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Estimating Prevalence of Autism Spectrum Disorders (ASD) in the Irish Population: A review of data sources and epidemiological studies

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This paper was compiled collaboratively by the R&D and Health Analytics Division and the Chief Medical Officer's Division of the Department of Health.

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Executive Summary

Autism spectrum disorders (ASDs) are a group of life-long neurodevelopmental conditions characterised by impairments in social interaction and communication, as well as restricted, repetitive and stereotyped patterns of behaviour. Although there have been many studies on the epidemiology of ASDs internationally, uncertainty remains about the true prevalence of ASD globally. This is largely attributed to differences in measurement techniques which makes direct comparisons difficult.

In the last twenty years, the reported prevalence of ASDs has increased. The most recent publication of the Centre for Disease Control (CDC) (April 2018) reported the prevalence of ASDs in the US as 1.68% in the surveillance year 2014 but ASDs have been reported to be as high as 2.6% in South Korea and as low as 0.39% in China.

Variations and changes in ASD prevalence rates reported may have several explanations: changes or broadening of the diagnostic criteria, differences in the methods used to study prevalence (sampling procedures, application of statistical methods), as well as an increased awareness among parents, professionals and the general public.

This paper examines available data and evidence relating to prevalence of ASDs in children and adults in Ireland for burden of diseases analysis and policy-making decisions.

Methods

As there is no specific autism register in Ireland, a number of administrative databases were explored to determine if there was any useful data to inform incidence/ prevalence rates. In addition, a literature search was conducted to identify any research or epidemiological studies relating to autism prevalence that have been undertaken over the past 20 years in Ireland. Prevalence rates of autism in other countries were sourced and analysed for comparison purposes. These included analyses of data from registries and epidemiological studies.

Key findings

- In autism epidemiology, point or period prevalence is more useful than incidence, as the disorder starts long before it is diagnosed, and the gap between initiation and diagnosis is influenced by many factors unrelated to risk. Therefore, this paper concentrates on the prevalence literature.
- Given the complexity of the classification of autism and the subsequent changes made to the DSM and ICD diagnostic categories over the last number of decades, estimating prevalence of the condition is fraught with methodological difficulties.
- While significant progress has been made over the past two decades in the development of screening and diagnostic instruments for autism spectrum disorders, accurate tools for screening and diagnosis of ASD are limited by the lack of a true test for ASD, which remains a behaviourally-defined disorder.
- The most reliable source of information would be a disease register where all individuals diagnosed with the disorder (using standardised definition and diagnostic instruments) would be recorded. While this does not exist in Ireland, this paper highlights other data sources which can help with estimating a prevalence rate.
- The National Council for Special Education (NCSE) collects data on students with ASD with resource teaching support or in special classes and special schools. Their analysis indicates a prevalence rate of 1.55 per cent and this figure is currently used for planning purposes. The Department of Education have recently changed their model whereby professional and other medical assessment or diagnosis of ASD will no longer be necessary for pupils to access educational teaching resources in schools. This is in line with an international shift away from diagnosis-driven access to services.
- Within the health sector, it is important to note that plans are advanced to merge and refine the existing disability information systems managed by the HRB into the NASS (National Ability Supports System). This will capture diagnosis, including ASD, and will be capable of recording more than one diagnosis to allow for co-morbidity. However, the NASS is still voluntary (not epidemiological in its collection), so will only record

those who have or need the services referred to above, and thus cannot be used for estimating prevalence rates.

- This paper highlights a number of national and epidemiological studies which sought to measure the prevalence of ASD. These studies employ different methodologies, diagnostic tools and age groups and as such are not directly comparable. The most recent data on the prevalence of ASD in Ireland is reported in a study carried out by Boilson and colleagues in Dublin City University (2016). This work is now part of an EU wide study, Autism Spectrum Disorders in the EU (ASDEU), which is designed to facilitate a common format for screening and diagnosing children with ASD across the EU with the aim of developing a standardised strategy for future surveillance. Their analysis reported a prevalence rate of 1% and when further analysis was conducted (to weight for a low response rate) a figure of 1.5% resulted. This is in line with prevalence estimates in other countries such as the UK, Finland, Norway, Denmark, Italy, Australia, Canada and USA. It is also similar to the Department of Education figure of 1.5% which is based on the numbers of children with a diagnosis of ASD who are availing of additional teaching supports in schools.
- Virtually all studies demonstrate a significant difference in the estimated prevalence rates of autism between the genders, with males four times more likely to be identified with autism than females. There is also evidence to suggest that ASD prevalence rates, and needs for services, are higher in disadvantaged areas.
- This review highlighted that there is no data available on prevalence of ASD in adults in Ireland. Indeed, there is only one study conducted internationally in the adult population. This community-based study carried out in 2011 in the UK found that ASD affects approximately 1% of the adult English household population. There was no evidence of a statistically significant reduction in prevalence of ASDs as a function of age.
- Based on all the above, and notwithstanding the absence of a single source of data to estimate ASD prevalence in Ireland, data compiled using a combination of methods suggests that there is a robust case for adopting an estimated prevalence rate of 1-1.5% for the purpose of planning policy and services.
- Repeating a prior study (such as the EU study led by DCU) using the same methodology and conducted in the same geographical area at different points in time, has potential to yield useful information on time trends provided that methods are kept relatively constant.

1.0 Introduction

Autism spectrum disorder (ASD) is a life-long developmental disability characterised by social and communication impairments and by restricted interests and repetitive behaviours.¹ An extremely wide range of individual differences is represented within this grouping, from individuals who also have a severe learning disability to those with average and above average intelligence. The Autism and Developmental Disabilities Monitoring (ADDM) Network shows that among children identified with ASD who had IQ scores available, about one third also have an intellectual disability.² In Ireland it is estimated that this figure is about 50%.³

All share the triad of difficulties in reciprocal social interaction, communication, and a lack of flexible thinking. ASDs impact on all areas of functioning and have significant implications throughout the lives of those affected across the entire ability range. Children displaying symptoms within all three areas of core autism symptomatology before 3 years of age are usually diagnosed with childhood autism, and it is estimated that about 30% of all ASD diagnoses relate to a diagnosis of childhood autism. The literature indicates a pattern of lifelong disorder, whereby individuals with an autistic spectrum disorder continue to be 'autistic' throughout their lives.

1.1 International classification of diseases

Mental and behavioural disorders are classified by two major nosological systems, the International Classification of Diseases - ICD (initiated in Paris in 1989),⁴ and the Diagnostic and Statistical Manual of Mental Disorders - DSM (DSM-I was published in the USA in 1952).⁵ The ICD has since gone through ten revisions, with ICD-10 the latest to be published (World Health Organization 1989). DSM has been revised more frequently, with successive revisions in 1987, 1994, 2000, and more recently in May 2013 (American Psychiatric Association 1987, 1994, 2000, 2013).

¹ American Psychiatric Association. Diagnostic and statistical manual of mental disorders. 4th ed. Text revision. Washington, DC: American Psychiatric Association; 2000.

² Centers for Disease Control and Prevention (CDC). Prevalence of Autism Spectrum Disorders—Autism and Developmental Disabilities Monitoring Network, United States, 2008. Morbidity and Mortal Weekly Report (MMWR) 2012; Vol. 61(3).

³ Personal communication with Prof. Louise Gallagher, Department of Psychiatry, Trinity College Dublin.

⁴ World Health Organization (1989) ICD-10: Classification of Mental and Behavioural Disorders. WHO.

⁵ American Psychiatric Association (1980) Diagnostic and Statistical Manual of Mental Disorders (3rd ed.) (DSM-III). APA.

It is important to note that there is a lot of convergence between the two international systems of diagnosis, partly because of collaborative agreements between the two organisations. It is possible to convert the diagnoses of one system into another. Both classification systems take the modern view of autism: that 'there is a spectrum of autistic conditions and that they are disorders of development, not "psychoses"'. Over time the definitions of autism have changed as illustrated by the numerous diagnostic criteria that were used in both epidemiological and clinical settings.

1.2 History of classification of ASD

In 1943, Kanner first documented a syndrome of "autistic disturbances" in 11 children who shared previously unreported patterns of behaviour, including poor social interaction, obsessiveness, stereotypic movement, and echolalia.⁶ Autism, the prototypic pervasive developmental disorder (PDD), is characterized by an onset prior to three years of age and by a triad of behavioural signs and symptoms, including:

- (1) abnormal development in the use of language
- (2) lack of reciprocal social interaction and responsiveness, and
- (3) restricted, stereotypical, and ritualised patterns of interests and behaviour.⁷

Autism first manifests in childhood, with age of onset for a diagnosis being under the age of 3 years. This does not necessarily mean that a person is diagnosed before turning 3 years of age, only that symptoms were present at that developmental stage.

For many years after autism was first described in the 1940s, its prevalence was considered to be two to four cases per 10,000 children.⁸ During the 1950–60s, autism was widely regarded as early presentation of childhood schizophrenia, an emotional disturbance rooted in parent–child psychodynamics. Consequently, in DSM I (1952) autism was classified as Schizophrenic reaction, childhood type.

This classification continued with the DSM II (1968), where autism appeared only under the following category: 295.8 Schizophrenia, childhood type. This category was for cases in which schizophrenic symptoms appear before puberty. The condition was considered to be manifested by autistic, atypical and withdrawn behaviour; failure to develop identity separate from the mother's; and general unevenness, gross immaturity and inadequacy of development. It was acknowledged that these developmental defects may result in mental retardation, which should also be diagnosed.

⁶ Kanner L. Autistic disturbances of affective contact. *Nerv Child*. 1943; 2:217–250.

⁷ Bailey A, Phillips W, Rutter M. Autism: towards an integration of clinical, genetic, neuropsychological, and neurobiological perspectives. *J Child Psychol Psychiatry*. 1996; 37:89–126.

⁸ Wing L, Potter D. The epidemiology of autistic spectrum disorders: is the prevalence rising? *Ment Retard Dev Disabil Res Rev*. 2002;8: 151–161.

1.3 Change in classification

By the 1970s a change in classification occurred and autism was understood as biologic in origin and no longer incompatible with mental retardation. Subsequently, the DSM-III and the DSM-111R (1987) provided a more complex definition of autistic disorder that required meeting 8 of 16 criteria among the three domains of social interaction, communication, and restricted interest or activities, dropping the requirement for early onset in life and providing a new category, “Pervasive Developmental Disorder, Not Otherwise Specified (PDD-NOS),” for children meeting some but not all diagnostic criteria for autistic disorder.

Revisions in the DSM-III-R definition of autism meant that although sensitivity was now very high, specificity still was quite low. In other words, a large number of children previously not diagnosed now met the diagnostic criteria for autism. In 1994, the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition (DSM-IV) introduced revised diagnostic criteria and five subtypes of autism under the category of pervasive developmental disorders. These included autistic disorder, Asperger disorder, pervasive developmental disorder—not otherwise specified (PDD-NOS), childhood disintegrative disorder, and Rhetts’s disorder.⁹ The first three subtypes comprise autism spectrum disorder (ASD), whereas the latter two conditions belong to the wider category of pervasive developmental disorders.

The fifth edition of DSM was published in 2013 and collapsed autism, Asperger syndrome and pervasive developmental disorder-not otherwise specified into a single diagnosis of Autism Spectrum Disorder (ASD).¹⁰ It is believed that individuals with ASD are best represented as a single diagnostic category because they show similar types of symptoms and are better differentiated by clinical specifiers (i.e., dimensions of severity) and associated features (i.e., known genetic disorders, epilepsy, and intellectual disability). An additional change to the DSM-5 includes synthesising the social and communication deficits section into one domain.

Autistic Disorder

Autistic disorder (DSM-IV-TR) or childhood autism (ICD 10) has an onset before the age of three and shows evidence of a cluster of features including abnormal functioning in social interaction, communication, and imagination and thought as evidenced through restricted, repetitive behaviour which cannot be solely explained on the basis of low cognitive functioning. It is much more common in boys. Autistic disorder is also known as “Kanner's Syndrome”.

⁹ American Psychiatric Association. Diagnostic and statistical manual of mental disorders. 4th ed. Washington, DC: American Psychiatric Association; 1994.

¹⁰ American Psychiatric Association. Diagnostic and statistical manual of mental disorders. 5th ed. Arlington, VA: American Psychiatric Association; 2013.

Asperger's Disorder (DSM-IV-TR) / Asperger's Syndrome (ICD-10)

While this disorder has an 'uncertain nosological validity', it shows the same kind of qualitative abnormalities of reciprocal social interaction as autistic disorder does with a restricted, stereotyped, repetitive repertoire of interests and activities. In international diagnostic terms, the main difference from autistic disorder has been that there is no clinically significant delay or retardation in cognitive development or in language acquisition, e.g. 'single words used by age two years' (DSM-IV-TR). More importantly, persons with Asperger Syndrome (AS) have communication difficulties (regardless of structural language skill). Children with Asperger Syndrome typically present for assessment relatively late in development. Asperger Syndrome appears to have a later onset or at least tends to be recognized at a later stage.

Atypical Autism or Pervasive Developmental Disorder-Not Otherwise Specified (PDDNOS)

This diagnostic category is used when there is a severe and pervasive impairment in the development of reciprocal social interaction associated with impairment in either verbal or non-verbal communication skills or with the presence of stereotyped behaviour, interests and activities, but not meeting the diagnostic criteria for specific pervasive developmental disorders, e.g. autistic disorder or Asperger's Syndrome. PDD-NOS includes atypical autism, i.e. presentations that are characterised by atypical symptomatology, or subthreshold symptomatology, or late onset, or all of these.

Impact of changing diagnostic criteria on reported prevalence rate

It is understandable, given the complexity of the classification of autism and the subsequent changes made to the DSM and ICD diagnostic categories over the last number of decades, that estimating prevalence of the condition is fraught with methodological difficulties. This is best illustrated in the work undertaken by Kielinen et al. (2000)¹¹ who applied different diagnostic criteria to the same group of children (n=39, 216). Whilst administering Kanner's original criteria the rate of autism was 2.3 per 10,000; this increased to 6.1 per 10,000 using the ICD-10; the use of DSM-IV criteria further increased the rate to 7.6 per 10,000. These findings illustrate a 3-fold variation in prevalence rates based solely upon varying diagnostic criteria. Baird et al. in the UK in 2006 found a prevalence rate of 116.1 per 10,000, but when the authors confined the definition of childhood autism to a narrower definition, this provided a prevalence of 24.8 per 10,000 for the same population.¹²

¹¹ M. Kielinen, Linna S.L., Moilanen, I. (2000) Autism in Northern Finland. *Eur Child Adolesc Psychiatry*. Sep;9(3):162-7.

¹² Baird, et al. (2006) Prevalence of disorders of the autism spectrum in a population cohort of children in South Thames: the Special Needs and Autism Project (SNAP). *The Lancet*. Jul 15;368 (9531):210-5.

This is further highlighted in a 2014 study: 'Potential impact of DSM-5 criteria on autism spectrum disorder (ASD) prevalence estimates.'¹³ The researchers found that estimates of the number of children with ASD might be lower using the current Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5) criteria than using the previous Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, Text Revision (DSM-IV-TR) criteria.

1.4 Screening and diagnosis

In addition to changing classification systems, there are numerous screening and diagnostic instruments used in the diagnosis of ASD. Significant progress has been made over the past two decades in the development of screening and diagnostic instruments for autism spectrum disorders (ASD).¹⁴ However accurate tools for screening and diagnosis of ASD are limited by the lack of a true test for ASD, which remains a behaviourally-defined disorder.

Screening is the prospective identification of unrecognised disorder by the application of specific tests or examinations. Surveillance refers to the on-going and systematic collection of data relevant to the identification of a disorder over time by an integrated health system.

Several parameters of screening instruments are important in assessing their efficacy and utility:

- i. Sensitivity is the proportion of individuals with a disorder who have a positive screen result,
- ii. Specificity is the proportion of individuals with a disorder who have a negative screen result,
- iii. Positive predictive value (PPV) is the proportion of individuals with a positive screen result who have the disorder.

Sensitivity is required to be high in order that the screen misses few cases of the disorder (avoiding falsely reassuring parents and professionals). Specificity is required to be high in order that few cases without the disorder are screen positive (avoiding falsely alarming parents and costly referral for in-depth assessment). When the sensitivity and specificity of a screen remain constant, the PPV is lower the rarer a disorder is within the population.¹⁵ Hence, PPV will be lower in the population than in referred samples. Glascoe (1996) has estimated that acceptable sensitivity and specificity for developmental screening tests are

¹³ Matthew J. Maenner, et al. Potential Impact of DSM-5 Criteria on Autism Spectrum Disorder Prevalence Estimates. *JAMA Psychiatry*. 2014;71(3):292-300.

¹⁴ See Charman, et al. (2013) Measurement Issues: Screening and diagnostic instruments for autism spectrum disorders – lessons from research and practice. *Child Adolesc Ment Health*. Feb 1; 18(1): 52–63.

¹⁵ Clarke and R. Harrington (1999). On diagnosing rare disorders rarely: appropriate use of screening instruments. *J Child. Psychol. Psychiatry*. Feb: 40(2):287-90.

70% to 80%, reflecting the nature and complexity of measuring the continuous process of child development.¹⁶

ASD is heterogeneous in the presentation and time course of core deficits. It would therefore be important for a screening programme to administer ASD-specific screening tools periodically at differing ages to detect children at risk who, for a number of reasons, may have been missed on an earlier occasion. Searches identified 46 screening tools for ASD. Most are designed for children, while only few measures are available for adults, especially those with additional intellectual disabilities. Many instruments are under-researched, although a small number such as the Modified Checklist for Autism in Toddlers and the Social Communication Questionnaire have been widely examined in a variety of populations.¹⁷

Diagnostic instruments

ASD screening instruments function to identify children in need of further monitoring or diagnostic evaluation. At that point, standardised autism diagnostic instruments are often employed to structure the information-gathering from both parents and identified children within a diagnostic assessment. The existence of, and ongoing improvements to, such measures are associated with more accurate diagnosis of ASD, including the ability to reliably describe milder and younger cases, as well as increased comparability of research findings based on better agreement as to “caseness” across research teams.

However, as with screening tools, diagnostic instruments are often limited by inadequate power to correctly identify individuals with and without ASD. Further, the estimates of such performance validity for each particular measure are necessarily limited by the absence of an absolute test for ASD, and as such are influenced by clinical experience in diagnosing ASD, training and experience in using the diagnostic measure, and evolution within the field in terms of what is recognised and labelled “ASD.”

Therefore, ASD assessment and diagnosis relies on behavioural assessment. It is widely accepted that diagnosis should be conducted using a multidisciplinary approach evaluating cognitive functioning, speech and language ability and broader developmental concerns, as well as behavioural evaluation.

Lack of a universally accepted screening instrument had led to recommendations in some countries not to implement population screening. The 2012 HSE review concluded that it would be inappropriate to be prescriptive about the assessment instrument used as new and

¹⁶ F.P. Glascoe (1996). Developmental Screening. In Wolraich, M., ed. Disorders of development and learning: A practical guide. St Louis: MO: Mosby.

¹⁷ Sappok, et al. (2015). Screening tools for autism spectrum disorders. *Advances in Autism*, 1 (1):12-29.

more appropriate ones emerge.¹⁸ However, they advocated for use of a semi-structured instrument in combination with the Autism Diagnostic Observation Schedule (ADOS) as that which provides the best sensitivity and specificity in the diagnosis, and stability over time.

1.5 Legislation

The Education for Persons with Special Education Needs EPSEN Act (2004) provides for supporting the rights of children to an educational assessment, an individual education plan, and to an independent appeals process. It fits into a legislative framework which, inter alia, includes the Education Act (1998), the Education (Welfare) Act (2000), the Equal Status Act (2000) and the Disability Act (2005), under the overall umbrella of the Constitution, as well as various international agreements and human rights provisions. The EPSEN Act has not yet been implemented.

The purpose of the Disability Act is to promote the participation of people with disabilities in society by supporting the provision of disability specific services and improving access to mainstream public services. The Act establishes a right to an independent assessment of individual needs and a related service statement. On 1st June 2007, Part 2 of the Disability Act became law for children under the age of five years, giving them a right to an independent assessment of need. The Disability Act (2005) places legal obligations on the HSE regarding assessment of need and service statements for all individuals with a disability and on the autism spectrum. The terms of the Act indicate that an assessment of need should commence within three months.

¹⁸ See National Review of Autism Services: Past, Present and Future (HSE, 2012), pp. 19-20 and inclusion of a table summarizing the main screening instruments, with their pros and cons.

2.0 Review of data sources to estimate prevalence in Ireland

There is currently no reliable method of estimating prevalence of Autism Spectrum Disorder in Ireland. The most reliable source of information would be a disease register where all individuals diagnosed with the disorder (using standardised definition and diagnostic instruments) would be recorded. This does not exist in Ireland. However, there are other data sources which might help with estimating a prevalence rate.

2.1 Irish data sources

The Irish census

The census of population has been the primary source of information on numbers of people with disabilities in Ireland. In Census 2006 questions on disability were broadened to include learning difficulties, intellectual disabilities, and psychological and emotional conditions; this resulted in a rise in the prevalence rate for all developmental disorders from 2.1% of children in 2002 to 3.2% in 2006. A follow-up nested study, entitled The National Disability Survey (2008), and carried out with 16,000 people, reported that 11% of children aged 0-17 years reported having a disability. Working estimates of prevalence for disabilities by category based on the total population of 5-18s in the census of 2006 estimated the prevalence of autism spectrum disorder at 4,730, a rate of 0.6%.

The 2016 census showed that 66,611 people or 1.4% of the population suffered from an intellectual disability, 8,902 higher than in 2011, representing a 15.4% increase. As in previous census data, the greatest incidence by far was amongst 10 to 14 year old males, with 5,233 affected in this age group - more than double that of females (2,284). This information while useful, does not provide data on ASD prevalence as no nested study has been undertaken to date.

Health Research Board

The Health Research Board manages two national service-planning databases for people with disabilities on behalf of the Department of Health.

- National Intellectual Disability Database (NIDD), established in 1995
- National Physical and Sensory Disability Database (NPSDD), established in 2002

The disability databases aim to provide comprehensive and accurate information for decision making in relation to the planning of specialised health and personal social services for people with intellectual, physical or sensory disabilities.

The National Intellectual Disability Database (NIDD) provides information on specialised health services currently used or needed by people with intellectual disability. The database informs the regional and national planning of these services by providing information on trends in demographics, current service use and future service need.

The following information is provided:

- demographic profile of people with intellectual disability
- specialised health services received by people with intellectual disability
- waiting times for specialised health services.

The NIDD is not diagnosis-driven and therefore does not capture individuals with ASD. Individuals with intellectual disability and secondary diagnosis of ASD can be recorded on the NIDD, but ASD-specific data cannot be extracted.

The National Physical and Sensory Disability Database (NPSDD) provides information on specialised health services utilised by people with physical/sensory disabilities. As not every individual in Ireland who has a physical/sensory or speech and language disability is availing of, or requiring a specialised health and personal social service, and as the registration on to the database is voluntary, the NPSDD cannot provide any definitive epidemiological statement on the number of people with a particular type of disability. Therefore, the database may not cover a proportion of people living in Ireland who have a physical or sensory disability.

As both databases are voluntary, have specific criteria for inclusion, and do not collect diagnosis, it is clear that not all individuals with ASD will have their needs recorded on these national databases, and therefore they cannot assist in estimating incidence/ prevalence of ASD.

Plans are advanced to merge and refine the existing disability information systems into the NASS (National Ability Supports System). This will continue to collect data on the HSE funded specialist services for disability including day, respite, residential, multi-disciplinary, supports for daily living such as PA (personal assistance) and home support. It will capture diagnosis, including ASD. People can record more than one diagnosis to allow for comorbidity. The NASS will also be able to capture where in education those of school age are and are registered, which may make it possible to triangulate with National Council for

Special Education data at a later stage. However, the NASS is still voluntary (not epidemiological in its collection), so will only record those who have or need the services referred to above, and thus cannot be used for estimating prevalence rates.

2.2 Children in receipt of services

People with autism or autism spectrum disorders (ASD) may be served by public supports in a range of areas – education supports, disability or mental health supports, social welfare payments, free travel, housing or employment supports. Some of these programmes apply across a range of disabilities, but in education there is a range of services and provision that is specifically linked to a diagnosis of ASD.

National Council for Special Education (NCSE)

NCSE data on students with ASD with resource teaching support or in special classes and special schools indicate a prevalence rate of about 1.55 per cent and this figure is currently used for planning purposes. This calculation is based on school-aged children with ASD in state-funded schools between 4-18 years. It should be understood that this is a school population prevalence rate and is a best estimate based on administrative information available. It is not perfect, because while it excludes 3-year-olds in early intervention classes (170 children approximately), it doesn't take into the account the number of 4-year-olds that are not in school, or children aged 4 and older who are on the Home Tuition scheme and not included in overall school population figures:

- Total school population: (883,903 – 170 children aged 3 years) = 883,733
- Students with ASD: (13,873-170) = 13,703 (13,703 / 883,733) x 100 = 1.55%

Using data from the Growing Up in Ireland (GUI) study, the Economic and Social Research Institute (ESRI) analysed how the prevalence of special educational needs varies across social class and income groups. They found that the percentage of pupils reported by teachers to have special educational needs was significantly greater for those in schools serving disadvantaged areas. This has been taken into consideration by the Department of Education in a new model of additional teaching resources for schools (Dept. of Education Circular No. 0013/2017).

There has been a marked increase in the number of children accessing special education services (Table 1).

Table 1: Statistics on students in school diagnosed with ASD

	Mainstream Primary*	Mainstream Post Primary*	Special Classes	Special Schools	Total
2011/12	4,231	1,759	1,722	1,522	9,234
2012/13	4,919	2,148	2,113	1,866	11,046
2013/14	5,455	2,557	2,699	1,886	12,597
2014/2015	5,709	2,917	3,237	2,124	13,987
2015/16	6,487	3,341	3,983	2,283	16,094

**Mainstream figures are based on the number of students Accessing Low Incidence Teaching Hours with an ASD Diagnosis. Source: Department of Education (2017).*

In relation to the disability support for children with Special Educational Needs, in the past the Department of Education and Skills required an assessment and diagnosis for a number of reasons. The diagnosis clarified the condition and the assessment advised of the levels of severity and therefore informed decisions about the levels of supports which needed to be in place.

The three main support provisions were:

I. Placement

Decisions on whether the child can be supported in a mainstream class, a special class in a mainstream school or a special school were generally based on recommendations in the child's assessment. A diagnosis of autism would assist in placing the child in a special school or special class supporting children with autism.

II. Resource teaching

Where a child with autism attends a mainstream class s/he was entitled to a fixed level of resource teaching support provided to the school by the NCSE. Under the new model, the resources are provided directly to the school based on the level of needs of

the school. Accordingly, there is no longer a need for a diagnosis to drive an individual allocation to the school.

III. Special Needs Assistant (SNA) support

The SNA scheme is currently being reviewed to see whether children can be supported differently or better. The model for allocating SNAs is also under review to see whether the need for a diagnosis can be removed as was the case with the Resource teacher scheme. Other supports including Home Tuition and Assistive technology are also available to those with a diagnosis.

The Department of Education has introduced a revised model of allocation of additional teaching supports to schools. This model includes the use of a school's social context in the development of a school's educational profile, as the socioeconomic status of pupils is linked to the incidence of certain types of special educational needs. The Department also noted that international evidence clearly shows that there is a higher incidence of special educational needs among boys and this is taken account of also in the new allocation model.

The profiled allocation for schools from September 2017 is based on

- the number of pupils with complex needs enrolled to the school;
- the learning support needs of pupils as evidenced by standardised test results; and
- the social context of the school including disadvantage and gender.

Professional and other medical assessment or diagnosis of a particular condition will no longer be necessary for pupils to access educational teaching resources in schools, nor will there be a requirement for schools to submit assessments annually in order to apply for additional teaching resources. It is worth noting that, from the introduction of this model in September 2017, the NCSE will no longer have access to information on the number of students with ASD receiving additional teaching supports, i.e. the information on which its prevalence rate of 1.5% is based. It should also be noted that the allocation of individual Special Needs Assistants still requires that a child has been formally diagnosed.

Domiciliary Care Allowance

Domiciliary Care Allowance (DCA) is a monthly payment for a child aged under 16 with a severe disability who requires ongoing care and attention substantially over and above the care and attention usually required by a child of the same age. It is not means tested. The allowance is administered by the Department of Social Protection. The guidelines state that the payment is not based on the type of disability but on the resulting physical or mental

impairment, which means that the child requires substantially more care and attention than another child of the same age. See below for the rates of new beneficiaries to the DCA for autism from 2010. The rates in the table are per 1,000 children aged 0-15, as the payment is only made for children in this age group (Source: DEASP 2017).

Year of Decision	New Beneficiaries	Rate per 1,000 aged 0-15
2010	1387	1.4
2011	3444	3.3
2012	3619	3.5
2013	3550	3.4
2014	4615	4.4
2015	5695	5.4
2016	6858	6.4

The most remarkable observation on this data is the five-fold increase in the number of children eligible for DCA payment with a diagnosis of ASD in the seven-year period. This is most likely explained by an increase in awareness of the condition.

Other data sources were explored, including Hospital Inpatient Enquiry (HIPE), National Inpatient Psychiatric Reporting System (NIPRS), National Self Harm Registry and TILDA. The information recorded in these databases was not sufficient to establish a national prevalence rate.

Data from service providers was also assessed. This included referrals to speech and language therapy services of persons with a diagnosis of ASD. This information reflected the availability of service provision and could not be used to estimate the prevalence of ASD in the Irish population.

This section highlights the lack of availability of specific data to calculate a national prevalence rate for ASD which can be used for planning purposes. However, the rate of 1.5% used by the Department of Education represents a pragmatic approach used to estimate the need for extra resource teaching support. This figure is based on the number of children with a diagnosis of ASD within the school system and is similar to the rate estimated in a recent epidemiological study of 6-11-year old children.¹⁹

¹⁹ Sweeney and Staines, Autism Counts (DCU, 2016).

3.0 Epidemiological studies from Ireland

Several epidemiological studies have been identified in the literature which aim to measure the prevalence of ASD in Ireland. These studies employ different methodologies, diagnostic tools and age groups and as such are not directly comparable. The studies are summarised below.

3.1 Prevalence of ASD in Ireland

Fitzgerald et al. (1997) examined diagnostic, prevalence, psychosocial and service issues in relation to persons with autism in the Eastern Health Board (EHB) area in the period 1990-1992.²⁰ All centres in the EHB area with children and adults up to 25 years with special needs were identified and contacted regarding the study (n = 25). This study found 272 (4.9 per 10,000) persons in the age-range 6-25 who met diagnostic criteria for autistic disorder using the Autistic Disorders Diagnostic Checklist.²¹ DSM-IV-R and ICD-10 criteria gave a prevalence rate of 4-5 per 10,000. This rate is similar to rates from other countries in the same time period.

This study was updated by Fitzgerald et al. in 2001. Estimates of the prevalence rates for autism and ASD in Ireland, based on the number of persons availing of state, voluntary and private services in the Eastern region were 7.7 per 10,000. This represents a clear increase in the prevalence rate of 4.9/10,000 reported in 1992/1993 for the region. The prevalence rate found in these studies is based on cohorts of children attending clinics. This means the prevalence rate is based on children in receipt of services and does not take into consideration unmet need. In addition, it is also likely that some mild cases may have been missed and that staff may not have considered the possibility of autism in some persons with severe intellectual disability or indeed with normal intelligence, and also that some preschool children may not have come to anyone's attention with autism.

More recently, a study by Perry et al. (2007) assessed the feasibility of administering the Checklist for Autism in Toddlers (CHAT) at the 18-month developmental check to estimate the prevalence of diagnosed cases of autism.²¹ The CHAT was administered to 2117 infants. The overall prevalence of clinically diagnosed autism following this screening exercise was

²⁰ Fitzgerald, Michael, et al. (1997) Irish families under stress: Vol.6 Planning for the future of autistic persons: a prevalence and psychosocial study in the Eastern Health Board area of Dublin. Eastern Health Board. ²¹ Wing, L. (1997) The autistic spectrum. *The Lancet*. 350(9093):1761-1766.

²¹ Perry, I.J. et al. (2007) Screening for autistic spectrum disorder at the 18-month developmental assessment: a population-based study. *Irish Medical Journal*, 100 (8):565-567

33.1 per 10,000 (95% CI: 13.3 to 68.0).

CHAT, developed by Simon Baron-Cohen and colleagues, assesses simple joint attention and pretend play behaviours by parental report and health practitioner observation through direct testing. In the UK, Baird et al. screened 16,235 infants at 18 months using the CHAT instrument.²² Following the first administration of the instrument - 16,235 infants screened at 18 months - they reported a positive screening rate for autism (medium or high risk CHAT score) of 251 per 10,000 (95% CI: 226–274), a somewhat higher rate than that observed in the Irish study: 137.0 per 10,000 (95% CI: 91.9 to 196.1). Similar to the Irish study (Perry et al, 2007), a significant proportion of children who screened positive on first assessment did not return for a further assessment. In the UK, children who scored medium or high risk after two screenings (n=32) were given full clinical assessments at 42 months and 10 cases of autism were diagnosed. Thus, the UK screening exercise yielded 12 cases of autism per 10,000 children screened on at least one occasion (95% CI: 2.9 to 11.3) as compared with 33.1 per 10,000 (95% CI: 13.3 to 68.0) in the study by Perry et al. As a result, the yield in terms of previously undiagnosed cases in this 2007 study appears high relative to the earlier UK study. However, comparisons between the two studies are constrained by the differences in sampling strategies and dropout rates.

The most recent data on the prevalence of ASD in Ireland is reported in a study carried out by Dublin City University (Boilson et al. 2016).²³ This study used a protocol developed in Europe called the European Autism Prevalence Protocol (EPAP). The main purpose of the EPAP protocol, which was funded by DG Sante EU funding, was to facilitate a standardised approach to estimating ASD rates across Europe. Subsequently further EU commission funding permitted the establishment of the Autism Spectrum Disorders in Europe (ASDEU) project (<http://asdeu.eu/>). This trans-European programme involving universities, charities and expert institutions was established to increase understanding of autism.²⁴ The Irish study was the first to operationalise the screening phase of the protocol and validate the use of a screening instrument - the Social Communication Questionnaire (SCQ) - as a primary screener for ASDs among national school children.

In this research, a study booklet completed by the parents of eligible children aged 6-11 years was returned to the teacher for collection by the study team. There were (n=7,951) primary school children screened males 54% (n =4,268) females 46% (n=3,683), special education school children (n=189) males 66% (n=125) females 34% (n=64), in three regions:

²² Baird, et al. (2000) A screening instrument for autism at 18 months of age: a 6-year follow-up study.

²³ Boilson, A.M., et al. (2016) Operationalisation of the European Protocol for Autism Prevalence (EPAP) for Autism Spectrum Disorder Prevalence Measurement in Ireland *Journal of Autism and Developmental Disorders*, 46(9), 3054-3067.

²⁴ The European Autism Information Systems Project (Posada & Ramirez, 2008) highlighted the lack of systematic and reliable data on the prevalence of autism spectrum disorders in Europe. The EAIS project designed a protocol for the study of ASD prevalence at European level.

Galway, Waterford and Cork. Participation rates for parents of eligible children were 69% (n=5,457) for national schools, 36% (n=69) for special education schools.

The distribution of SCQ total scores for the national school sample were strongly skewed towards lower scores 4.65 ± 4.75 , range 0-36. The majority of children (92%) scored in the normal range (0 to 11) (n=5002), 4% scored in the moderate range (12-14) (n=225) and 4% scored in the high range (>15) score range (n=230). An optimal cut-off score (>13) differentiated ASD from other diagnosis [sensitivity 0.90, specificity 0.81, positive predictive value 0.43, and negative predictive value 0.98], [Test retest reliability mean interval: 15 months, Pearson's r of 0.77, df = 499, $p < 0.001$].

The authors concluded that the feasibility of screening children for ASDs with the EPAP protocol, using the SCQ in a non-clinical setting of Irish primary and special schools, was demonstrated. The importance of this study is that the screening questionnaire, i.e. the Social and Communication Questionnaire (SCQ), was validated for use in this population.

Furthermore, following this validation, the instrument was used to estimate the prevalence of ASD in a national school population aged 6-11 years. Both National and Special Education Schools were included in this study. Parents of almost 8,000 national school children in mainstream and special education schools across Cork, Waterford and Galway cities were administered the ASD screening questionnaire (SCQ) on behalf of their child. There was a 70% response rate. The total number of children identified with a diagnosis of Autism Spectrum Disorder was 63, giving a prevalence rate of 1.0%. Within Special Education schools in the study regions, 36 of the 69 children who participated were identified with an autism diagnosis, giving an estimated 52% prevalence rate for this population.

According to the authors of this study, the findings should be regarded as a minimum prevalence rate across the three study regions combined for the following reasons:

- while the overall response rates for National Schools were high, a significant number of children identified and invited to attend multi-disciplinary assessment did not avail of the assessment or were lost to follow-up
- response rates at the Special Education Schools were low at just 36% and therefore cannot be regarded as representative of the Special Education school population.

When further analysis was conducted (to weight for this low response rate) a figure of 1.5% resulted. The authors feel that this is a more accurate reflection of the prevalence rate in this particular cohort. This is similar to the Department of Education figure of 1.5% which is based on the numbers of children with a diagnosis of ASD who are availing of additional teaching supports in schools.

3.2 Prevalence of autism in Northern Ireland

'The Prevalence of Autism (including Asperger's Syndrome) in School Age Children in Northern Ireland 2018' was published in May 2018 by the UK Department of Health.²⁵ Statistics detailed within the publication were sourced from the Department of Education and include the number of school aged children identified with autism (including Asperger's) by Health and Social Care Trust area, Urban/Rural, Multiple Deprivation Measure, gender, school year and special educational needs assessment. The data shows a year-on-year increase in estimated prevalence of autism in school aged children in Northern Ireland.

- The estimated prevalence of autism within the school aged population in Northern Ireland has increased by 1.7 percentage points, from 1.2% in 2008/09 to 2.9% in 2017/18.
- There is a significant difference in the estimated prevalence rates of autism between the genders, with males four times more likely to be identified with autism (4.5%) than females (1.2%), in line with international findings.
- In general, prevalence across all school years was higher in 2017/18 compared with 2008/09. In 2017/18 the highest prevalence rate recorded was for those children aged 13 years and the lowest was for children aged 5 years. The results indicate that most identification of autism in school occurs when children are aged between 5 and 8 years old.
- The Northern Ireland urban population has a statistically significant higher prevalence rate than the rural population.
- Using the Northern Ireland Multiple Deprivation Measure (MDM) ranking 13% of the children identified with autism in 2017/18 were from the most deprived decile in Northern Ireland. The rate of autism in this most deprived decile was 31% higher than the Northern Ireland average.

There are several points worth noting about the above findings. Firstly, this report indicates an estimated prevalence of autism of 2.9% in school aged children in Northern Ireland, significantly higher than the 1.5% estimated in the ROI in the same age group. The difference in prevalence rate reported between Northern Ireland and the Republic of Ireland may be related to the introduction of the Autism Act (Northern Ireland) 2011. This was accompanied by an increase in awareness of autism via campaigns and consciousness raising events, and this may well have contributed to a rise in the number of assessments carried out and positive diagnoses processing through the system.

²⁵ The Prevalence of Autism (including Asperger Syndrome) in School Age Children in Northern Ireland 2018, Department of Health (Information and Analysis Directorate), May 2018 (available at <https://www.health-ni.gov.uk/topics/dhssps-statistics-andresearch>).

Secondly, the report highlights the much higher rates in children from lower socio-economic backgrounds. This, at first glance, might appear to be in contrast to results emanating from USA monitoring studies, which found a higher prevalence of autism in White children compared to Hispanic and other ethnic groups. However, this difference in the USA is thought likely to be related to the availability of diagnostic services rather than a true reflection of a higher incidence in White children. Socio-economic and gender differences in rates have been taken into consideration in the Department of Education's revised allocation model for additional teaching supports in schools. This revised model will be evaluated after two years when unmet need should be identified.²⁶

²⁶ Department of Education, Circular No. 0013/2017.

4.0 International databases

There are several international databases which have been reporting prevalence estimates over a period of time based on DSM and ICD criteria.

Autistic Developmental Disability Monitoring Network (ADDM)

One such database is the Autistic Developmental Disability Monitoring Network (ADDM) in the USA. The Children's Health Act (US) authorized CDC to create the ADDM Network in 2000. The ADDM Network is an active surveillance system that provides estimates of the prevalence and characteristics of ASD among children aged 8 years whose parents or guardians reside within 11 ADDM sites in the United States (selected counties or parts of counties in Arkansas, Arizona, Colorado, Georgia, Maryland, Missouri, New Jersey, North Carolina, South Carolina, Utah, and Wisconsin). The core surveillance activities in all ADDM Network sites focus on children aged 8 years because it has been demonstrated that this is the age of peak prevalence. While this database has limitations, e.g. only 11 States are included in the surveillance network and the population study is not representative of the total US population, it does nevertheless provide a comparable population-based ASD prevalence estimate from different sites every 2 years and evaluates how these estimates are changing over time.

Prevalence estimate from the ADDM

The most recent findings published in April 2018²⁷ estimate a prevalence rate of ASD in 8year olds of one in 59 (1.68%) for the surveillance year 2014. The findings from the ADDM Network in 2016 (for surveillance year 2012) had previously estimated that the percentage of children identified with autism spectrum disorder (ASD) was about 1 in 68 or 1.5%. This increase in estimated prevalence between 2012 and 2014 is reflective of a general increase across years since 2000, the first year of measurement. The estimated prevalence of ASDs increased 23% during 2006 to 2008 and 78% during 2002 to 2008. However, the two most recent surveys prior to the current findings had been suggestive of a plateauing prevalence at around 1.5%.

Fombonne (2018)²⁸, in an Editorial published after the CDC ADDM Network findings were released, set out a number of factors to consider when seeking to interpret the results. Firstly, he highlighted the fact that parents of children with autism have unusually high

²⁷ Baio et al., (April 2018) Prevalence Spectrum Disorder among children aged 8 years- Autism and Developmental Disabilities Monitoring Network, 11 sites, United States, 2014, Surveillance Summaries, 67 (6): 1-23

²⁸ E Fombonne (June 2018), Editorial: The rising prevalence of autism, Journal of Child Psychology and Psychiatry, Vol 59, Issue 7.

participation in surveys, making it plausible that non-participants have less autism than participants. In other words, he suggested that differential participation rates may have biased upwards prevalence estimates. Secondly, Fombonne noted that the CDC methodology excludes children without medical/education records, possibly resulting in under-ascertainment of cases. Despite being a unique attempt to maintain a consistent survey methodology across areas and over time, CDC results have consistently pointed to under-ascertainment in some States (e.g. Alabama), in ethnic minority groups and in lower socio-economic regions. Finally, Fombonne suggests that another limitation relates to the fact that case status confirmation does not require a direct assessment of participants with ASD. Record review is a procedure which can be employed to confirm diagnosis but in two previous validation studies conducted, 65% of subjects met the diagnostic criteria when directly assessed with standardised clinical tools in one study, whereas there was near full agreement in the other validation study. Taken together, Fombonne concludes that the upward trend in prevalence rates in the CDC Surveys are complex and difficult to interpret. Furthermore, he points out that, even though not stated explicitly in the most recent report, a preliminary estimate of 1.45% can be seen for estimation of prevalence of ASD determined by DSM 5 behavioural criteria, an 18% reduction compared to that reported using DSM-IV criteria in the same sub-sample.

iCARE – International

Data from six countries is collected by the International Collaboration for Autism Registry Epidemiology (iCARE). Denmark is project lead in this partnership, which collects national health registry data from Western Australia, Denmark, Finland, Norway, Sweden and Israel. The data encompasses 'Autism geographical and temporal heterogeneity, phenotype, family and life course patterns, etiology, birth weight, birth order, and age of diagnosis'.²⁹ The nature of the data gathered in each country is outlined in Appendix A (Schendel, 2013). Information sourced from Denmark, Finland and Sweden is from government-maintained registries that record diagnoses from in- or out-patient clinics or hospital contact. Israel, Norway and Western Australia retrieve diagnostic information from government-maintained registries which record those receiving services or benefits. (The data from Israel and Western Australia does not include non-Jewish and Aboriginal populations respectively.³⁰)

- The Danish National Psychiatric Register (DPR) holds electronic records since 1970 and outpatient records since 1995. ASD diagnosis is based on ICD-10. Previous prevalence estimates of ASD in Denmark were reported to be in the region of 1% but the most recent findings which were published in November 2018 found an autism

²⁹ Louise Gallagher, et al. (2014) Report of the Consultation for a National Registry and Biobank for Autism and Neurodevelopmental Disorders.

³⁰ D.E. Schendel (2013). The International Collaboration for Autism Registry Epidemiology (iCARE): Multinational Registry-Based Investigations of Autism Risk Factors and Trends. *J Autism Dev Disord.* Nov; 43(11): 2650–2663.

prevalence of 1.65% in 10-year olds (for 2016)³¹. The study authors mined the DPR and patient registries for every autism diagnosis in people born between 1980 to 2012, and they followed these individuals through to 2016. Between 1980 and 2012 2,055,928 people were born in Denmark, 31,961 of whom received an autism diagnosis. In addition to an overall increase in the estimated prevalence of autism to 1.65%, the findings indicate that the prevalence of autism seems to increase with age as they tracked people into adulthood. However, many global commentators have cautioned about interpretation of the findings which are suggestive of an apparent surge of diagnoses observed beyond childhood – 0.5 per cent of people born in Denmark from 1990-1991 received an autism diagnosis by 10 years of age. But, by the time individuals are 26 years of age the prevalence rate increases to 1.3 per cent, suggesting almost twice as many diagnoses are made between the ages of 10 and 26 as they are before age 10. Some international researchers have called for a clinical evaluation to confirm adult diagnoses to assist in the interpretation of the findings.

- The Finnish data is from the Hospital Discharge Registry 1969-1993 and Care Register for Health Care (1993+). Diagnoses are based on ICD-9 and ICD-10. Cumulative prevalence of ASD has been reported as over 1% in Finland.³²³³
- Norway is conducting an Autism Birth Cohort (ABC) study, which is a sub-study of the Norwegian Mother and Child Cohort (MoBa) study. This examines data on 114,500 children born between 1999 and 2009, with questionnaires conducted through pregnancy and childhood, and recorded blood samples. The Medical Birth Registry of Norway is the primary source for MoBa, but the Norwegian Patient Registry (which is mandated by law, with no consent or opt-out) is the major source of ASD cases in the ABC study. Diagnoses are per ICD-10. A 2012 study by the Norwegian Institute of Public Health estimates the cumulative incidence of autism to be 0.7% for children aged 6-11 years.³⁴ Since then, it has conducted a large record review study to determine the quality of recorded diagnoses, finding that 85% of diagnosed children meet the diagnostic criteria (DSM-IV) for autism spectrum disorders. Recent updates of the registry data indicate that the cumulative incidence is higher than estimated in 2012, with estimates between 1.0% and 1.2% from age 12 years and higher.³⁴
- The National Patient Register (Sweden) includes all inpatient data since 1987 and all outpatient data since 2001. Diagnosis is based on ICD-9 and ICD-10. A comparative

³¹ Schendel DE & Thorsteinsson E (2018) Cumulative incidence of autism into adulthood for birth cohorts in Denmark, 1980-2012, *JAMA* 320: 1811-1813

³² Atladóttir et al. (2015). The increasing prevalence of reported diagnoses of childhood psychiatric disorders: a descriptive multinational comparison. *Eur Child Adolesc Psychiatry*. 24:173-183. ³⁴ Surén, et al. 'Autism Spectrum Disorder, ADHD, Epilepsy, and Cerebral Palsy in Norwegian Children,' *Pediatrics*. 2012.

³³ .e152.

³⁴ Information supplied by Pål Surén, Norwegian Institute of Public Health.

paper published in 2015 suggests that Sweden's prevalence of ASD is 0.7% and of childhood autism is 0.3%.³⁵

- The Western Australia Register for ASD is established since 2013, and uses DSM-IV and DSM-V. It collects simple demographic and diagnostic information, such as date of birth, gender, primary language at home; diagnostic criteria used; diagnostic methods; IQ (verbal and non-verbal) and/or developmental abilities; other cognitive assessments; comorbidity; language assessments and adaptive behaviour. The 2015 comparative paper suggests Western Australia has a prevalence rate of 0.5% for ASD and 0.4% for childhood autism.³⁶

ASD-EU

Autism Spectrum Disorders in Europe (ASDEU) is a three-year programme run by a consortium of 20 groups from 14 countries (see Appendix B). ASDEU has received 2.1 million euros from the Directorate-General of Health and Consumers of the European Commission (DG-SANCO) to increase understanding of and improve responses to autism. It will study the prevalence of autism in 12 countries in the European Union; analyse the economic and social costs of autism; review existing arrangements and develop proposals for early detection programmes; train professionals; validate biomarkers for the disorder; and improve understanding of diagnosis, comorbidity, and effective care and support for adults and senior citizens with autism.

The lack of mechanisms to obtain consistent and reliable information about ASD trends at the European level is an important obstacle for the development of better and more equitable services. Hence, prevalence estimation across Europe and the development of a standardized strategy to be used for future surveillance of the ASD figures is a key objective of the ASD-EU programme.

Dublin City University is participating from Ireland and the results of the prevalence study (estimated prevalence of 1-1.5%) have been discussed in an earlier section (see Boilson et al (2016), p. 20).

Under the auspices of the ASDEU Study, the first population-based ASD prevalence study conducted in Italy indicates a prevalence of ASD in children aged 7-9 years of one in 87 (0.86%).³⁷

³⁵ Attladottir et al. (2015) The increasing prevalence of reported diagnoses of childhood psychiatric disorders: a descriptive multinational comparison. *Eur Child Adolesc Psychiatry*. 24:173-183.

³⁶ Ibid.

³⁷ Narzisi et al (2018) Prevalence of Autism Spectrum Disorder in a Large Italian Catchment Area- a School Based Population Study within the ASDEU Project, *Epidemiology and Psychiatric Sciences* (First View Published Online on 6 September 2018)

ASD-UK

ASD-UK is a research database administered in Newcastle University since 2011. It registers children aged 2-16 years at point of diagnosis (those with a diagnosis of autism, atypical autism, autism spectrum disorder, pervasive developmental disorder, or Asperger syndrome). The register is entirely voluntary, and a comparable adult database is in the process of being established. The information held includes the child's name and date of birth, parents'/carers' names and dates of birth, address and contact details, the type of ASD diagnosis and other medical conditions, information about the child's communication, developmental skills and behaviour. The register also asks for information on the school the child attends (if applicable) and on family members, such as siblings' names and dates of birth, and other family members with a diagnosis of ASD.

The ASD-UK website (<http://www.asd-uk.com/research/>) lists nine completed projects using this data source but none relate to incidence or prevalence (most probably because of the voluntary nature of the database).

Autism bio-collections

A bio-collection is a large set of biologically characterised samples, such as blood or tissue collected from a group of individuals who typically have a specific medical condition. Biocollections are useful as a dedicated resource to generate clinical and scientific data for the analysis of medical conditions on a large scale, as well as to create functional disease models to explore the biology of clinical conditions.³⁸ Large-scale bio-collections and associated comprehensive data that can aid the interrogation of the relationship between the genotype and phenotype effects at the individual and group levels can address the issue of heterogeneity. Bio-collections have been shown as valuable resources and have enabled large-scale studies on ASD. The recent genetic studies have begun to reveal de novo mutations on major cellular pathways. Examples of international autism bio-collections include Autism Genetic Resource Exchange (AGRE), Simons Simplex Collection (SSC), Danish Newborn Screening (NBS), Biobank Autism Inpatient Collection (AIC), and Autism Tissue Programme (ATP)/ Autism BrainNet.

³⁸ Lochmüller H, Schneiderat P. (2010) Biobanking in rare disorders. *Adv Exp Med Biol.* 686:105–13.

5.0 International studies of the prevalence of autism

The first studies of the prevalence of autism were published in the 1960s and 1970s, when autism was thought to be a very severe condition, usually accompanied by intellectual disability. These studies reported the prevalence to be approximately four to five cases per 10,000 children.³⁹ Appendix C includes a list of studies from around the world which have been conducted across five decades (1966-2016) to estimate the prevalence of ASDs. It is clear from the information that there has been a marked increase in the reported rates of ASD since the early 1990s. Furthermore, this paper has provided details of survey findings published in more recent times across the world, and their current estimates of prevalence. This includes:

- The most recent findings of the CDC ADDM Network, published in April 2018, estimate a prevalence rate of ASD of one in 59 of children aged 8 (1.68%) for the surveillance year 2014. The findings from the ADDM Network in 2016 (for surveillance year 2012) had previously estimated that the percentage of children identified with autism spectrum disorder (ASD) was about 1 in 68 or 1.5%.
- Previous prevalence estimates of ASD in Denmark were reported to be in the region of 1% but the most recent findings which were published in November 2018 found an autism prevalence of 1.65% in 10-year olds (for 2016).
- Cumulative prevalence of ASD has been reported as over 1% in Finland
- Recent updates of the registry data in Norway indicate that the cumulative incidence is estimated between 1.0% and 1.2% from age 12 years and higher.
- Sweden's prevalence of ASD in 2015 was 0.7%.
- Under the auspices of the ASDEU Study, the first population-based ASD prevalence study conducted in Italy indicates a prevalence of ASD in children aged 7-9 years of one in 87 (0.86%).
- Data from the UK Millennium Cohort Study, which is a sample of more than 19,000 children, representative of the UK population show that 1.7 % of parents reported that children aged 6-8 had been identified as having an Autism Spectrum Disorder.
- The estimated prevalence of autism within the school aged population in Northern Ireland is 2.9% in 2017/18.

³⁹ Gillberg C, Wing L. (1999). Autism: not an extremely rare disorder. *Acta Psychiatr Scand.* 99:399–406.

In addition to those studies previously described, the National Autism Spectrum Disorder Surveillance System (NASS) in Canada, which is a federally funded initiative set up to estimate and monitor the number of individuals with ASD, have recently reported their findings from 2015⁴⁰, focusing initially on those aged 5-17 years from six provinces and one territory. They reported that the combined prevalence of ASD is 1 in 66. In order to compare these results with the recently published CDC ADDM Network findings for 8 year olds in the United States, the Canadian authors analysed their prevalence estimates at 8 years of age and reported that this was slightly lower than the rate in the US, at 1 in 63.

In July 2018, Autism Spectrum Australia (ASPECT)⁴¹ revised its autism prevalence rates upwards from 1 in 100 to an estimated 1 in 70 to reflect recent changes in diagnostic criteria and new international research. They noted that the increase does not necessarily mean that autism is on the rise, but that increased awareness and more accurate diagnosis is at least partially important in explaining the rise.

To examine the hypothesis of a secular increase in the prevalence of autism, it is important to note that prevalence estimates will be inflated when case definition is broadened and case ascertainment is improved. Time trends in prevalence can, therefore, only be gauged in investigations that hold these parameters under strict control over time. These methodological requirements must be borne in mind while reviewing the evidence for a secular increase in the prevalence of ASDs. The discussion in the following section is adapted from an article by Fombonne and colleagues in 2009⁴², where they reviewed and discussed the various methodologies, surveys and prevalence figures reported.

Study descriptions

Studies have been conducted in many countries and over half of the results have been published since 2000. Most study populations are urban based. The age range of the population included in the studies range from birth to early adult life but most surveys have included school-age samples with an overall median age of eight years. There is a large variation in the size of the population surveyed. Studies with small sample sizes tended to yield higher prevalence rates than studies with larger sample sizes.⁴³

⁴⁰ Ofner et al. (2018) Autism Spectrum Disorder among children and youth in Canada 2018- A report of the National Autism Spectrum Disorder Surveillance System, Public Health Agency of Canada

⁴¹ Available at <http://autismspectrum.org.au>

⁴² Fombonne E, Quirke S & Hagen A (2009) Prevalence and interpretation of recent trends in rates of pervasive developmental disorders, *McGill J Med*, 12(2): 73

⁴³ Fombonne, E. (2005) Epidemiology of autistic disorder and other pervasive developmental disorders. *J Clin Psychiatry* 66:3-8.

Study designs

Some studies have relied on existing administrative databases⁴⁴ or on national registers⁴⁵ for case identification. Most investigations of ASD have relied on a two-stage or multistage approach to identify cases in underlying populations. The first screening stage of these studies often consisted of sending letters or brief screening scales requesting school and health professionals to identify possible cases of autism. Each investigation varied in several key aspects of this screening stage. First, the coverage of the population varied enormously from one study to another. In addition, the surveyed areas varied in terms of service development as a function of the specific educational or health care systems of each country and of the year of investigation. Second, the type of information sent out to professionals invited to identify children varied from simple letters including a few clinical descriptors of autism-related symptoms or diagnostic checklists rephrased in nontechnical terms, to more systematic screening based on questionnaires or rating scales of known reliability and validity. Third, participation rates in the first screening stages were variable although refusal rates tended to be very low.

Few studies provided an estimate of the reliability of the screening instrument. The sensitivity of the screening methodology is also difficult to gauge in autism surveys, as the proportion of false negatives was usually not estimated. Prevalence estimates must, therefore, be seen as underestimates of “true” prevalence proportions.

Participation rates in second stage assessments were also generally high. The source of information used to determine a case usually involved a combination of informants and data sources, with a direct assessment of the person with autism in about half of the studies. However, surveys of large populations⁴⁶ did not include a direct diagnostic assessment of subjects. Nevertheless, the methods developed by the Centre for Disease Controls for recent large surveys, including the ADDM Network, rely on a multisource, multi-informant screening of the population, abstraction of key developmental data, and review by panels of expert clinicians who apply best-estimate procedures of known reliability and validity.

The assessments were conducted with various diagnostic instruments, ranging from a classical clinical examination to the use of a variety of standardized measures that included, in the most recent studies, gold standard diagnostic tools such as the Autism Diagnostic Interview-Revised (ADI-R) or the Autism Diagnostic Observational Schedule (ADOS). The

⁴⁴ Croen, L. A., et al. (2002) The changing prevalence of autism in California. *J Autism Dev Disord* 32:307-215; Gurney, J.G., et al. (2003) Analysis of prevalence trends of autism spectrum disorder in Minnesota. *Arch Pediatr Adolesc Med* 157:622-627; Fombonne E, et al. (2006) Pervasive developmental disorders in Montreal, Quebec, Canada: prevalence and links with immunizations. *Pediatrics* 118: e139-e150.

⁴⁵ Madsen, K. M., et al. (2002) A Population-based study of measles, mumps, and rubella vaccination and autism. *N Engl J Med* 347:1477-1482.

⁴⁶ Autism and Developmental Disabilities Monitoring Network Surveillance Year 2000 Principal Investigators; Centre for Disease Control and Prevention 2007 Prevalence of autism spectrum disorders – autism and developmental disabilities monitoring networks, 6 sites, United States, 2000. *MMWR Surveill Summ* 56:1-11; Autism and Developmental Disabilities Monitoring Network Surveillance Year 2002 Principal Investigators; Centre for Disease Control and Prevention 2007 Prevalence of autism spectrum disorders – autism and developmental disabilities monitoring network, 14 sites, United States, 2002. *MMWR Surveill Summ* 56:12-28.

precise diagnostic criteria retained to define a case varied according to the study and, to a large extent, reflected historical changes in classification systems. Thus, Kanner's criteria, Lotter's, and Rutter's definitions were used in surveys conducted before 1980, whereas DSM-based definitions took over thereafter as well as ICD-10 since 1990. As described earlier, Kielinen et al. (2000)⁴⁷ have shown that a 2- to 3-fold variation in prevalence of autism can result from applying different diagnostic criteria to the same survey data.

In a recent publication from China, where authors conducted a meta-analysis of the pooled prevalence of ASD in the general population in China (with mean age from 1.6-8 years), the pooled prevalence of ASD was 39.23 per 10,000, lower than reported by all other countries to date⁴⁸. However, it is mooted that this lower estimate might be explained, in part, by the use of different screening tools. The most common screening tool used in China is the CABS, but this is rarely, if ever, used in Western studies (where the ADOS or ADI-R are widely used).

Referral statistics

Increasing numbers of children referred to specialist services or known to special education registers have been taken as evidence for an increased incidence of autism spectrum disorders. However, trends over time in referred samples are confounded by many factors such as referral patterns, availability of services, heightened public awareness, decreasing age at diagnosis, and changes over time in diagnostic concepts and practices, to name only a few.

Strong evidence of “diagnostic switching” was produced in California⁴⁹ and in all US states,⁵⁰ indicating that a relatively high proportion of children previously diagnosed as having intellectual disability are now identified as having an ASD diagnosis. Decreased age at diagnosis has also been shown to contribute to the rising numbers of children diagnosed with ASD.⁵¹ In the United Kingdom, Jick et al. (2003)⁵² have shown that the incidence of specific developmental disorders (including language disorders) decreased by about the same amount that the incidence of diagnoses of autism increased in boys born from 1990 to 1997. A more recent UK study⁵³ has shown that up to 66% of adults previously diagnosed with developmental language disorders would meet diagnostic criteria for a broad definition

⁴⁷ Kielinen, M. et al. (2000) Autism in Northern Finland. *Eur Child Adolesc Psychiatry* 9:162-167.

⁴⁸ Fei Wang et al (2018) The Prevalence of Autism Spectrum Disorders in China: a comprehensive meta-analysis. *Int J Biol Sci*, 14 (7): 717-725.

⁴⁹ Eagle, R.S. (2004) Commentary: Further commentary on the debate regarding increase in autism in California. *J Autism Dev Disord* 34:87-88.

⁵⁰ Shattuck, P.T. (2006) Diagnostic substitution and changing autism prevalence. *Pediatrics* 117:1438-1439.

⁵¹ Wazana, A., et al. (2007) The autism epidemic: fact or artefact? *J Am Acad Child Adolesc Psychiatry* 46:721-730

⁵² Jick, H., et al. (2003) Epidemiology and possible causes of autism: changes in risk of autism in the UK for birth cohorts 1990-1998. *Pharmacotherapy* 23:1524-1530.

⁵³ Bishop, D.V., et al. (2008) Autism and diagnostic substitution: evidence from a study of adults with a history of developmental language disorder. *Dev Med Child Neurol* 50:341-345.

of ASD. Overall, evidence from these referral statistics is very weak and proper epidemiologic studies are needed to assess secular changes in the incidence of a disorder.

Several analyses of special educational disability in the United States⁵⁴ showed increased numbers of ASD children in schools, but the increase was not specific to autism. These analyses also showed a marked period effect that identified the early 1990s as the period where the prevalence estimates started to go up in all ages and birth cohorts, coinciding closely with the inclusion of ASDs in the federal Individual with Disabilities Educational Act (IDEA) funding and reporting mechanism in the United States.

International collaboration

The iCARE partners published a comparative prevalence paper in 2015; this used a population-based cohort including all live-born children in Denmark, Finland, Sweden and Western Australia, from January 1, 1990 through December 31, 2007, and followed through December 31, 2011.⁵⁵ The main outcome measure was age-specific prevalence of diagnoses reported to population-based registry systems in each country. The geographical comparison was enhanced by harmonization of the study period and data prior to analysis (including diagnostic codes used to define the conditions) and application of a uniform statistical analytic approach across the multiple datasets. The authors observed that the cumulative prevalence of ASD in the oldest cohorts was well over 1% in Finland and Sweden, and over 1.5% in Denmark, with little evidence for a plateau in the prevalence curves even in the oldest cohorts. The proportion of childhood autism observed in Denmark, Sweden and Finland was comparable to what was previously reported (30%). Western Australia displayed a higher proportion of childhood autism; over 80% of the ASD cases from Australia were diagnosed with childhood autism. This pattern may relate to the availability of funding for therapy services for younger children (less than 5 years) with ASD in Western Australia.

A comparison of autism prevalence trends in Denmark and Western Australia was also reported as part of this collaboration.⁵⁶ This study compared prevalence statistics for two distinct geographical regions, Denmark and Western Australia, both of which have had population-based registers and consistent classification systems operating over the past decade. Overall ASD prevalence rates were higher in Denmark (68.5 per 10,000 children) compared with Western Australia (51.0 per 10,000 children), while the diagnosis of childhood autism was more prevalent in Western Australia (39.3 per 10,000 children) compared with Denmark (21.8 per 10,000 children). The authors concluded that these differences are

⁵⁴ Gurney, J.G., et al. (2003) Analysis of prevalence trends of autism spectrum disorder in Minnesota. *Arch Pediatr Adolesc Med* 157:622-627; Shattuck, P.T. (2006) Diagnostic substitution and changing autism prevalence. *Pediatrics* 117:1438-1439.

⁵⁵ Atladóttir, et al. (2015) The increasing prevalence of reported diagnoses of childhood psychiatric disorders: a descriptive multinational comparison. *Eur Child Adolesc Psychiatry*. 24: 173-183.

⁵⁶ Parner et al. (2011) A comparison of autism prevalence trends in Denmark and Western Australia. *Autism Dev. Disord.* Dec. 41(12): 1601-8.

probably caused by local phenomena affecting case ascertainment but influence from biological or geographical factors may exist.

Another study which uses pooled data from many studies attempts to develop global and regional prevalence models and estimate the global burden of ASDs.⁵⁷ A systematic review was conducted for epidemiological data (prevalence, incidence, remission and mortality risk) of autistic disorder and other ASDs. Data were pooled using a Bayesian meta-regression approach while adjusting for between-study variance to derive prevalence models. The study found that in 2010 there were an estimated 52 million cases of ASDs, equating to a prevalence of 7.6 per 1000 or one in 132 persons. After accounting for methodological variations, there was no clear evidence of a change in prevalence for autistic disorder or other ASDs between 1990 and 2010. Worldwide, there was little regional variation in the prevalence of ASDs. Globally, autistic disorders accounted for more than 58 DALYs (disability-adjusted life years) per 100 000 population and other ASDs accounted for 53 DALYs per 100 000.

Comparison of cross-sectional epidemiologic studies

As discussed earlier, epidemiologic studies of ASD each possess unique design features which could account almost entirely for between-studies variations in prevalence proportions. The time trends in the prevalence of autism are, therefore, difficult to gauge from published prevalence estimates. The significant correlation between prevalence of ASD and year of publication could merely reflect increased efficiency over time in case identification methods used in surveys as well as changes in diagnostic concepts and practices.⁵⁸ In studies using capture-recapture methods, it is apparent that up to one third of prevalent cases may be missed by an ascertainment source, even in recently conducted studies.⁵⁹

The most convincing evidence that method factors could account for most of the variability in published prevalence estimates comes from a direct comparison of eight recent surveys conducted in the United Kingdom and the United States.⁶⁰ In each country, four surveys were conducted around the same year and with similar age groups. As there is no reason to expect huge between-area differences in prevalence, prevalence estimates should, therefore, be comparable within each country. However, there was a 6-fold variation in prevalence for UK surveys and a 14-fold variation in US figures. In each set of studies, high

⁵⁷ A.J. Baxter, et al. (2015) The Epidemiology and Global Burden of Autism Spectrum Disorders. *Psychological Medicine*. Feb. 45(3): 601-613.

⁵⁸ Kielinen, M. et al. (2000) Autism in Northern Finland. *Eur Child Adolesc Psychiatry* 9:162-167; Webb, E.V., et al. (1997) The changing prevalence of autistic disorder in a Welsh health district. *Dev Med Child Neurol*. 39:150-152; Magnusson, P., et al. (2001) Prevalence of autism in Iceland. *J Autism Dev Disord* 31:153-163; Shattuck, P.T. (2006) Diagnostic substitution and changing autism prevalence. *Pediatrics* 117:1438-1439; Bishop, D.V., et al. (2008) Autism and diagnostic substitution: evidence from a study of adults with a history of developmental language disorder. *Dev Med Child Neurol* 50:341-345.

⁵⁹ Harrison, M.J., et al. (2006) Prevalence of autistics spectrum disorders in Lothian, Scotland: an estimate using the "capture-recapture" technique. *Arch Dis Child* 91:16-19.

⁶⁰ Fombonne, E. (2005) Epidemiology of autistic disorder and other pervasive developmental disorders. *J Clin Psychiatry* 66:3-8.

estimates derived from surveys where intensive population-based screening techniques were used, whereas lower prevalence proportions were obtained from studies relying on passive administrative methods for case finding. Because no passage of time was involved, the magnitude of these gradients in prevalence can only be attributed to differences in case identification methods across surveys.

Even more convincing evidence comes from the large survey by the CDC on 408,000 US children aged 8 and born in 1994⁶¹ where an average prevalence of 66/10,000 was reported for 14 US states. However, there was more than a 3-fold variation in state-specific prevalence proportions that ranged from a low 33/10,000 for Alabama to a high of 106/10,000 in New Jersey. These substantial differences reflected ascertainment variability across sites in a study that was otherwise performed with the same methods and at the same time, and in children born in the same year. Thus, no inference on trends in the incidence of ASDs can be derived from a simple comparison of prevalence estimates over time, since studies conducted at different periods are likely to differ even more with respect to their methodology.

Repeat studies in defined geographical areas

Repeated studies, using the same methodology and conducted in the same geographical area at different points in time, can potentially yield useful information on time trends provided that methods are kept relatively constant. The Göteborg studies⁶² provided three prevalence estimates, which increased over a short period of time. However, different age groups were included in each survey. Other factors such as improved detection among the intellectually disabled, cases born to immigrant parents, change in local services, and a progressive broadening of the definition of autism over time were hypothesized by the authors to account for the trend.⁶³ Similarly, studies conducted in Japan at different points in time in Toyota⁶⁴ and Yokohama⁶⁵ showed rises in prevalence that their authors interpreted as reflecting the effect of both improved population screening of pre-schoolers and of a broadening of diagnostic concepts and criteria.

⁶¹ Ibid. [Note, the latest figures from the CDC (2014) estimate that 14.7 per 1,000 8 year olds have been identified with ASD.]

⁶² Gillberg, C. (1984) Infantile autism and other childhood psychoses in a Swedish urban region. Epidemiological aspects. *J Child Psychol Psychiatry* 25:35-43; Gillberg, C., et al. (1991) Is autism more common now than ten years ago? *Br J Psychiatry* 158:403-409.

⁶³ Gillberg (1991).

⁶⁴ Kawamura Y., et al. (2008) Reevaluating the incidence of pervasive developmental disorders: impact of elevated rates of detection through implementation of an integrated system of screening in Toyota, Japan. *Psychiatry Clin Neurosci* 62:152-159.

⁶⁵ Honda, H., et al. (1996) Cumulative incidence and prevalence of childhood autism in children in Japan. *Br J Psychiatry* 169:228-235; Honda, H., et al. (2005) No effect of MMR withdrawal on the incidence of autism: a total population study. *J Child Psychol Psychiatry* 46:572-579.

Successive birth cohorts

In large surveys encompassing a wide age range, increasing prevalence among most recent birth cohorts could be interpreted as indicating a secular increase in the incidence of the disorder, provided that alternative explanations can confidently be ruled out. This analysis was used in two large French surveys.⁶⁶ The surveys included birth cohorts from 1972 to 1985 (735,000 children, 389 of whom had autism) and, pooling the data of both surveys, age-specific prevalence showed no upward trend.

Two separate surveys of children born 1992–1995 and 1996–1998 in Staffordshire in the United Kingdom⁶⁷ were performed with rigorously identical methods for case definition and case identification. The prevalence for combined ASDs was comparable and not statistically different in the two surveys,⁶⁸ suggesting no upward trend in overall prevalence of ASDs during the studies time interval.

Data from the UK Millennium Cohort Study, which is a sample of more than 19,000 children, representative of the UK population show that 1.7 % of parents reported that children aged 68 had been identified as having an Autism Spectrum Disorder.

Incidence studies

Incidence can be difficult to measure with rarer chronic diseases such as autism. In autism epidemiology, point or period prevalence is more useful than incidence, as the disorder starts long before it is diagnosed, and the gap between initiation and diagnosis is influenced by many factors unrelated to risk. Subsequently, on reviewing the literature on the subject we have concentrated on the prevalence literature.

However, the few incidence studies which have been conducted showed an upward trend in incidence over short periods of time. In one of the largest study of 1,410 subjects, there was a 10-fold increase in the rate of first recorded diagnoses of Pervasive Developmental Disorders (DSM IV) in United Kingdom general practice medical records from 1988–1992 to 2000–2001.⁶⁹ The increase was more marked for PDDs other than autism, but the increase in autism was also obvious. However, none of these studies' investigations could determine

⁶⁶ Fombonne E., et al. (1992) Prevalence of infantile autism in four French regions. *Soc Psychiatry Psychiatr Epidemiol* 27:203210; Fombonne, E., et al. (1997) Autism and associated medical disorders in a French epidemiological survey. *J Am Acad Child Adolesc Psychiatry* 36:1561-1569.

⁶⁷ Chakrabarti, S., et al. (2001) Pervasive developmental disorders in preschool children. *JAMA* 285:3093-3099.

⁶⁸ Chakrabarti, S., et al. (2005) Pervasive developmental disorders in preschool children: confirmation of high prevalence. *Am J Psychiatry* 162:1133-1141.

⁶⁹ Smeeth, L., et al. (2004) MMR vaccination and pervasive developmental disorders: a case-control study. *Lancet* 364:963-969.
⁷¹ Powell, J.E., et al. (2000) Changes in the incidence of childhood autism and other autistic spectrum disorders in preschool children from two areas of the West Midlands, UK. *Dev Med Child Neurol* 42:624-628; Barbaresi, W.J., et al. (2005) The incidence of autism in Olmsted Country, Minnesota 1976-1997: results from a population-based study. *Arch Pediatr Adolesc Med* 159:3744; Honda, H., et al. (2005); Wong, V.C., et al. (2008) Epidemiological study of autism spectrum disorder in China. *J Child Neurol* 23:67-72.

the impact of changes over time in diagnostic criteria, improved awareness and service availability on the upward trend. The same conclusions apply to other incidence studies.⁷¹

In a population study using the UK General Practice Research Database (GPRD), Taylor et al. (2013) estimated the annual autism prevalence rates for children aged 8 years in 2004-2010.⁷⁰ Annual prevalence rates for each year were steady at approximately 3.8/1000 boys and 0.8/1000 girls. Annual incidence rates each year were also steady at about 1.2/1000 boys and 0.2/1000 girls. The authors concluded that following a reported fivefold increase in the annual incidence rates of autism during the 1990s in the UK, the incidence and prevalence rates in 8-year-old children reached a plateau in the early 2000s and remained steady through 2010.

Current evidence does not strongly support the hypothesis of a secular increase in the incidence of autism but power to detect time trends is seriously limited in existing datasets. Although it is clear that prevalence estimates have gone up over time, this increase most likely represents changes in the concepts, definitions, service availability, and awareness of autistic-spectrum disorders in both the lay and professional public. To assess whether the incidence has increased, method factors that account for an important proportion of the variability in prevalence must be tightly controlled. The possibility that a true change in the underlying incidence has contributed to higher prevalence figures remains, however, to be adequately tested.

Prevalence of autism in adults

Although early ASD research focused primarily on children, there is increasing recognition that ASD is a lifelong neurodevelopmental disorder. A community study carried out in 2011 by Brugha et al. found that autism spectrum disorder affects approximately 1% of the adult English household population.⁷¹ There was no evidence of a statistically significant reduction in prevalence of ASDs as a function of age. There are no similar studies in the literature to compare the findings, but the authors conclude that the study demonstrates for the first time in the general population that the rate of ASD is not significantly associated with age, suggesting that the causes of autism are temporally constant. This supports the argument that the reported increase in prevalence rate of autism internationally is more likely to be due to increased recognition of ASD and subsequently an increase in the rate of diagnosis.

In addition, an interesting paper has recently been published by Hirvikoski and colleagues (2016) from Sweden.⁷² This paper demonstrates increased mortality from almost all causes in persons diagnosed with ASD compared to the general population. This leads the authors

⁷⁰ Taylor, B. et al. (2013) Prevalence and incidence rates of autism in the UK: time trend from 2004-2010 in children aged 8 years. *BMJ Open*. Oct 16;3(10).

⁷¹ Brugha, et al. (2011) Epidemiology of Autism Spectrum Disorders in Adults in the Community in England. *ARCH GEN PSYCHIATRY*. 68(5), May.

⁷² Hirvikoski, et al. (2016) Premature mortality in autism spectrum disorder. *Br J Psychiatry*. Mar;208(3):232-8.

to conclude that persons with ASD may have increased biological vulnerability, as well as insufficient awareness, diagnosis and treatment of comorbid diseases within the health care system.

6.0 Discussion

The previous sections have highlighted the challenges associated with estimating the prevalence of ASD in a population. Studies conducted from many countries have demonstrated the reported increase in prevalence of ASD diagnosis over the past two decades.

However, direct comparisons between countries and studies is limited by the variations in populations and age groups studied, in screening and diagnostic tools employed which have not been validated, and the lack of follow-up in many studies which is necessary to increase positive predictive value.

6.1 Possible reasons for a reported increase in prevalence of autism

In many countries, a diagnosis of autism gives children greater access to specialised services and special education services than do diagnoses of other conditions. This benefit makes clinicians more likely to diagnose a child with autism, even those who are on the borderline of the clinical criteria. In 1991, the U.S. Department of Education ruled that a diagnosis of autism qualifies a child for special education services. The change may have encouraged families to get a diagnosis of autism for their child. The number of children who have both a diagnosis of autism and intellectual disability has also risen steadily over the years.

As described earlier in the report, access to resource teaching and a special needs assistant in the Irish education system required a diagnosis. It is possible that this has also been a factor in the increase in the reported prevalence in the Irish context.

Policy changes may have also played a role. In 2006, the American Academy of Paediatrics recommended screening all children for autism during routine paediatrician visits at 18 and 24 months of age. This move may have led to diagnoses for children who would otherwise have slipped under the radar.

6.2 Reported increase in prevalence rates in Ireland

It is clear that policy changes and associated legislation have in part contributed to the reported increase in prevalence of ASD globally. A similar trend has occurred in Ireland.

Fitzgerald, et al.'s 1990-92 study found a rate of 4.9 per 10,000 persons in age range 6-25 years. This had risen to 7.7 per 10,000 in 2001 in the same group.⁷³ A study in 2007 by Perry, et al., using a different age cohort and methodology, found a prevalence value of 33.1 per 10,000, and more recently a study by DCU found a rate of 1.0% in 6-11-year olds.⁷⁴

In Ireland there have been several commissioned reports over the past number of years which have examined the issue of ASD. This has resulted in greater parental and professional awareness.⁷⁵ As highlighted throughout this paper, changes in reported ASD prevalence over time may have several explanations: broadening diagnostic criteria and differences in the methods used to study prevalence, e.g. sampling procedures, application of statistical methods.

Despite these challenges it is important to agree a national prevalence rate in order to plan services (social, educational, health) for this group of individuals. The Department of Education has used a prevalence rate of 1.5% based on the number of children with a diagnosis of ASD who are currently accessing special education services. This is a pragmatic approach, and it is similar to international rates quoted in the literature. In addition the most recent epidemiological study conducted in Ireland to estimate the prevalence of ASD (Boilson et al.) has reported a similar rate. As discussed earlier, this study uses a screening instrument which has been validated in the population used and is part of a European wide project. This may provide a valid method to monitor ASD prevalence rates in Ireland in the future.

There is currently no reliable method of estimating prevalence of Autism Spectrum Disorder in Ireland. In this paper, we have used a combination of methods to arrive at a figure which may be used for assessing need and planning appropriate services. However, it is important that an accurate method of determining prevalence of ASDs is agreed, and that the prevalence rate is kept under review.

6.3 Policy implications

An extremely wide range of individual differences is represented within the Autistic Spectrum Disorder categorization, from individuals who also have a severe learning disability to those with average and above average intelligence. All share the triad of difficulties in reciprocal social interaction, communication, and a lack of flexible thinking. ASDs impact on all areas of

⁷³ Fitzgerald, Michael, et al. (1997) Irish families under stress: Vol.6 Planning for the future of autistic persons: a prevalence and psychosocial study in the Eastern Health Board area of Dublin. Eastern Health Board.

⁷⁴ Perry, I.J. et al. (2007) 'Screening for autistic spectrum disorder at the 18-month developmental assessment: a populationbased study'. Irish Medical Journal, 100 (8):565-567

⁷⁵ The Current State of Autism Services in Laois/Offaly (LOFFA, 2015); National Review of Autism Services: Past, Present and Way Forward (HSE, 2012); Review of Services for Persons with Autistic Spectrum Disorder (ERHA, 2002); Keenan, et al. Meeting the Needs of Families Living with Children with Autism Spectrum Disorder (2007).

functioning and have significant implications throughout the lives of those affected across the entire ability range.

It is crucial to accurately identify children so that they can access evidence-based early interventions. However, there are also significant economic and social costs if children are allocated services based on a diagnosis without consideration of their severity level or specific needs.

Unmet need of adults with ASD requires to be documented in Ireland.

Table 2

Characteristics of the data contributing sites in the International Collaboration for Autism Registry Epidemiology (iCARE)

Site	Denmark	Finland	Israel	Norway	Sweden	Western Australia (WA) ^a
Catchment area	Nation-wide	Nation-wide	Nation-wide	Nation-wide	Nation-wide	State-wide
Average births/year	62,000	60,000	86,000	61,000	107,000	24,000
Country of birth or ethnic profile	90 % Danish	95 % born in Finland 3 % born else-where in Europe 2 % born outside Europe	98 % born in Israel 2 % born elsewhere	88 % Norwegian 12 % other	> 90 % born in Sweden < 10 % born elsewhere	93 % Caucasian 7 % other
Health care provision	Public	Public	Public	Public	Public	Public and private
Type of study population	Birth cohort	Birth cohort	Birth cohort	Birth cohort	Birth cohort	Birth cohort
Data source of study population	Danish Medical Birth Registry	Finnish Medical Birth Registry	Israel Birth Register	Medical Birth Registry of Norway	Swedish Patient Registry	Midwives' Notification System
<i>Characteristics of data source of autism spectrum disorders (ASDs) diagnosis for individuals in iCARE birth cohort study populations</i>						
Name	Danish Psychiatric Central Registry	Finnish Hospital Discharge Registry	Israeli Ministry of Social Affairs	Norwegian National Insurance Scheme (Norwegian Patient Register to be used when available)	Swedish Hospital Discharge Register	1 Disability Services Commission of WA 2 WA Register for ASD
Type	Medical discharge registry	Medical discharge registry	Service registry	Benefit registry	Medical discharge registry	1 Service registry 2 Diagnosis registry
Steward	Government agency	Government agency	Government agency	Government agency	Government agency	1 Government agency 2 Telethon Institute for Child Health Research
Diagnostic system for iCARE cohorts	ICD-8: 1987-1993 ICD-10: 1994-present	ICD-9: 1987-1995 ICD-10: 1996-present	DSM-IV	ICD-9: 1987-1992 ICD-10: 1992-present	ICD-9: 1987-96 ICD-10: 1997-present	DSM-IV (individuals diagnosed under earlier diagnostic systems were reclassified to DSM IV by the data sources)
Types of contact reported to source	Inpatient: 1987-present Outpatient: 1995-present Does not include contacts with general practitioners (the primary care providers)	Inpatient: 1987-present Outpatient: 1998-present Does not include contacts within primary health care (before 2011) and private outpatient visits outside hospitals contacts	All registrations for services; likely includes more severely impaired individuals Individuals with ASD not receiving benefits are not recorded	All registrations for benefits Individuals with ASD not receiving benefits are not recorded.	Inpatient: 1987-present Outpatient: 2000-present Does not include contacts with general practitioners (the primary care providers)	All registrations for services

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Author	Year	Country	Period Studied	Age range	No. of children	Criteria	Methodology	Prevalence (per 1000)
Lotter	1966	England	1964	8 to 10	78,000	Kanner	Case enumeration & direct exam	0.45 (0.31-0.62)
Brask	1970	Denmark	1962	2 to 134	46,500	Kanner	Case enumeration	0.43 (0.26-0.66)
Treffert	1970	USA	1962-67	3 to 12	899,750	Kanner	Case enumeration	0.07-0.31 (0.0-1.0)
Wing & Gould	1979	England	1970	0 to 14	35,000	Kanner	Case enumeration & direct exam	0.49 (0.29-0.78)
Hoshino et al. (1)	1982	Japan	1977	0 to 17	234,039	Kanner	Case enumeration & direct exam	0.23 (0.19-0.27)
Ishii & Takahashi	1983	Japan	1981	6 to 12	35,000	Rutter	Case enumeration & direct exam	1.6 (1.2-2.8)
Bohman et al.	1983	Sweden	1979	0 to 20	69,000	Rutter	Case enumeration & direct exam	0.3 (0.2-0.5)
McCarthy et al.	1984	Ireland	1978	8 to 10	65,000	Kanner	Case enumeration & direct exam	0.43 (0.29-0.59)
Gillberg	1984	Sweden	1980	4 to 18	128,584	DSM-III	Case enumeration & direct exam	0.20 (0.13-0.30)

Author	Year	Country	Period Studied	Age range	No. of children	Criteria	Methodology	Prevalence (per 1000)
Steffenburg & Gillberg	1986	Sweden	1984	<10	78,413	DSM-III	Case enumeration & direct exam	0.45 (0.31-0.62)
Matsuishi et al.	1987	Japan	1983	4 to 12	32,834	DSM-III	Case enumeration & direct exam	1.55 (1.16-1.64)
Burd et al.	1987	USA	1985	2 to 18	180,986	DSM-III	Case enumeration & direct exam	0.12 (0.00-0.20)
Bryson et al.	1988	Canada	1985	6 to 14	20,800	DSM-III	Case enumeration and direct exam	1.01 (0.62-1.54)
Bryson et al.	1988	Canada	1985	6 to 14	20,800	DSM-III	Case enumeration and direct exam	1.01 (0.62-1.54)
Tanoue et al.	1988	Japan	1977-85	3 to 7	95,394	DSM-III	Case enumeration	1.38 (1.16-1.64)
Ciadella & Mamelle	1989	France	1986	3 to 9	135,180	DSM-III	Case enumeration	0.51 (0.39-0.63)
Sugiyama & Abe	1989	Japan	1979-84	2 to 5	12,263	DSM-III	Population screen & direct exam	1.3 (0.7-2.1)
Ritvo et al.	1989	USA	1984-88	8 to 12	184,822	DSM-III	Case enumeration & direct exam	0.40 (0.31-0.50)
Gillberg et al.	1991	Sweden	1988	4 to 13	78,106	DSM-III-R	Case enumeration & direct exam	0.95 (0.74-1.95)

Author	Year	Country	Period Studied	Age range	No. of children	Criteria	Methodology	Prevalence (per 1000)
Honda et al.	1996	Japan	1994	1.5 to 6	8,537	ICD-10	Population screen & direct exam	2.11 (1.25-3.33)
Fombonne et al.	1997	France	1992-93	6 to 16	325,347	ICD-10	Case enumeration & direct exam	0.54 (0.46-0.62)
Arvidsson et al.	1997	Sweden	1994	3 to 16	1,941	ICD-10	Population screen & direct exam	3.10 (1.14-6.72)
Webb et al.	1997	Wales	1992	3 to 15	73,300	DSM-III-R	Case enumeration & direct exam	0.72 (0.54-0.95)
Sponheim & Skjeldae	1998	Norway	1992	3 to 14	65,688	ICD-10	Case enumeration & direct exam	0.38 (0.25-0.56)
Kadesjo et al.	1999	Sweden	1992	6.7 to 7.7	826	ICD-10	Case enumeration & direct exam	6.0 (1.97-14.1)
Baird et al.	2000	England	1998	1.5 to 8	16,235	ICD-10	Population screen & direct exam	3.1 (2.29-4.06)
Powell et al.	2000	England	1995	1 to 4	29,200	DSM-III-R or DSM-IV	Case enumeration	0.96 (0.64-1.39)
Magnusson & Saemundsen	2000	Iceland	1997	5 to 14	43,153	ICD-10	Population screen & direct exam	0.86 (0.60-1.18)
Chakrabarti & Fombonne	2001	England	1998	2.5 to 6.5	15,500	DSM-IV	Population screen & direct exam	1.68 (1.1-2.46)

Author	Year	Country	Period studied	Age range	No. of children	Criteria	Methodology	Prevalence (per 1000)
Bertrand et al.	2001	USA	1998	3 to 10	8,996	DSM-IV	Case enumeration & direct exam	4.0 (2.8-5.5)
Croen et al.	2001	USA	1987-99	0 to 21	4.6m	DSM-III-R or DSM-IV	Case enumeration	1.1 (1.06-1.14)
Allsopp et al. (2)	2003	USA	1996	3 to 10	290,000	DSM-IV	Case enumeration	3.4 (3.2-3.6)
Gurney et al. (2)	2003	USA	1981-82, 2001-02	6 to 17	N/A	DSM-IV	Case enumeration	4.4 (4.3-4.5)
Lingam et al.	2003	UK	2000	5 to 14	186,206	ICD-10	Case enumeration	1.5 (1.3-1.7)
Icasiano et al.	2004	Australia	2002	2 to 17	45,153	DSM-IV	Case enumeration	3.9 (3.3-4.5)
Lauritsen et al.	2004	Denmark	2001	0 to 9	682,397	ICD-10	Case enumeration	1.2 (1.1-1.3)
Fombonne et al.	2006	Canada	1987-98	5 to 21	27,749	DSM-IV	Case enumeration	2.16 (1.65-2.78)
Baird et al.	2006	UK	1990-91	9 to 10	56,946	ICD-10	Case enumeration, screen and direct exam	3.89 (3.39-4.43)
CDC Network (1)	2007	USA	2000	8	187,761	DSM-IV	Case enumeration and	6.7 (6.3-7.0)

Author	Year	Country	Period studied	Age range	No. of children	Criteria	Methodology	Prevalence (per 1000)
							record review	
CDC ADMM Network (1)	2007	USA	2002	8	444,050	DSM-IV	Case enumeration and record review	6.6 (6.3-6.8)
Oullette-Kuntz et al.	2007	Canada	1996-2004	4 to 9	2,240,537	Special education classification	Case enumeration from special education classification	1.2 (1996), 4.3 (2004)
Wong et al. (1)	2008	Hong Kong	1986-2005	0 to 14	4,247,206	DSM-IV	Case enumeration	1.6
Williams et al.	2008	Australia	2003-04	6 to 12	5,459	DSM-IV	Questionnaires	1.0 (0.8-1.0) to 4.1 (3.8-4.4)
Montiel-Nava et al.	2008	Venezuela	2005-06	3 to 9	254,905	DSM-IV	Case enumeration	1.7 (1.3-2.0)
Baron-Cohen et al.	2009	UK	2003-04	5 to 9	5,484	Special ed. needs register	Case enumeration from survey and direct exam	15.7 (9.9-24.6)
CDC ADMM Network (1)	2009	USA	2004	8	172,335	DSM-IV	Case enumeration and record review	8.0 (7.6-8.4)
CDC ADMM Network (1)	2009	USA	2006	8	308,038	DSM-IV	Case enumeration and record review	9.0 (8.6-9.3)
Al-Farsi et al.	2010	Oman	2009	0 to 14	798,913	DSM-IV	Case enumeration	0.1 (0.1-0.2)

								0.2
Parner et al.	2011	Denmark	1994-99	N/A	404,816	DSM-IV	Case enumeration	6.9 (6.5-7.2)
Parner et al.	2011	Western Australia	1994-99	N/A	152,060	DSM-IV	Case enumeration	5.1 (4.7-5.5)
Chien et al.	2011	Taiwan	1996-2005	0 to 18	372,642	ICD-9	Case enumeration	2.9
Windham et al.	2011	USA	1994, 1996	0 to 8	82,153 (1994), 80,249(1996)	DSM-IV	Case enumeration	4.7 (4.2-5.1) (1994), 4.7 (4.2-5.2) (1996)
Kim et al.	2011	South Korea	2005-09	7 to 12	55,266	DSM-IV	Case enumeration from survey and direct exam	26.4 (19.1-33.7)
Zimmerman et al.	2012	USA	2002, 2006, 2008	8	26,213 (2002), 29,494 (2006), 33,757 (2008)	ICD-9 and special education classification	Case enumeration	6.5 (2002), 10.2 (2006), 13.0 (2008)
Kocovska et al.	2012	Faroe Islands	2002, 2009	7 to 16 (2002), 15 to 24 (2009)	7,122 (2002), 7,128 (2009)	DSM-IV, ICD-10	Screening and direct exam	5.6 (2002), 9.4 (2009)

Author	Year	Country	Period studied	Age range	No. of children	Criteria	Methodology	Prevalence (per 1000)
Blumberg et al.	2013	USA	2011-12	6 to 17	95,677	Parent report	Telephone survey	2%
Zablotsky et al.	2015	USA	2011-14	3 to 17	43,283	Parent report	Household survey	2.40%
Christensen et al. (1)	2016	USA	2012	8	346,978	DSM-IV	Case enumeration and record review	14.6 (8.2-24.6)

Notes

- (1) The prevalence reported represents the average.
- (2) The prevalence study provided overall rate only.