EDITORIAL

Hidden in plain sight

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This issue of IJE includes a theme on autism edited by two of us (Ezra Susser and Michaeline Bresnahan). Our colleague Guohua Li joins us to write this introduction.

In choosing this topic we venture into territory marked by controversy and conflict. The heat of the conflict is reflected in two recently published books,1,2 reviewed here by Stein3 and by Grinker.4 One of the flashpoints of controversy pertains to time trends in autism and autism spectrum disorders (ASDs). ASDs are variously defined in epidemiological studies, but usually include ICD-10 Childhood Autism, Atypical Autism, Other Pervasive Developmental Disorder, Pervasive Developmental Disorder Unspecified and Asperger’s Syndrome. Recent reports suggest that ~0.4% of children are affected with autism, and ~1% affected with ASD.5 This is about a 10-fold higher prevalence for autism than was found in the earliest research such as the classic 1966 study by Lotter,6 and 5-fold higher than was found for autism in most studies as recently as the 1990s.7 A similarly dramatic time trend is found when attention is extended to the broader category of ASD. These figures have generated intense debate over whether the observed increase is mainly ‘real’ (increasing incidence) or ‘artefactual’ (increasing ascertainment).

The original research piece presented by King and Bearman,8 commentaries on this article by leaders in the field9–12 and the paper by Naaser et al13 contribute important data and thoughtful perspectives to this debate. Naaser and colleagues draw on the registries of Western Australia to illustrate the concurrence of changes in diagnostic criteria and available services with periods of increase in ascertainment, providing a type of ecological evidence for factors other than increasing incidence playing an important role in the autism and ASD time trends. King and Bearman take a quite different approach. They draw on the California Department of Developmental Services, a source that has figured large in the controversy over time trends in autism. They use the individual-level data, available from this source, to estimate the proportion of the increased prevalence of autism that could be due to diagnostic substitution and accretion. Based on observed changes in diagnosis, the paper infers that a significant part of the increase can be attributed to a shift away from mental retardation alone towards autism (with or without mental retardation). The commentary by Charman et al9 also presents intriguing data that illustrate the impact of varying diagnostic criteria. In this commentary they include a remarkable comparison of autism diagnoses made by the old Lotter criteria, which focus on social aloofness and rituals, and newer ICD-10 criteria for the same children. The experts applying Lotter criteria included Lorna Wing14 and Susan Bryson who were familiar with the use of the criteria from the early time period. A significantly lower proportion of children were diagnosed with autism by Lotter criteria than by ICD-10 criteria.

Although all of these articles present evidence that some of the time trend is related to increasing ascertainment, they also concur that a significant portion of the time trend remains unexplained. If any part of this represents an increase in incidence—as seems likely—we are pressed to intensify the search for environmental contributions to aetiology. As Rutter points out, this investment must include studies with the means to identify these factors in the relevant time frame—prospective research beginning in gestation with careful case identification and ongoing follow-up.12 Such methods are already being pursued in some studies such as the Autism Birth Cohort nested in the Norwegian Mother and Child Study,
and considered in forthcoming studies such as the United States National Children’s Study.

In this commentary, we want to draw attention to another profound implication of the increasing prevalence of diagnosed childhood autism. Under any plausible scenario we can infer: first, at present the majority of adults with autism were either misdiagnosed or undiagnosed as children; and secondly, there is a large cohort of children with autism who will soon become adults. This represents a daunting challenge to public health.

What we know about autism in adulthood is minimal. Longitudinal studies of individuals diagnosed as children are generally limited by small numbers, selection of cases from specialized clinics and/or follow-ups that do not extend into mid-life. Moreover, we lack reliable diagnostic methods for adults. Three recent studies15–17 address these limitations to some degree. Their results suggest that autistic children with a low childhood IQ rarely attain independent living or social relationships, but also that those without intellectual disability exhibit considerable diversity and a few do very well. These studies could not, of course, include children with autism who were not diagnosed. The lives of the misdiagnosed or undiagnosed adults with autism remain a mystery. We suspect that some could be found in the intellectual disabilities service system, the mental health service system and other social services systems. Others may be found in shelters for homeless people or in prison. Still others will be found living in their parents’ homes or living independently in the community.

We know from published biographies what some individual outcomes look like. In Bay Alone,18 the latest instalment in the telling of the life of Noah Greenfeld and his family, Noah’s brother gives an unvarnished view of the life of a low-functioning middle-aged man with autism. Noah is ‘living proof of the obvious: autism is not a terminal disease’ (p. 341). With the benefit of strong parent advocates, Noah received the most credible treatments available during childhood. As an adult he has had to rely on the limited public system. He now bears the scars of mistreatment, abuse and neglect at the hands of strangers. ‘He was a lost boy then, and he is a lost grown-up now’ (p. 344).

Public health implications

Even the wealthiest societies will find that they will need to make a substantial investment to meet the need of adults with autism. First, there is a practical need for a very large increase in service capacity. As we learned with children, expanding capacity will bring individuals into the system who would have otherwise remained unidentified.

Secondly, we need to know more about what services these adults need. The needs of individuals with autism can be quite distinct from others with developmental disabilities. For instance, sensory disturbances—oversensitivity to sound, light, touch—are common in children with autism, and often persist into adulthood. These may be sufficiently intense that an environment well suited to the needs of individuals with other developmental disabilities (e.g. Down’s syndrome, cerebral palsy) would be aversive to an individual with autism. Another example is the need for the sameness characteristic of autism but not these other disorders. So noted, our responses should be evidence based; this requires a significant expansion of mental health services research.

Thirdly, as epidemiologists, we need to adopt a life-course approach to autism and other developmental disabilities, where we usually study only children. Reliable diagnostic approaches tailored to adults must be developed. Understanding the ASDs across the life cycle will be essential for developing treatment and services programmes to support these individuals throughout their life. A life-course approach may also be invaluable for understanding aetiology, deepen our understanding of the factors determining different trajectories of the disorder, and reveal the possibilities and the limits of development for autistic individuals within a long-term supportive environment.

Finally, and perhaps above all, our societies need to make an ethical commitment to address the problem. While we are beginning to make this commitment to disabled children—who are already in a dependent class by virtue of age—we seem to be less willing to make this commitment to adults. If ignorance is no longer an excuse, is neglect an option? We think not.

Conflict of interest: None declared.

References


10 Fombonne E. Commentary on King and Bearman. *Int J Epidemiol* 2009;38:1241–42.


