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Changes in the frequency and characteristics of children diagnosed with autistic disorder in two Norwegian cohorts: 1992 and 2009

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Abstract

Background: Is the increasing prevalence of autistic disorder (AD) a well-documented trend or merely a reflection of the wider recognition of AD among both the public at large and health care professionals? Data from relevant studies are frequently compromised by comparisons of different sites and different diagnostic methods.
Objectives: To explore changes over time, we reviewed the following: 1) the frequency of AD diagnoses; 2) the characteristics of the diagnosed children; and 3) the ages of the children when initial concerns were addressed and AD diagnoses made.
Method: We compared the case records of children between the ages of 1 and 17 years who were residing in Nordland County, Norway, and who were diagnosed with AD during two different data collection periods: 1992 (Cohort 1) and 2009 (Cohort 2).
Results: In Cohort 1, 28 children were diagnosed with AD; 71 children in Cohort 2 received AD diagnoses. The increase was greatest among children with intelligence quotient (IQ) values of at least 70. The proportion of children with genetic syndromes was around 20% in both cohorts. Median age at AD diagnosis did not differ between the two cohorts (4.5 vs. 5.0 years, respectively). When the two cohorts were combined, children with IQ values of 70 or more without a genetic syndrome and those with IQ values of less than 50 with genetic syndromes were diagnosed at approximately the same age (5.5 and 5.3 years, respectively). Both groups were significantly older at diagnosis as compared with children with IQ values of less than 50 without genetic syndromes (3.5 years).
Conclusions: The increase in the number of children diagnosed with AD is consistent with findings from international studies. Contrary to predictions, the age at diagnosis was not reduced over time. A higher proportion of children with IQ values in the average range in the latter cohort may have contributed to this. A delayed diagnosis of AD among children with genetic syndromes may indicate that early autism symptoms are attributed to the genetic condition. Clinical implications are discussed.

Keywords: autistic disorder; time trend; early diagnosis; genetic syndromes; intellectual disability

Introduction

Prevalence estimates for autistic disorder (AD) (core autism) have varied widely over time, from around 4.7 per 10,000 in studies published between 1966 and 1993 (1) to around 74 per 10,000 in more recent epidemiological studies (2). For the whole autism spectrum, current prevalence estimates exceed 1%. A recent report from the U.S. Centers for Disease Control and Prevention reported that 1 in 68 children received ASD diagnoses (3), although the study methods used in that report have been criticized (4). It is clear that autism is not the very rare condition that it was once considered to be. It is likely that factors such as the broadening of the concept to

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† E. Grindheim has sadly passed away before this paper was finished.

autism spectrum disorder (ASD) (5), the expansion of the diagnostic criteria, the development of services, and the improved awareness of the condition have played major roles in the rise in prevalence estimates (6).

Among the many variables that potentially contributed to changes in prevalence estimates is the recognition that autism can be associated with a range of specific genetic syndromes. However, estimates of comorbidity also vary widely. For example, ASD has been reported in 21% to 50% of boys with fragile X syndrome and in 24% to 60% of individuals with tuberous sclerosis (7). Comorbidity figures for autism among patients with Cornelia de Lange syndrome range from 32% to 62% (8). Among children with Down syndrome, AD was reported in 6%, and ASD was reported in 18% (9); similar figures 8% and 19% respectively, were reported by Moss and colleagues (10). Warner and colleagues (11) reported that the proportion of children with Down syndrome who met the cutoff for autism on the Social Communication Questionnaire (12) was 16.5%.

Earlier diagnosis is another factor that affects prevalence estimates (13). AD can reliably be diagnosed by experienced clinicians in children 3 years old or younger (14). However, the average age at AD diagnosis varies across studies from 3.1 years (15) to 4.8 years (16) to 5.2 years (17) in accordance with sample characteristics and ascertainment methods (18,19). Parents are initially concerned about social development, but they also have worries about general delays and other specific problems (20,21). In addition, many of them have to wait a long time for diagnostic confirmation (22,23).

Geographical factors are also considered important. For example, although recent U.S. figures have suggested rates of more than 1% for ASD (3) and a South Korean study reported prevalence estimates of 0.9% for AD and 2.6% for ASD (24), an epidemiological study in Norway (25) found rates of only 0.14% for AD and 0.5% for ASD.

Some researchers argue that increased prevalence rates are influenced by "diagnostic substitution": in recent years, children with intellectual disability, attention-deficit/hyperactivity disorder, or language disorder have been more likely to be diagnosed with AD or ASD in addition to these other conditions rather than intellectual disability, attention-deficit/hyperactivity disorder, or language disorder alone (6,26,27). Overshadowing has also been described in relation to coexisting attention-deficit/hyperactivity disorder and ASD, which suggests that an initial ADHD diagnosis may be associated with a delayed ASD diagnosis (28). The increased frequency of the occurrence of ASD with other medical, developmental, and behavioral disorders increases the possibility of "over-shadowing" in that all additional problems are attributed to the medical condition (7,28,29).

Finally, terminology and diagnostic criteria have also changed over time. In the past, the term *autistic disorder* was used to describe children who met criteria for core autism according to the criteria presented in the *Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition* (DSM-IV) (30) or the *International Statistical Classification of Diseases and Related Health Problems, 10th Revision* (ICD-10) (31). However, with the recent changes of the DSM-5 classification (5), the *term autism spectrum disorder* is now used to include all children who previously would have been diagnosed as having either AD, Asperger syndrome, atypical autism, or pervasive developmental disorder – not otherwise specified.

Because of the variability in the estimates in different countries and over time, Fombonne (6) has argued that repeated studies using the same methodology and conducted in the same geographical area at different points in time are important to obtain more reliable information about time trends. Two studies from Nordland County in Norway were used in the present study to address this issue by comparing two groups of children diagnosed with AD and residing in the county at the times of data collection. The data were collected 17 years apart using records from the central hospital responsible for the diagnostic assessment and follow-up of all children with AD in the county.

The main focus of the current study was to examine the change in the proportion of children between the ages of 1 and 17 years who had been diagnosed with AD (ICD-10: F84.0/DSM-IV-TR: 299.0) and who were living in Nordland County, Norway, during two separate data collection periods: 1992 and 2009. We also examined differences in child characteristics over time, including intelligence quotient (IQ) values and the presence of genetic syndromes. The referral procedures in Nordland County for children who were suspected of having AD were the same for both years studied, and the children were diagnosed and registered at the same specialized services in the county during both time periods. Health services in Norway are free of charge for all children, and children are routinely offered developmental check-ups at well-baby clinics throughout the first 4 years of life.

The aims of the study were to determine the following: 1) the change in the proportion of children living in Nordland County and registered with an AD diagnosis at the time of the two data collections; 2) the changes in child characteristics with respect to developmental level and genetic conditions; 3) the changes over time in the age at which parents first became concerned about their child's development

and the child's age at diagnostic confirmation; and 4) the associations between the child's characteristics and the ages of first concerns and diagnosis.

In the light of previous studies that have investigated changes in the rates of AD diagnosis, it was predicted that there would be a significantly greater number of children diagnosed with AD in 2009 as compared with 1992. It was also expected that there would be more children with normal range intellectual ability and AD in the 2009 data collection and that the proportion of children with both genetic syndromes and AD would increase. The age of parental first concern and age of diagnosis were expected to be lower in the data collected in 2009 as compared with 1992 as a result of a greater focus on early identification (32,33). The presence of a genetic syndrome was expected to reduce both the age of first concern and the age at AD diagnosis, although children with IQ values in the normal to high range were predicted to be older when parents reported their first concern as well as when the diagnosis occurred.

Method

Participants

In 1992, a retrospective record-based study was conducted including all children between 1 and 17 years old who had been diagnosed with AD and who were living in Nordland County at the time of the data collection (34); this group will hereafter be referred to as *Cohort 1*. Case records from the hospital were used to retrieve information related to birth, medical conditions, and diagnostic procedures. A replication of the study that used the same method to retrieve information was carried out in 2009 to result in two non-overlapping cohorts; the group identified in that study will hereafter be called *Cohort 2*. In both studies, all children had been assessed and diagnosed by the hospital's Autism Team in collaboration with the Paediatric Ward. Cohort 1 comprised 28 children born between January 1, 1975, and December 31, 1991; Cohort 2 comprised 71 children born between January 1, 1992, and December 31, 2008.

All children included in the two data collections lived in rural areas or small towns and attended mainstream kindergarten or schools. The majority of the children (92%) were ethnic Norwegians. During the time period that covered the birth years of the children in both cohorts, there was a decrease in the general population with regard to the number of children between the ages of 1 and 17 years from approximately 65,610 in 1975 to 56,808 in 1992 to 53,686 in 2009. The method of only using hospital records to identify children with AD does not allow for the calculation of true prevalence; however, since the catchment area, children's ages, and method of

referral were the same, estimates of the proportion of AD diagnoses among the referred children across time can be compared.

Ethics

The study was registered by the Medical Director at the Central Hospital of Nordland as a quality assurance study and approved by the Ombudsman for Data Protection and the Northern Regional Ethical Committee.

Diagnostic assessments

For Cohort 1, diagnoses were made by a specialized autism team that included a psychologist and a pediatrician on the basis of diagnostic criteria presented in the DSM-III-R (35). For Cohort 2, specialists from the autism team diagnosed the children in accordance with ICD-10 criteria (31), which are comparable to the criteria for AD in the DSM-IV. All children were also observed at home and at kindergarten/school. The Autism Diagnostic Observation Schedule (36) was routinely used for children who were diagnosed after 2007 and for some who had been diagnosed earlier (n = 14).

Genetic syndromes were diagnosed by genetic experts at the university hospitals in Tromsø and Oslo with the use of the best available screening techniques at the time. In Cohort 1, 22 children (79%) were assessed with either chromosomal analysis (n = 19) or computed tomography/magnetic resonance scanning (n = 3). Comparable numbers for Cohort 2 revealed the presence of 55 such children (77%): 52 who were diagnosed with chromosomal analysis and three who were diagnosed with computed tomography/magnetic resonance scanning.

Cognitive assessments

In Cohort 1, almost all children (n = 25; 89%) were assessed with the Vineland Adaptive Behavior Scales (VABS) (37). One child's cognitive level was assessed with the Bayley Scales of Infant Development (38), one was obtained via clinical assessment, and data were missing for one child. In Cohort 2, all but 2 children (n = 69; 97%), were formally assessed with at least one of the following tests: Bayley Scales, Wechsler IQ measures, Leiter-R, or Vineland Adaptive Behavior Scales (37). The IQ scores were categorized as normal (IQ ≥ 70), mild intellectual disability (IQ = 50-69), moderate intellectual disability (IQ = 30-49), and severe intellectual disability (IQ < 30).

Statistical analysis

Data analysis was carried out with the use of SPSS version 23 (39). Kolmogorov-Smirnov testing for skewness and kurtosis (40) indicated that the

distribution of data for the age of first concern and for the diagnosis of AD deviated significantly from the normal curve, so nonparametric analyses (i.e., Mann–Whitney U test and Kruskal-Wallis one-way analysis of variance) based on median scores were used. For each p value, the corresponding z-scores are given in the parenthesis. Associations between the age of first parental concern and the age of diagnosis were calculated using Pearson's correlations (two-tailed). The chi-squared test (X^2) was used to assess differences in frequency across time. When cell frequencies were small, Fisher's exact test was used. The significance level was set at $p \leq .05$.

Results
Cohort comparisons

TABLE 1. Intellectual levels of children in Cohorts 1 and 2

Intellectual level	Cohort 1 (1992) (N = 27*)	Cohort 2 (2009) (N = 71)
	N (%)	N (%)
Normal (IQ > 70)	0 (0)	16 (23)
Mild intellectual disability (IQ 50-69)	3 (11)	13 (18)
Moderate intellectual disability (IQ 30-49)	3 (11)	22 (31)
Severe intellectual disability (IQ < 30)	21 (78)	20 (28)

IQ, Intelligence quotient.
*Data missing for one individual

TABLE 2. Genetic syndromes in Cohorts 1 and 2

Genetic syndromes	Cohort I (1992) (N = 28)	Cohort 2 (2009) (N = 71)
	N (%)	N (%)
Down	0	3 (4.2%)
Fragile X	0	1 (1.4%)
DiGeorge	0	1 (1.4%)
Williams	0	1 (1.4%)
Cornelia de Lange	0	2 (2.8%)
Distal trisomy 15q	0	1 (1.4%)
Smith-Lemli-Opitz	1 (3.5%)	0
Tuberous sclerosis	3 (11%)	1 (1.4%)
Other chromosomal deviations	2 (7%)	2 (2.8%)
Malformations	0	1 (1.4%)
Total	6 (21%)	13 (18%)

Frequency of autistic disorder diagnoses
In 1992 (Cohort 1), the total number of children between the ages of 1 and 17 years with a diagnosis of AD residing in Nordland county was 28 (21 boys [75%]), which corresponded to about 0.05%[1] of the total child population of 56,808. In 2009 (Cohort 2), 71 children were diagnosed (55 boys [77%]), which corresponded to about 0.13% of the total child population of 53,686. Thus, the proportion of children with AD had significantly increased across time (X^2[1, N = 110,494] = –4.61; p < .001).

Child characteristics: intelligence quotient and the presence of genetic syndromes
Children's developmental level
In Cohort 1, the majority of children (89%) had moderate or severe intellectual disability (IQ < 50); none had intellectual levels in the normal range (IQ $\geq$ 70). In Cohort 2, 42 children (59%) had moderate

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[1] This is not the population prevalence, but it provides the proportion of AD diagnoses among children 1 to 17 years old relative to the child population from which they were recruited. This for the comparison of Cohort 1 versus Cohort 2.

or severe intellectual disability; 16 (23%) had IQ scores within the normal range (Table 1). There were significantly more children with intellectual levels in the normal range in Cohort 2 (Fisher's exact test; p = .04).

Genetic syndromes

In Cohort 1, 6 children (21%) had diagnosed genetic syndromes; in Cohort 2, the corresponding number was 13 (18%); there was no significant difference (Fisher's exact test; p = .78). The types of genetic

syndromes were identified differed across both cohorts, with only tuberous sclerosis occurring in both (Table 2).

For two children in Cohort 1, the age at genetic syndrome diagnosis was missing. In all other cases, the genetic diagnosis was made at an earlier age than the diagnosis of AD. All four affected children in Cohort 1 and nine of the 16 affected children in Cohort 2 received their genetic syndrome diagnoses before the age of 12 months.

TABLE 3. Parents' first concerns about their children's development

Main presenting problem*	Social impairment	Ritualistic/repetitive behaviors	Delay/loss of language	General developmental delay	Epilepsy
Cohort 1 (N = 25)	13 (52%)	0	4 (16%)	7 (28%)	1 (4%)
Cohort 2 (N = 67)	43 (64.2%)	6 (9%)	8 (11.9%)	9 (13.4%)	1 (1.5%)

*Data missing for three individuals in Cohort 1 and four individuals in Cohort 2

Age and type of parents' first concerns

For both cohorts, parental concerns about their children's development were often expressed before the age of 12 months (59% in Cohort 1; 39% in Cohort 2), and almost all parents reported concerns by the time their children reached the age of 3 years (93% in Cohort 1; 84% in Cohort 2).[2] There was no significant difference across cohorts with regard to the age in months at first concerns: for Cohort 1, the median was 9 months, and for Cohort 2, the median was 18 months (z = 1.10; Mann–Whitney U test, p = .27).

Most parents reported more than one type of concern. However, in both cohorts, initial worries fell into four main categories. Concerns about social development (e.g., lack of eye contact, reduced interest in social interaction) were most prevalent, and these were often noted in combination with ritualistic behavior, language delay, or general developmental delay. These issues were noted before the age of 12 months in eight children (31%) in Cohort 1 and in 17 children (24%) in Cohort 2. The next most frequent concerns involved general developmental delay, and this was followed by a delay or loss of language and the presence of ritualistic/repetitive behaviors. Seizures as the sole

cause of early concern occurred in only one case in each cohort (Table 3).

Age at autistic disorder diagnosis

There was no significant cohort difference in age in months at the time of AD diagnosis: for Cohort 1, the median age was 54 months, and it was 60 months for Cohort 2 (z = 0.24; Mann–Whitney U test, p = .81). The percentage of children who received a diagnosis of AD before the age of 36 months was similar in the two cohorts: three (11%) in Cohort 1 versus nine (13%) in Cohort 2 (Fisher's exact test; p = .58). The majority of children—16 (59%) in Cohort 1 and 50 (70%) in Cohort 2—were diagnosed before the age of 6 years. The same proportion of children in both cohorts (18%) was diagnosed at the age of 8 years or older.

The relationship between the age at first concern and the age at AD diagnosis was investigated. In Cohort 1, the age of first parental concern was positively correlated with the age at AD diagnosis (r[25] = .41; p = .03): the earlier the concerns were noted, the earlier the age at AD diagnosis. In Cohort 2, there was no significant relationship between initial parental concerns and age at AD diagnosis (r[65] = –.15; p = .23).

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[2] Age data were missing for one child in Cohort 1 and four children in Cohort 2.

Combined cohorts

Intelligence quotient and genetic syndrome in relation to age of first concerns and autism diagnosis

There were no significant differences between cohorts with respect to age of first concerns and AD diagnosis. The two cohorts were thus combined to increase the sample size to explore the association between the presence of genetic syndromes or intellectual disability and the age of first concern and AD diagnosis.

Table 4 summarizes the ages of first concerns and AD diagnosis according to intellectual and genetic status. The children were divided into four mutually exclusive groups: 1) IQ < 50 with genetic syndromes; 2) IQ < 50 without genetic syndromes; 3) IQ = 50 to 69 without genetic syndromes; and 3) IQ ≥ 70 without genetic syndromes. As predicted, children with IQ values in the normal range (≥70) were significantly older than children with IQ values of 50 or less (irrespective of genetic syndrome) when they were first suspected of showing atypical development (comparison with genetic syndrome: z = 3.00; Mann–Whitney U test, $p < .001$; with no genetic syndrome: $z = 2.50$; Mann–Whitney U test, $p < .015$). As predicted, the age at which parents first became concerned was significantly earlier for children with genetic syndromes. These children—all of whom had IQ values of less than 50—were diagnosed with AD significantly later than children at the same intellectual level without genetic syndromes (z = 2.21; Mann–Whitney U test, $p = .027$).

TABLE 4. Age of first parental concerns and age of autism diagnosis in relation to intellectual level and the presence of genetic syndromes for Cohorts 1 and 2 (N = 94)

Subgroups*	Age of concern in months		Age of diagnosis in years	
	Median	Range	Median	Range
IQ > 70 (N = 16)	24.0	4.0-36.0	5.5	3.0-15.0
IQ = 50-69 (N = 15)	18.0	5.0-48.0	4.5	1.0-11.0
IQ <50, no syndrome (N = 44)	17.0	0.0-60.0	3.5	1.7-11.0
IQ < 50, with syndrome (N = 19)†	2.0	0.0-42.0	5.3	3.3-12.0
Total sample‡	15.0	0.0-60.0	4.9	1.0-15.0
Cohort 1	9.0	1.0-60.0	3.9	1.7-11.0
Cohort 2	18.0	0.0-48.0	5.0	1.0-15.0

IQ, Intelligence quotient

*Only children with values for both *Age of concern* and *Age of diagnosis* are included (N = 94)

†All children with genetic syndromes had intelligence quotients of less than 50

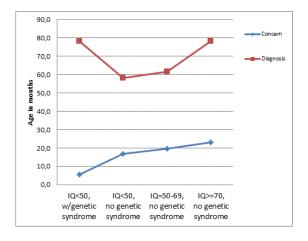‡The N value varies depending on the variable analyzed, missing from 1 to 4 children

FIGURE 1. **Delays between the age of first parental concerns and age of autism diagnosis for Cohorts 1 and 2 (N = 94)**

Children with IQ values of 70 or more were significantly older when they received their AD diagnoses as compared with children without genetic syndromes with IQ values of less than 50 (median, 3.5 vs. 5.5 years, respectively; z = 2.50; Mann–Whitney U test, $p = .015$). Children with genetic syndromes were diagnosed with AD at almost the same age as children with IQ values of 70 or more (median, 5.3 vs. 5.5 years, respectively).

Figure 1 illustrates the gap between the age of parents' first concerns and the age at AD diagnosis. The delay in receiving a diagnosis was greatest for children with genetic disorders and for children with IQ values in the normal range.

Discussion

The aim of the present study was fourfold: 1) to explore whether two studies conducted 17 years apart show a change in the number of children diagnosed with AD; 2) to compare child characteristics in the two cohorts; 3) to compare changes over time in children's ages at parents' initial concerns and AD diagnosis; and 4) to document the

association between child characteristics and age at first concern and diagnosis.

As predicted, we found a significant increase in the frequency of diagnosis, from 28 children in 1992 (about 0.05% of the total child population) to 71 children in 2009 (about 0.13% of the total child population). There were more children with IQ values in the average range in the latter cohort. Contrary to our expectations, children in Cohort 2 were somewhat older than those in Cohort 1 both when they were suspected of deviant development (median, 18 vs. 9 months, respectively) and when they received a diagnosis of AD (median, 5.0 vs. 4.5 years, respectively), although the differences were not significant. The proportion of children with genetic syndromes remained stable at 21% and 18%, respectively. As predicted, parental concerns about deviant development emerged earlier for children with genetic syndrome, but contrary to predictions, these same children were among the oldest to receive their AD diagnoses (median, 5.3 years). As predicted, the other group that received later AD diagnoses (median, 5.5 years) consisted of the more intellectually able children (i.e., those with IQ values in the average range or higher).

Awareness of autism has increased among the general public as well as among teachers and health care personnel. We therefore predicted—in line with the findings of Fombonne and colleagues (6)—that some of the increase in frequency in Cohort 2 could be explained by younger children being diagnosed with AD. However, the results of our study indicated that there was no significant change in median age at diagnosis; in fact, rather than decreasing, the age at diagnosis had tended to increase by about 6 months. Another expectation was that there would be more children with genetic disorders in Cohort 2 due to the growing awareness of the increased risk of ASD in children with these conditions and the more sophisticated techniques used to identify genetic disorders. However, the proportion of children with such conditions had not changed significantly over time.

The one factor that seems to be associated with the change in diagnostic rates is IQ values. The present study documents a significant increase (20%) in the number of children with IQ values in the normal range in Cohort 2. These children tend to show less severe autistic symptoms than children with additional intellectual disabilities, and they often represent more of a diagnostic challenge. The increased awareness of AD across the intellectual spectrum may have played an important role in increasing the rates of AD diagnosis in this particular subgroup.

In both cohorts, the majority of parents suspected that there were problems with their children's development before the age of 36 months. The most frequent concerns focused on core autism features (e.g., social or communication impairments, ritualistic behaviors), often in combination with language delay or general delay (20,41).There was often a substantial delay between the emergence of early parental concerns and the diagnosis of AD. Moreover, the time gap reported by Crane and colleagues (41) did not seem to decrease during the past few decades in Nordland County. The failure to find a decrease in the average age at diagnosis over time was particularly disappointing, especially because early diagnosis is important for access to appropriate education and intervention. Although several studies now suggest a decline in the age at diagnosis (18,19), considerable disparities remain. A few of these were pointed out in a review by Daniels and Mandell (32), which indicated that a general increase in awareness does not automatically lead to a decrease in diagnostic age (41).

Delays in obtaining an AD diagnosis were particularly marked in children with higher IQ values. Their often more subtle aberrances, together with a lack of a substantial delay in language development, can lead to parents' early concerns being disregarded by professionals, which may result in repeated referrals until diagnosis is confirmed (21). This not only delays the provision of services, but it also negatively affects parental perceptions of the diagnostic process and of the clinicians involved and increases parental stress (41). These effects were also reported in studies by Crane and colleagues (21) and Guinchat and colleagues (20).

By contrast, children with severe cognitive impairments (IQ < 50) were more likely to receive an early diagnosis. This has also been noted in other studies (14,17), and it may be due to the fact that young children with delayed developmental milestones are more likely to be referred to specialist pediatric services with greater expertise in recognizing and diagnosing AD at an earlier age.

Fombonne (6) has argued that data showing an increase in AD diagnoses over time are often confounded by factors such as referral patterns, the availability of services, public awareness, and changes in diagnostic concepts and practices. In the present study, referral patterns, the availability of services, and the service organization in Nordland County had changed little over the relevant decades, so these factors are less likely to be major contributors to the observed increase in the proportion of AD diagnosis. Another possible contributing factor is that the diagnoses in the two cohorts were based on somewhat different diagnostic systems (i.e., the DSM-III-R in Cohort 1 and the ICD-10 in Cohort 2). Volkmar (42) argues that the DSM-III-R seems to inflate the number of cases diagnosed as compared

with either the ICD-10 or expert clinical judgment. The diagnostic criteria for AD (i.e., core autism), which is the focus of the present study, have not changed greatly over the studied period and have tended to remain more stable than the criteria for other diagnoses on the spectrum.

Unfortunately, our data could not be used to determine whether diagnostic substitution had occurred or to what extent in the two cohorts studied. It has been shown by Suren and colleagues (43) that, across various counties in Norway, it is likely that diagnostic substitution occurs. However, in our study, all assessments were done by the same hospital in Nordland County and concerned only AD, thereby reducing the likelihood of this being a key factor in the differences found between the two cohorts.

The other group for whom an AD diagnosis was most likely to be delayed was children with genetic syndromes. It might be expected that autism in these children would be identified early, because they, too, are usually involved with specialist pediatric health services from an early age (7). Instead, they were among the oldest to receive their AD diagnoses. Several studies have pointed out that, when autistic symptoms are "overshadowed" by genetic conditions, the affected children are likely to be referred for examination for possible AD much later than other children are (7,29,44). This situation may prevent them from receiving appropriate interventions from an early age.

To summarize, our findings have indicated a significant increase in AD diagnoses in Nordland County over a 17-year period, and they corroborate the conclusions drawn by studies from other countries. Although this was not an epidemiological study, the data suggest a rise in prevalence rates; the increase cannot be attributed to major changes in local referral patterns, clinical diagnostic practices, or the availability of services. However, it is possible that the rise is influenced by a greater awareness among professionals of autism in children of average IQ, who were less likely to be recognized in earlier years. Unfortunately, any increased awareness of autism and AD among relevant professionals in the county did not result in a reduction in the age at diagnosis, and the gap between parents' early concerns and children's AD diagnoses did not decrease over time. Delays in the age at diagnosis were particularly marked in children with IQ values in the average range and in those with genetic disorders.

Although the reasons for the increase in the number of cases diagnosed over time remain unclear, these findings do have important implications for diagnosis.

Strengths and limitations of the study
The main strength of the study is that it includes *all* children diagnosed with AD who were registered with the Specialist Health Service in Nordland County and residing in the county in 1992 and 2009. The children in the two cohorts were born during two non-overlapping time periods, and the referral patterns and services have not changed.

Nevertheless, it also suffers from a number of methodological problems that limit the conclusions that can be drawn. First, the sample size is small. Although there was no evidence of substantial changes in diagnostic practice over time, standardized diagnostic instruments (e.g., the Autism Diagnostic Observation Schedule) were used to confirm diagnoses in only 14 cases. Second, the study compares two clinically referred samples, but children with high levels of autistic traits may have received educational services without being referred to specialized services for a possible diagnosis. Third, the use of the DSM-III-R criteria in Cohort 1 and the ICD-10 criteria in Cohort 2 may be related to the change in diagnostic frequency. However, this would be expected to lead to a *decrease* in diagnostic rates (42). Fourth, the results relate only to children residing in Nordland County at the two time points examined and thus may not be generalizable to other counties or other time periods. Finally, the ICD-10 classification of children into subgroups within the autism spectrum can be problematic (45) and can lead to misclassification. This might have influenced the number of registered children with AD in Nordland County. However research has shown relatively high agreement for the diagnosis of AD specifically (46).

Clinical significance
In children with genetic syndromes, there seems to be a risk of the genetic syndrome "overshadowing" the autistic symptom patterns. Results from this study show that there is a considerable delay in confirming an AD diagnosis in this group, which results in the postponing of appropriate intervention and support. The potential effects of this delay in diagnosis are considerable; they not only affect children's treatment and educational provisions, but they may possibly alter long-term outcomes. Crane (41) has reported a significant negative impact on affected families. Mental health providers should ensure that children with genetic syndromes in combination with autistic traits are thoroughly assessed to clarify whether an AD (or other) diagnosis is warranted. If autistic traits are consistently described in children with IQ values in the normal range, there is a particular need for a highly expert diagnostic assessment, because their

symptoms may be subtle or atypical. Greater awareness among clinicians of the presentation of autism in these groups could have a significant and positive impact on the affected children's age at diagnosis and on the provision of appropriate interventions and support.

Conflicts of Interest
The authors declare no conflicts of interest.

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