic Traits Questionnaire (ESAT; Dietz, Swinkels et al., 2006; Swinkels, van Daalen, van Engeland, & Buitelaar, 2006), 2) using a specific referral protocol, and 3) building a multidisciplinary diagnostic team. The programme was evaluated in a controlled study involving children in two regions (N = 2793, range 0–11 years). The main outcome variables were a difference in mean age at ASD diagnosis and a difference in the proportion of children diagnosed before 36 months. Results: ASD was diagnosed 21 months (95% CI 9.6, 32.4) earlier in the experimental region than in the control region during the follow-up period, with the mean age at ASD diagnosis decreasing by 19.5 months (95% CI 10.5, 28.5) from baseline in the experimental region. Children from the experimental region were 9.4 times (95% CI 2.1, 41.3) more likely than children from the control region to be diagnosed before age 36 months after correction for baseline measurements. Most of these early diagnosed children had narrowly defined autism with mental retardation. Conclusions: The integrated early detection programme appears to be clinically relevant and led to the earlier detection of ASD, mainly in children with a low IQ. Keywords: Early detection, autism spectrum disorder, ESAT, screening, guidelines, implementation. Abbreviations: ASD: autism spectrum disorder; ESAT: Early Screening of Autistic Traits Questionnaire.

Autism and related conditions develop before 3 years of age, with most affected children showing signs of abnormality in the first year of life (Chawarska & Volkmar, 2005). Although many parents of children with autism spectrum disorders (ASD) are concerned about the social and communicative development of their children at an early age (Howlin & Asgharian, 1999), ASD generally remains undiagnosed until late preschool years or thereafter (Filipek et al., 1999). While progress has been made in the earlier identification of children with ASD (Charman & Baird, 2002), there is still a difference between age at diagnosis in research-based screening projects versus age at diagnosis in daily clinical practice (Holzer et al., 2006).

The availability of educational and behavioural interventions for children with ASD (see overview National Research Council, 2001) emphasises the importance of timely diagnosis. Although there is currently inadequate empirical evidence to show that earlier intervention is more beneficial than later intervention, most clinicians consider early intervention important (Filipek et al., 1999) as it may provide a better long-term outcome for children and their families. Moreover, early detection facilitates educational planning and accurate monitoring of children's development (National Research Council, 2001). It deters parents from 'shopping for health care' and offers an opportunity to provide parents with realistic information about the genetic risk of having another child with ASD. It may also be cost effective, because late identification means that expensive treatment programmes may be needed for secondary behaviour problems that could have been prevented or at least treated more effectively and at lower costs during the preschool years (Barnett & Escobar, 1989).

Screening in primary care is a sensitive issue because false positive results may cause parents unnecessary anxiety or may result in a cascade of avoidable further assessments, while false negative results may falsely reassure parents and delay an early start of interventions. Two models have been proposed for the early detection of ASD. The first...
model is based on a systematic population screening in which autism-specific screens are applied to all children at certain ages in addition to routine developmental surveillance (Johnson, Myers, & the Council on Children with Disabilities, 2007). Williams and Brayne (2006) reviewed the evidence for this approach, using the UK National Screening Committee’s (2000) framework, and concluded that screening at a population level could not be recommended because (a) few longitudinal studies describing the natural history of ASD include data on early identified cases, (b) no screening test is available that fully meets validation criteria, and (c) there is insufficient evidence regarding the effectiveness of interventions. The second model of early detection is based on a two-stage screening approach in which a specific screening instrument for ASD is applied only to those children who are found to have a deviant developmental path during routine developmental surveillance (Filipek et al., 1999).

Even though primary care is well organised in many countries, generally systematic attention is not paid to the early screening and detection of ASD. Moreover, knowledge of the early signs of autism, screening and diagnostic assessment procedures, and treatment possibilities is inconsistent amongst primary care providers (Dosreis, Weiner, Johnson, & Newschaffer, 2006; Heidgerken, Geffken, Modi, & Frakey, 2005).

To our knowledge, only a few, uncontrolled, studies have reported on specific approaches for the implementation of guidelines for the early identification of ASD. Chakrabarti, Haubus, Dugmore, Orgill, and Devine (2005) described a stepwise screening and diagnosis programme with four stages (from universal screening to in-depth assessment of children who were strongly suspected to have ASD) in which primary care workers and a multidisciplinary Child Development Team played an important role. They reported an average age at initial diagnosis of 41 months (range 21–78 months). The method used in California and reported by Koegel, Nefdt, Koegel, Bruinsma, and Fredeen (2006) was based on raising community awareness of early signs of autism. Before the intervention, children did not receive services until age 48–60 months old, whereas after the intervention, the average age of children screened and later diagnosed as having autism was 29 months (range 16–55 months). In another study, Holzer et al. (2006) developed practice parameters for ASD and circulated these to all child and adolescent psychiatrists practising in the Swiss canton of Vaud. In addition, early child professionals were familiarised with early signs of autism and a specific screening tool, the Checklist for Autism in Toddlers (Baron-Cohen, Allen, & Gillberg, 1992). Results indicated a significant decrease of about 17 months in the mean age at diagnosis, from 72 to 53 months; however, this effect was not sustained 2 years after implementation of the practice parameters. These three studies all used an implementation strategy roughly in line with Filipek et al.’s (1999) guidelines and reported promising results. However, the lack of a control group meant that it is not known whether the interventions per se led to the earlier detection of ASD. Without a control group other a-specific factors (e.g., media items on autism) cannot be ruled out. For this reason, the present study included a control group.

The current study is part of a larger clinical and research project, the DIANE project (Diagnosis and Intervention study on Autism in the NETHERlands), and must be viewed in the context of the local health care setting. In the Netherlands, as in several other European countries, health care for young children is delivered through community well-baby clinics that provide routine developmental surveillance and vaccinations. Babies come to the clinic 9 times in the first 15 months of life and thereafter 4 times until they are 4 years old (Laurent de Angulo, Brouwers-de Jong, & Bulk, 2005). Well-baby clinics are free of charge and are attended by virtually all parents in their child’s first year of life and by about 80% until their child is 4 years old (Frenken, 2005). Since 1994, specific independent infant–toddler development teams, which work in close collaboration with doctors and nurses of the well-baby clinics, have also been set up. These multidisciplinary teams provide parents who may have specific concerns about their child’s development with easily accessible first-line care. They also carry out case-management, investigate children’s developmental problems in general (but provide no specific diagnostic assessments), and when necessary refer children to secondary or tertiary health care services for diagnostic assessment and treatment.

The first aim of the current study was to develop a clinically relevant, integrated early detection programme based on the two-stage screening approach suggested by Filipek et al. (1999). This programme uses the 14-item Early Screening of Autistic Traits Questionnaire (ESAT; Dietz et al., 2006; Swinkels et al., 2006) to screen infants and toddlers for ASD. The ESAT (14 items) is an empirically based screening instrument for use in high-risk populations. The second aim was to evaluate this screening approach, to determine whether ASD could be detected earlier, preferably before 36 months.

Methods

Study design and setting

A study in two regions (experimental vs. control) at baseline and during programme implementation (follow-up) was designed to evaluate the effect of the
integrated early detection programme. The baseline period was defined as January to December 2003 and the follow-up period as January 2004 to December 2006. The experimental and control regions are both mainly rural areas, with middle-sized cities and smaller villages with relatively few immigrants. The experimental region consisted of the province of Gelderland in the east of the Netherlands and smaller parts of four adjacent provinces. The control region consisted of the provinces of Friesland, Groningen, and Drenthe in the north of the Netherlands. Data were collected by expert centres with extensive expertise in ASD in both regions: by Karakter Child and Adolescent Psychiatry University Centre Nijmegen in the experimental region and the Child and Adolescent Psychiatry Unit of the University Medical Centre Groningen and the Autism Team Northern Netherlands in the control region. While other institutions in both regions assess and diagnose school-aged children with ASD, preschool children at risk of ASD are almost exclusively assessed at these expert centres. For reasons of feasibility, it was not possible to randomise the areas included in the experimental and control regions.

In general, in the Netherlands children at all ages can be referred to specific psychiatric assessment centres by general practitioners, medical specialists (e.g., neurologist or paediatrician), by professionals from other mental health care services or from institutions for language development, and by primary care workers (i.e., doctors of well-baby clinics and members of the specific infant–toddler development teams, see introduction).

Participants

The sample included 2793 participants who were referred for clinical psychiatric evaluation in the baseline and follow-up years in the experimental and control regions and was limited to children younger than 12 years at referral. Very few children living outside the experimental or control regions were referred to the expert centres participating in this study. Of the children from the experimental region who were not diagnosed with ASD, about 60% had externalising disorders, about 13% had internalising disorders, and about 27% had other disorders. In the control region, about 78% of the non-ASD cases had externalising disorders, about 12% of them had internalising disorders, and a further 10% had other disorders. These diagnoses did not change substantially throughout the study period in either region. See Table 1 for more demographics.

Integrated early detection programme

The integrated early detection programme is known as the ‘DIANE project’ and consisted of three elements: (a) training of primary care workers to recognise early signs of autism and to use the ESAT (Dietz et al., 2006; Swinkels et al., 2006), (b) use of a specially designed referral protocol, and (c) formation of a multidisciplinary team.

Training.

In order to raise awareness and to familiarise healthcare professionals with the early signs of autism and with the ESAT, an opinion leader (SS) developed and organised training sessions from autumn 2003 until the end of 2006 in the experimental region. Of 39 training sessions delivered, 22 focused specifically on training primary care workers, because these professionals are most likely to refer young children at high risk of ASD and were considered the main target group. Training was given to relatively small groups of 10 to 30 professionals with interactive participation through discussions and questions. Primary care umbrella organisations in the region made attendance compulsory for primary care workers, who were awarded with CME (Continuing Medical Education) points.

All training sessions lasted about 2 hours and were usually held in evening hours. The introduction included: (a) review of ASD criteria (DSM-IV; APA, 2000), (b) prevalence rates, (c) explanatory models of ASD, and (d) information on early intervention and the importance of early detection. The main part of the training session included a review of early signs of autism and all ESAT items, illustrated by video clips showing children with abnormal or absent behaviour as well as video clips of typically developing children, to clarify what could be expected of a young child at a certain age. Lastly, the referral protocol was outlined (see below). Larger groups (n = 50 to >80) of other interested professionals (such as general practitioners, paediatricians, speech and language therapists) attended other additional lectures (17 in total) with a lower level of interactive training. Information about the DIANE project could be obtained from brochures or the Radboud University/Karakter website and the ESAT could be downloaded free of charge.

Referral protocol.

Professionals wishing to refer a child younger than 36 months for assessment of ASD were first required to complete the ESAT (with the assistance of the parents). Children that screened positive with the ESAT were always invited for further assessment. If a child screened negative with the ESAT, the referring professional had to provide additional information showing the child to be at high risk. Untrained professionals who had heard about the assessment possibilities for young children could also refer children for further investigation but the ESAT still had to be completed. In some cases, members of our team completed the ESAT with the parents’ assistance on the phone. Within 2 weeks of referral, parents were invited to bring their child for assessment. They also received questionnaires to be completed, including a questionnaire about personal/family situation and the child’s development, and the Child Behaviour Checklist (Achenbach & Rescorla, 2000).

Multidisciplinary diagnostic team.

A multidisciplinary team for infant psychiatry that is specialised in the early diagnosis of ASD was set up at the Karakter Child and Adolescent Psychiatry University Centre. This team consisted of two experienced child-psychotherapists, four psychologists, and a psycho-diagnostic employee. Each child referred was assigned to a case-manager, a psychologist. A coordinator, a manager, and two administrative assistants supported the team. The psychologists had been trained to administer and
score the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000) and/or the Autism Diagnostic Interview-Revised (ADI-R; Lord, Rutter, & Le Couteur, 1994). More information about the integrated early detection programme can be obtained from the first author.

Measures and procedure

For this study, we used measures that are routinely included in the screening and diagnostic procedures for children with suspected ASD.

Screening. In the experimental region, children younger than 36 months who had been referred for possible ASD were screened with the ESAT (Dietz et al., 2006; Swinkels et al., 2006). The ESAT consists of 14 easy-to-administer items measuring early social-communication skills, play, and restricted and repetitive behaviours, answered with yes or no. Children who failed three or more items are considered to be at risk for ASD.

Clinical diagnosis. The diagnostic protocol usually included a psychiatric evaluation, administration of the ADOS and/or the ADI-R, and assessment of cognitive and language skills. IQ scores were assessed with age-appropriate psychometric tests, most frequently the Mullen Scales of Early Learning (Mullen, 1995), the Psycho-Educational Profile-Revised (Schopler, Reichler, Bashford, Lansing, & Marcus, 1990), the Snijders–Oomen Nonverbal Intelligence Test (Tellegen, Winkel, Wijnberg-Williams, & Laros, 1998), and the Wechsler tests (Wechsler, 1997, 2002). In addition, standardised parent–child play was observed in most preschool children. Certified child psychologists or psychiatrists performed and administered all assessments. Shortly after diagnosis, specific care was organised for all children and their parents. Preschool children diagnosed with ASD were referred for further neuro-paediatric screening.

Control region

In 2005 and 2006, the Autism Team Northern Netherlands organised five training sessions (lasting about 3 hours each) to instruct primary care workers in the control region about the early signs of autism; however, systematic screening for ASD with a specific screening instrument and use of an explicit referral protocol were not mentioned. There was ample time for questions and discussion. These lectures were attended by about 20–50 primary care workers. As in the experimental region, a multidisciplinary team assessed children referred for possible ASD to one of the specialist centres in the control region using parent–child play observation, the ADOS, ADI-R, cognitive, or language skills measures, as indicated.

Data analysis

Chi-square statistics were used to compare the characteristics of the participants in both regions at baseline and during follow-up and to investigate differences in the distribution of IQ (<70, 70–89, ≥90) by age, type of ASD diagnosis, region, and time (baseline or during follow-up). The primary outcome variable to identify an effect of the integrated early detection programme was a change from baseline in mean age at ASD diagnosis between the experimental and control regions. Univariate analyses of covariance were used to analyse the variation in age at ASD diagnosis. An interaction effect of region and measurement time was specified to identify the impact of the programme in the experimental region compared with the control region. Based on clinical impression, a change in age at diagnosis of ≥12 months was considered to be clinically meaningful. To detect this difference, power calculations showed that two ASD-diagnosed groups of at least n = 221 were needed (estimated SD = 45 months at baseline, p = .05, power = .80). The secondary outcome variable was the number of children diagnosed with ASD before the age of 36 months as a percentage of the total number of children newly diagnosed with ASD (new cases). Binary logistic regression modelling was used to analyse the secondary outcome variable, with type of ASD diagnosis, sex, and IQ as covariates.

Results

Comparison of demographics

Comparison between the two regions showed that age at referral was differently distributed in both periods (baseline: Χ²(3) = 17.93, p < .001; follow-up: Χ²(3) = 184.79, p < .001). Significantly more children aged 0–2 years were referred for evaluation in the experimental region than in the control region. See Figure 1 for the (change in) percentage of children referred before 36 months of age by region and period, with three time points during follow-up.

Within the experimental region, significantly more children aged 0–2 years were referred in the follow-up period compared to the baseline period (Χ²(3) = 17.96, p < .001), and significantly more children of this age than of other ages were diagnosed with ASD (Table 1). In contrast, within the control region, age at referral was not different between the two periods.

Of the children younger than 36 months referred for an ASD evaluation in the experimental region in the follow-up period, about two-thirds were diag-
nosed with ASD and one-third was not diagnosed with ASD but instead had other developmental disorders that needed professional help. Only two of these children functioned normally (for more details see Oosterling et al., 2009).

As expected, and as shown in Figure 2, more referrals of children aged 0–2 years in the experimental region were made by primary care workers (professionals exposed to the training programme) during the follow-up period than at baseline. However, in the control region other medical professionals remained the core referral group.

Change in mean age at ASD diagnosis

The primary outcome variable, the mean age at ASD diagnosis, dropped dramatically in the follow-up period in the experimental region but remained relatively stable in the control region (see Table 2). There was a significant difference of −21.0 months (95% Confidence Interval = 32.4 – 9.6 months, p < .001) between the experimental and control regions from baseline to follow-up. This was considered a clinically meaningful result as the change in mean age was ≥12 months. Corrected for potential confounders, such as type of ASD diagnosis, sex, and IQ, the difference was −16.5 months (95% Confidence Interval = 28.6 – 4.3 months, p < .01).

Change in proportion of children diagnosed before 36 months

The secondary outcome variable, the proportion of children diagnosed before 36 months, was similar in the two regions at baseline (see Table 3). However, during the follow-up period in the experimental region, the proportion of children diagnosed with ASD before 36 months increased by 22.4%, whereas in the control region this proportion decreased by 1.7% (see Table 3). The binary logistic regression model with an interaction term defined as region · period showed that for the experimental region the Odds Ratio of the follow-up versus baseline period was significantly higher than for the control region (9.4, 95% Confidence Interval = 2.1–41.3, p < .01). Corrected for potential confounders such as type of ASD diagnosis, sex, and IQ, this Odds Ratio was 9.0 (95% Confidence Interval = 1.3–61.4, p < .05).

IQ, age, and ASD diagnoses

The level of cognitive functioning within a certain type of ASD diagnosis (autism or non-autism ASD) was similar in the two regions at both times and across age groups (see Table 4), as all X²-values were not significant (p > .05). Moreover, of the children aged 0–2 years referred for clinical assessment during the follow-up period in the experimental region and diagnosed with ASD, 67% (90/135) had autism and 33% (45/135) had non-autism ASD. Most

### Table 1 Within and between region comparison of the referral rates by age, sex, and ASD diagnosis

<table>
<thead>
<tr>
<th>Age at referral</th>
<th>Experimental region</th>
<th>Control region</th>
<th>Experimental region</th>
<th>Control region</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Baseline (n = 260)</td>
<td>Follow-up (n = 967)</td>
<td>Baseline (n = 238)</td>
<td>Follow-up (n = 1328)</td>
</tr>
<tr>
<td>Preschool 0–2 years</td>
<td>n</td>
<td>%</td>
<td>n</td>
<td>%</td>
</tr>
<tr>
<td>ASD-diagnosis</td>
<td>11</td>
<td>33.3</td>
<td>138</td>
<td>58.7</td>
</tr>
<tr>
<td>Male</td>
<td>26</td>
<td>78.8</td>
<td>177</td>
<td>75.3</td>
</tr>
<tr>
<td>3 years</td>
<td>14</td>
<td>41.2</td>
<td>46</td>
<td>26.2</td>
</tr>
<tr>
<td>ASD-diagnosis Male</td>
<td>28</td>
<td>82.4</td>
<td>88</td>
<td>81.5</td>
</tr>
<tr>
<td>School-age 4–7 years</td>
<td>82</td>
<td>31.5</td>
<td>298</td>
<td>30.8</td>
</tr>
<tr>
<td>ASD-diagnosis Male</td>
<td>59</td>
<td>72.0</td>
<td>222</td>
<td>74.5</td>
</tr>
<tr>
<td>8–11 years</td>
<td>111</td>
<td>42.7</td>
<td>326</td>
<td>33.7</td>
</tr>
<tr>
<td>ASD-diagnosis Male</td>
<td>89</td>
<td>80.2</td>
<td>235</td>
<td>72.1</td>
</tr>
</tbody>
</table>

*p < .01; ns = not significant

### Figure 2 Percentage of children referred before 36 months of age by type of referrer, period and region.

*Note. Missing value = 4; PCW = Primary Care Worker (trained type of referrer in follow-up period in experimental region); GP = General Practitioner; MS = Medical Specialist; MHCS = Mental Health Care Service; ILD = Institution for Language Development
children in this specific age group and diagnosed with autism had mental retardation (IQ < 70; 75–87%), whereas in the same age group children with non-autism ASD were higher functioning ($C^2(2) = 45.95, p < .001$). The same results were found for children aged 0–2 years referred in the baseline period in the experimental region ($C^2(2) = 6.52, p < .05$), and in the follow-up period in the control region ($X^2(2) = 6.77, p < .05$), but not in the baseline period in the control region, which was probably due to the limited size of this sub-sample ($X^2(2) = 2.63, p > .05$).

### Discussion
This study describes an integrated programme, based on the two stage-screening approach of Filipek et al. (1999), for the early detection of ASD. During follow-up in the experimental region, the mean age at ASD diagnosis was significantly, and meaningfully, lower than at baseline or compared with the control region. Moreover, the proportion of ASD diagnoses made in children younger than 3 years increased in the experimental region compared with the control region, and most of the referrals for ASD evaluation

#### Table 2 (Difference in) mean age in months at ASD diagnosis

<table>
<thead>
<tr>
<th></th>
<th>Baseline mean age (SD)</th>
<th>Follow-up mean age (SD)</th>
<th>Difference in mean age (95%-CI)</th>
<th>p</th>
</tr>
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<tbody>
<tr>
<td>Experimental region</td>
<td></td>
<td></td>
<td></td>
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</tr>
<tr>
<td>mean age</td>
<td>82.9 (36.4)</td>
<td>63.5 (36.9)</td>
<td>−19.5 (−28.5−10.5)</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>Control region</td>
<td></td>
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<tr>
<td>mean age</td>
<td>82.9 (29.7)</td>
<td>84.4 (31.5)</td>
<td>1.5 (−5.7−8.8)</td>
<td>.68</td>
</tr>
</tbody>
</table>

**Note.** CI = Confidence Interval

<table>
<thead>
<tr>
<th></th>
<th>Baseline</th>
<th>Follow-up</th>
<th>OR(95%-CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Experimental region</td>
<td>6.3%</td>
<td>28.7%</td>
<td>6.04(2.37–15.39)</td>
</tr>
<tr>
<td>Control region</td>
<td>4.7%</td>
<td>3.0%</td>
<td>0.64 (.20-2.02)</td>
</tr>
</tbody>
</table>

**Note.** CI = Confidence Interval; OR = Odds Ratio

<table>
<thead>
<tr>
<th></th>
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<tr>
<td>mean age</td>
<td>82.9</td>
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Table 2 includes data on the difference in mean age in months at ASD diagnosis by region and study period.

### Table 3 Proportion of children newly diagnosed with ASD younger than 36 months and Odds Ratios for the difference between follow-up and baseline by region

<table>
<thead>
<tr>
<th></th>
<th>Baseline</th>
<th>Follow-up</th>
<th>OR(95%-CI)</th>
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<tr>
<td>Experimental region</td>
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<tr>
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<tr>
<td>mean age</td>
<td>82.9</td>
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**Note.** CI = Confidence Interval; OR = Odds Ratio

Table 3 presents the proportions of children newly diagnosed with ASD younger than 36 months and Odds Ratios for the difference between follow-up and baseline by region.

### Table 4 Intelligence Quotient by age at referral (new cases) and by type of ASD diagnosis, period and region

<table>
<thead>
<tr>
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<th>Follow-up</th>
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<td>Control region</td>
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<tr>
<td>mean age</td>
<td>82.9</td>
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</tbody>
</table>

**Note.** Aut = Autism; Non-Aut ASD = Non-Autism ASD

Table 4 includes intelligence quotients by age at referral (new cases) and by type of ASD diagnosis, period and region.
were made by trained primary care workers as opposed to non-specific referrers in the control region.

The mean age at ASD diagnosis of 63.5 months found in this study is still quite old compared to the mean age at ASD diagnosis reported in other studies (range 29–53 months; Chakrabarti et al., 2005; Holzer et al., 2006; Koegel et al., 2006). However, differences remain hard to interpret, because results greatly depend on the upper age limits in those studies. Our study included children up to 12 years of age, but other studies were not very explicit about the age ranges included. Nevertheless, convincingly supportive to the effectiveness of the integrated early detection programme described, we found that almost 30% of the children from the experimental region diagnosed with ASD during the follow-up period were younger than 36 months.

Holzer et al. (2006) reported that the improvements in earlier detection achieved with their Practice Parameters programme were not sustained 2 years later. As their programme is comparable to ours in many ways (e.g., based on training of relevant primary care workers, use of a specific screening tool and standardised diagnostic measures), it is probably important to ensure that primary care professionals keep up to date and remain motivated, so that this improvement in early diagnosis of ASD is sustained.

The results of our study suggest that the increase in the number of children diagnosed at an early age is associated with their level of cognitive functioning. In line with previous research, the advantage of stimulating the earlier detection and diagnosis of ASD apparently lowers the age of ASD diagnosis predominantly in low-functioning cases, so that higher-functioning children with ASD are still likely to be detected and diagnosed later. The finding that about 75–87% of the children referred before 36 months with narrowly defined autism had mental retardation (IQ < 70) is consistent with the findings of Baird et al. (2006): they reported IQ rates below 70 in 73% of the cases meeting the narrow definition of childhood autism.

Unexpectedly, and despite the use of comparable diagnostic assessment procedures performed by similarly qualified health professionals, there were diagnostic differences between the two regions. Both at baseline and during the follow-up period relatively more children were diagnosed with autism than with pervasive developmental disorder – not otherwise specified (PDD-NOS) in the experimental region, whereas PDD-NOS was the main diagnosis in the control region. Thus, between regions, there could be differences in assigning some diagnoses in the interface to either autism or PDD-NOS. However, this does not detract from the finding that the diagnosis of ASD occurred earlier after implementation of the integrated early detection programme.

Some aspects concerning the potential large-scale implementation of the integrated early detection programme should be mentioned. In our view, the early detection of ASD is undoubtedly facilitated by a well-functioning primary care system providing general developmental surveillance and easy accessible additional care. However, barriers to the implementation of the programme can be expected at different levels (Dosreis et al., 2006; Grol & Grimshaw, 2003; Holzer et al., 2006; Pinto-Martin, Dunkle, Earls, Fliedner, & Landes, 2005; Zwaigenbaum et al., 2007). At an organisational level, lack of reimbursement and lack of time to administer and interpret the screening instruments may hinder programme implementation. At a professional level, disagreement over 'cookbook' guidelines may be involved, as well as low expectations concerning results, unfamiliarity with screening instruments and procedures, inconsistent knowledge about ASD among primary care providers, and fear of positive results. At a patient level, resistance to asking for help and late or non-compliance may be an obstacle to the early detection and diagnosis of ASD (Dietz, Swinkels, van Daalen, van Engeland, & Buitelaar, 2007).

A limitation of this study is that results cannot be generalised to other regions and countries, given the possible differences in local health care services. Moreover, there may be differences in attitude towards early diagnosis, in healthcare-seeking behaviour, and in referral. Because the outcome data were collected while the programme was ongoing, it has yet to be determined whether the improvements in early diagnosis are sustained in the long term.

Conclusion

In general, the results of this controlled study support those of earlier, uncontrolled, studies showing that the availability of an early identification tool and primary care workers’ knowledge of early signs of ASD, and ongoing involvement in a screening programme can lead to earlier detection, referral, and diagnosis of ASD. If such a programme is to be implemented, then it is essential that there are follow-up services and facilities for children who are at risk of ASD and appropriate autism-specific interventions for parents and children at the ages being screened (Pivalizza, 2007). In our study, if an early diagnosis was made, regardless of the type of diagnosis, all children and their parents were brought into contact with treatment and counselling facilities. Further implementation of Filipek et al.’s (1999) guidelines should be combined with additional research to justify the need for early detection of ASD by validating the evidence-base for early interventions. Special attention should be given to specific risk groups, such as the children of parents who choose not to participate in screening programmes and/or diagnostic assessments for the early diagnosis of ASD or other developmental disorders.
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Key points

• This paper describes an integrated programme to improve the early detection of ASD by translating guidelines as suggested by Filipek et al. (1999) into clinical practice.
• Programme efficacy was tested in a large controlled study involving two regions with baseline and follow-up measurements over 4 years.
• Training primary care workers to recognise early signs of autism and to use an early screening tool for ASD (ESAT) is necessary to achieve earlier detection of ASD.
• This paper is important for advancing health policy regulations and in guiding the development of training relevant professionals.

References


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