Developing better information about the health and health care of people with developmental disabilities in Scotland

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A thesis submitted in December 2021 to the University of Glasgow for the Degree of Doctor of Philosophy by published work,[1] incorporating papers arising from research carried out at the Institute of Health and Wellbeing, College of Medical, Veterinary, and Life Sciences, University of Glasgow, Gartnaval Royal Hospital, 1055 Great Western Road, G12 0XH.

Acknowledgments

The papers in this thesis include data which I have worked on since January 2015 when I joined the Institute of Health and Wellbeing at the University of Glasgow, as a researcher, within the research programme, the Scottish Learning Disabilities Observatory.

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I would like to acknowledge my advisors, mentors and other colleagues within the Institute of Health and Wellbeing, especially researchers within the Scottish Learning Disabilities Observatory team. I would like to say a special thanks to Professor Sally-Ann Cooper (OBE) for all the guidance and support she has provided to me in the past six years, including giving me the opportunity to work within the Scottish Learning Disabilities Observatory and carry out such important research, to help address inequalities experienced by people with learning disabilities and autism in Scotland. This was a life changing opportunity for me, which has made this thesis possible.

Finally, I would like to thank my family for all their support and encouragement. I dedicate this thesis to my three lovely children; Leon, Eva and Chloe.

List of Contents

Glossary of terms

OR	Odds ratio
HR	Hazards ratio
CI	Confidence interval
DOI	Digital object identifier
PMID	PubMed identifier
N	Number
P	Probability value
R^2	Coefficient of determination

Abstract

Introduction

Neurodevelopmental disorders are a group of disorders that manifest early in development, and include intellectual disabilities and autism, among others. Intellectual disabilities refer to impairments in intellectual functioning (an intelligence quotient <70), together with deficits in adaptive functioning (need for support for daily personal independence and social functioning), with onset during the developmental period. Autism is characterised by persistent deficits in social communication and social interactions across multiple contexts, and restricted, repetitive patterns of behaviour, interests or activities, with onset of these symptoms in the early developmental period. People with intellectual disabilities and people with autism are thought to experience high levels of physical and mental health problems and earlier mortality than other people on average. Yet, there is a dearth of empirical evidence about the health of people with intellectual disabilities and people with autism which presents a barrier to understanding the complex factors that produce differential health outcomes. Ensuring adults with intellectual disabilities or autism live not only longer but healthier lives is a priority for the World Health Organisation and the Scottish Government.

Methods

The portfolio of publications (n=15) presented in this mixed methods PhD thesis, represents a selection of my peer reviewed publications in international scientific journals since 2015. I [Laura Hughes-McCormack] am the lead author on 4 of these publications and co-author on 11 publications (including being second author on 8 of these) which were prepared from research I undertook within the Scottish Learning Disabilities Observatory (SLDO) at the Institute of Health and Wellbeing, University of Glasgow. This research programme was funded by the Scottish Government in 2014 to provide evidence on the health of people with

intellectual disabilities and autism in Scotland, and thus to inform the development of public policy. The research presented includes systematic reviews, quantitative research, data linkage research, and is presented in three themes throughout this thesis, as follows. Theme I. Health of people with intellectual disabilities and autism; seven of the nine quantitative studies presented under this theme/section analysed data from the Scottish Census, relating to people with intellectual disabilities or autism from 94% of the Scottish population (n=5,269,054) who responded to the Census in 2011. Two further studies (a quantitative study and a systematic review study) are reported, which investigated sedentary behaviour and oral health. Theme II. Health care of people with intellectual disabilities and Down syndrome; to quantify the management of long-term conditions, data for a population-based cohort of people with intellectual disabilities (n=721) was compared using an established evidence-based approach to measuring the quality of primary health care for all people without intellectual disabilities (n=764,672) in the largest health board in Scotland, throughout 2007-2010. A further systematic review study investigated hospital admissions for physical health conditions in adults with intellectual disabilities. Theme III. Survival/ death of people with intellectual disabilities and Down syndrome; two systematic reviews are reported, investigating deaths in people with intellectual disabilities and people with Down syndrome, and a further data linkage study which investigated birth and death rates and hospitalisations (throughout a 25-year period) among children/young people with Down syndrome in Scotland. Each theme includes a clear overview of the background/ rationale, methodology, results and impact of this body of work in relation to the development of better information on health and health care of people with intellectual disabilities and autism in Scotland.

Results

Theme I: Health of people with intellectual disabilities and autism

Findings of the Scottish Census 2011 studies (I-VII), show that poor health was more common for people with intellectual disabilities (odds of 43 in statistically predicting poor general health) and they were seven times more likely to report a current mental health condition than people without intellectual disabilities. Autism had odds of 11.3 in predicting poor general health in children and young people, and odds of 7.5 in adults. Comorbidities were found to be common among people with autism. Other studies presented under this theme (VIII, IX) show adults with intellectual disabilities have higher levels, and different correlates, of sedentary behaviour and poorer oral health.

Theme II: Health care of people with intellectual disabilities and Down syndrome

Findings from two studies (X, XI) show, people with intellectual disabilities were receiving lower quality health care compared to other people across all long-term conditions investigated on 38/57 (66.7%) quality indicators. A follow up study, comparing baseline data to data in 2014 found adults with intellectual disabilities still had a lower proportion of indicators met than the general population; but by 2014, the healthcare inequality gap had reduced compared with 2007-10. A systematic review (XII), further investigating the quality of health care of people with intellectual disabilities, found people with intellectual disabilities were admitted to hospital more frequently than the general population for physical conditions which if managed effectively at the primary care level, should not lead to hospital admission, although evidence is limited.

Theme III: Survival/ death of people with intellectual disabilities and Down syndrome

Findings from the two systematic reviews (XIII, XIV) found mortality rates are higher in (1) the intellectual disabilities population and (2) the Down syndrome population, compared with

the general population. For the intellectual disabilities' population, an average age of death of 20 years lower was found compared to the general population. For the Down syndrome population, compared with the general population, an average age of death of 28 years lower was found although survival rates have improved over time. Patterns of cause of death were different for people with intellectual disabilities and people with Down syndrome compared to the general population. The data linkage study (XV) found the incidence of Down syndrome live-births was 1.0/1,000 births over the last 25 years. More children and young people with Down syndrome died (n=92; 7.4%) over the 25-year period compared to controls (n=23; 0.4%); that is 18.5 times more. There was increased risk of hospitalisation as more of the Down syndrome group had at least one admission (1,105 [89.5%] versus 3,305 [53.5%]; adjusted HR=1.84 [1.68, 2.01]). Re-admissions, emergency admissions and length of stay were also higher for the Down syndrome group.

Conclusions

This thesis has presented for each of the three inter-related themes, a range of my peer reviewed publications from international journals. A previous dearth of empirical evidence about the health and health care of people with intellectual disabilities and people with autism has presented barriers to understanding the complex factors that produce differential health outcomes. The research presented in this thesis has provided robust evidence on the extent of poor health, health care, and premature mortality among people with intellectual disabilities, and people with autism which has not previously been quantified in research. This evidence has led to shaping Scottish policy and practice to support the needs of people with intellectual disabilities, for example, the most recent learning disabilities strategy in Scotland, The Keys to Life, updated in 2019, includes input from my research.

List of publications

The full text of the articles (except those published in journals without a Creative Commons license) are available in Appendix 1. Note that these articles have been reproduced solely for inclusion in this thesis, and should not be disseminated or distributed further without ascertaining agreement from the original copyright holders. Table 1 indicates my personal contribution to each of these manuscripts.

Theme I: Health of people with intellectual disabilities and autism:

- **Hughes-Mccormack, L. A.,** Rydzewska, E., Henderson, A., MacIntyre, C., Rintoul, J., and Cooper, S.-A. (2018a) Prevalence and general health status of people with intellectual disabilities in Scotland: a total population study. *Journal of Epidemiology and Community Health*, 72(1), pp. 78-85. (doi:10.1136/jech-2017-209748) (PMID:29070675)
- **Hughes-McCormack, L. A.,** Rydzewska, E., Henderson, A., MacIntyre, C., Rintoul, J., and Cooper, S.-A. (2018b) Prevalence of mental health conditions and relationship with general health in a whole-country population of people with intellectual disabilities compared with the general population. *BJPsych Open*, 3(5), pp. 243-248. (doi:10.1192/bjpo.bp.117.005462) (PMID:29034100) (PMCID:PMC5620469)
- Rydzewska, E., **Hughes-McCormack, L. A.,** Gillberg, C., Henderson, A., MacIntyre, C., Rintoul, J., and Cooper, S.-A. (2018) Prevalence of long-term health conditions in adults with autism: observational study of a whole country population. *BMJ Open*, 8(8), e023945. (doi:10.1136/bmjopen-2018-023945) (PMID:30173164) (PMCID:PMC6120653)

- Rydzewska, E., **Hughes-McCormack, L.,** Gillberg, C., Henderson, A., MacIntyre, C., Rintoul, J., and Cooper, S.-A. (2019a) Age at identification, prevalence and general health of children with autism: observational study of a whole country population. *BMJ Open*, 9, e025904. (doi:10.1136/bmjopen-2018-025904) (PMID:31289063)
- Rydzewska, E., **Hughes-McCormack, L. A.,** Gillberg, C., Henderson, A., MacIntyre, C., Rintoul, J., and Cooper, S.-A. (2019b) Prevalence of sensory impairments, physical and intellectual disabilities, and mental health in children and young people with self/proxy-reported autism: observational study of a whole country population. *Autism*, 23(5), pp. 1201-1209. (doi:10.1177/1362361318791279) (PMID:30328695)
- Rydzewska, E., **Hughes-Mccormack, L. A.,** Gillberg, C. , Henderson, A., MacIntyre, C., Rintoul, J., and Cooper, S.-A. (2019c) General health of adults with autism spectrum disorders - a whole country population cross-sectional study. *Research in Autism Spectrum Disorders*, 60, pp. 59-66. (doi:10.1016/j.rasd.2019.01.004)
- Kinnear, D., Rydzewska, E., Dunn, K., **Hughes-McCormack, L. A.,** Melville, C., Henderson, A., and Cooper, S.-A. (2019) The relative influence of intellectual disabilities and autism on mental and general health in Scotland: a cross-sectional study of a whole country of 5.3 million children and adults. *BMJ Open*, 9(8), e029040. (doi:10.1136/bmjopen-2019-029040) (PMID:31462474)
- Melville, C.A., McGarty, A., Harris, L., **Hughes-Mccormack, L.,** Baltzer, M., McArthur, L., Morrison, J., Allan, L. and Cooper, S.-A. (2018) A population-based, cross-sectional study of the prevalence and correlates of sedentary behaviour of adults with intellectual disabilities. *Journal of Intellectual Disability Research*, 62(1), pp. 60-71. (doi:10.1111/jir.12454) (PMID:29214701)

- Ward, L.M., Cooper, S.-A., **Hughes-Mccormack, L.,** Macpherson, L., and Kinnear, D. (2019) Oral health of adults with intellectual disabilities: A systematic review. *Journal of Intellectual Disability Research*, 63(2), pp. 1359-1378. (doi:10.1111/jir.12632) (PMID:31119825)

Theme II: Health care of people with intellectual disabilities and Down syndrome:

- Cooper, S-A., **Hughes-McCormack, L.,** Greenlaw, N., et al. (2018) Management and prevalence of long-term conditions in primary health care for adults with intellectual disabilities compared with the general population: A population-based cohort study. *Journal of Applied Research in Intellectual Disabilities*. 31(Suppl. 1): 68– 81. (doi.org/10.1111/jar.12386) (PMID:28984406)
- **Hughes-McCormack, L-A.,** Cooper, S-A., Greenlaw, N., McConnachie, A., Allan, L., Baltzer, M., McArthur, L., Henderson, A., Melville, C., Morrison, J., Ross, K (2021) Changes over time in the management of long-term conditions in primary health care for adults with intellectual disabilities, and the health care inequality gap compared with the general population. *Journal of Applied Research in Intellectual Disabilities.* 34(2), pp. 634-647. (doi: 10.1111/jar.12833) (PMID:33283349)
- Dunn, K., **Hughes-Mccormack, L.,** and Cooper, S.-A. (2018) Hospital admissions for physical health conditions for people with intellectual disabilities: systematic review. *Journal of Applied Research in Intellectual Disabilities*, 31(S1), pp. 1-10. (doi:10.1111/jar.12360) (PMID:28467010)

Theme III: Survival/ death of people with intellectual disabilities and Down syndrome:

- O'Leary, L., Cooper, S-A., **Hughes-McCormack, L.** (2018) Early death and causes of death of people with intellectual disabilities: A systematic review. *Journal of*

Applied Research in Intellectual Disabilities. 31(3), 325– 342. (doi.org/10.1111/jar.12417) (PMID: 28984406)

- O'Leary, L., **Hughes-Mccormack, L.,** Dunn, K., and Cooper, S.-A. (2018) Early death and causes of death of people with Down syndrome: a systematic review. *Journal of Applied Research in Intellectual Disabilities,* 31(5), pp. 687-708. (doi:10.1111/jar.12446) (PMID:29573301)
- **Hughes-McCormack, L-A.,** McGowan.R, Pell, J.,.,Mackay, D., Henderson, A., O'Leary, L., Cooper S-A. (2020). Birth incidence, deaths, and hospitalisations of children and young people with Down syndrome, 1990-2015: birth cohort study. *BMJ Open.* 10(e033770). (doi: 10.1136/bmjopen-2019-033770) (PMID:32241786)

Table 1: Contribution to projects

Years	Title of study	Activities			
		Design	Data analysis	Data interpretation	Manuscript writing
Theme I: health of people with intellectual disabilities and autism					
2016-2017	Prevalence and general health status of people with intellectual disabilities in Scotland-a total population study.	**C**	**L**	**L**	**L**
2016-2017	Prevalence of mental ill-health and relationship with physical health in a whole country population of 26,349 people with intellectual disabilities, compared with the general population.	**C**	**L**	**L**	**L**
2016-2018	General health of adults with autism spectrum disorders - a whole country population cross-sectional study.	**C**	**C**	**C**	**C**
2016-2018	Prevalence of long-term health conditions in adults with autism: observational study of a whole country population	**C**	**C**	**C**	**C**
2016-2018	Prevalence of sensory impairments, physical and intellectual disabilities, and mental health in children and young people with self/proxy-reported autism - observational study of a whole country population.	**C**	**C**	**C**	**C**
2016-2018	Prevalence, age at identification, and general health status of children and young people known to have autism spectrum disorder - a whole country population cross-sectional study.	**C**	**C**	**C**	**C**
2018-2019	The relative influence of intellectual disabilities and autism on mental and general health in Scotland: a cross-sectional study of a whole country of 5.3 million children and adults.	**-**	**C**	**C**	**C**
2016-2017	A population-based, cross-sectional study of the prevalence and correlates of sedentary behaviour of adults with intellectual disabilities.	**C**	**-**	**C**	**C**
2018-2019	Oral health of adults with intellectual disabilities: a systematic review.	**-**	**C**	**C**	**C**
Theme II: health care of people with intellectual disabilities and Down syndrome					
2015-2018	Changes over time in the management of long-term conditions in primary health care for adults with intellectual disabilities, and the health care inequality gap compared with the general population.	**C**	**C**	**L**	**L**
2016-2017	Management and prevalence of long-term conditions in primary health care for adults with intellectual disabilities compared with the general population: a population based cohort study.	**C**	**C**	**C**	**L**
2015-2017	Hospital admissions for people with intellectual disabilities: systematic review.	**-**	**C**	**C**	**C**
Theme III: survival/death of people with intellectual disabilities and Down syndrome					
2015-2018	Life expectancy and causes of death of people with intellectual disabilities: a systematic review.	**-**	**C**	**C**	**C**
2015-2018	Early death and causes of death of people with Down syndrome: a systematic review.	**-**	**C**	**C**	**C**
2017-2019	Prevalence, deaths, and hospitalisations of people with Down syndrome, 1990-2015: birth cohort study.	**C**	**L**	**L**	**L**

L=lead; C=contributed; -= no contribution or not applicable

This explanatory essay has a word count of 11,935 of the required limit of 10,000-12,000 words.

Explanatory essay

Introduction

Neurodevelopmental disorders:

Neurodevelopmental disorders are a group of disorders that typically manifest early in development and are characterised by developmental deficits that produce impairments of personal, social, academic, or occupational functioning (Bitta et al, 2018). They include intellectual disabilities and autism among others. Intellectual disabilities refer to impairments in intellectual functioning (an intelligence quotient <70), together with deficits in adaptive functioning (need for support for daily personal independence and social functioning), with onset during the developmental period (WHO, 1992). Autism shares deficits in three core domains: social interaction, communication, and restricted interests/repetitive behaviours, with onset of these symptoms in the early developmental period (WHO, 1992). Intellectual disabilities and autism are quite common (Cooper et al., 2016). However, accurate prevalence rates have been difficult to identify from previous research due to methodological differences and limitations (Cooper et al., 2016). Co-occurring intellectual disabilities and autism is also common, occurring in around 2.58/1000 children/young people and 0.74/1000 adults (Dunn et al., 2019). There are additional challenges identifying the prevalence of autism, as the definition has now broadened considerably beyond original descriptions, and clinicians also now base their diagnosis on fewer symptoms than a decade ago (Kanner, 1968; Asperger, 1944; Arvidsson et al., 2018). As such, the prevalence of autism in adults and children may be substantially different, possibly reflecting that the concept of autism spectrum has broadened in recent years. It is essential to have accurate information on the proportion of children and young people who are known to have intellectual disabilities, autism, or co-

occurring autism and intellectual disabilities and their health status, in order to accurately plan appropriate prevention and intervention measures, and provision of resources for those people who may influence demand for services designed for people with intellectual disabilities or autism.

Health inequalities of people with intellectual disabilities:

More people with intellectual disabilities now live in the community and enjoy longer lives than in the past, due to the policies of deinstitutionalisation and closure of long stay hospitals in the late 20th and early 21st century, and improving access to health care. However, the profile of health problems experienced by people with intellectual disabilities remains different from that of the general population. People with intellectual disabilities experience a more complex set of health needs, including both physical and mental health conditions, and premature mortality (NHS, 2004; Ouelette-Kuntz et al., 2004; Emerson et al., 2007; Oesburg et al., 2011; Fujiura, 2012; Emerson et al., 2014; Cooper et al., 2015; Robertson et al., 2015). It has been suggested that early mortality of people with intellectual disabilities is to a large extent avoidable (Heslop et al., 2013; Glover and Emerson, 2013). However, the body of existing research is limited.

Health inequalities of people with autism:

Children and adults with autism are thought to have poorer general and mental health than other people although the quantity of previous research is limited (Simonoff et al., 2008; Croen et al., 2015; National Longitudinal Transition Study 2, 2017). Moreover, the relative extent to which being autistic or having intellectual disabilities accounts for their poorer population health is unclear. The largest study to examine this in adults in a general community population found no difference in rates of mental ill health in adults with co-

occurring autism and intellectual disabilities, compared with age-gender-Down syndrome-level of ability-matched adults with intellectual disabilities but no autism (Melville et al., 2008). More research is needed, given the frequent co-occurrence of these conditions. Moreover, it is important to understand health in both child and adult populations, given the more recent changes in diagnosis and level of awareness, which have may influenced an increasing diagnosis of autism in younger populations.

The Scottish Learning Disabilities Observatory

In 2015, the Scottish Learning Disabilities Observatory was set up to develop a programme of research, designed to collect information and evidence to build understanding of the health and health inequalities experienced by people with learning disabilities and people with autism in Scotland. The research in this thesis arose from my contribution to this programme of work over a five-year period. The Scottish Learning Disabilities Observatory have used a range of methodologies to meet the strategic goals, including reviewing existing evidence through systematic reviews and meta-analyses, analysing available statistical data including routinely collected health and population census data, monitoring and assessing health trends for people with intellectual disabilities. We contribute to the increased visibility of people with intellectual disabilities in public health data by working with people with intellectual disabilities, their families, carers and supporters to include their voices in our activities and outputs. A large portion of the work included in this thesis arose from investigating population census data which was one key strand of research in the Scottish Learning Disabilities Observatory, specifically involving data from Scotland's 2011 Census. Once every 10 years the Scottish Government holds a national census to provide a snapshot of all the people in Scotland on one night. For the first time, Scotland's Census 2011 (on 27.3.11), gave people the chance to say whether or not they had intellectual disabilities and/or autism. Despite some limitations, including a lack of

information on whether the responses were completed by proxies or the person with intellectual disabilities, this dataset has an extremely high completion rate and contained a very clear question to identify whom had intellectual disabilities. We have no reason to believe the results are not generalisable to other high-income countries. This provided the Scottish Learning Disabilities a unique opportunity to analyse and present information about people with learning disabilities and people known to have autism, compared with the whole population.

Building an evidence base to inform health and health care policy:

Addressing the health inequalities of people with intellectual disabilities is a major challenge for policy makers and service providers, as highlighted in the Scottish learning disabilities strategy, 'The keys to life', launched in 2013, updated in 2015 and again in 2019 (Scottish Government, 2019). In an umbrella review of systematic reviews on the heath and health care of people with intellectual disabilities, Roberson et al (2015) found most of the systematic reviews identified reported serious issues with the methodology of the studies they were reviewing, such that clear conclusions could not be drawn. Robertson et al (2015) highlight how responding to health inequalities of people with intellectual disabilities is a critically important issue for primary and secondary healthcare services and how this demands action on several fronts, including building a more robust evidence base of the scale and determinants of health inequalities to help compensate for the previous under-investment in high-quality research on the health of people with intellectual disabilities.

A healthy life is one of the core priorities at the heart of the Scottish strategy for autism which was first published in 2011 and updated in 2018 (Scottish Government, 2018). However, it is clear from previous research that health inequalities of people with autism

have been under investigated. In a scoping review of health and health system quality (e.g., high quality care delivery, adequate care access) disparities in autism (Bishop-Fitzpatrick et al., 2017), only 9 studies were identified. Most studies (77.8%) identified racial disparities in access to general medical services for children with autism. However, no studies examined disparities in health outcomes or included older adults. Bishop-Fitzpatrick et al (2017) recommended that additional work should examine health disparities, and their causal pathways, in autism, particularly for older adults.

The published work:

The publications presented here represent a selection of my published work that arose from research conducted within the Scottish Learning Disabilities Observatory, in my role as a researcher in the Institute of Health and Wellbeing. Each of the publications presented in this thesis are peer-reviewed publications in international scientific journals. My research, in line with the remit of the Scottish Learning Disabilities Observatory, has generated better information across the areas of the health and the health care of people with intellectual disabilities (with and without Down syndrome) and people with autism. To provide the evidence for this, including a clear overview of the rationale, methodology, results and impacts of this body of work in the areas of health and health care, the research presented in this thesis is structured according to three relevant themes, which are as follows: I. Health of people with intellectual disabilities and autism, II. Health care of people with intellectual disabilities and Down syndrome, and III. Survival/ death of people with intellectual disabilities and Down syndrome.

My research was intended to generate evidence and build understanding of the causes of poor health and health inequalities experienced by people with intellectual disabilities and people

with autism. This reflects some of the policy recommendations in the recently updated Scottish Health Needs Assessment (Truesdale et al., 2017) for people with intellectual disabilities.

The published works presented here encompass numerous key policy areas of health and health care including morbidity, management of health, health care utilisation, mortality and life expectancy that are central to tackling the health inequalities experienced by people with intellectual disabilities (with, and without, Down syndrome) and people with autism. From the papers and work presented here, I have demonstrated that my work has gone some way to develop the evidence-base on the health needs of people with intellectual disabilities and people with autism, increasing their visibility in routinely collected data, and outlines where change needs to be made. This work is helping to inform actions, practice and policy to reduce health inequalities for these groups of people.

The published work is presented in three themes to represent the three important elements of research activity required to develop evidence informed policy noted earlier. The overarching research questions for each theme are presented below each sub section:

Theme I: Health of people with intellectual disabilities and autism

Research questions:

- What is the prevalence of intellectual disabilities, autism and co-occurring intellectual disabilities by age and gender?
- What is the reported general health status of children, young people and adults with intellectual disabilities, autism and co-occurring intellectual disabilities compared with the general population and associations with age and gender?

- What is the reported mental health status of children, young people and adults with intellectual disabilities, autism and co-occurring intellectual disabilities compared with the general population and associations with age and gender?
- What is the prevalence and correlates of sedentary behaviour in adults with intellectual disabilities?
- What is the prevalence and most common measurements of poor oral health among people with intellectual disabilities?

Theme II: Health care of people with intellectual disabilities and Down syndrome

Research questions:

- How are long-term conditions managed within primary health care for adults with intellectual disabilities compared with the general population, using indicators of best practice from the Quality and Outcomes Framework?
- Have there been improvements in the management of long-term conditions over time within primary health care for adults with intellectual disabilities compared with the general population, using indicators of best practice from the Quality and Outcomes Framework?
- Are people with intellectual disabilities admitted to hospital more frequently than the general population (including for ambulatory sensitive conditions), and do any differences in admission rate persist when between-group disease prevalence differences are adjusted for?
- Are people with intellectual disabilities admitted to hospital for the same reasons as the general population?

Theme III: Survival/ death of people with intellectual disabilities and Down syndrome

Research questions:

- What is the extent of early mortality among people with intellectual disabilities compared with the general population, any trends in terms of the extent of this health inequality and the pattern of cause of death?
- What is the extent of early mortality among people with Down syndrome compared with the general population, any trends in terms of the extent of this health inequality and the pattern of cause of death?
- What is the incidence of live births of people with Down syndrome over a 25-year period?
- What is the frequency of deaths of children and young people with Down syndrome compared with matched controls over a 25-year period?
- What is the hospital admission frequency and duration, emergency admissions and readmissions of children and young people with Down syndrome compared with matched controls over a 25-year period?

Theme I: Health of people with intellectual disabilities and autism

During my time as a researcher with the Institute of Health and Wellbeing, I have worked with data from Scotland's Census 2011 (National Records of Scotland). Scotland's census provides statistical information on the number and characteristics of Scotland's population and households at the census day, 27 March 2011. The census is undertaken every 10 years. It includes people living in communal establishments (such as care homes and student halls of residence) as well as people living in private households. The most recent census in 2011,

included 94% of the Scottish population (n=5,269,054), close to the highest ever compliance rate. This census included a new question added in 2011, question 20, which made it possible to identify people with intellectual disabilities and/ or autism. This question asked respondents: 'Do you have any of the following conditions which have lasted, or are expected to last, at least 12 months? Tick all that apply'. There was a choice of 10 response options: (1) deafness or partial hearing loss, (2) blindness or partial sight loss, (3) learning disability (e.g., Down's syndrome), (4) learning difficulty (e.g., dyslexia), (5) developmental disorder (e.g., autistic spectrum disorder or Asperger's syndrome), (6) physical disability, (7) mental health condition, (8) long-term illness, disease or condition, (9) other condition and (10) no condition. There was an additional option for reporting any 'other condition', in which a prompt was included for the respondent to report the type of 'other' condition. Under 'other', if a person indicated that they had one of the conditions previously specified, this was recoded so that this person was counted as if they had responded to the relevant option of the question. For example, if a person indicated they were blind under 'other', this would be recoded under option 2 of this question.

Learning disability (e.g., Down syndrome) is the term used in Scotland for the internationally used term of intellectual disabilities. Importantly, the census distinguished between intellectual disability (learning disability) and learning difficulty (e.g., dyslexia) and developmental disorders (e.g., autism spectrum disorder or Asperger's syndrome). Whilst the response option for autism was the broad term developmental disorder, it only prompted respondents to reply with regards to autistic spectrum disorder or Asperger's syndrome. As part of the methodological preparations for Scotland's Census, 2011, cognitive question testing was undertaken on the question on these conditions, to investigate whether it was answered accurately and willingly, and what changes might be required to improve data

quality. Retrospective probing was undertaken with 102 participants with a mix of gender and age, both with and without the conditions or with more than one of the conditions, and including people with autism, intellectual disabilities, dyslexia, dyspraxia, speech impairment, mental health conditions (both milder and more serious), and other long-term conditions. This resulted in a redesign of the question on autism to the version used. The other questions did not require any modification. We therefore interpret responses to this question as relating to people who know they have autism. The studies presented include the population who selected intellectual disabilities or autism. However, within intellectual disabilities or autistic populations, there is an overlap between these two conditions. For example, within the intellectual disabilities' population, a proportion of this group will also have autism. A proportion of the autistic population also have intellectual disabilities. As with a wide body of previous research, each of these populations tends to be studied separately, so the relative extent to which being autistic, or having intellectual disabilities may account for their poor population health is not clear. This is important to understand, given the frequent co-occurrence of these conditions, and is important to understand in both child and adult populations, given the more recent change in co-occurrence due to higher frequency of diagnosis of autism. This dataset presented a rare opportunity to investigate the intersectionality in the health of the population with intellectual disabilities, the population with autism, the population with intellectual disabilities without autism and the population with autism without intellectual disabilities. All but two of the studies that follow in this theme/section are based on data from the Scottish census, relating to people with intellectual disabilities or autism or those with intellectual disabilities without autism and those with autism without intellectual disabilities. The additional two papers are included to provide a wider context on the health inequalities of people with intellectual disabilities in two important areas, oral health, and sedentary behaviors. Oral health is fundamental to general

health and well-being, and this may be particularly true for people with intellectual disabilities who experience poorer health in general (e.g., Robertson et al, 2015). General health is investigated in the research questions from the Scottish Census 2011, also presented under this theme. This provides a wider picture of the health profiles and health needs of people with intellectual disabilities. Poor oral health (namely periodontal disease) is also associated with long term conditions, some of which may be more prevalent or complex in people with intellectual disabilities. This includes obesity, which is consistently found to be much more common in people with intellectual disabilities. The final paper was included as it relates to understanding the correlates of sedentary behaviour in people with intellectual disabilities, which might impact on obesity. This again can help aid our understanding of the health amongst people with intellectual disabilities by taking a wider viewpoint on health inequalities. I was lead author on two of these papers and contributed substantially to the work of the other papers. In particular, I played a lead role in developing the methods of analysis of the census data. The individual level data from the census was not held by the research team, it was held by National Records of Scotland, and only aggregate data in the form of tables was available. I helped adapt the analysis from more traditional methods to suit the secondary data we had and to be able to run inferential statistical procedures rather than provide only a descriptive analysis of the findings. For all the papers that will be discussed, this involved breaking down the data set into numerous categories, from which the number of people who were in each associated category was identified. Each of the categories was entered into a statistical package (SPSS), and once the numbers of people were identified for each category, a frequency variable was created to correspond to each category. This meant that a frequency weight could be applied, and the data could be analysed in the same way as if it were individual level data. This also meant we could conduct more complex analyses,

such as logistic regressions, investigating the influence of other variables such as age and gender in the analysis. More details about each specific paper follows:

Paper I **Hughes-McCormack, L-A.,** Rydzewska, E., Henderson, A., MacIntyre, C., Rintoul, J., Cooper S-A. (2017). Prevalence and general health status of people with intellectual disabilities in Scotland-a total population study. *Journal of Epidemiology and Community Health*. (doi:10.1136/jech-2017-209748) (PMID:29070675)

This manuscript reports the prevalence of intellectual disabilities (in childhood, adulthood and older age) and the reported general health status of people with and without intellectual disabilities across the lifespan. This area of research was important as little is known about the reported general health of people with intellectual disabilities. Ratings of general health have been found to be a valid marker of morbidity and mortality in the general population (Idler & Benyamini, 1997; Lee, 2000; Young et al, 2010; Mewton & Andrews, 2013). Additionally, few studies had previously reported on age of identification of intellectual disabilities.

Data from the Scottish population (n=5,269,054) who responded (94%) to the census in 2011 were used for the manuscript. Specifically, those who responded to the individual questions section, question 20 as having intellectual disabilities compared to those who did not report any intellectual disabilities. In total, 26,349 people with intellectual disabilities were identified from the census, which is 0.5% of the total population. We investigated general health, which was collected through the question, 'How is your health in general?' with a five point response scale (*very good, good, fair, bad, very bad*) which was redesigned to become a

binary response of ‘poor health’ (from fair, bad, very bad health) or ‘good health’ (from very good, good health). We conducted logistic regressions to determine the Odds Ratios of intellectual disabilities predicting poor health and associations with age and gender.

The findings provide important knowledge about the prevalence of intellectual disabilities across all ages, which is vital for informing resource allocation and financial planning for people with intellectual disabilities. It also demonstrated, in a total population, that people with intellectual disabilities experience poor general health. Intellectual disabilities had odds of 43 in statistically predicting poor health, and the health difference with the general population is more marked for children/young people than adults. The 16–24 year-old group had better health than those aged 0–15 years, as did, to a lesser extent, the individuals aged 25–34 years. This suggests that some children with the most complex health needs do not reach adult ages. I concluded in this paper that the findings demonstrate an urgent need to focus on improvements in healthcare and supports and the wider determinants of health in this population, which differ from the general population.

Paper II **Hughes-McCormack L.,** Rydzewska, E., Henderson, A., MacIntyre, C., Rintoul, J., Cooper S-A. (2017). Prevalence of mental ill-health and relationship with physical health in a whole country population of 26,349 people with intellectual disabilities, compared with the general population. *British Journal of Psychiatry Open.* (doi:10.1192/bjpo.bp.117.005462) (PMID:29034100) (PMCID:PMC5620469)

Building upon the research conducted in the previous paper, in addition to general health, we were also able to investigate another very important aspect of health from the Census 2011,

specifically mental health. This paper specifically sought to determine the prevalence of mental health conditions and relationships with general health in a total population with and without intellectual disabilities with data gathered again from Scotland's 2011 Census. We initiated this project as mental ill health is unevenly distributed across the population and appeared to be more prevalent among people with intellectual disabilities compared with the general population. However, most previous studies had serious limitations. National Institute for Health and Care Excellence (NICE) clinical guideline development group (NICE, 2016) reviewed the epidemiological evidence on prevalence of mental ill health in people with intellectual disabilities compared with the general population, and concluded they were not very confident in the results, in view of the variation in quality of the studies, and the need to include administrative samples because of limited population-based cohorts. We felt investigating associations with mental and general health were also important having already demonstrated that poor general health is more common among people with intellectual disabilities, meaning these groups of people are at high risk of comorbid physical and mental ill health. In the general population, this combination is associated with poorer outcomes, such as premature death.

To investigate the prevalence of mental ill health in the population with and without intellectual disabilities, and the relationship of mental and physical health in the population with intellectual disabilities, we included the same sample from **Paper I** (census responses from 94% of the Scottish population (n=5,269,054) and people identified with intellectual disabilities (n=26,349)). The data was categorised by age (8 bands) and sex, we reported frequency data on mental health and we conducted logistic regressions to determine the odds ratios (OR) of intellectual disabilities predicting poor mental health and associations with general health, controlling for age and sex.

This Scottish total country study is the largest and most complete population study we have identified investigating reported mental health of people with intellectual disabilities compared with people in the general population across the lifespan. It also reports novel data on general health associations with mental ill health. The findings provide vital knowledge that people with intellectual disabilities of all ages have substantially higher rates of mental health conditions than the rest of the population, and their mental health conditions are often associated with poor general health. These are essential findings for general psychiatrists and mainstream psychiatric services, as well as for carers, who need to be aware of the substantial burden of mental health conditions and its co-existence with poor general health. Without this information, the potential for misdiagnosis is high, and inadequate or inappropriate treatment is a risk. Moreover, these findings provide information about the extent of mental ill-health in children and adults separately. Importantly, mental health in children and young people with intellectual disabilities is not well understood, with a lack of regular and up-to-date data on the prevalence of mental health problems during this stage being widely criticised. Consistent with previous UK research (Hassiotis and Turk, 2012) in England, in which detailed individual clinical assessments of mental health were conducted, the findings of this research found there is an increased prevalence of mental disorders in children and young people with intellectual disabilities in the total population of Scotland, compared to the age matched general population. This adds to the currently limited body of knowledge about mental ill health in children and young people with intellectual disabilities. We concluded that given the associations we report, more research is needed to determine relationships between specific mental health problems, physical conditions and associated factors, for example, adversity, among people with intellectual disabilities, to help influence the development of appropriate interventions, and health and social care policy.

This manuscript was discussed in an editorial commentary for the British Journal of Psychiatry Open (Sheehan et al, 2018) which read: *"The recent publication in this journal of a study reporting the prevalence of mental illness and its association with mental health conditions in people with intellectual disability is a timely reminder of the additional mental and physical health needs of this group. The authors use data collected in the 2011 Scottish census, thereby achieving almost universal population coverage, demonstrating once again the power of exploiting routinely collected data in health research. The analysis showed that people with intellectual disability were seven times more likely to self-report (or their caregiver to proxy-report) a current mental health condition than those in the comparison group without intellectual disability. This apparent higher rate of mental ill health exists across the lifespan and is associated with significant additional personal, social and economic costs. The authors highlight the relative paucity of research evidence for effective psychotherapeutic interventions in this group and highlight the continuing need to improve individual outcomes and quality of life."*

Paper III Rydzewska, E., **Hughes-McCormack, L-A.,** Gillberg, C., Henderson, A., MacIntyre, C., Rintoul, J., Cooper, S-A. (2018). General health of adults with autism spectrum disorders - a whole country population cross-sectional study. *Research in Autism Spectrum Disorders.* (doi:10.1016/j.rasd.2019.01.004)

This next paper utilised similar research questions and methodology as described previously, with whole country data, drawn from Scotland's Census, 2011, this time, to investigate general health status in an adult population with autism compared with other adults. This study focused on adults only due to the reasons previously discussed, in relation to potential

differences in the adult and child populations, due to broadening of the concept of autism spectrum in recent years. The data for children and young people was investigated separately and is presented later (**Paper IV**). We identified people with autism from the individual Census questionnaire, from Question 20, on those who responded 'yes' to developmental disorder. We identified autism in 6,649/3,746,584 (0.2%) adults aged 25+years, of whom 46.8% (N=3,111) had poor general health, compared with 23.7% (N=887,878) of other people. Autism had OR = 5.1 (4.9–5.4, 95% CI) for predicting poor general health, or OR = 7.5 (6.9–8.2, 95% CI) when the interaction with age was included. Poor health was common across the entire lifespan for adults with autism but was more common at older age, and for women. Recommendations from this paper include that poor general health merits attention across the full life for adults with autism. We highlight that health practitioners need to be alert to the burden of potential health problems to seek them out to be addressed, and so the health agenda can turn towards potential mechanisms for prevention and better support for adults who may call upon services for people with autism.

Paper IV Rydzewska, E., **Hughes-McCormack, L-A.,** Gillberg, C., Henderson, A., MacIntyre, C., Rintoul, J., Cooper, S-A. (2019). Age at identification, prevalence and general health of children with autism: observational study of a whole country population. *BMJ Open*. e025904. (doi:10.1136/bmjopen-2018-025904) (PMID:31289063)

As an extension of **Paper III,** this paper reports the findings of the prevalence of autism, age of reporting/identifying autism, and general health status of children/young people with autism. This paper was focused only on children and young people aged 0-24 with and without autism. This includes a sample of 25,063 children and young people with autism out

of 1,548,819 (1.6%) of the total population of children and young people in Scotland. Autism had OR=11.3 (11.0 to 11.7) in predicting poor health. Autistic females had poorer health than autistic males, OR=1.6 (1.5 to 1.8). The study demonstrates that children and young people with autism reported poor general health, compared with children and young people without autism. These findings contribute to the currently limited evidence base on the general health of children and young people with autism. Accurate information on the proportion of children with autism, and their health status, is essential to accurately plan appropriate prevention and intervention measures, and provide resources for those who may put demand upon services designed for people with autism.

Paper V Rydzewska, E., **Hughes-McCormack, L-A.,** Gillberg, C., Henderson, A., MacIntyre, C., Rintoul, J., Cooper, S-A. (2018). Prevalence of long-term health conditions in adults with autism - observational study of a whole country population. *BMJ Open.* e023945. (doi: 10.1136/bmjopen-2018-023945) (PMID:30173164) (PMCID:PMC6120653)

We established in the previous papers discussed that people with intellectual disabilities and people with autism have significantly poorer health across the lifespan compared to people without these conditions. In this paper, we sought to further investigate some specific long term conditions in adults with autism. This paper investigated the prevalence of comorbid mental health conditions, sensory impairments and physical disabilities in the population of adults aged 25+ with and without reported autism (sample as described in **Paper III)**. Some conditions, such as epilepsy and intellectual disabilities are known to be common among people with autism. There is a general dearth of research on other long-term conditions such as hearing and visual impairments and physical disabilities. We investigated the prevalence

of six comorbidities: deafness or partial hearing loss, blindness or partial sight loss, intellectual disabilities, mental health conditions, physical disability, and other condition; odds ratios (95% confidence intervals) of autism predicting these comorbidities, adjusted for age and gender; and odds ratios for age and gender in predicting comorbidities within the population with reported autism. All comorbidities were more common for adults with autism. Autism statistically predicted all of the conditions: OR 3.3 (95% CI 3.1 to 3.6) for deafness or partial hearing loss, OR 8.5 (95% CI 7.9 to 9.2) for blindness or partial sight loss, OR 94.6 (95% CI 89.4 to 100.0) for intellectual disabilities, OR 8.6 (95% CI 8.2 to 9.0) for mental health conditions, OR 6.2 (95% CI 5.8 to 6.6) for physical disability and OR 2.6 (95% CI 2.5 to 2.8) for other conditions. We recommend that clinicians need heightened awareness of comorbidities in adults with autism to improve detection and suitable care, especially given the added complexity of assessment in this population and the fact that hearing and visual impairments may cause additional difficulties with reciprocal communication which are also a feature of autism; hence posing further challenges in assessment.

Paper VI Rydzewska, E., **Hughes-McCormack, L-A.,** Gillberg, C., Henderson, A., MacIntyre, C., Rintoul, J., Cooper, S-A. (2018). Prevalence of sensory impairments, physical and intellectual disabilities, and mental health in children and young people with self/proxy-reported autism - observational study of a whole country population. *Autism.* (doi: 10.1177/1362361318791279) (PMID:30328695)

As an extension of the previous paper, the purpose of this paper was to investigate comorbid conditions in a whole-country population of children/young people with autism compared to children/young people without autism. If individual long-term conditions are indeed more

common in children and young people with autism, then a greater awareness of them would be important. Existing studies are mostly small and few are contemporary with regards to the prevalence of intellectual disabilities and mental health conditions in children and young people with autism; results are particularly limited with regards to visual and hearing impairments, and we found no studies on physical disabilities. With the data from the Scottish census, we were able to investigate these understudied comorbid conditions in a total population study.

The same cohort of children and young people with and without autism aged 0-24 years as described in **Paper IV** was included for the current paper. We calculated the number and percentage of children and young people with and without autism reporting deafness or partial hearing loss, blindness or partial sight loss, intellectual disabilities, mental health conditions, physical disability, and other conditions. As also found for adults with autism, each of the long-term conditions investigated was substantially more common in the population of children/young people with autism than in other children/young people. Autism had an odds ratio of 5.4 (5.1–5.6) for predicting deafness/partial hearing loss, odds ratio of 8.9 (8.1–9.7) for blindness/partial sight loss, odds ratio of 49.7 (38.1–64.9) for intellectual disabilities, odds ratio of 15.7 (13.4–18.5) for mental health conditions, odds ratio of 15.8 (14.1–17.8) for physical disability and odds ratio of 3.9 (3.8–4.0) for other conditions. This finding is very important, considering each of these conditions are disabling, and can significantly impact upon learning, functioning and longer-term quality of life when they occur as single conditions; clearly their coexistence with autism adds extra complexity. We recommend from the findings that practitioners need a heightened response to the possibilities of comorbidities with autism, particularly of hearing and visual impairments and physical disabilities.

Paper VII Kinnear, D., Rydzewska, E., Dunn, K., **Hughes-McCormack L-A.,** Melville, C., Henderson A., Cooper S-A. (2019). Relative influence of intellectual disabilities and Autism on mental and general health in Scotland: a cross-sectional study of a whole country of 5.3 million children and adults. *BMJ Open***.** e029040. (doi: 10.1136/bmjopen-2019-029040) (PMID:31462474)

In this paper based on Scottish census 2011 data, the aim was to determine the relative extent that autism and intellectual disabilities are independently associated with poor mental and general health, in children and adults. We identified people with intellectual disabilities or autism from the individual census questionnaire, as previously discussed from Question 20. In children/youth, intellectual disabilities (OR 7.04, 95% CI 6.30 to 7.87) and autism (OR 25.08, 95% CI 23.08 to 27.32) both independently predicted mental health conditions. In adults, intellectual disabilities (OR 3.50, 95% CI 3.20 to 3.84) and autism (OR 5.30, 95% CI 4.80 to 5.85) both independently predicted mental health conditions. In children/youth, intellectual disabilities (OR 18.34, 95% CI 17.17 to 19.58) and autism (OR 8.40, 95% CI 8.02 to 8.80) both independently predicted poor general health. In adults, intellectual disabilities (OR 7.54, 95% CI 7.02 to 8.10) and autism (OR 4.46, 95% CI 4.06 to 4.89) both independently predicted poor general health. This large study is the first study to have reported on the extent to which autism and intellectual disabilities are independently associated with poor mental and general health, in children and adults. These findings have implications for health services and supports, as they indicate that poor mental and general health is not just related to the coexistence of these conditions (intellectual disabilities and autism), or due to just having intellectual disabilities.

Paper VIII Melville, C., McGarty, A., Harris, L., **Hughes-McCormack, L.A.,** Baltzer, M, McArthur, L., Morrison, J., Allan, L., Cooper, S-A. (2017). A population-based, cross-sectional study of the prevalence and correlates of sedentary behaviour of adults with intellectual disabilities. *Journal of Intellectual Disability Research*. (doi:10.1111/jir.12454) (PMID:29214701)

This paper was the first study to publish population-based data on the prevalence and correlates of sedentary behaviour in adults with intellectual disabilities. Unhealthy dietary patterns and low levels of physical activity have been shown to contribute to the increased prevalence of obesity (National Institute for Health and Care Excellence 2014), diabetes (Balogh et al. 2015) and mental ill-health (Cooper et al., 2007) experienced by adults with intellectual disabilities. Investigating unhealthy dietary patterns and low levels of physical activity is therefore important to understanding the overall health of adults with intellectual disabilities. In this study, adults with intellectual disabilities (from NHS Greater Glasgow and Clyde Health Board area) were invited to take part in a comprehensive health check programme. Demographic and health data were collected during semi-structured interviews and physical examination. Individuals who opted into the health check were invited to consent to the health check data being used for research purposes. Screen time was used as a proxy measure of sedentary behaviour. Bivariate and multivariate statistical modelling examined correlates of screen time. There was a high participation rate in the research as consent was gained for 725 of 836 (87.0%) invited adults. The findings show compared with adults who do not have intellectual disabilities, adults with intellectual disabilities have higher levels, and different correlates, of sedentary behaviour. Fifty per cent of the 725 participants reported four or more hours of screen time per day. Male gender, higher levels of intellectual ability, mobility problems, obesity, not having hearing impairment and not having

epilepsy were all significantly associated with higher screen time in the final multivariate model (R^2 = 0.16; Hosmer–Lemeshow goodness of fit statistic P = 0.36). We recommend that supporting adults with intellectual disabilities to make positive lifestyle behaviour changes is a priority to reduce health inequalities. These findings provide a better understanding of sedentary behaviour among adults with intellectual disabilities that can be used to inform the design of effective behaviour change programmes.

Paper IX Ward, L., Cooper, S.-A., **Hughes-Mccormack, L-A**., Macpherson, L., and Kinnear, D. (2019). Oral health in adults with intellectual disabilities: a systematic review. *Journal of Intellectual Disabilities Research*. 63(11), pp. 1359-1378. (doi: 10.1111/jir.12632) (PMID:31119825)

Oral health is an important factor in overall general health and wellbeing, and some previous reports indicated that people with intellectual disabilities experience poorer oral health compared to the general population (Crowley et al., 2005; Mac Giolla Phádraig et al., 2014) but quantification of its current extent was less clear. This systematic review reported here was conducted to quantify its extent. This systematic review was prospectively registered with the International Prospective Register of Systematic Reviews (PROSPERO) and was completed according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA; Shamseer et al., 2015). 33 of the 3,958 retrieved articles were selected for inclusion, based on pre-defined inclusion criteria. The conclusions of the review were that adults with intellectual disabilities still experience a high burden of poor oral health, and this is also related to systemic diseases. The recommendations have important implications for raising awareness of oral hygiene needs amongst carers of people with intellectual disabilities and professionals treating these groups of people.

Summary of Theme I

People with intellectual disabilities and people with autism are thought to experience serious, life limiting health inequalities compared to the general population (NHS Scotland, 2004; Ouelette-Kuntz et al., 2004; Simonoff et al., 2008; Oesburg et al., 2011; Cooper et al., 2015; Croen et al., 2015; National Longitudinal Transition Study 2, 2017). Despite this, there were significant evidence gaps related to their health and health care (Robertson et al., 2015; Bishop-Fitzpatrick et al., 2017). The publications discussed in theme I, presented a wide array of evidence to quantify the extent that people with intellectual disabilities and people with autism experience poorer health compared to the general population. The findings of the studies presented here are a useful resource to inform policy decisions.

As with any study, there are limitations with each of the studies discussed. The Census data responses were completed by heads of households; and some will have been self-reports and some proxy-reports, though we cannot identify which was which. The responses in the census questionnaire also reflect subjective reports, rather than objective measurements of health. Despite these limitations, the response options in question 20 of the census questionnaire makes this a rare data set, and possibly the only country census that identifies people with intellectual disabilities (living in private households or communal establishments) and distinguishes these from specific learning difficulties, such as dyslexia, and also from autism. This made it possible for us to identify a wealth of information about people with intellectual disabilities and people with autism. This is particularly important considering research of mental or general health involving people with intellectual disabilities/autism is often very limited by problems such as a failure to include people with severe/profound intellectual disabilities, low response rates/small sample sizes, inclusion of private households only,

unsuitable methods of identifying intellectual disabilities/autism and restricted ages of adults or children included (NICE, 2016; Emerson et al., 2014).

My involvement with the census data goes beyond the writing of manuscripts as I led on developing the 'learning disabilities' data content for our Scottish Learning Disabilities Observatory website. I personally added 489 infographics, and 102 data tables relating to census data. There is evidence that this information is making an impact, as since 2016, the webpages have had 14,387 unique visitors worldwide and 22,566 overall visits, with 3.52 pages viewed each time on average. I have presented the information widely at academic (e.g., international conferences) and non-academic contexts (e.g., to professionals, parents/carers and people with intellectual disabilities and people with autism).

Theme II: Health care of people with intellectual disabilities and Down syndrome

Leading on from **Theme I,** given that people with intellectual disabilities experience poorer health, it has been recognised that access to high quality health care is vital and that improvements are needed (Sheehan et al., 2016). Despite this recognition, there has been a serious lack of studies investigating how health is managed in health care settings for people with intellectual disabilities compared to the general population. This type of research is highly indicated as people with intellectual disabilities face barriers in accessing health services (Hanlon et al., 2018), compounded by communication difficulties, and organisational and social support limitations (Ouelette-Kuntz et al., 2004). To reduce inequities, primary health care providers need to effectively manage long-term conditions in keeping with best practice. However, available evidence suggests this may not always occur. No previous

research had quantified the extent of differences in the management of long term health conditions in people with intellectual disabilities compared to the general population. The first two related papers, **Paper X** and **XI** presented here are the first studies to quantify the management of long term condition in primary care from a population based cohort study. Following this, a systematic review is presented, investigating further the quality of health care of people with intellectual disabilities, specifically, of hospitalisation for physical conditions.

Paper X Cooper, S-A., **Hughes-McCormack, L-A.,** Greenlaw, N., McSkimming, P., McConnachie, A., Allan, L., Baltzer, M., McArthur, L., Henderson, A., Melville, C., Morrison, J. (2018). Management and prevalence of long-term conditions in primary health care for adults with intellectual disabilities compared with the general population: a population based cohort study. *Journal of Applied Research in Intellectual Disabilities*. (doi:10.1111/jar.12386) (PMID:28730746)

This study utilised indicators of best practice from the "Quality and Outcomes Framework" (QOF)[2] to measure the management of long term conditions and the extent of disease prevalence in people with intellectual disabilities (n=721). Data was collected manually from primary care health records following consent for each person with intellectual disabilities in keeping with Scottish law. This was compared to findings of the routinely collected data from the Quality Outcomes Framework in the same time frame for all people without intellectual

[2] In the UK (and in Scotland until 2016), the contract between general practitioners and the government health departments includes pay for performance on specific indicators of management of long-term conditions which are considered to be evidence-based best-practice (the "Quality and Outcomes Framework").

disabilities (n=764,672) in Greater Glasgow and Clyde health board (NHS GG&C) throughout 2007-2010.

Adults with intellectual disabilities were significantly more likely than adults in the general population to have epilepsy (28.2% versus 0.8%, $p < .0001$), psychosis (7.6% versus 0.9%, $p < .0001$), asthma (9.2% versus 5.3%; $p < .0001$), diabetes (6.4% versus 3.4%; $p < .0001$), heart failure (2.5% versus 0.9%, $p < .001$) and hypothyroidism (5.3% versus 2.8%, $p = .0001$). No significant difference was found between the two populations in the rates of chronic obstructive pulmonary disease, coronary heart disease, chronic kidney disease or stroke. Whilst hypertension was common amongst the people with intellectual disabilities (12.8%), it occurred at a similar rate as in the general population. Despite having a higher prevalence across a number of long-term conditions from the Quality and Outcomes framework, the findings from this publication report that long term conditions are managed poorly for people with intellectual disabilities compared to the general population. Adults with intellectual disabilities received significantly poorer management of all long-term conditions on 38/57 (66.7%) indicators. Achievement was high (75.1%–100%) for only 19.6% of adults with intellectual disabilities, compared with 76.8% of the general population. Implications from the study include finding practical and implementable means of supporting general practices in providing equitable care for people with intellectual disabilities.

Paper XI **Hughes-McCormack, L-A.,** Greenlaw, N., McSkimming, P., McCowan, C., Ross, K., Allan, L., Baltzer, M., McArthur, L., Henderson, A., Melville, C., Morrison, J., Cooper, S-A. (2018). Changes over time in the management of long-term conditions in primary health care for adults with intellectual disabilities, and the health care inequality gap compared with the general

population. *Journal of Applied Research in Intellectual Disabilities.* 34(2), pp.634-647. (doi: 10.1111/jar.12833) (PMID:33283349)

This study is the follow up to **Paper X.** It was conducted to investigate the extent of, and changes in, the primary health-care inequality gap over time, regarding management of long-term conditions. In 2014, individual patient data were extracted electronically from GP records of all practices participating in the NHS Greater Glasgow and Clyde intellectual disabilities enhanced service (n=3,638). Comparisons at both times were made with the NHS GG&C's general population.

In 2014, the extent of the health-care inequality gap had reduced for people with intellectual disabilities compared to the general population. Specifically, the inequalities between adults with intellectual disabilities and the general population was greatest in 2007-2010 ($p<0.001$). In adults with intellectual disabilities, there was a significant difference between the time points, indicating that there has been an improvement in the proportion of indicators met over time ($p < .001$), whereas there is no difference over time for the general population ($p = .718$). Group effects were examined within each time point, and adults with intellectual disabilities were less likely to have a high proportion of indicators met than the general population at both time points; however, the higher proportion of indicators met for adults with intellectual disabilities were 5.32 times more likely in 2014 than in 2007-2010 (95% CI (2.69, 10.55). So, although there appeared to be a significant healthcare inequality gap between the two populations in 2014, the extent of the gap had been reduced from 2007-2010.

The study suggests NHS GG&C effected a step-change in health-care of adults with intellectual disabilities and reduced health-care inequality. Health indicators are process measures, but considered as best practice, and the pathway to improved health, so they are important. In NHS GG&C, several initiatives were underway in the intervening period to support primary health-care services in delivery of health-care, including a programme of health checks for adults with intellectual disabilities, and dedicated support from the intellectual disabilities health service to primary care. It is possible that these initiatives contributed to the results we report, and if so, demonstrate that poor health-care is not inevitable for people with intellectual disabilities and improvements can be made. The local health check programme is a plausible explanation for the positive changes observed: randomised control trials (RCTs) have demonstrated health checks for adults with intellectual disabilities versus treatment as usual are clinically and cost effective (Cooper et al, 2014). In this publication the remaining inequalities highlight the need still for further action, such as annual health checks.

Paper XII Dunn, K., **Hughes-McCormack, L-A.,** Cooper, S-A. (2017). Hospital admissions for people with intellectual disabilities: Systematic Review. *Journal of Applied Research in Intellectual Disabilities.* (doi:10.1111/jar.12360) (PMID:28467010)

This purpose of this paper was to investigate if people with intellectual disabilities have inequalities in hospital admissions for physical health conditions compared with the general population. Rates of hospitalization for "ambulatory care-sensitive conditions," have been used as an indicator of access to, and quality of primary care. Ambulatory care-sensitive conditions are defined as conditions which, if managed effectively at the primary care level,

should not lead to a hospital admission, for example, epilepsy and diabetes. However, from previous research, it is unclear if people with intellectual disabilities are admitted to hospital more frequently than the general population for ambulatory care-sensitive conditions.
A systematic review was conducted, searching six databases using terms on intellectual disabilities and hospital admission. This systematic review was prospectively registered with the International Prospective Register of Systematic Reviews (PROSPERO) and was completed according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA; Shamseer et al., 2015). Papers were selected based on pre-defined inclusion/exclusion criteria, data extracted, tabulated and synthesized and quality assessed. Seven of 29,613 papers were included. This research found people with intellectual disabilities are admitted to hospital more frequently than the general population for ambulatory care-sensitive conditions. Asthma and diabetes admission data suggest suboptimal primary health care for people with intellectual disabilities compared with the general population, although evidence is limited. This supports the previous findings of **Paper X**, that people with intellectual disabilities are not receiving the appropriate treatment in primary care settings.

Summary of Theme II

As discussed in detail within each of the published papers, there are limitations to each study. **Paper X** and **XI** included only one health board, although this is the largest health board in Scotland. A further limitation is that the long-term conditions included in the Quality and Outcomes Framework may not include all those most relevant to people with intellectual disabilities. Despite limitations, using an established evidence based approach to measuring the quality of primary health care, findings indicate people with intellectual disabilities are receiving lower quality health care compared to other people. This is particularly concerning

as we know that people with intellectual disabilities experience poorer health and higher levels of multimorbidity compared to peers (NHS Scotland, 2004; Ouelette-Kuntz et al., 2004; Oesburg et al, 2011; Cooper et al., 2015). The failings in the management of long-term conditions have implications for both policy and practice. There appears to be an urgent need for staff training in primary care about the specific health needs of people with intellectual disabilities. This is supported by previous research exploring practice nurses' perceptions and experiences of delivering an anticipatory health check for adults with intellectual disabilities in primary care. The study involving 11 practice nurses (from health check intervention practices in NHS Greater Glasgow and Clyde) found that the nurses often reported modifying the health check to respond to individual patients' needs, and as a result, this limited the identification of new or potentially unrecognised health needs - instead the health check process was dominated by previously recognised health 'problems' or issues (MacDonald et al., 2018). We recommended from these studies that the quality of healthcare for people with intellectual disabilities could be enhanced with introduction of specific interventions such as further training in primary care including training on health checks for people with intellectual disabilities which could make a big improvement to the poor health of people with intellectual disabilities.

Theme III: Survival/ death of people with intellectual disabilities and Down syndrome

Since the 1970s, there has been growing evidence to suggest people with intellectual disabilities experience premature deaths compared with the general population (Heslop et al, 2014). The following papers were initiated in response to the need for a robust appraisal of the evidence to gain a better understanding of the life expectancy, mortality rates and cause of

deaths of people with intellectual disabilities and Down syndrome, so that actions to reduce premature deaths could be identified.

Paper XIII O'Leary, L., **Hughes-McCormack, L-A.,** Cooper, S-A. (2017). Early death and causes of death of people with intellectual disabilities: a systematic review. *Journal of Applied Research in Intellectual Disabilities.* 31(3), pp. 325-342. (doi: 10.1111/jar.12417) (PMID:28984406) *(*doi:10.1111/jar.12417)

This systematic review investigated mortality among people with intellectual disabilities compared to the general population to identify any trends in terms of the extent of the health inequality and the pattern of cause of death. This systematic review was prospectively registered with the International Prospective Register of Systematic Reviews (PROSPERO) and was completed according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA; Shamseer et al, 2015). Of 19,111 retrieved articles, 27 met criteria. This systematic review found mortality rates are higher in the intellectual disabilities population, compared with the general population, with an average age of death of up to 20 years lower. Respiratory disease and circulatory diseases were found to be the main common causes of death. However, cancer was less common, and cancer profile differed from the general population. An interesting finding emerged from the results; mortality rates between the population with intellectual disabilities and the general populations were found to be greater for females than males.

Paper XIV O'Leary, L., **Hughes-McCormack L-A,** Dunn, K., Cooper S-A. (2017). Early death and causes of death of people with Down syndrome: a systematic review. *Journal of Applied Research in Intellectual Disabilities.* (doi:

10.1111/jar.12446) (PMID:29573301)

Related to **Paper XIII,** this systematic review used the same methodology to investigate mortality among people with Down syndrome compared to the general population and found 34 articles that met criteria. This systematic review was prospectively registered with the International Prospective Register of Systematic Reviews (PROSPERO) and was completed according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA; Shamseer et al, 2015). Findings indicate mortality rates are higher in the Down syndrome population, compared with the general population, with an average age of death - up to 28 years lower. Despite this, survival rates have improved over time, particularly for those with congenital heart anomalies. It is hypothesised that this is in part, due to improvements in health and social care and attitudes to disability in recent decades (Bull et al, 1985; Hijii et al, 1997; Carothers et al, 1999; Jensen, 2014; Jacobs et al, 2016). This led to an investigation of birth rates and death rates (throughout a 25-year period) among children/young people with Down syndrome in Scotland, presented in the following **Paper XV,** to investigate any changes in rates over time.

Paper XV **Hughes-McCormack, L-A.,** McGowan, R, Pell, J., Mackay, D., Henderson, A., O'Leary, L., Cooper S-A. (2019). Birth incidence, deaths, and hospitalisations of children and young people with Down syndrome in Scotland, 1990-2015: birth cohort study. *BMJ Open.* e033770. (doi: 10.1136/bmjopen-2019-033770) (PMID:32241786)

There is a lack of up-to-date research which investigates live-birth rates, deaths and hospital admission needs of children/young people with Down syndrome in Scotland. This makes it

challenging to plan for the support that children with Down syndrome and their families need. In this population-based cohort of linked birth and hospital admissions, we found the Scottish incidence of Down syndrome live-births to be 1.0/1,000 births over the last 25 years, with it being possibly higher now than in the early 1990s. The Scottish birth rate has fallen overall, but not for babies with Down syndrome. Also, consistent with previous literature (**Paper XIV**), there were more deaths among children and young people with Down syndrome (n=92; 7.4%) over the 25-year period compared to children and young people without Down syndrome (n=23; 0.4%); that is 18.5 times more than controls. We found Down syndrome among children and young people to be associated with increased risk of hospitalisation as reported in the limited body of previous research (Zhu et al, 2013; Fitzgerald et al, 2013; Ting et al, 2016). More of the Down syndrome group had at least one admission (1,105 [89.5%] versus 3,305 [53.5%]; adjusted HR=1.84 [1.68, 2.01]), and re-admissions (945 [85.5%] versus 1,685 [51.0%]; adjusted HR=2.56 [2.08, 3.14]). More of their admissions were emergencies (78.1% versus 71.6%; first emergency admission adjusted HR=2.87 [2.61, 3.15]), and longer duration (5.03 days [Median=2], versus 1.78 days [Median=1]) by 3.25 days; adjusted (log transformed) Coefficient=0.25 (CI 0.22-0.28). Admission rates increased from 1990-2003 to 2004-2015 for the Down syndrome group (90.7% to 92.2%) and decreased for controls (63.3% to 44.8%). The recommendations of this study highlight a need to develop tailored packages of support for families and/or carers with children and young people with Down syndrome to cope with the evident higher health care needs of their offspring, and support for the impact of hospitalisation on the developing child with Down syndrome.

Summary of Theme III

The evidence suggests that there are disproportionately poor outcomes of mortality for people with intellectual disabilities, with, and without Down syndrome. Whilst lifespan has increased, the gap with the general population does not appear to have decreased other than for people with Down syndrome, who still on average die earlier. In these studies, the pattern of cause of death also differs from the general population, with respiratory disease and circulatory disease being the most common underlying causes of death for people with intellectual disabilities. However, a lack of detailed information regarding immediate and contributory causes of death is a limitation of previous research. These studies show that reducing the risk of early death among people with intellectual disabilities should be a priority area of research to address the unequal health experienced by this population. It is recommended from these studies, that more robust, standardized data in relation to identification and characteristics of people with intellectual disabilities, and standard age bandings and definition of intellectual disabilities across countries is necessary to understand leading factors that influence specific causes of death in people with intellectual disabilities. Moreover, as indicated previously in this thesis, people with intellectual disabilities experience poorer management of their long-term conditions in primary care settings and more avoidable hospital admissions, which also may be an indicator of lower quality primary care. While access to good quality and timely healthcare is vital for optimal health, the potential influence of the quality of primary health care and access to services among people with intellectual disabilities in relation to premature death is currently unknown and should be investigated in future research. This is necessary to identify factors that can be modified to improve health, healthcare and reduce premature mortality in people with intellectual disabilities.

Concluding remarks:

This thesis has presented for each of the three inter-related themes, a range of my peer reviewed publications from international journals. A previous dearth of empirical evidence about the health and health care of people with intellectual disabilities and people with autism has presented barriers to understanding the complex factors that produce differential health outcomes. The research presented in this thesis has provided robust evidence on the extent of poor health, health care, and premature mortality among people with intellectual disabilities, and people with autism which has not previously been quantified in research. The Keys to Life, was most recently updated in 2019, with input from my research, and research conducted by others in the Institute and other partners. The updated strategy has set out plans to further develop the vision around nurturing healthy lives, choice and control, independence and active citizenship for people with intellectual disabilities in Scotland (Scottish Government, 2019).

A key feature of the work presented is that it contributes to health improvement by providing information, data, and intelligence on the health and healthcare of people with intellectual disabilities and people with autism. The collection of work summarised demonstrates that I have made a substantial contribution to research evidence that will help raise awareness of health inequalities experienced by people with intellectual disabilities and people with autism, to help inform healthy public policy. Specifically, the evidence in the first theme of this thesis, *health of people with intellectual disabilities and autism,* has provided novel information, from a total population dataset, based on the Scottish Census in 2011, which is the first country Census (to our knowledge) that identified people with intellectual disabilities (separately from specific learning difficulties) and people with autism. From this unique dataset, the prevalence of mental health and general health experienced by people with intellectual disabilities and people with autism was quantified, including long-term conditions

for people with autism and general health associations with mental ill health in people with intellectual disabilities. The findings provide vital knowledge that people with intellectual disabilities of all ages have substantially higher rates of mental health conditions than the rest of the population, and their mental health conditions are often associated with poor general health. Research investigating the prevalence of long-term conditions in people with autism was previously very limited, and this research has provided new knowledge on the extent of the inequalities faced by this population in specific relation to deafness/partial hearing loss, blindness/partial sight loss, intellectual disabilities, mental health conditions, physical disability, and other conditions. Each of the long-term conditions investigated was substantially more common in the population with autism than in other people. These are findings which are very important, especially considering each of these conditions can be disabling.

The research in the second theme of this thesis regarding ***health care of people with intellectual disabilities and Down syndrome***, reported evidence from a study which utilised indicators of best practice from the "Quality and Outcomes Framework" (QOF) to measure the extent of inequality across a wide range of long-term conditions and health-promoting actions in the largest health board in Greater Glasgow and Clyde for people with intellectual disabilities compared to the general population. This study found that people with intellectual disabilities experience poorer quality of primary health care compared to the general population. A systematic review study also found people with intellectual disabilities were admitted to hospital more frequently than the general population for ambulatory care-sensitive conditions, which further indicates suboptimal primary health care for people with intellectual disabilities compared with the general population. However, in a follow up study (post a health check intervention) to the study in Greater Glasgow and Clyde (pre health

check), there was a step-change in primary health-care of adults with intellectual disabilities and reduced health-care inequality (on indicators of best practice from the "Quality and Outcomes Framework"). There was a significant difference between the time points for adults with intellectual disabilities, indicating that there has been an improvement in the proportion of quality indicators met over time, whereas there was no difference over time for the general population. While remaining inequalities between the population with intellectual disabilities and the general population were still apparent, highlighting further action is still necessary, the findings clearly demonstrated that poor health-care is not inevitable for people with intellectual disabilities and improvements can and should be made. This study provides extremely important evidence, as it is the first study (to our knowledge) to have demonstrated improvements in health care for adults with intellectual disabilities relative to the general population over time. This is very important, as good health care is one of the pathways to better health, and, as evidenced in this thesis, poor health and premature death are substantial problems for people with intellectual disabilities. This finding is additionally important, as despite public health services having worked for decades to reduce health and healthcare inequalities in minority populations (Marmot, 2010), there have been few successful examples such as this (Mackenbach, 2010; National Audit Office, 2010). Health checks have been shown to be clinically effective in detecting unmet need and identifying potentially treatable conditions – many of which have been shown by the Observatory to be highly prevalent in the intellectual disabilities population; and supporting targeted action to address health needs. Therefore, annual health checks are recommended as good practice for addressing the mental and physical health care needs of people with learning disabilities. These have not been routinely available in Scotland, although they are now currently being rolled out. As previously discussed, the research in this thesis relates only to one health board in Scotland. It is important that we investigate the impact of health checks on people with

intellectual disabilities more widely across Scotland in the future to identify their impact. There is still a lot more to be understood about the impact of health checks on people with intellectual disabilities, in relation to diagnostic overshadowing and premature mortality, for example. These types of further investigations are necessary to help identify the strengths and limitations of health checks, and where further action may still need to be taken.

The third theme of research, *survival/ death of people with intellectual disabilities and Down syndrome*, that was presented in this thesis, found that, not only do people with intellectual disabilities have poorer physical and mental health, but they also have higher mortality rates compared with the general population, with an average age of death up to 20 years lower. Other significant findings from this research theme were that respiratory disease and circulatory diseases were much more common causes of death in people with intellectual disabilities compared to the general population, with over-representation of deaths that could be considered avoidable, including mortality amenable to optimal quality health care or mortality that is preventable by public health interventions focusing on wider determinants of public health, such as behaviour and lifestyle factors, and environmental factors. Amenable mortality from respiratory related illness among people with intellectual disabilities, highlighted from the evidence in this thesis, is a particular area of concern.

Leading on from the research in this thesis, the Scottish Learning Disabilities Observatory have continued to develop novel research, including data-linkage studies, designed to help reduce health morbidity and its associated impact on quality of life on people with intellectual disabilities and autism, and the risk of early mortality among people with intellectual disabilities. There has been a strong focus on quantifying the level of avoidable mortality in both adults and children with intellectual disabilities and understanding the specific causes of

this (Cooper et al., 2020; Smith et al., 2020). Our team recently conducted a systematic review and meta-analysis (Truesdale et al., 2021) which found people with intellectual disabilities face a 10-fold rate of respiratory death and 26-fold rate of death from pneumonia compared to the general population. A higher rate of respiratory deaths (55 times greater) was found among children and young people compared to their peers, than was found for adults with intellectual disabilities compared to peers. In relation to this, I have collaborated with an organisation in Scotland that works solely with people with profound and multiple learning disabilities (PMLD) and their families for a better life (PAMIS: Promoting a more inclusive society), along with another colleague from the Scottish Learning Disabilities Observatory. People with PMLD, have a number of complex healthcare needs, and may be at higher risk of poor health outcomes, including premature mortality (O'Leary et al., 2017). We worked together on a public engagement/ knowledge exchange project (developing a video with a bereaved parent of a child with intellectual disabilities, primary care clinician, speech and language therapist and a young person with a learning disability), to highlight practical ways, to help reduce avoidable respiratory deaths in children and young people with intellectual disabilities. This led to us being invited to contribute to the new Scottish Postural Care Strategy. We had input in relation to "Ambition 1. Everyone at risk of body shape deterioration will be empowered to self-manage their posture by being an equal partner in their care through a high-quality postural care service which encompasses training, information and support.". We helped to shape recommendation 7. That is, to generate high quality research to validate the impact of protection of body shape and translate and disseminate these research findings to inform future actions, policy and practice to benefit people at risk of body shape deterioration. Suggested priority areas for research included: long term outcomes in physical health, mental health and wellbeing, sleep, as well as the

social impact of postural care interventions, integration of postural care across different settings, e.g., education, acute care, accessibility of assessment and referrals.

The research in this thesis has provided a wide body of new knowledge about the health and health care of people with intellectual disabilities and autism. The research has found that people with intellectual and disabilities and autism experience more physical and mental health problems compared to the general population, and people with intellectual disabilities experience avoidable mortality, and lower quality of primary care at a much higher rate when compared to the general population. This research has provided a strong foundation for future research to further investigate specific factors, that are potentially modifiable, and that place people intellectual disabilities and people with autism at risk of developing avoidable illnesses, that can be life threatening. Further investigating this is vital to help identify the roots of any differences for people with intellectual disabilities and people with autism compared to the general population that increases risk of avoidable premature mortality and factors which may be amenable to changes, such as better healthcare (e.g. targeted health checks) and/ lifestyle/behavioural factors (e.g. risk factors for specific diseases, diet, exercise), staff training (e.g. better recognition of persons at risk of aspiration, and feeding posture for persons at risk of regurgitation/choking).

To reduce avoidable ill health and mortality among people with intellectual disabilities, there is an assumption that this may be possible by increasing access to health services as well as providing high-quality health care (Hanlon et al, 2018). However, health morbidity, healthcare, and mortality research have often been investigated separately, as individual strands of research, to date for people with intellectual disabilities. The potential contribution of primary health care quality and access factors in relation to avoidable ill health and

mortality for people with intellectual disabilities compared to the general population is unknown. There is a need now for longitudinal research, linking health care and avoidable mortality, using primary care data to investigate how a wide range of potentially modifiable primary care quality and access factors in primary care impact on avoidable (particularly amenable) mortality in people with intellectual disabilities compared to the general population across the lifespan. Until this is properly captured, primary care interventions that seek to improve health outcomes of people with intellectual disabilities are unlikely to work, due to the complexity of the health needs and experiences of health care among people with intellectual disabilities and their families. Addressing these issues may be crucial to reducing the risk of poor health outcomes, such as premature mortality, among this population. This type of research has potential to inform and strengthen policy guidance to improve NHS practice around primary care management of people with intellectual disabilities. This could also provide important avenues for future research, identify structural/ organisational changes in health services needed to overcome the substantial barriers to access, inform how to provide practical support and/ or training to health care professionals and to be able to communicate and support people with intellectual disabilities. There is an urgent need to find ways to best help and support people with intellectual disabilities and their families to experience better access to health care and higher quality of care from these vital community health services.

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Appendix 1: The publications included in the thesis:

Paper I (Full reference and link to the manuscript below)

Hughes-McCormack LA, Rydzewska E, Henderson A, MacIntyre C, Rintoul J, Cooper SA. Prevalence and general health status of people with intellectual disabilities in Scotland: a total population study. J Epidemiol Community Health. 2018 Jan;72(1):78-85. doi: 10.1136/jech-2017-209748. Epub 2017 Oct 25. Erratum in: J Epidemiol Community Health. 2018 Apr 28;: PMID: 29070675.

Prevalence and general health status of people with intellectual disabilities in Scotland: a total population study | Journal of Epidemiology & Community Health (bmj.com)

Paper II

BJPsych Open (2017)
3, 243–248. doi: 10.1192/bjpo.bp.117.005462

Prevalence of mental health conditions and relationship with general health in a whole-country population of people with intellectual disabilities compared with the general population

Laura A. Hughes-McCormack, Ewelina Rydzewska, Angela Henderson, Cecilia MacIntyre, Julie Rintoul and Sally-Ann Cooper

Background
There are no previous whole-country studies on mental health and relationships with general health in intellectual disability populations; study results vary.

Aims
To determine the prevalence of mental health conditions and relationships with general health in a total population with and without intellectual disabilities.

Method
Ninety-four per cent completed Scotland's Census 2011. Data on intellectual disabilities, mental health and general health were extracted, and the association between them was investigated.

Results
A total of 26 349/5 295 403 (0.5%) had intellectual disabilities. In total, 12.8% children, 23.4% adults and 27.2% older adults had mental health conditions compared with 0.3, 5.3 and 4.5% of the general population. Intellectual disabilities predicted mental health conditions; odds ratio (OR)=7.1 (95% CI 6.8–7.3). General health was substantially poorer and associated with mental health conditions; fair health OR=1.8 (95% CI 1.7–1.9), bad/very bad health OR=4.2 (95% CI 3.9–4.6).

Conclusions
These large-scale, whole-country study findings are important, given the previously stated lack of confidence in comparative prevalence results, and the need to plan services accordingly.

Declaration of interest
None.

Mental ill health is unevenly distributed across the population and appears to be more prevalent among people with intellectual disabilities compared with the general population.[1,2] An adult prevalence of 22.4% has been reported excluding problem behaviours and autism or 40.9% including problem behaviours and autism;[1] and a prevalence of 36% including problem behaviours has been reported in children and young people, compared with 8% in those without intellectual disabilities.[2] However, most studies have limitations. Indeed, a recent National Institute for Health and Care Excellence (NICE) clinical guideline development group[3] reviewed the epidemiological evidence on prevalence of mental ill health in people with intellectual disabilities compared with the general population, and concluded they were not very confident in the results, in view of the variation in quality of the studies, and the need to include administrative samples because of limited population-based cohorts. For example, the most detailed, population-based study of adults with intellectual disabilities to date included only 1023 and no direct general population comparison group,[1] and the most comprehensive study of children and young people with intellectual disabilities, which did include a general population comparison group, included only 641 with intellectual disabilities, and was restricted to just those living in private households. Both studies had high recruitment uptake. Although other studies have also reported prevalence rates, they tend to be limited by non-representative samples and/or small study sizes.[4,5]

Poor physical health is more common among people with intellectual disabilities.[6–9] People with intellectual disabilities are, therefore, at high risk of comorbid physical and mental ill health. In the general population, this combination is associated with poorer outcomes, such as premature death.[10–12] Hence an improved understanding of the prevalence of mental ill health and its relationship with general health in the population with intellectual disabilities, undertaken on a large scale, is highly indicated.

This study's aims were to (1) describe the prevalence of mental ill health in the population with intellectual disabilities compared with those without intellectual disabilities, (2) investigate the odds of intellectual disabilities on having mental ill health when adjusted for age and gender, and (3) investigate the relationship of mental and physical health in the population with intellectual disabilities.

Method

Data source

The data source was Scotland's Census 2011. This provides statistical information on the number and characteristics of Scotland's population and households at the census day, 27 March 2011.[13] The census is undertaken every 10 years. It includes people living in communal establishments (such as care homes and student halls of residence) as well as people living in private households. In 2011, Scotland's Census was estimated to have achieved a 94% response rate, which is close to the highest ever historic compliance rate. A coverage assessment and adjustment process was used to take account of non-response in the final Census outputs. Scotland's Census is probably one of few country censuses that identifies

people with intellectual disabilities and distinguishes these from specific learning disabilities such as dyslexia, and from autism; indeed it may be unique in this regard. Full details of the methodology and other background information on Scotland's Census 2011 are available at: www.scotlandscensus.gov.uk/supporting-information.

The Census requires the form to be completed by the head of household or joint head of household on behalf of all occupants in private households, and the manager is responsible on behalf of all occupants in communal dwellings. It is a legal requirement to complete the Census, and the Census form clearly states this, and that a head of household not completing it or supplying false information can be fined £1000. The Census team follow up non-responders and also provide help to respond when that is needed (there was not an easy-read version), hence the high 94% completion rate.

Census variables

Intellectual disabilities, mental health condition and other conditions

People with intellectual disabilities were identified by self-/proxy-reporting on intellectual disabilities from the individual questions section; question 20: 'Do you have any of the following conditions which have lasted, or are expected to last, at least 12 months? Tick all that apply'. There was a choice of 10 response options, which included:

(1) deafness or partial hearing loss
(2) blindness or partial sight loss
(3) learning disability (e.g. Down syndrome)
(4) learning difficulty (e.g. dyslexia)
(5) developmental disorder (e.g. autism spectrum disorder or Asperger syndrome)
(6) physical disability
(7) mental health condition
(8) long-term illness, disease or condition
(9) other condition and
(10) no condition.

For 'other condition', a prompt was included for the respondent to report the type of 'other condition'. Importantly, the question distinguishes between intellectual disabilities (for which the term 'learning disability' is used in Scotland), specific learning disabilities and autism.

General health

General health status was collected through the question, 'How is your health in general?', with a five-point response scale as follows: (1) very good, (2) good, (3) fair, (4) bad and (5) very bad.

Procedures

Following Scottish Government approval, data from Scotland's Census 2011 were analysed under the auspices of a collaborative research project with the National Records of Scotland at its premises in Ladywell House, Edinburgh. All resulting raw frequency tables of Census data were checked to ensure they did not breach statistical disclosure control thresholds and were published on the Scotland's Census website, available under the Health topic at: www.scotlandscensus.gov.uk/ods-web/data-warehouse.html#additionaltab

Data analysis

We calculated the number and percentage of people with intellectual disabilities, compared with the general population. We compared gender and age structure by Pearson's χ^2 test. We calculated the number and percentage of people with intellectual disabilities reporting a mental health condition and compared this with the general population by Pearson's χ^2 test. We then used two binary logistic regressions to calculate the odds ratios (95% confidence intervals (CIs)) of intellectual disabilities referenced to no intellectual disabilities, in statistically predicting the dependent variable 'mental health condition', adjusted: (1) for gender and age (given the different gender and age structure in the two populations), and (2) for gender, age and general health status. Age was categorised into the following groups: 0–15, 16–24, 25–34, 35–44, 45–54, 55–64, 65–74 and 75+ years. The age group 0–15 years was the reference group for age, male was the reference gender group and good/very good health was the reference group for general health status. Next, within the population with intellectual disabilities, two binary logistic regressions were undertaken to investigate: (1) the extent to which age and gender were independent predictors of having a mental health condition, and (2) the extent to which age, gender and general health status were statistically independent predictors of having a mental health condition. The enter method was used. All analyses were conducted with SPSS version 22.

Results

Participant characteristics

Scotland's Census 2011 includes records on 5 295 403 people; 26 349 (0.5%) with intellectual disabilities and 5 269 054 without intellectual disabilities. There were 15 149 (57.5%) males and 11 200 (42.5%) females with intellectual disabilities, compared with 2 567 444 (48.7%) males and 2 727 959 (51.8%) females without intellectual disabilities (χ^2=860.5; d.f.=1; P<0.001). The population with intellectual disabilities were younger than those without, with 5234 (19.9%) aged 0–15 years, 18 660 (70.8%) aged 16–64 years and 2455 (9.3%) aged 65+ years, compared with 911 097 (17.3%) aged 0–15 years, 3 470 078 (65.9%) aged 16–64 years and 887 879 (16.9%) aged 65+ years in those without intellectual disabilities (χ^2=1083.2; d.f.=2; P<0.001). The prevalence of intellectual disabilities was highest in childhood, reaching its peak by age 9 years (0.7–0.8%), being 0.5% at 25–34 years, and then progressively falling from age 55–64 years to 0.2% at age 75+ years.

The population with intellectual disabilities were significantly more likely to have physical disability (32.6% *v.* 6.6%; χ^2=28316.0; d.f.=1; P<0.001), blindness or partial sight loss (13.1% *v.* 2.3%, χ^2=13034.7; d.f.=1; P<0.001), deafness or partial hearing loss (12.4% *v.* 6.6%, χ^2=1421.6; d.f.=1; P<0.001) and autism (21.7% *v.* 0.5%, χ^2=197451.8; d.f.=1; P<0.001).

Prevalence of mental health conditions

Mental health conditions were reported more often for people with, compared with people without, intellectual disabilities: 668 (12.8%) *v.* 2653 (0.3%) aged 0–15 years (χ^2=22415.3; d.f.=1; P<0.001); 4370 (23.4%) *v.* 184 191 (5.3%) aged 16–64 years (χ^2=11907.5; d.f.=1; P<0.001); and 668 (27.2%) *v.* 40 393 (4.5%) aged 65+ years (χ^2=2857.7; d.f.=1; P<0.001). Table 1 and Fig. 1 show the prevalence of mental health conditions by age and gender. There are significant differences between people with and without intellectual disabilities at all ages and for both genders, most markedly so for children and young people.

Adjusting for age and gender, given the different distributions in the two populations, intellectual disabilities had an odds ratio of 7.1 (95% CI 6.8–7.3) in statistically predicting mental health conditions (Table 2). Good/very good health, fair health had an odds ratio of 10.4 (95% CI 10.3–10.6) and bad/very bad health had an odds ratio of 25.7 (95% CI 25.4–26.0) in statistically predicting mental health conditions, when referenced against good/very good health (Table 2).

Relationship between general health and mental health

Within the intellectual disabilities population, female gender reduced the likelihood of mental health conditions. Each progressive

Table 1 Prevalence of mental ill health in the populations with and without intellectual disabilities

Age group	Intellectual disabilities, *n*/*N* (%)			Other people, *n*/*N* (%)		
	Male *N*=15 149	Female *N*=11 200	All *N*=26 349	Male *N*=2 552 295	Female *N*=2 716 759	All *N*=5 269 054
0–15	450/3253 (13.8)	218/1981 (11.0)	668/5234 (12.8)	1739/465 853 (0.4)	914/445 244 (0.2)	2653/911 097 (0.3)
16–24	492/2501 (19.7)	281/1661 (16.9)	773/4162 (18.6)	5664/314 382 (1.8)	9113/313 944 (2.9)	14 777/628 326 (2.4)
25–34	436/2025 (21.5)	286/1450 (19.7)	722/3475 (20.8)	13 552/326 582 (4.1)	19 330/337 270 (5.7)	32 882/663 852 (4.9)
35–44	529/2209 (23.9)	398/1650 (24.1)	927/3859 (24.0)	22 004/355 461 (6.2)	27 633/375 434 (7.4)	49 637/730 895 (6.8)
45–54	653/2456 (26.6)	490/1845 (26.6)	1143/4301 (26.6)	22 723/382 061 (5.9)	29 384/400 394 (7.3)	52 107/782 455 (6.7)
55–64	442/1539 (28.7)	363/1324 (27.4)	805/2863 (28.1)	16 581/325 383 (5.1)	18 207/339 167 (5.4)	34 788/664 550 (5.2)
65–74	212/805 (26.3)	208/762 (27.3)	420/1567 (26.8)	6776/224 557 (3.0)	8106/255 668 (3.2)	14 882/480 225 (3.1)
75+	111/361 (30.7)	137/527 (25.9)	248/888 (27.9)	7741/158 016 (4.9)	17 770/249 683 (7.1)	25 511/407 654 (6.3)
All ages	3325/15 149 (21.9)	2381/11 200 (21.3)	5706/26 349 (21.7)	96 780/2 552 295 (3.8)	130 457/2 716 759 (4.8)	227 237/5 269 054 (4.3)

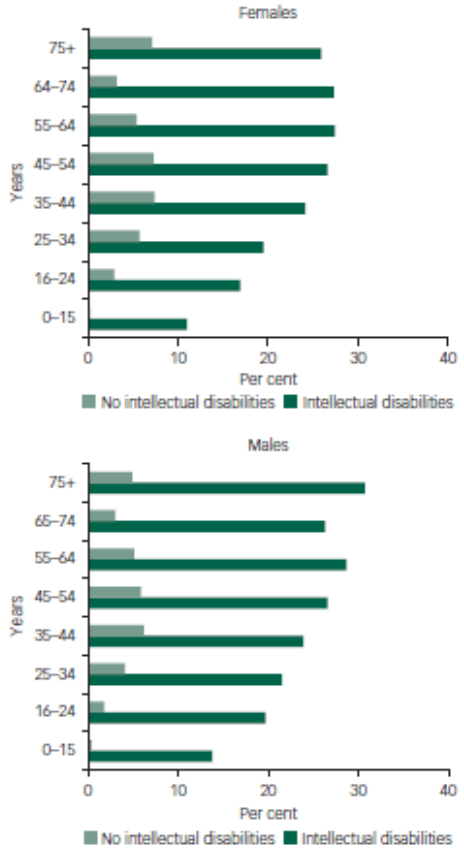

Fig. 1 Prevalence of a mental health condition by gender and age group.

increase in age group, up to 64 years, predicted mental health conditions compared with childhood; thereafter, the odds ratio plateaued (Table 3). Fair health had an odds ratio of 1.8 (95% CI 1.7–1.9) and bad/very bad health had an odds ratio of 4.2 (95% CI 3.9–4.6) in statistically predicting mental health conditions, when referenced against good/very good health (Table 3).

Discussion

Principle findings and interpretation

This large-scale study is the first of a whole-country population to investigate mental health and the relationship with general health in the population with intellectual disabilities at all ages, including children. It found a substantially higher prevalence of mental health conditions at all ages compared with the population without intellectual disabilities. These findings are important, given the previously stated lack of confidence as to whether people with intellectual disabilities have higher rates of mental health conditions, when excluding problem behaviours and autism.[3] Few previous studies have reported mental health in comparison with the general population, and those that did had either small sample sizes or reported from general population cohort studies. The general population cohort studies are limited through being focused on mild intellectual disabilities as very few people with more severe intellectual disabilities are included in existing cohorts, and typically are restricted to private households, whereas people with intellectual disabilities are more likely to live in communal establishments than are the general population.

We found mental health conditions to be related to poorer general health status. In the whole population, this was the case for fair health compared with good/very good health and was more markedly so for people with bad/very bad health. We found the same pattern in the population with intellectual disabilities, but to a lesser extent. Contrary to the general population, however, males rather than females were more likely to have mental health conditions, highlighting the importance of specifically studying the health of people with intellectual disabilities, to provide health intelligence that is relevant to them.

The 23.4% prevalence of mental health conditions we report for adults with intellectual disabilities is very similar to the 22.4% (excluding problem behaviours and autism) reported in the previously largest study of 1023 adults who all had individual detailed assessments.[1] The Census prevalence of mental health conditions also excludes autism, as autism was separately enquired about, but it did not specifically enquire about problem behaviours, so it is unclear whether or not respondents included problem behaviours within their understanding of mental health conditions. Regarding children, the previously reported rate of 36% in 641 surveyed children with intellectual disabilities did include problem behaviours.[2] This is higher than the 12.8% with mental health conditions in Scotland's Census data, which may well be accounted for by problem behaviours; the substantial difference compared with the general population is apparent in both these data-sets.

Table 2 Independent predictors of mental ill health in the whole population

	Regression 1		Regression 2	
Characteristic	Odds ratio	95% CI	Odds ratio	95% CI
Ability				
No intellectual disabilities (reference)	–	–	–	–
Intellectual disabilities	7.058	6.843–7.280	3.187	3.077–3.300
Gender				
Male (reference)	–	–	–	
Female	1.238	1.227–1.248	1.243	1.232–1.255
Age				
0–15 (reference)	–	–	–	
16–24	6.908	6.652–7.172	6.175	5.943–6.415
25–34	14.655	14.139–15.189	11.292	10.888–11.710
35–44	20.437	19.729–21.171	11.987	11.564–12.426
45–54	20.063	19.369–20.782	8.451	8.152–8.761
55–64	15.628	15.079–16.197	4.484	4.322–4.651
65–74	9.083	8.747–9.433	2.016	1.939–2.096
75+	18.456	17.796–19.139	2.979	2.869–3.093
General health status				
Very good/good (reference)			–	
Fair			10.445	10.328–10.563
Bad/very bad			25.702	25.387–26.021
Constant	0.003	–	0.002	

Table 3 Independent predictors of mental ill health in the population with intellectual disabilities

	Regression 1		Regression 2	
Characteristic	Odds ratio	95% CI	Odds ratio	95% CI
Gender				
Male (reference)	–	–	–	
Female	0.917	0.864–0.974	0.893	0.839–0.950
Age				
0–15 (reference)	–	–	–	
16–24	1.562	1.396–1.748	1.660	1.479–1.863
25–34	1.799	1.603–2.019	1.925	1.710–2.166
35–44	2.171	1.945–2.423	2.253	2.014–2.522
45–54	2.485	2.236–2.763	2.485	2.230–2.770
55–64	2.694	2.401–3.023	2.556	2.271–2.876
65–74	2.572	2.220–2.902	2.389	2.072–2.753
75+	2.699	2.291–3.193	2.450	2.061–2.913
General health status				
Very good/good (reference)			–	
Fair			1.783	1.663–1.912
Bad/very bad			4.210	3.887–4.559
Constant	0.151	–	0.092	

Within the population with intellectual disabilities, increasing age was associated with increasing odds of mental health conditions, until age 64 years when the odds plateaued, giving some credence to a healthy survivor effect in this population.[14] As the older adults start to acquire mental health conditions related to ageing (dementia), one would expect their extent of mental health conditions to increase further. This is not observed in the data, although prevalence rates are still substantially greater than in the general population and then in the population of children with intellectual disabilities. This may be because of the premature death of people with severe/profound intellectual disabilities who have higher levels of mental health conditions, relative to people with mild intellectual disabilities.

The population with intellectual disabilities also had higher rates of physical disability, blindness or partial sight loss, deafness or partial hearing loss and autism. Despite the considerably greater burden of both mental health and physical health that we have reported in the population with intellectual disabilities, it is notable the extent to which their needs have been neglected by researchers, as shown by the limited evidence base. Clearly, more research is needed to better understand the health and healthcare needs of people with intellectual disabilities.

Strengths and limitations

This Scottish total country study is the largest and most complete population study we have identified investigating reported mental health of people with intellectual disabilities compared with people in the general population across the lifespan, and reports on general health associations with mental ill health. The advantage of Scotland's Census data is that it covers communal establishments as well as private households, and specifically, systematically, asked about the presence of intellectual disabilities for each person, distinguishing this from specific learning disabilities and autism; lack of these features has previously been highlighted as a limitation of previous research on people with intellectual disabilities conducted with large data-sets.[15] Additionally, it provides a whole-country coverage, with a 94% response. It may well be unique in having all these features and, therefore, is the strength of the study.

The ascertained prevalence of intellectual disabilities by Scotland's Census is similar for adults (0.5%) to that reported in a recent meta-analysis of population-based studies of prevalence (4.94/1000),[16] giving further credence to the ascertainment in the Census. However, we recognise that the country of study, sample population, age range, definition of intellectual disabilities and method of ascertainment can all impact on identified rate.[17] Cohort effects result in prevalence varying with time, such that past and more recent studies of the same age ranges may well provide different rates.[17] These factors become lost within a meta-analysis. For children and young people we found the prevalence in Scotland's Census varied by age, in view of the time taken for identification in childhood, making comparisons with published studies difficult. Regarding proxy-reporting for presence of intellectual disabilities, limitations have been reported with young children (e.g. age 3),[15] and this is apparent in Scotland's Census data as ascertainment increases up to age 9. Adults would, of course, have been diagnosed in childhood.

We have no reason to believe the study results are not generalisable to other high-income countries.

A limitation is the lack of information on whether the responses were completed by proxies or the person with intellectual disabilities. Given the style and questions on the Census, we consider it very unlikely that people with intellectual disabilities would have been able to complete the form without help. In private households, the head of household completing the Census was most likely to be a parent of the people with intellectual disabilities, in supported living a support worker, and in communal establishments the manager. The data are, therefore, likely to be proxy-report rather than self-report for the great majority of people with intellectual disabilities. We recognise that proxy ratings could differ from self-ratings with regard to health ratings,[15] but without them would have no information on people unable to self-report because of severe/profound intellectual disabilities. We note that proxy-reporting is the basis for much of the healthcare provided for people with intellectual disabilities who cannot self-report.

A further limitation is the cross-sectional design of the study. Hence, we are merely reporting associations, and causality cannot be assumed. Indeed, in the general population there is literature to suggest that mental ill health can lead to poor general ill health, and also the reverse, with poor general health leading to mental ill health.[11,12] A longitudinal study would be needed to untangle the preceding and precipitating factors.

Implications

People with intellectual disabilities of all ages have substantially higher rates of mental health conditions than the rest of the population, and their mental health conditions are often associated with poor general health. Their health patterns differ from those seen in the general population.[9] These conditions are often not identified or are misattributed to the persons' learning disabilities (diagnostic overshadowing). Additionally, people with intellectual disabilities are known to experience difficulties accessing mainstream services.[6,7] Globally, no country has comprehensive coverage of intellectual disability psychiatric services except the UK, and in the UK, there is increasing 'mainstreaming' of psychiatric care. Knowledge on intellectual disabilities and the poor general health associated with mental health is, therefore, essential for general psychiatrists and mainstream psychiatric services, as well as for carers. Without awareness of the substantial burden of mental health conditions and its co-existence with poor general health, the potential for misdiagnosis is high, and inadequate or inappropriate treatment is a risk. Health burden is also important to know for resource allocation, service planning, and to support individuals and their carers.

Clearly, given the associations we report, more research is needed to determine relationships between specific mental health problems, physical conditions and associated factors, for example, adversity, among people with intellectual disabilities, to help influence the development of appropriate interventions, and health and social care policy.

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Paper III

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General health of adults with autism spectrum disorders – A whole country population cross-sectional study

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ABSTRACT

Background: General health status in adult populations with autism spectrum disorders has been little studied. We aimed to investigate general health status and predictors of poor health in adults with autism spectrum disorders compared with other adults.

Method: Whole country data were drawn from Scotland's Census, 2011. We calculated and compared the frequencies of health status in adults with and without autism spectrum disorders. We then used logistic regressions to calculate odds ratios (OR) with 95% confidence intervals (95% CI) of autism predicting poor general health in the whole population, adjusted for age and gender, and OR (95% CI) of age and gender predicting poor general health within the autism spectrum disorders population.

Results: Autism spectrum disorders were reported for 6649/3,746,584 (0.2%) adults aged 25+years, of whom 46.8% (N = 3111) had poor general health, compared with 23.7% (N = 887,878) of other people. Poor health was common across the entire lifecourse for adults with autism spectrum disorders. Autism had OR = 5.1 (4.9–5.4, 95% CI) for predicting poor general health, or OR = 7.5 (6.9–8.2, 95% CI) when the interaction with age was included. Poorer health was more common at older age, and for women.

Conclusions: Poor general health merits attention across the full lifecourse for adults with autism. Health practitioners need to be alert to the burden of potential health problems to seek them out to be addressed, and so the health agenda can turn towards potential mechanisms for prevention and better support for adults who may call upon services for people with autism.

1. Introduction

Autism spectrum disorders are common, yet most health evidence relates to children and young people rather than adults. This is a serious gap in the evidence-base, as autism spectrum disorders are lifelong, and health status is an integral component of quality of life. Additionally, there is some evidence to suggest that the health needs of people with autism spectrum disorders are sometimes

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overlooked, therefore, resulting in low level of satisfaction with healthcare services (Nicolaidis et al., 2012) further exacerbated by barriers in accessing medical care (Saqr, Braun, Porter, Barnette, & Hanks, 2017). This potentially puts adults with autism spectrum disorders at risk of not receiving effective treatments, or receiving potentially harmful treatments that are not required. People with autism spectrum disorders and health practitioners, therefore, need to be aware of the extent of health needs/health status, to ensure quality of life through availability and accessibility of health preventions, management, and support that is appropriately tailored to meet the needs of this population.

Adults with autism spectrum disorders appear to experience a wide range of additional health conditions, although their mental health has been more commonly studied than physical health. In view of this, it might be expected that they have poorer general health than other people. For example, systematic reviews suggest that depression (Stewart, Barnard, Pearson, Hasan, & O'Brien, 2006; Wigham, Barton, Parr, & Rodgers, 2017), bipolar disorder (Skokauskas & Frodl, 2015; Vannucchi et al., 2014), schizophrenia (Kincaid, Doris, Shannon, & Mulholland, 2017; Lugo Marín et al., 2018), suicidal thoughts/behaviour (Segers & Rawana, 2014; Zahid & Upthegrove, 2017), and non-affective psychosis (Padgett, Miltsiou, & Tiffin, 2010) may be more common in adults with autism spectrum disorders than other people. There are fewer systematic reviews on physical health, but sleep problems (Van de Wouw, Evenhuis, & Echteld, 2012), and atopy (Billeci et al., 2015; Su et al., 2016) may be more common in people with autism, and epilepsy is more common, especially in the presence of additional intellectual disabilities (Amiet et al., 2008). In a study of 378 adults with autism spectrum disorders, 88% who self-reported, and 74% who had carer reporting, described two or more physical health conditions, with the most common being sleep problems, allergies, and gastrointestinal conditions, and 57% and 45% respectively reported two or more mental health conditions (Gotham et al., 2015). In another study of 92 adults with autism spectrum disorders most commonly reported medical conditions were seizures, obesity, insomnia, and constipation, and the median number of medical conditions per person was 11 (Jones et al., 2016). A further study of 1507 adults with a record of autism spectrum disorders in their medical records found that 54% had a record of mental health conditions, and 14% had autoimmune conditions, 36% allergy, 35% gastrointestinal disorders, 18% sleep disorders, 12% epilepsy, 26% hypertension, 8% diabetes, 34% obesity, and 7% thyroid disease (Croen et al., 2015). In addition, in a study of 6649 adults with autism spectrum disorders, 14% had deafness or partial hearing loss, 12% had blindness or partial sight loss, 33% had mental health conditions, and 24% had physical disability (Rydzewska et al., 2018).

However, despite these reports on specific conditions, we have identified only two published studies investigating general health status in adults with autism spectrum disorders. A small Taiwanese study of 30 men and 11 women with autism without intellectual disabilities aged 20–37 years, compared their health ratings with a non-autism age, gender matched control group, using a 5-point Likert scale. Five (12.2%) adults with autism rated their health as extremely bad, 6 (14.6%) as bad, 16 (39.0%) as not bad/not good, 7 (17.1%) as good and 7 (17.1%) as extremely good. This compared with ratings amongst the control group of none scoring extremely bad (0%), 3 (7.3%) bad, 13 (31.7%) not bad/not good, 19 (46.3%) good and 6 (14.6%) extremely good (Lin, 2014). The group with autism, therefore, rated their general health as poorer, but the study was small scale, and its recruitment methods (via a hospital clinic and autism groups) may have resulted in a non-representative sample, and explains its inclusion of only young adults. A longitudinal study in USA of 180/406 adults with autism aged 23–60 years, 52% of whom also had intellectual disabilities, analysed data collected from mothers in 2011 to 2012. Physical health was recorded as poor or good/very good; 144 (80.0%) reported good/very good physical health, but there was no general population comparison data, limiting the conclusions that can be drawn (Bishop-Fitzpatrick et al., 2016). We are not aware of any other studies which have investigated the general health status of adults with autism spectrum disorders, nor compared this with the general population.

This study aimed to investigate the general health status of adults with autism spectrum disorders compared with other adults, the odds ratio (OR) with 95% confidence interval (95% CI) of autism predicting poor general health within the whole population, and the OR of age and gender predicting poor general health within the population with autism spectrum disorders.

2. Methods

2.1. Participants

Participants were drawn from Scotland's Census, 2011. People with autism spectrum disorders were identified from the question on the Census which asked: 'Do you have any of the following conditions which have lasted, or are expected to last, at least 12 months? Tick all that apply'. There was a choice of 10 response options, which included the following three: developmental disorder (for example, autistic spectrum disorder or Asperger's syndrome); learning disability (for example, Down's syndrome); learning difficulty (for example, dyslexia). Importantly, the question on developmental disorder only prompted respondents to reply with regards to autistic spectrum disorder or Asperger's syndrome. We, therefore, interpret responses to this question as relating to people who know they have autism spectrum disorders. Additionally, the question distinguished autism spectrum disorders from learning disability, which in the UK is synonymous with the international term 'intellectual disabilities', and learning difficulties such as dyslexia.

As part of the methodological preparations for Scotland's Census, 2011, cognitive question testing was undertaken on the question on these conditions, to investigate whether it was answered accurately and willingly, and what changes might be required to improve data quality. Retrospective probing was undertaken with 102 participants with a mix of gender and age, both with and without the conditions or with more than one of the conditions, and including people with autism spectrum disorders, intellectual disabilities, dyslexia, dyspraxia, speech impairment, mental health conditions (both milder and more serious), and other long-term conditions. This resulted in a redesign of the question on autism spectrum disorders to the version used. The other questions did not require any modification.

2.2. Measures & design

Scotland's Census, 2011 provides information on the number and characteristics of Scotland's population and households on the census date, 27 March 2011. The census is undertaken every 10 years. It includes the whole Scottish population: people living in communal establishments (such as care homes and student halls of residence) as well as people living in private households. The 2011 Census required the questions to be completed by the head of household or joint head of household on behalf of all occupants in private households, and the manager on behalf of all occupants in communal dwellings. It is a legal requirement to complete the Census, and failure to complete, or supplying false information can attract a fine of £1000. The 2011 Census team followed up non-responders, and also provided help to respond when that was needed, hence the high completion rate, estimated at 94% (National Records of Scotland (NRS, 2013). Scotland's Census, 2011 is probably one of the few country censuses which identifies people with autism spectrum disorders, indeed it may be unique in this regard.

Approval was gained from the Scottish Government for secondary analysis of the Census data. Full details of the methodology and other background information on Scotland's Census 2011 are available at:

http://www.scotlandscensus.gov.uk/supporting-information.

Information on general health status of adults with and without autism spectrum disorders was collected through the question: 'How is your health in general?', which invited responses on a 5-point Likert scale: (1) very good, (2) good, (3) fair, (4) bad, (5) very bad.

2.3. Procedures and analysis

We calculated the number and percentage of adults aged 25+ years reported to have autism spectrum disorders, by age and gender. We also calculated the number and percentage of adults with and without autism spectrum disorders reporting very good, good, fair, bad, and very bad health, and compared differences using chi-square tests. Within the whole population of adults in Scotland, we then used a binary logistic regression to calculate odds ratios (OR) with 95% confidence intervals (95% CI) of autism predicting a derived, dichotomised variable of poor health (fair, bad, or very bad health) versus good health (very good or good health), adjusted for age and gender. Age was categorised into groups of 25–34, 35–44, 45–54, 55–64 and 65+, with the 25–34-year-old age group being the reference group. Gender was bivariate, with men being the reference group. Within the population with autism spectrum disorders, we then calculated the OR (95% CI) for age and gender predicting poor health. All analyses were conducted with SPSS software version 22.

3. Results

3.1. Number of adults with autism by age and gender

Scotland's Census, 2011 includes records on 3,746,584 people aged 25 years and over, of whom 6649 (0.2%) were reported to have autism spectrum disorders. 4610 (69.3%) of the 6649 adults with autism spectrum disorders were men, and 2039 (30.7%) were women. 1953 (29.4%) of adults with autism spectrum disorders were also recorded to have intellectual disabilities. Table 1 provides the age and gender characteristics of adults with and without autism spectrum disorders. The proportion of the population with autism spectrum disorders was lower at older age groups over 55 years.

3.2. General health

3111 (46.8%) of adults with autism spectrum disorders had poor (fair, bad, or very bad) general health, compared with 887,878 (23.7%) of other adults ($\chi^2 = 1945.2$, df = 1, $p < 0.001$). Table 2 and Figs. 1–3 show general health status of adults with and without autism spectrum disorders in Scotland, by age group and gender. This shows adults at all ages with autism spectrum disorders being more likely to report poor general health compared to other adults; at age 25–34 years, $\chi^2 = 3084.1$, df = 1, $p < 0.001$; at age 35–44 years, $\chi^2 = 1554.4$, df = 1, $p < 0.001$; at age 45–54 years, $\chi^2 = 723.1$, df = 1, $p < 0.001$; at age 55–64 years, $\chi^2 = 273.3$, df = 1, $p < 0.001$; at age 65+ years, $\chi^2 = 118.8$, df = 1, $p < 0.001$. The proportion of people with autism spectrum disorders

Table 1
Prevalence of autism spectrum disorders in adults aged 25+ years.

Age group	All adults			Adults with autism		
	Total N = 3,746,584	F N = 1,965,129	M N = 1,781,455	Total N = 6649	F N = 2039	M N = 4610
25-34y	667,327	338,720	328,607	2389 (0.36%)	636 (0.19%)	1753 (0.53%)
35-44y	734,754	377,084	357,670	1588 (0.22%)	471 (0.12%)	1117 (0.31%)
45-54y	786,756	402,239	384,517	1267 (0.16%)	377 (0.09%)	890 (0.23%)
55-64y	667,413	340,491	326,922	707 (0.11%)	233 (0.07%)	474 (0.14%)
65+y	890,334	506,595	383,739	698 (0.08%)	322 (0.06%)	376 (0.10%)

Table 2
General health status of people with and without autism spectrum disorders.

All adults aged 25 +						
N = 3,746,584						
General health	Autism			Without autism		
	Total 6649 (100%)	F 2039 (100%)	M 4610 (100%)	Total 3,739,935 (100%)	F 1,963,090 (100%)	M 1,776,845 (100%)
Very good	1,327 (20.0%)	349 (17.1%)	978 (21.2%)	1,548,794 (41.4%)	799,671 (40.7%)	749,123 (42.2%)
Good	2,211 (33.3%)	634 (31.1%)	1577 (34.2%)	1,303,263 (34.8%)	678,544 (34.6%)	624,719 (35.2%)
Fair	1,909 (28.7%)	625 (30.7%)	1284 (27.9%)	601,767 (16.1%)	330,755 (16.8%)	271,012 (15.3%)
Bad	783 (11.8%)	276 (13.5%)	507 (11.0%)	217,741 (5.8%)	117,531 (6.0%)	100,210 (5.6%)
Very bad	419 (6.3%)	155 (7.6%)	264 (5.7%)	68,370 (1.8%)	36,589 (1.9%)	31,781 (1.8%)
25-34 y						
N = 667,327						
General health	Autism			Without autism		
	Total 2389 (100%)	F 636 (100%)	M 1753 (100%)	Total 664,938 (100%)	F 338,084 (100%)	M 326,854 (100%)
Very good	593 (24.8%)	133 (20.9%)	460 (26.2%)	421,660 (63.4%)	210,543 (62.3%)	211,117 (64.6%)
Good	891 (37.3%)	224 (35.2%)	667 (38.0%)	192,928 (29.0%)	100,786 (29.8%)	92,142 (28.2%)
Fair	602 (25.2%)	176 (27.7%)	426 (24.3%)	37,707 (5.7%)	20,113 (5.9%)	17,594 (5.4%)
Bad	192 (8.0%)	64 (10.1%)	128 (7.3%)	10,071 (1.5%)	5296 (1.6%)	4775 (1.5%)
Very bad	111 (4.6%)	39 (6.1%)	72 (4.1%)	2572 (0.4%)	1346 (0.4%)	1226 (0.4%)
35-44 y						
N = 734,754						
General health	Autism			Without autism		
	Total 1588 (100%)	F 471 (100%)	M 1117 (100%)	Total 733,166 (100%)	F 376,613 (100%)	M 356,553 (100%)
Very good	334 (21.0%)	85 (18.0%)	249 (22.3%)	400,806 (54.7%)	206,222 (54.8%)	194,584 (54.6%)
Good	526 (33.1%)	158 (33.5%)	368 (32.9%)	238,948 (32.6%)	121,246 (32.2%)	117,702 (33.0%)
Fair	451 (28.4%)	133 (28.2%)	318 (28.5%)	63,017 (8.6%)	33,142 (8.8%)	29,875 (8.4%)
Bad	192 (12.1%)	69 (14.6%)	123 (11.0%)	23,654 (3.2%)	12,406 (3.3%)	11,248 (3.2%)
Very bad	85 (5.4%)	26 (5.5%)	59 (5.3%)	6741 (0.9%)	3597 (1.0%)	3144 (0.9%)
45-54 y						
N = 786,756						
General health	Autism			Without autism		
	Total 1267 (100%)	F 377 (100%)	M 890 (100%)	Total 785,489 (100%)	F 401,862 (100%)	M 383,627 (100%)
Very good	230 (18.2%)	67 (17.8%)	163 (18.3%)	351,549 (44.8%)	181,055 (45.1%)	170,494 (44.4%)
Good	419 (33.1%)	119 (31.6%)	300 (33.7%)	284,266 (36.2%)	141,915 (35.3%)	142,351 (37.1%)
Fair	376 (29.7%)	118 (31.3%)	258 (29.0%)	96,095 (12.2%)	49,986 (12.4%)	46,109 (12.0%)
Bad	164 (12.9%)	45 (11.9%)	119 (13.4%)	40,591 (5.2%)	22,004 (5.5%)	18,587 (4.8%)
Very bad	78 (6.2%)	28 (7.4%)	50 (5.6%)	12,988 (1.7%)	6902 (1.7%)	6086 (1.6%)
55-64 y						
N = 667,413						
General health	Autism			Without autism		
	Total 707 (100%)	F 233 (100%)	M 474 (100%)	Total 666,706 (100%)	F 340,258 (100%)	M 326,448 (100%)
Very good	102 (14.4%)	24 (10.3%)	78 (16.5%)	221,444 (33.2%)	116,828 (34.3%)	104,616 (32.0%)
Good	207 (29.3%)	66 (28.3%)	141 (29.7%)	256,769 (38.5%)	129,784 (38.1%)	126,985 (38.9%)

(continued on next page)

Table 2 (continued)

55-64 y						
N = 667,413						
General health	Autism			Without autism		
	Total 707 (100%)	F 233 (100%)	M 474 (100%)	Total 666,706 (100%)	F 340,258 (100%)	M 326,448 (100%)
Fair	224 (31.7%)	82 (35.2%)	142 (30.0%)	120,750 (18.1%)	60,958 (17.9%)	59,792 (18.3%)
Bad	112 (15.8%)	33 (14.2%)	79 (16.7%)	51,151 (7.7%)	24,781 (7.3%)	26,370 (8.1%)
Very bad	62 (8.8%)	28 (12.0%)	34 (7.2%)	16,592 (2.5%)	7907 (2.3%)	8685 (2.7%)
65 + y						
N = 890,334						
General health	Autism			Without autism		
	Total 698 (100%)	F 322 (100%)	M 376 (100%)	Total 889,636 (100%)	F 506,273 (100%)	M 383,363 (100%)
Very good	68 (9.7%)	40 (12.4%)	28 (7.4%)	153,335 (17.2%)	85,023 (16.8%)	68,312 (17.8%)
Good	168 (24.1%)	67 (20.8%)	101 (26.9%)	330,352 (37.1%)	184,813 (36.5%)	145,539 (38.0%)
Fair	256 (36.7%)	116 (36.0%)	140 (37.2%)	284,198 (31.9%)	166,556 (32.9%)	117,642 (30.7%)
Bad	123 (17.6%)	65 (20.2%)	58 (15.4%)	92,274 (10.4%)	53,044 (10.5%)	39,230 (10.2%)
Very bad	83 (11.9%)	34 (10.6%)	49 (13.0%)	29,477 (3.3%)	16,837 (3.3%)	12,640 (3.3%)

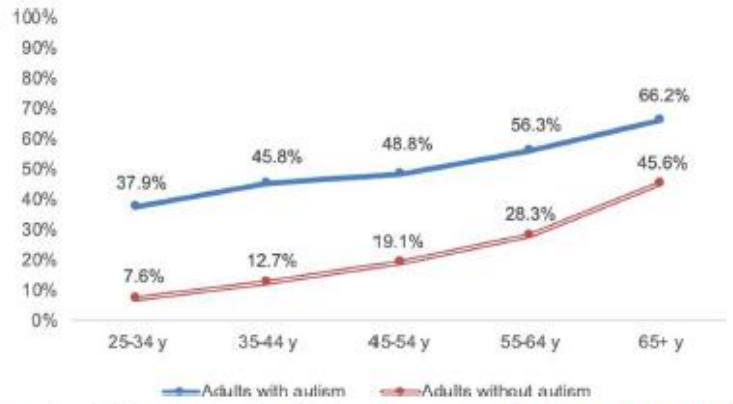

Fig. 1. Proportion of adults aged 25+ with and without autism spectrum disorders reporting poor health by age.

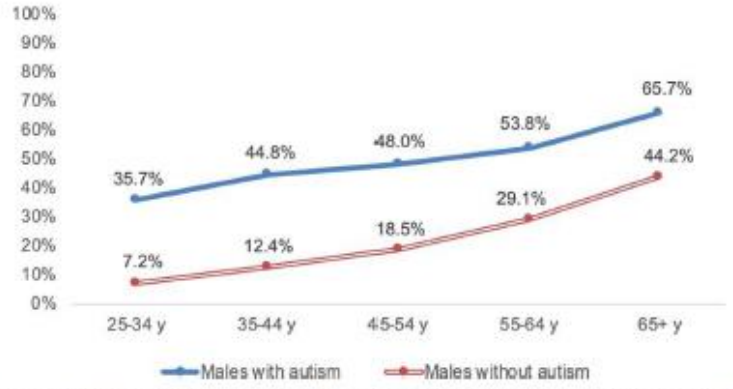

Fig. 2. Proportion of males aged 25+ with and without autism spectrum disorders reporting poor health by age.

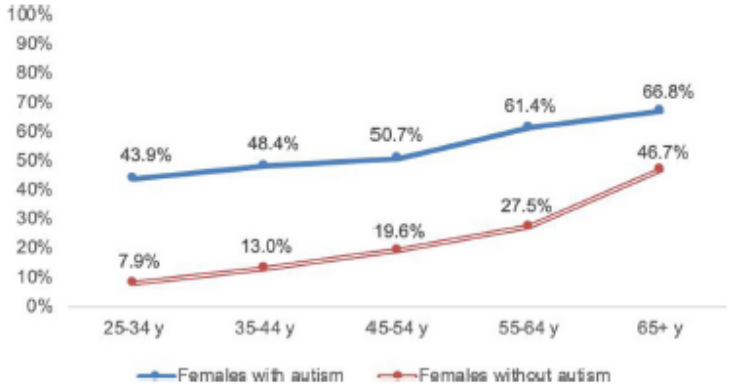

Fig. 3. Proportion of females aged 25+ with and without autism spectrum disorders reporting poor health by age.

rating their general health as poor increased with age, from 37.9% at 25–34 years, to 66.2% at 65+ years, compared respectively with 7.6% to 45.6% in the population without autism. Younger females with autism spectrum disorders aged 25–34 were more likely to have poor general health than males with autism, but at other age groups the difference was not statistically significant: at age 25–34 years, $\chi^2 = 13.2$, df = 1, $p < 0.001$; at age 35–44 years, $\chi^2 = 1.8$, df = 1, $p > 0.1$ ($p = 0.183$); at age 45–54 years, $\chi^2 = 0.8$, df = 1, $p > 0.1$ ($p = 0.382$); at age 55–64 years, $\chi^2 = 3.6$, df = 1, $p > 0.05$ ($p = 0.056$); at age 65+ years, $\chi^2 = 0.1$, df = 1, $p > 0.1$ ($p = 0.764$).

Given the different age and gender distributions in the populations with and without autism spectrum disorders, we adjusted for both these variables, and found that autism had an odds ratio of 5.1 (4.9–5.4, 95% CI) in predicting poor health (Table 3). Within the whole population, a gradient is seen across age categories, with older adults being more likely to have poor health, as were women. When the interaction term is added (age × autism), it can be seen that autism is shown to have more marked association with the outcome of poor health (OR = 7.5; 6.9–8.2, 95% CI) and more so at older age groups. Females had poorer health than males. Within the population with autism spectrum disorders, the influence of age was less marked for older age, whereas it was more marked for women (Table 4).

4. Discussion and implications

Adults with autism spectrum disorders have substantially poorer general health than other adults, across the entire adult life-course. Indeed, we have quantified poor health to be more than five times more likely in adults with autism spectrum disorders compared with other people. The influence of autism spectrum disorders on poor health is even greater than that of older age in the whole population, as including the interaction term of age x autism results in an ORs for autism spectrum disorders of 7.5. Women had poorer health status than men, particularly so women with autism spectrum disorders. These findings are important, as health practitioners need to be alert to the burden of potential health problems so that they are sought out to be addressed, and so that the health agenda can also start to turn towards potential mechanisms for prevention and better support.

Table 3
Independent predictors of poor health in the whole population.

		Regression 1		Regression 2 (including the interaction term: age x autism)	
Variable		Odds ratio	95% confidence interval	Odds ratio	95% confidence interval
Autism	No autism (reference)	–		–	
	Autism spectrum disorders	5.149	4.891-5.420	7.537	6.935-8.190
Age group	25-34 (reference)	–		–	
	35-44	1.775	1.755-1.796	1.782	1.762-1.802
	45-54	2.859	2.829-2.890	2.873	2.843-2.904
	55-64	4.788	4.738-4.839	4.811	4.761-4.862
	65+	10.167	10.067-10.269	10.216	10.114-10.318
Gender	Male (reference)	–		–	
	Female	1.051	1.046-1.057	1.052	1.046-1.057
Age x autism	25-34 (reference)	–		–	
	35-44	–		.778	.684-.885
	45-54	–		.543	.473-.623
	55-64	–		.438	.369-.519
	65+	–		.311	.261-.372
Constant		.080		.080	

Table 4
Independent predictors of poor health within the population with autism spectrum disorders.

Variable		Odds ratio	95% confidence interval
Age group	25-34 (reference)	–	
	35-44	1.380	1.213-1.570
	45-54	1.553	1.353-1.782
	55-64	2.088	1.761-2.475
	65 +	3.089	2.585-3.693
Gender	Male (reference)	–	
	Female	1.235	1.110-1.374
Constant		.576	

There has been little previous focus on general health of adults with autism spectrum disorders despite adulthood comprising the majority of a person's lifespan. We identified only two published reports on this topic, only one of which drew comparisons with the general population but was conducted on a small scale, with a sample that may not have been representative (the authors acknowledge its bias), as it excluded those with additional intellectual disabilities, and included only a young adult age group (Lin, 2014). Results are not, therefore, directly comparable with our study findings. However, as outlined in the introduction, there is a broad range of health conditions that appear to be common in people with autism spectrum disorders and that are likely to contribute to their poor general health status. For example, a health insurance data study found that 1507 autistic adults compared with 15,070 non-autistic adults had higher rates of almost all physical and mental health conditions investigated in the study (Croen et al., 2015). Health conditions comorbid with autism are, therefore, clearly a topic that requires further study, and raised awareness amongst health practitioners and carers.

Existing research shows that adults with autism spectrum disorders are more likely to report unmet medical needs and dissatisfaction with their care than the general population (Nicolaidis et al., 2012) as well as barriers in accessing medical care (Saqr et al., 2017) though other studies showed levels of satisfaction with healthcare comparable to the general population (Gerber et al., 2017). Adults with autism spectrum disorders are also likely to be hospitalised for life-threatening conditions and undergo major surgeries (Jones et al., 2016), indicating a significant medical comorbidity burden. They also experience access barriers to healthcare and primary care treatment (Lunsky, Klein-Geltink, & Yates, 2013). Additionally, medical providers report lack of confidence in caring for their adult patients with autism spectrum disorders (Erickson-Warfield, Crossman, Delahaye, Der Weerd, & Kuhlthau, 2015; Unigwe et al., 2017; Zerbo, Massolo, Qian, & Croen, 2015), but little has been published on how to improve healthcare access and delivery for adults with autism. As the adolescent and adult populations with autism spectrum disorders continue to grow, it becomes increasingly important that we seek better understanding of their medical needs.

4.1. Strengths and limitations

This study is both novel, and has considerable methodological strengths. It included the whole adult population of Scotland, and had a high completion rate (94%) as well as being large in scale. General health status was systematically enquired about for everyone, as was the presence or absence of autism spectrum disorders. Whilst the term 'developmental disorders' was used, the Census prompted responses only for autistic spectrum disorder or Asperger's syndrome, and carefully worded the question to capture autism spectrum disorders following cognitive testing of the question. Those adults, for whom a diagnosis of autism spectrum disorder was recorded, will reflect the contemporary diagnostic practices in place during their childhood; the concept of the autism spectrum has broadened in recent years. We do not know if this accounts for the lower prevalence of autism spectrum disorders recorded in the adults over the age of 55 years, or if this has been impacted upon by suicides or other causes of premature deaths, as the study is cross-sectional in design. The broadening of the autism spectrum concept in diagnostic practice reflects that seen in other high-income countries; hence we believe the results of this study are generalisable to other high-income countries. Census responses were completed by heads of households; hence some will have been self-reports and some proxy-reports. These reflect subjective reports of general health, rather than objective measurements of health.

4.2. Implications

This study fills a significant gap in existing research on general health, an important component of quality of life, of adults with autism spectrum disorders, and has important implications. It is essential to have accurate information on health status in order to raise awareness, and accurately plan appropriate prevention and intervention measures, and provision of resources for people who may put demand upon services designed for people with autism spectrum disorders. The poor general health ratings observed in the population of adults with autism spectrum disorders demonstrate a clear need to focus on improvements in healthcare and supports, and on the wider determinants of health in this population which may well differ from the general population.

Conflicts of interest

None declared.

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Paper IV

Open access Research

BMJ Open

Age at identification, prevalence and general health of children with autism: observational study of a whole country population

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ABSTRACT

Objectives Reported childhood prevalence of autism varies considerably between studies and over time, and general health status has been little investigated. We aimed to investigate contemporary prevalence of reported autism by age, and general health status of children/young people with and without autism.

Design Secondary analysis of Scotland's Census, 2011 data. Cross-sectional study.

Setting General population of Scotland.

Participants All children (n=916 331) and young people (n=632 488) in Scotland.

Main outcome measures Number (%) of children/young people reported to have autism and their general health status; prevalence of autism; prevalence of poor health (fair, bad and very bad health); odds ratios (95% confidence intervals) of autism predicting poor health, adjusted for age and gender and OR for age and gender in predicting poor health within the population with reported autism.

Results Autism was reported for 17 348/916 331 (1.9%) children aged 0–15, and 7715/632 488 (1.2%) young people aged 16–24. The rate increased to age 11 in boys and age 10 in girls, reflecting age at diagnosis. Prevalence was 2.8% at age 10 (4.4% for boys; 1.1% for girls), and 2.9% at age 11 (4.5% for boys; 1.1% for girls). 22.0% of children and 25.5% of young people with autism reported poor health, compared with 2.0% and 4.4% without autism. Autism had OR=11.3 (11.0 to 11.7) in predicting poor health. Autistic females had poorer health than autistic males, OR=1.6 (1.5 to 1.8).

Conclusion Accurate information on the proportion of autistic children and their health status is essential plan appropriate prevention and intervention measures and provide resources for those who may put demand on services designed for autistic people.

Strengths and limitations of this study

- Large, whole country population study.
- High response rate of 94%, and a systematic enquiry of everyone regarding autism and their general health status.
- Results are generalisable to other child and young people populations in high-income countries.
- Autism and general health status were self/proxy reported by respondents rather than each person having a clinical assessment.
- Six per cent of records were imputed.

INTRODUCTION

Reports on the prevalence of autism inevitably depend on the criteria used. The concept of autism spectrum disorders has now broadened considerably beyond original descriptions,[1 2] and clinicians also now base their diagnosis on fewer symptoms than a decade ago.[3] Additionally, there is now increased awareness about autism; hence the reported prevalence of autism has increased. Several systematic reviews have attempted to synthesise research studies on prevalence, with overall prevalence varying, dependent on the studies included, for example, their age-ranges, years the studies were conducted in (and hence criteria), data-collection methods, size and representativeness of included studies. Even when restricted to studies published since 2000, studies selected for inclusion in the reviews have shown wide ranges in reported prevalence.[4–7] Recent reviews are summarised in table 1.

The included age range in studies is likely to be critical in these reported rates, related to the age at which children are diagnosed. This, however, seems to be little investigated. A California, USA, study demonstrated that as well as rates of diagnosis of autism increasing, this was particularly so among preschool children,[8] while a large Swedish study found that the number of autism symptoms in children diagnosed with autism had fallen in children diagnosed at age 7–12 years, but not at age 1–6 years.[3] In the National Survey of Children's Health, USA, 259 (24.6%) of children with autism were diagnosed at <3 years of age,

Table 1 Examples of findings from systematic reviews of recent studies on childhood/youth prevalence of autism

Review		No. of studies	Publication dates of studies	Median prevalence/1000	Range/1000
Autistic disorder					
French *et al*., 2013[4]	Autistic disorder	26	2000–2011	2.2	0.8–9.4
	Asperger syndrome*	13	1998–2011	2.1	0.5–2.8
Elsabbagh *et al*., 2012[5]	Northern European	16	2000–2008	1.9	0.7–3.9
	Western Pacific	12	2000–2011	1.2	0.3–9.4
	South East Asia/East Mediterranean	1	-	-	-
	Americas	7	2001–2010	2.2	1.1–4.0
	Overall			1.7	0.3–9.4
Tsai, 2014[6]		43	2001–2013	2.8	0.3–19.0
Pervasive developmental disorder					
French *et al*., 2013[4]		34	2000–2011	6.2	0.6–26.4
Elsabbagh *et al*., 2012[5]	Northern Europe	14	2000–2011	6.2	3.0–11.6
	Western Pacific	4	2004–2011	-	1.6–19.0
	South East Asia/East Mediterranean	4	2007–2012	-	0.1–10.7
	Americas	13	2001–2010	6.5	1.3–11.0
	Overall			6.2	0.1–19.0
Tsai, 2014[6]		61	2000–2014	7.0	0.2–26.4
Adak and Halder, 2017[7]		25	2005–2015	9.2	0.7–26.4

*The authors comment on dubious quality of results.

479 (44.5%) at 3–5 years and 383 (30.9%) at >5 years of age.[9] A review has suggested there remains considerable variation in age at diagnosis.[10] Further current data are clearly needed.

One reason why it is important to understand prevalence of autism is that the health profile of children and young people with autism is thought to differ from that of typically developing children and requires interventions and supports.[11–13] Therefore, these combined factors, that is, knowledge of prevalence and health profile of autistic children, are essential for planning and delivery of services.

Subjective general health status is commonly measured in general population studies, and has been demonstrated to be extremely valid, with a strongly predictive linear gradient across health status (from best to poorest) being associated with subsequent number of medical appointments, hospital admissions and mortality.[14–17] It is, therefore, important to measure if there are general health status differences in children and young people with autism compared with other children. However, in terms of general health status of children and young people with autism, there has been very little research. A study in USA reported parent-rated general health for 895 young people with autism aged 13–25 years at baseline, at five time points across 2001–2009, but did not include a general population comparison group.[18] General health was rated as excellent, very good, good or fair/poor. Fair/poor ratings were reported for 6.6% in 2001, 6.4% in 2003, 7.6% in 2005, 6.1% in 2007 and 6.6% in 2009.[18] A large study presenting data from the 2011–2012 National Survey of Children's Health identified 1188/56 746 children with autism under the age of 18, who were found to have significantly lower log odds of health (−1.30, p<0.001) compared with all other children.[19]

To our knowledge, no other studies have investigated reported general health status of children and young people with autism, nor drawn direct comparisons with the general population. This appears to be a major gap in our knowledge.

This study aimed to investigate, on a large scale (the entire population of a country—Scotland) (1) the prevalence of autism, and age of reporting/identifying autism in childhood and (2) the general health status of children and young people with autism compared with those without autism.

METHODS

Procedures

Approval was gained from the Scottish Government for secondary analysis of Scotland's Census, 2011 data under the auspices of a collaborative research project with National Records of Scotland.

Data source

Scotland's Census, 2011, provides information on the number and characteristics of Scotland's population and households on the census day, 27 March 2011. The census is undertaken every 10 years. It includes the whole Scottish population: people living in communal establishments (such as care homes and student halls of residence) as well as people living in private households. Scotland's Census is one of the few country censuses, and indeed it may be unique, in identifying people with autism. One householder on behalf of all occupants in private households, and manager on behalf of all occupants in communal dwellings, was required to complete the Census information. In the great majority of cases, this was, therefore, a parent of the child/young person. The Census form clearly stated that it is a legal requirement to complete the Census, and that not completing it, or supplying false information, can result in a £1000 fine. The Census team conducted a follow-up of non-responders, and provided help to respond when that was needed, hence the high completion rate of 94%.[20] For 2011, the UK Census Offices endorsed CANCEIS (Canadian Census Edit and Imputation System) as the cornerstone of the 2011 Census Editing Strategy. CANCEIS performs robust, cost-effective editing and imputation while incorporating methodological best practice. The Census team used a Census Coverage Survey, including around 40 000 households, to estimate numbers and characteristics of the missing 6%. The Coverage Survey and Census records were deterministically matched using automated and clerical matching to check for duplicates. Individuals estimated to have been missed from the Census were then imputed using a subset of characteristics from real individuals, including information on their health. The edit and imputation methodology was adapted from the rigorous and systematic guidelines of the Office for National Statistics, which is the UK's largest independent producer of official statistics and the recognised national statistical institute in the UK.[21] Two further Scottish Government reports provide information on the estimation and adjustment process used to produce census population estimates for Scotland[22] as well as full details of the methods and other background information.[23]

Census variables

People with autism were identified from Census question 20, which asked, 'Do you have any of the following conditions which have lasted, or are expected to last, at least 12 months? Tick all that apply'. There was a choice of 10 response options, which included developmental disorder (eg, autistic spectrum disorder or Asperger's syndrome), learning disability (eg, Down's syndrome), learning difficulty (eg, dyslexia) and mental health condition.

During the methodology development for Scotland's Census, 2011, Ipsos MORI Scotland was commissioned to undertake cognitive question testing on question 20 on long-term health conditions and disabilities. This was to test whether the questions were answered accurately and willingly by respondents, and to identify any changes needed to improve data quality and/or the acceptability of the response options for the Scottish population. Cognitive interviewing is a widely used approach to critically evaluate and improve survey questionnaires.[24] It enables researchers to modify survey material to enhance clarity. Retrospective probing was selected as the most appropriate technique. The questions were tested with 102 participants with a mix of gender, age and health conditions and disabilities (including people with more than one of the conditions), to ensure accurate and willing completion.[25] They included people with autism, intellectual disabilities, dyslexia, dyspraxia, speech impairment, mental health conditions (both milder and more serious) and other long-term conditions. This resulted in a redesign of the question on autism, to 'developmental disorder (eg, autism spectrum disorder or Asperger's syndrome)' in order to accurately capture specifically the data on autism. The questions on the other conditions tested (some of which, from a medical perspective, can be considered as developmental disorders) did not require any modification.

Thus, the choice of wording of the question on autism was informed and carefully considered. The term 'developmental disorder' was used and prompted respondents to reply with regard to only autistic spectrum disorder or Asperger syndrome. The question also distinguished autism from learning disability (which in the UK is synonymous with the international term 'intellectual disabilities'), learning difficulties such as dyslexia, and mental health conditions.

The Census team imputed answers for the 14.7% who did not tick any of the boxes in question 20, based on their free text answers for this question and answers to other health questions in the Census, which increased the completion rate to 97.4%. For the remaining 2.6%, the Census team assumed the most plausible explanation was that the person had no long-term condition but did not see the 'No condition' check box at the end of the question, and hence recorded them as such.[26]

Information on general health status was collected through question 19 which had a 5-point response scale: 'How is your health in general?' (1) very good, (2) good, (3) fair, (4) bad, (5) very bad. Similarly, as for question 20, question 19 was tested during the cognitive question testing during the development of the Census. The question was found to not require any modification.

Data analysis

We calculated the number and percentage of children reported to have autism, by age and gender. We also calculated the number and percentage of children and young people with and without autism reporting very good, good, fair, bad and very bad health, and compared differences using χ^2 tests. Within the whole population of children and young people in Scotland, we then used a logistic regression to calculate odds ratios (with 95%

CI) of autism predicting a derived, dichotomised variable of poor health (fair, bad or very bad health) versus good health (very good or good health), adjusted for age and gender. Age was categorised into groups of 0–15 years (children), or 16–24 years (youth), with the 0–15 year-olds being the reference group. These age groups were selected as in Scotland full legal capacity, with some limitations, is granted to people aged ≥16. Gender was binary; the reference group was male. We then calculated the ORs of age and gender in predicting poor health within the population with autism. All analyses were conducted with SPSS software V.22.

Patient and public involvement

The question on autism was included in Scotland's Census, 2011, at the behest of third sector organisations for people with autism. People with autism took part in the cognitive question testing during the planning of the Census. This study was undertaken by the Scottish Learning Disabilities Observatory, which has a specific remit for people with autism; its steering group includes partners from third sector organisations. Results from this study will be disseminated for people with autism in easy-read version via the Scottish Learning Disabilities Observatory website and newsletters.

RESULTS

Number (%) of children and young people with autism by age and gender

Scotland's Census, 2011, includes records on 916 331 children aged 0–15 years and 632 488 young people aged 16–24 years. Autism was reported for 17 348 (1.9%) of the children, and 7715 (1.2%) of the young people. Table 2 and figure 1 show the age and gender distribution of the children with and without autism. As expected, there are more males than females with autism; 13 841/17 348 (79.8%) children with autism were male. The rate of reported autism increased to age 11 in boys and age 10

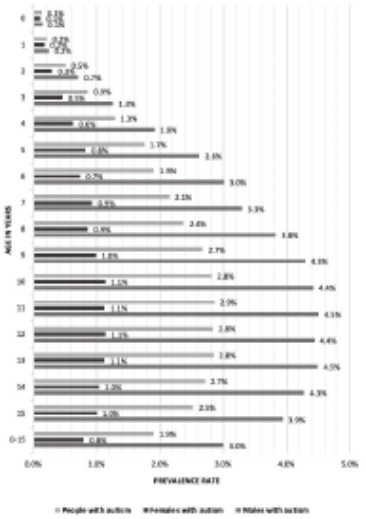

Figure 1 Identified childhood prevalence of autism by age and gender.

Table 2 Identified prevalence of childhood autism by age and gender

	All children			Children with autism		
Age in years	Total	Female	Male	Total	Female	Male
0	58 715	28 823	29 892	76 (0.1%)	34 (0.1%)	42 (0.1%)
1	59 556	29 188	30 368	126 (0.2%)	52 (0.2%)	74 (0.2%)
2	58 909	28 936	29 973	301 (0.5%)	87 (0.3%)	214 (0.7%)
3	58 764	28 735	30 029	509 (0.9%)	132 (0.5%)	377 (1.3%)
4	56 877	27 915	28 962	730 (1.3%)	176 (0.6%)	554 (1.9%)
5	55 224	26 910	28 314	966 (1.7%)	223 (0.8%)	743 (2.6%)
6	55 236	26 872	28 364	1053 (1.9%)	200 (0.7%)	853 (3.0%)
7	53 786	26 172	27 614	1154 (2.1%)	244 (0.9%)	910 (3.3%)
8	52 325	25 665	26 660	1243 (2.4%)	222 (0.9%)	1021 (3.8%)
9	53 046	26 022	27 024	1418 (2.7%)	257 (1.0%)	1161 (4.3%)
10	55 067	26 950	28 117	1549 (2.8%)	306 (1.1%)	1243 (4.4%)
11	56 769	27 699	29 070	1623 (2.9%)	313 (1.1%)	1310 (4.5%)
12	58 656	28 412	30 244	1665 (2.8%)	324 (1.1%)	1341 (4.4%)
13	59 971	29 353	30 618	1705 (2.8%)	330 (1.1%)	1375 (4.5%)
14	61 152	29 586	31 566	1658 (2.7%)	307 (1.0%)	1351 (4.3%)
15	62 278	29 987	32 291	1572 (2.5%)	300 (1.0%)	1272 (3.9%)
0–15	916 331	447 225	469 106	17 348 (1.9%)	3507 (0.8%)	13 841 (3.0%)

Table 3 General health status of children and young people with and without autism

	Age in years											
	0–15 years n=916 331						16–24 years n=632 488					
	Autism			Without autism			Autism			Without autism		
General health	Total 17 348 (100%)	F 3507 (100%)	M 13 841 (100%)	Total 898 983 (100%)	F 443 718 (100%)	M 455 265 (100%)	Total 7715 (100%)	F 1676 (100%)	M 6039 (100%)	Total 624 773 (100%)	F 313 929 (100%)	M 310 844 (100%)
Very good	7470 (43.1%)	1291 (36.8%)	6179 (44.6%)	758 328 (84.4%)	376 945 (85.0%)	381 383 (83.8%)	3070 (39.8%)	531 (31.7%)	2539 (42.0%)	459 492 (73.5%)	223 178 (71.1%)	236 314 (76.0%)
Good	6073 (35.0%)	1178 (33.6%)	4895 (35.4%)	122 814 (13.7%)	58 499 (13.2%)	64 315 (14.1%)	2683 (34.8%)	605 (36.1%)	2078 (34.4%)	137 956 (22.1%)	75 489 (24.0%)	62 467 (20.1%)
Fair	2892 (16.7%)	718 (20.5%)	2174 (15.7%)	14 760 (1.6%)	6800 (1.5%)	7960 (1.7%)	1451 (18.8%)	367 (21.9%)	1084 (17.9%)	22 102 (3.5%)	12 507 (4.0%)	9595 (3.1%)
Bad	651 (3.8%)	204 (5.8%)	447 (3.2%)	2367 (0.3%)	1159 (0.3%)	1208 (0.3%)	375 (4.9%)	125 (7.5%)	250 (4.1%)	4237 (0.7%)	2279 (0.7%)	1958 (0.6%)
Very bad	262 (1.5%)	116 (3.3%)	146 (1.1%)	714 (0.1%)	315 (0.1%)	399 (0.1%)	136 (1.8%)	48 (2.9%)	88 (1.5%)	986 (0.2%)	476 (0.2%)	510 (0.2%)

in girls, being relatively similar across ages 9–15 years for both genders, reflecting the ages at which the autism was diagnosed in the population. Prevalence was 2.8% at age 10 years (4.4% for boys and 1.1% for girls) and 2.9% at age 11 years (4.5% for boys and 1.1% for girls).

General health

Table 3 shows reported general health status of children and young people with and without autism in Scotland. The children and young people with autism reported poorer health; 22.0% of children and 25.5% of young people with autism reported poor (fair, bad or very bad) general health, compared with only 2.0% of children and 4.4% of young people without autism (χ^2=29 365.6; df=1; p<0.001 for children, and χ^2=7652.1; df=1; p<0.001 for young people). Table 3 shows that the discrepancy between those with and without autism was greater for females than males, for children rather than young people and was even more prominent when comparing bad/very bad health (as opposed to fair/bad/very bad health), for example, 9.1% of girls with autism had bad/very bad health compared with only 0.4% of girls without autism.

Table 4 shows the results from the regression with the whole population data. Autism had OR=11.3 (95% CI 11.0 to 11.7) in predicting poor health, adjusted for gender and age. Young people were more likely to have poor health than children, as were females. This pattern was also seen within the autistic population, more markedly so for females, and less so for increasing age when compared with the whole population (table 5). Female gender had OR=1.6 (95% CI 1.5 to 1.8), and age 16–24 years had OR=1.2 (95% CI 1.1 to 1.3) in predicting poor health within the autistic population.

Table 4 OR of autism, age and gender in predicting poor health* in the whole population

Variable		OR	95% CI
Autism	No autism (reference)	-	
	Autism	11.339	10.983 to 11.707
Age	0–15 (reference)	-	
	16–24	2.137	2.098 to 2.176
Gender	Male (reference)	-	
	Female	1.126	1.106 to 1.147
Constant		0.020	

*Fair, bad or very bad health.

DISCUSSION

Principle findings and interpretation

We identified the prevalence of reported autism to be 1.9% in children aged 0–15 years overall, and that the reported rate increased with age up to 10 years in girls and 11 years in boys, reflecting the age at which it was identified/diagnosed. Almost all were identified by age 9 years, with the majority before primary school. Prevalence was 2.8% at age 10 years and 2.9% at age 11 years, higher than when the rate is reported for all children overall. This is

Table 5 OR of age and gender in predicting poor health* in the population with autism

Variable		OR	95% CI
Age	0–15 (reference)	-	
	16–24	1.206	1.133 to 1.284
Gender	Male (reference)	-	
	Female	1.635	1.527 to 1.750
Constant		0.252	

*Fair, bad or very bad health.

of importance when interpreting prevalence studies, as autism in early childhood will clearly be underreported, thus lowering the overall reported childhood prevalence, unless detailed individual assessments are undertaken, which is not realistic in large-scale population-based research. Our study is the only whole-country population study we are aware of to-date to report prevalence of autism using current concepts of the autism spectrum and is highly representative as autism was systematically enquired about for the entire population, with a 94% response rate. Of considerable significance, we report that children/young people with autism were more than 11 times more likely to have poor health than the rest of the population. This inequality was greater for females than males, and more so than in the general population.

Comparison with existing literature

We found a higher rate of autism than that in the most recent systematic reviews on the subject. This finding most likely reflects that the data are more recent (2011) compared with the most recent reviews, which included data from studies completed a decade earlier, and that we report by year of age, rather than just for all children combined. More comparable studies include the Stockholm Youth Cohort which reported rates of autism in 2011 of 0.40% at age 0–5 years, 1.74% at age 6–12 years, 2.46% at age 13–17 years and 1.76% at age 18–27 years; and of 1.44% at ages 0–17 years overall.[27] The Data Resource Center for Child & Adolescent Health findings for 2014[28] and 2016[9] report higher prevalence of autism at 2.2% (n=243) and 2.5% (n=1131) in all 3–17 year olds but are on a smaller scale. The Autism and Developmental Disabilities Monitoring Network, in 11 sites in the USA, provides estimates of the prevalence of autism in 8-year-old children.[29] In 2014, this varied across sites from 1.3% to 2.9%, with a combined prevalence of 1.7%.[29]

Reported general health was substantially poorer for children and young people with autism compared with the general population. However, there is limited previous research with which to compare our findings; indeed, we believe we are the first to study general health status compared directly with the general population in a whole country population of children and young people with autism. Our findings of poor (fair, bad or very bad) health in 2.0% of children and 4.4% of young people without autism are similar to those reported in a National Health Interview Survey in 2014 which found fair/poor health for 1.6% (n=234) of children aged 0–17 years.[28] However, the study did not report health status separately for children with autism. A further US study reported lower rates of fair/poor health than the 25.5% we found in the young people with autism.[18] It reported fair/poor health in 6.6% in 2001, 6.4% in 2003, 7.6% in 2005, 6.1% in 2007 and 6.6% in 2009 of 895 young people with autism aged 13–25 years at baseline, but did not have a general population comparison group.[18] It also used measures of health not directly comparable with our study, using a 4-point scale of excellent, very good, good and fair/poor health.[18] Our findings of OR of 11.3 for autism predicting poor general health in the whole population of children and young people are not directly comparable with the findings from the National Survey of Children's Health from 2011 to 2012, due to differences in the scales used, though the results are in the same direction.[19]

Young people with autism had poorer health than children with autism, but the extent of this difference was much less than that seen in the general population. The difference in the extent of influence of age category between the people with and without autism lies in the substantial inequalities in general health that are associated with having autism, regardless of age. Our findings show that children and young people with autism of all ages are more likely to experience poorer general health compared with the rest of the population. We are unable to explain the reasons for this, but note that it is in addition to, and may be related to, their increase in comorbidities compared with other children and young people.[11–13] This requires further investigation.

Strengths and limitations

This large-scale study covers the whole population of Scotland, and we believe it is currently unique in being a whole country study in which every citizen was systematically enquired about regarding having autism and their general health status. It also had a high completion rate of 94%, suggesting the results are highly representative and likely to be generalisable to other high-income countries. Limitations include the use of the term of 'developmental disorders' in the Census. However, it prompted responses only for the examples of autistic spectrum disorder or Asperger's syndrome, and was tested prior to its use in the Census. Furthermore, the developmental disorders category was distinguished from intellectual disabilities, learning difficulties, and mental health conditions, which are important distinctions. The wording of the question on autism was informed in advance by the cognitive question testing procedure. Therefore, we consider that respondents will have replied accordingly, that is, regarding autism. However, we have no means to check this. Respondents reported whether or not each child/young person was known to have autism rather than each person having an assessment for autism. We are unable to report on the age that each child/young person received their diagnosis; hence we report instead the number of children at each age who have received the diagnosis. They are the proportion at each age who will call on services for children/young people with autism, so this information is important for service planning. Some reporting error is possible, but we are unable to check this. The majority of reports were proxy-reports by parents, but we do not know the extent of proxy versus self-reports for the young people. Neither do we know the extent to which proxy-reporting of general health status compares with an individual's report. The general health status responses were subjective measurements, which might have been influenced by a variety of factors such

as carer burden. It is controversial as to whether autism can be diagnosed in very young children. We found that a small number did report it in the first 2 years. While there may be some reporting error, differences in development in autistic children have been reported to be apparent from as early as 6 months, and widespread by 18 months.[30] The data from this study were collected in 2011, so it will not have captured any changes that have occurred since then. While we described the imputation process, we cannot state with certainty whether the imputed 6% of records contained similar, higher or lower proportion of children and young people with reported autism but note that this missing 6% is a small proportion overall. Imputation of zero by the Census team on the 2.6% of the population with missing data on long-term conditions was not tested, though considered to be the most plausible explanation. Despite these limitations, we believe the results of this study are generalisable to other high-income countries and fill a significant gap in existing research on general health status of children and young people with autism.

Implications for clinicians

It is essential to have accurate information on the proportion of children and young people who are known to have autism, and their health status, in order to accurately plan appropriate prevention and intervention measures, and provision of resources for those people who may put demand on services designed for people with autism. This requires a full understanding of age differences, and age at diagnosis. The poor general health status observed in the population of children and young people with autism demonstrates a clear need to focus on improvements in healthcare and supports, and the wider determinants of health in this group, which may well differ from the general population.

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Contributors ER analysed the data, jointly interpreted it and wrote the first draft of the manuscript. LAH-M, CG and AH jointly interpreted the data, and contributed to the manuscript. CM and JR worked on the Census, jointly interpreted the data and contributed to the manuscript. S-AC conceived the project, interpreted the data and contributed to the manuscript. All authors approved the final version of the manuscript. S-AC is the study guarantor. S-AC confirms the manuscript is an honest, accurate and transparent account of the study being reported, that no important aspects of the study have been omitted and there has been no discrepancies from the study as planned.

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Paper V

BMJ Open

Prevalence of long-term health conditions in adults with autism: observational study of a whole country population

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ABSTRACT

Objectives To investigate the prevalence of comorbid mental health conditions and physical disabilities in a whole country population of adults aged 25+ with and without reported autism.

Design Secondary analysis of Scotland's Census, 2011 data. Cross-sectional study.

Setting General population.

Participants 94% of Scotland's population, including 6649/3 746 584 adults aged 25+ reported to have autism.

Main outcome measures Prevalence of six comorbidities: deafness or partial hearing loss, blindness or partial sight loss, intellectual disabilities, mental health conditions, physical disability and other condition; ORs (95% CI) of autism predicting these comorbidities, adjusted for age and gender; and OR for age and gender in predicting comorbidities within the population with reported autism.

Results Comorbidities were common: deafness/hearing loss—17.5%; blindness/sight loss—12.1%; intellectual disabilities—29.4%; mental health conditions—33.0%; physical disability—30.7%; other condition—34.1%. Autism statistically predicted all of the conditions: OR 3.3 (95% CI 3.1 to 3.6) for deafness or partial hearing loss, OR 8.5 (95% CI 7.9 to 9.2) for blindness or partial sight loss, OR 94.6 (95% CI 89.4 to 100.0) for intellectual disabilities, OR 8.6 (95% CI 8.2 to 9.0) for mental health conditions, OR 6.2 (95% CI 5.8 to 6.6) for physical disability and OR 2.6 (95% CI 2.5 to 2.8) for other condition. Contrary to findings within the general population, female gender predicted all conditions within the population with reported autism, including intellectual disabilities (OR=1.4).

Conclusions Clinicians need heightened awareness of comorbidities in adults with autism to improve detection and suitable care, especially given the added complexity of assessment in this population and the fact that hearing and visual impairments may cause additional difficulties with reciprocal communication which are also a feature of autism; hence posing further challenges in assessment.

Strengths and limitations of this study

- ► Unique study of comorbidity in adults with reported autism in a whole country population.
- ► High response rate of 94% and systematic enquiry of everyone regarding autism and comorbidities (deafness, blindness, intellectual disabilities, mental health conditions, physical disability and other condition).
- ► Results of the study are generalisable to other adult populations in high-income countries.
- ► Findings are limited by the broad survey reporting of comorbidities, rather than detailed examinations.

INTRODUCTION

In the last 20 years, there has been a considerable increase in awareness of autism, but research on the comorbid conditions that adults with autism experience is limited.[1] It has been suggested that some comorbidities are more common in children with autism than in the general population,[2] but little research has been conducted with adults. Given the communicative and other problems that are a prominent feature of autism, the detection and management of comorbid conditions in people with autism is more complex than for other people. Therefore, it is important to know whether or not health problems are more common than in the general population. Empirically founded information about autism comorbidity would help to raise clinicians' awareness, and in turn increase identification and appropriate management.

Mental health has been studied more than physical health in adults with autism. However, systematic reviews reveal wide variation in reported prevalence of mental ill-health between studies. This is partly because almost all studies are based on clinical populations. Therefore, findings cannot be generalised with confidence, and additionally most study samples are small in size, and very few have drawn comparisons with the general population. It has

been suggested that depression,[3] bipolar disorder,[4] suicidal thoughts/behaviour,[5] non-affective psychosis[6] and attention-deficit hyperactivity disorder[7 8] may be more common in adults with autism. A further study in North California, USA used medical records from a single health delivery provider of inpatient and outpatient medical and mental health services to identify 1507/1 578 658 (0.1%) adults with autism, who were age and gender matched with controls without recorded autism.[9] The study found that 19.2% of the adults with autism also had a record of intellectual disabilities, and 54% also had a record of one or more mental health conditions; with rates of individual mental conditions being 3–22 times higher for the adults with autism than their controls, and higher in the women with autism than in the men with autism.[9] The study does, however, reflect the sampling frame; only those individuals with an existing record of autism in their medical records were identified as having autism.

Blindness/sight loss, deafness/hearing loss and physical disabilities may be more common in adults with autism than in other people, but most of the existing literature is drawn from non-representative and/or small populations, and without general population comparisons; hence leaving significant doubts as to the actual degree of over-representation. One exception is the North California study of a wide range of conditions recorded in medical records which found 16 (1.1%) adults with autism to have low vision or blindness (OR=7.85) and 71 (4.7%) with hearing impairment (OR=2.35).[9] A further large study across 25 states in USA included 1002 adults known to have autism, but was drawn from the population receiving intellectual and developmental disabilities services; hence, it is clearly not representative of the population of adults with autism.[10] Indeed, only 97 (9.7%) participants did not have intellectual disabilities, so while 9.4% had visual impairments, 5.7% had hearing impairments and 6.0% had physical disability, these rates cannot be generalised to the wider population with autism. In a study of 92/305 adults aged 23–50 who had been identified to have autism in childhood in the 1980s, 11 of whom were deceased, participants answered questions on medical conditions and symptoms.[11] Of the 92, 73% had intellectual disabilities, 12% had hearing impairment and 25% visual impairment.[11] Neither of these two studies included a general population comparison group. We were unable to identify any other studies on these conditions in adults with autism.

This study aimed to investigate the prevalence and predictors of deafness or partial hearing loss, blindness or partial sight loss, intellectual disabilities, mental health conditions, physical disability and other condition, in a whole country population of adults with reported autism aged 25+ compared with their peers without autism.

METHODS

Data source

Scotland's Census, 2011 provides information on Scotland's population on the Census date, 27 March 2011. Approval was gained from the Scottish Government for secondary analysis of the Census data. The Census includes the whole Scottish population, whether living in communal establishments (such as care homes and student halls of residence) or private households. Scotland's Census is one of the few country censuses that asks every person in the country whether or not they have autism, indeed it may be unique in this regard. One householder on behalf of all occupants in private households (the household reference person), and manager on behalf of all occupants in communal dwellings, was required to complete the Census information. The Census team also followed up non-responders and provided help to respond when needed. The Census form clearly states that it is a legal requirement to complete the form and non-completion or supplying false information attracts a £1000 fine. The Census is conducted every 10 years. In 2011, it was estimated to have achieved a 94% response rate.[12] During the original data processing, the Census team adjusted for the 6% of the total population of Scotland for whom there was not completed Census data. This used a Census Coverage Survey (including around 40 000 households) to estimate numbers and characteristics. The Coverage Survey and Census records were matched using automated and clerical matching. All Census individuals, including individuals reporting long-term health conditions, were deterministically matched to check if any records were duplicated. Individuals estimated to have been missed from the Census were then imputed, using a subset of characteristics from real individuals, including information on their health, to reach the 100% dataset completeness rate. The process of development of the Scotland's Census 2011 Edit and Imputation Methodology was adapted from the Office for National Statistics rigorous and systematic guidelines, which are available here: http://webarchive.nationalarchives.gov.uk/20160108193745/http://www.ons.gov.uk/ons/guide-method/method-quality/survey-methodology-bulletin/smb-69/index.html.

Further details on how the Census population estimates were arrived at are also available here: http://www.scotlandscensus.gov.uk/documents/censusresults/release1b/rel1bmethodology.pdf.

Full details of the methodology and other background information on Scotland's Census 2011 are available at: http://www.scotlandscensus.gov.uk/supporting-information.

CENSUS VARIABLES

Self-reporting/proxy reporting was used to identify people with autism and other long-term conditions from the Census questionnaire, question 20: 'Do you have any of the following conditions which have lasted, or are expected to last, at least 12 months? Tick all that apply'. Respondents were given a choice of 10 response options: (1) deafness or partial hearing loss, (2) blindness or partial

sight loss, (3) learning disability (eg, Down's syndrome), (4) learning difficulty (eg, dyslexia), (5) developmental disorder (eg, autistic spectrum disorder or Asperger's syndrome), (6) physical disability, (7) mental health condition, (8) long-term illness, disease or condition (9) other condition, (10) no condition. Following internal requirements for all Scotland's Census 2011 outputs stipulated by the National Records of Scotland, options 8 (long-term illness, disease or condition) and 9 (other condition) were merged and coded as one category of 'other condition'; thus, this term is used henceforth when referring to both these categories.

Importantly, while question 20: 'Do you have any of the following conditions which have lasted, or are expected to last, at least 12 months?', included the broad term developmental disorder, it only provided reference to 'autistic spectrum disorder' and 'Asperger's syndrome'. For the purpose of this study, we, therefore, interpreted responses to this question as relating to people who know they have these conditions, henceforth referred to as autism. Additionally, the question distinguished autism from learning disability (which in the UK is synonymous with the international term 'intellectual disabilities'), learning difficulty (which in the UK is synonymous with the international term 'specific learning disability' such as dyslexia) and mental health conditions, which are important distinctions.

As part of the methodological preparations for Scotland's Census, 2011, the General Register Office for Scotland commissioned Ipsos MORI Scotland to undertake cognitive question testing of the question 20 on long-term health conditions and disabilities. The aim was primarily to test whether the questions were answered accurately and willingly by respondents, and what changes might be required to improve data quality and/or the acceptability of the response options. Cognitive interviewing is a widely used approach to critically evaluate survey questionnaires.[13] It enables researchers to modify survey material to enhance clarity. Retrospective probing was deemed to be the most appropriate of the different techniques for the Census. The questions were tested with 102 participants with a mix of gender and age, both with and without the health conditions and disabilities (including people with more than one of the conditions), to ensure accurate and willing completion, and included people with autism, intellectual disabilities, dyslexia, dyspraxia, speech impairment, mental health conditions (both milder and more serious) and other long-term conditions. This resulted in a redesign of the question on autism to 'developmental disorder, for example, autism spectrum disorder or Asperger's syndrome' in order to accurately capture specifically the data on autism. The questions on the other conditions tested (some of which, from a medical perspective, can be considered as developmental disorders) did not require any modification. Further information can be found at: http://www.scotlandscensus.gov.uk/documents/research/2011-census-health-disability-questions.pdf http://www.scotlandscensus.gov.uk/documents/legislation/changes-to-gov-statement-report.pdf

DATA ANALYSIS

We calculated the numbers and percentages of people with and without reported autism reporting deafness or partial hearing loss, blindness or partial sight loss, intellectual disabilities, mental health conditions, physical disability and other condition. We compared differences between the populations with and without reported autism using χ^2 tests. Within the whole population, we then used six binary logistic regressions to calculate OR (95% CI) of autism predicting having each of the six types of additional health conditions, adjusted for age group and gender. We then calculated the ORs for age group and gender in predicting each of the six comorbidities within the population with reported autism. All analyses were conducted with SPSS software V.22.

PATIENT AND PUBLIC INVOLVEMENT

The question on autism was included in Scotland's Census, 2011 at the behest of third sector organisations for people with autism. This study was undertaken by the Scottish Learning Disabilities Observatory, which has a specific remit for people with autism; its steering group includes partners from the third sector organisations. Results from this study will be disseminated for people with autism in easy-read version via the Scottish Learning Disabilities Observatory website and newsletters.

RESULTS

Participant characteristics

Scotland's Census 2011 included records on 5 295 403 people. There were 6649/3 746 584 (0.2%) adults aged 25+ recorded to have autism as defined here, 4610 (69.3%) of whom were men and 2039 (30.7%) women compared with 1 776 845 (47.5%) men and 1 963 090 (52.5%) women in the adult population without autism (table 1). The rate of autism was lowest in the oldest age groups (autism may be associated with reduced life expectancy).

Prevalence of reported comorbidities

The adult population with reported autism was significantly more likely to have each of the additional health conditions when compared with the population without reported autism, with each at the p<0.001 level (table 2).

Table 3 shows the OR (95% CI) of autism predicting each of the six conditions: OR 3.3 (3.1 to 3.6) for deafness or partial hearing loss, OR 8.5 (7.9 to 9.2) for blindness or partial sight loss, OR 94.6 (89.4 to 100.0) for intellectual disabilities, OR 8.6 (8.2 to 9.0) for mental health conditions, OR 6.2 (5.8 to 6.6) for physical disability and OR 2.6 (2.5 to 2.8) for other condition. Table 4 shows the OR (95% CI) of age and gender in predicting comorbidities

Table 1 Number and proportion of adults with autism by age and gender

Age	Gender, n (%)	Autism, n (%)	Without autism, n (%)
All adults aged 25+	Male n=1 781 455 (100)	4610 (0.3)	1 776 845 (99.7)
	Female n=1 965 129 (100)	2039 (0.1)	1 963 090 (99.9)
	All n=3 746 584 (100)	6649 (0.2)	3 739 935 (99.8)
25–34 years	Male n=328 607 (100)	1753 (0.5)	326 854 (99.5)
	Female n=338 720 (100)	636 (0.2)	338 084 (99.8)
	All n=632 488 (100)	2389 (0.4)	664 938 (99.6)
35–44 years	Male n=357 670 (100)	1117 (0.3)	356 553 (99.7)
	Female n=377 084 (100)	471 (0.1)	376 613 (99.9)
	All n=734 754 (100)	1588 (0.2)	733 166 (99.8)
45–54 years	Male n=384 517 (100)	890 (0.2)	383 627 (99.8)
	Female n=402 239 (100)	377 (0.1)	401 862 (99.9)
	All n=786 756 (100)	1267 (0.2)	785 489 (99.8)
55–64 years	Male n=326 922 (100)	474 (0.1)	326 448 (99.9)
	Female n=340 491 (100)	233 (0.1)	340 258 (99.9)
	All n=667 413 (100)	707 (0.1)	666 706 (99.9)
65+ years	Male n=383 739 (100)	376 (0.1)	383 363 (99.9)
	Female n=506 595 (100)	322 (0.1)	506 273 (99.9)
	All n=890 334 (100)	698 (0.1)	889 636 (99.9)

within the population with autism aged 25+. As one would expect, in the whole population, older age group statistically predicted blindness, deafness, physical disability and other condition, while age over 55 reduced the likelihood of intellectual disabilities (presumably due to early death), as did the 65+ age group for mental health conditions. Female gender predicted blindness, mental health conditions, physical disability and other condition, while male gender predicted deafness and intellectual disabilities. Within the population with reported autism, older age group also statistically predicted blindness, deafness, physical disability and other condition, but not intellectual disabilities and mental health conditions. Contrary to findings in the general population, female gender predicted all conditions within the population with reported autism.

DISCUSSION

Principal findings and comparison with existing literature

Comorbidity is substantially greater in adults with reported autism than in other people; with ORs of 95 for intellectual disabilities, 9 for mental health conditions, 9 for deafness or partial hearing loss, 6 for physical disability, 3 for blindness or partial sight loss and 3 for other condition. All these conditions were common in adults with reported autism. These findings are important given the gap in evidence, as clinicians need to have heightened awareness of potential comorbidities in order to provide suitable investigation and management to maximise functioning and therefore improve quality of life. Findings on hearing and visual impairments for people with reported autism are perhaps particularly important, given the impact of these impairments on reciprocal communication, which is also an integral underlying impairment in autism. Clinical assessments of people with autism are more complex and take longer than for the average person. Nevertheless, our findings have demonstrated that investment in such assessments is necessary and important given the much higher prevalence of comorbidities.

We found mental health conditions in 33% of all adults with reported autism (range 23%–37% depending on age group; 27%–37% for men and 30%–40% for women). This high rate is lower than a previous report of 54%,[9] but their sample may have been biased to a more severely affected/complex population given their sampling, as shown by their lower identification rate for autism. We found 14% with hearing impairment (range 7%–46% depending on age group; 5%–44% for men and 11%–47% for women), and 12% with visual impairments (range 7%–30% depending on age group; 7%–27% for men and 10%–35% for women), notably higher than the rates recorded in medical records reported in the North California study (4.7% and 1.1% respectively, though ORs were not dissimilar)[9] likely reflecting the different study methodologies. In the study of 92 adults with autism, 12% had hearing impairment and 25% visual impairment,[11] respectively, though the study was much smaller and of limited age range than in our study. A high rate of intellectual disabilities in children with autism has been described previously; we have now quantified the extent of this—29% (25%–32% depending on age group; 22%–35% for men and 31%–42% for women)—in

Table 2 Prevalence of comorbidities in adults with and without autism by age and gender

Age group	Condition	Autism			Without autism		
		Men N=4610 (100%)	Women N=2039 (100%)	Total N=6649 (100%)	Men N=1 776 845 (100%)	Women N=1 963 090 (100%)	Total N=3 739 935 (100%)
All adults aged 25+	Deafness/partial hearing loss	583 (12.6)	356 (17.5)	939 (14.1)	178994 (10.1)	160495 (8.2)	339489 (9.1)
	Blindness/partial sight loss	503 (10.9)	304 (14.9)	807 (12.1)	52351 (2.9)	65198 (3.3)	117549 (3.1)
	Intellectual disabilities	1254 (27.2)	699 (34.3)	1953 (29.4)	8141 (0.5)	6859 (0.3)	15000 (0.4)
	Mental health condition	1468 (31.8)	728 (35.7)	2196 (33.0)	90292 (5.1)	121584 (6.2)	211876 (5.7)
	Physical disability	973 (21.1)	626 (30.7)	1599 (24.0)	150896 (8.5)	188347 (9.6)	339243 (9.1)
	Other condition	1402 (30.4)	864 (42.4)	2266 (34.1)	407090 (22.9)	489875 (25.0)	896965 (24.0)
		Men N=1753 (100%)	Women N=636 (100%)	Total N=2 389 (100%)	Men N=326 854 (100%)	Women N=338 084 (100%)	Total N=664 938 (100%)
25–35 years	Deafness/partial hearing loss	94 (5.4)	68 (10.7)	162 (6.8)	4341 (1.3)	3651 (1.1)	7992 (1.2)
	Blindness/partial sight loss	118 (6.7)	62 (9.7)	180 (7.5)	2382 (0.7)	1698 (0.5)	4080 (0.6)
	Intellectual disabilities	391 (22.3)	211 (33.2)	602 (25.2)	1634 (0.5)	1239 (0.4)	2873 (0.4)
	Mental health condition	466 (26.6)	188 (29.6)	654 (27.4)	13522 (4.1)	19428 (5.7)	32950 (5.0)
	Physical disability	253 (14.4)	163 (25.6)	416 (17.4)	5616 (1.7)	5200 (1.5)	10816 (1.6)
	Other condition	420 (24.0)	218 (34.3)	638 (26.7)	23726 (7.3)	31470 (9.3)	55196 (8.3)
		Men N=1117 (100%)	Women N=471 (100%)	Total N=1588 (100%)	Men N=356 553 (100%)	Women N=376 613 (100%)	Total N=733 166 (100%)
35–44 years	Deafness/partial hearing loss	83 (7.4)	44 (9.3)	127 (8.0)	8442 (2.4)	7067 (1.9)	15509 (2.1)
	Blindness/partial sight loss	94 (8.4)	46 (9.8)	140 (8.8)	3664 (1.0)	2498 (0.7)	6162 (0.8)
	Intellectual disabilities	304 (27.2)	146 (31.0)	450 (28.3)	1905 (0.5)	1504 (0.4)	3409 (0.5)
	Mental health condition	377 (33.8)	187 (39.7)	564 (35.5)	22156 (6.2)	27844 (7.4)	50000 (6.8)
	Physical disability	216 (19.3)	112 (23.8)	328 (20.7)	12711 (3.6)	12727 (3.4)	25438 (3.5)
	Other condition	318 (28.5)	190 (40.3)	508 (32.0)	43670 (12.2)	54825 (14.6)	98495 (13.4)

Continued

Table 2 Continued

		Men N=890 (100%)	Women N=377 (100%)	Total N=1267 (100%)	Men N=383 627 (100%)	Women N=401 862 (100%)	Total N=785 489 (100%)
45–54 years	Deafness/partial hearing loss	116 (13.0)	58 (15.4)	174 (13.7)	19 115 (5.0)	13565 (3.4)	32 680 (4.2)
	Blindness/partial sight loss	113 (12.7)	46 (12.2)	159 (12.5)	6753 (1.8)	4554 (1.1)	11307 (1.4)
	Intellectual disabilities	268 (30.1)	133 (35.3)	401 (31.6)	2188 (0.6)	1712 (0.4)	3900 (0.5)
	Mental health condition	316 (35.5)	140 (37.1)	456 (36.0)	23060 (6.0)	29734 (7.4)	52794 (6.7)
	Physical disability	195 (21.9)	110 (29.2)	305 (24.1)	22783 (5.9)	24340 (6.1)	47 123 (6.0)
	Other condition	283 (31.8)	152 (40.3)	435 (34.3)	74773 (19.5)	86373 (21.5)	161 146 (20.5)
		Men N=474 (100%)	Women N=233 (100%)	Total N=707 (100%)	Men N=326 448 (100%)	Women N=340 258 (100%)	Total N=666 706 (100%)
55–65 years	Deafness/partial hearing loss	123 (25.9)	35 (15.0)	158 (6.7)	35743 (10.9)	21889 (6.4)	57 632 (3.4)
	Blindness/partial sight loss	77 (16.2)	39 (16.7)	116 (6.5)	9193 (2.8)	6640 (2.0)	15833 (1.2)
	Intellectual disabilities	158 (33.3)	98 (42.1)	256 (22.7)	1381 (0.4)	1226 (0.4)	2607 (0.4)
	Mental health condition	175 (36.9)	87 (37.3)	262 (22.9)	16848 (5.2)	18483 (5.4)	35331 (5.3)
	Physical disability	150 (31.6)	85 (36.5)	235 (15.1)	36100 (11.1)	37034 (10.9)	73134 (4.7)
	Other condition	199 (42.0)	114 (48.9)	313 (24.6)	106 897 (32.7)	109001 (32.0)	215898 (16.4)
		Men N=376 (100%)	Women N=322 (100%)	Total N=698 (100%)	Men N=383 363 (100%)	Women N=506 273 (100%)	Total N=889 636 (100%)
65+ years	Deafness/partial hearing loss	167 (44.4)	151 (46.9)	318 (45.6)	111353 (29.0)	114323 (22.6)	225676 (25.4)
	Blindness/partial sight loss	101 (26.9)	111 (34.5)	212 (30.4)	30359 (7.9)	49808 (9.8)	80167 (9.0)
	Intellectual disabilities	133 (35.4)	111 (34.5)	244 (35.0)	1033 (0.3)	1178 (0.2)	2211 (0.2)
	Mental health condition	134 (35.6)	126 (39.1)	260 (37.2)	14706 (3.8)	26095 (5.2)	40901 (4.6)
	Physical disability	159 (42.3)	156 (48.4)	315 (45.1)	73686 (19.2)	109046 (21.5)	182732 (20.5)
	Other condition	182 (48.4)	190 (59.0)	372 (53.3)	158024 (41.2)	208206 (41.1)	366230 (41.2)

Table 3 Results of six regressions showing independent predictors of comorbid conditions in the whole adult population

Condition	Variable	OR (95% CI)
Deafness or partial hearing loss	Autism	
	No autism (reference)	–
	Autism	3.32 (3.075 to 3.585)
	Age	
	25–34 (reference)	–
	35–44	1.768 (1.721 to 1.817)
	45–54	3.55 (3.464 to 3.638)
	55–64	7.742 (7.563 to 7.926)
	65+	28.621 (27.987 to 29.269)
	Gender	
	Male (reference)	–
	Female	0.683 (0.678 to 0.688)
	Constant	0.015
Blindness or partial sight loss	Autism	
	No autism (reference)	–
	Autism	8.514 (7.861 to 9.220)
	Age	
	25–34 (reference)	–
	35–44	1.36 (1.308 to 1.414)
	45–54	2.335 (2.254 to 2.419)
	55–64	3.882 (3.752 to 4.016)
	65+	15.769 (15.287 to 16.267)
	Gender	
	Male (reference)	–
	Female	1.018 (1.006 to 1.030)
	Constant	0.006
Intellectual disabilities	Autism	
	No autism (reference)	–
	Autism	94.571 (89.409 to 100.032)
	Age	
	25–34 (reference)	–
	35–44	1.101 (1.050 to 1.154)
	45–54	1.187 (1.134 to 1.243)
	55–64	0.958 (0.910 to 1.008)
	65+	0.631 (0.598 to 0.665)
	Gender	
	Male (reference)	–
	Female	0.812 (0.788 to 0.838)
	Constant	0.005
Mental health condition	Autism	
	No autism (reference)	–
	Autism	8.595 (8.163 to 9.050)
	Age	
	25–34 (reference)	–
	35–44	1.404 (1.384 to 1.424)

Continued

Table 3 Continued

Condition	Variable	OR (95% CI)
	45–54	1.383 (1.364 to 1.403)
	55–64	1.076 (1.060 to 1.093)
	65+	0.913 (0.899 to 0.926)
	Gender	
	Male (reference)	–
	Female	1.247 (1.236 to 1.258)
	Constant	0.046
Physical disability	Autism	
	No autism (reference)	–
	Autism	6.21 (5.841 to 6.603)
	Age	
	25–34 (reference)	–
	35–44	2.138 (2.091 to 2.186)
	45–54	3.786 (3.708 to 3.866)
	55–64	7.311 (7.164 to 7.460)
	65+	15.288 (14.994 to 15.587)
	Gender	
	Male (reference)	–
	Female	1.064 (1.056 to 1.072)
	Constant	0.016
Other condition	Autism	
	No autism (reference)	–
	Autism	2.64 (2.502 to 2.786)
	Age	
	25–34 (reference)	–
	35–44	1.709 (1.690 to 1.728)
	45–54	2.839 (2.810 to 2.868)
	55–64	5.269 (5.217 to 5.323)
	65+	7.671 (7.597 to 7.745)
	Gender	
	Male (reference)	–
	Female	1.068 (1.063 to 1.074)
	Constant	0.088

a much larger study of adults. There are few other studies with which we can draw comparisons, and we identified none on physical disability in adults with autism with which we could compare the high rate of 24% for all adults aged 25+ (range 15%–45% depending on age group; 14%–42% for men and 24%–45% for women).

A view has been expressed that autism is currently underdiagnosed in more intellectually able females compared with males.[14] We found that 34% of women compared with only 27% of men with autism reported accompanying intellectual disabilities, so the female population with autism was intellectually less able than the male population with autism. Our findings may, therefore, provide some evidence to support the view of underdiagnosis of autism in the more intellectually able women. Alternatively, women and men with autism may actually be intellectually different.

We have previously reported Census findings on comorbidities for people with intellectual disabilities.[15] Many conditions are related to intellectual level, with there being a gradient across the whole spread of intelligence (not just intellectual disabilities).[16] Given the lower average intelligence we found in the autistic women than the autistic men, one might expect more comorbidities to be found in the women than the men. Indeed, the women with autism had higher rates of all

Table 4 Results of six regressions showing independent predictors of comorbid conditions in the adult population with autism

Condition	Variable	OR (95% CI)
Deafness or partial hearing loss	Age	
	25–34 (reference)	–
	35–44	1.189 (0.934 to 1.514)
	45–54	2.178 (1.738 to 2.731)
	55–64	3.92 (3.088 to 4.975)
	65+	11.179 (8.972 to 13.929)
	Gender	
	Male (reference)	–
	Female	1.169 (1.001 to 1.365)
	Constant	0.07
Blindness or partial sight loss	Age	
	25–34 (reference)	–
	35–44	1.179 (0.936 to 1.485)
	45–54	1.75 (1.397 to 2.192)
	55–64	2.378 (1.851 to 3.056)
	65+	5.148 (4.117 to 6.438)
	Gender	
	Male (reference)	–
	Female	1.232 (1.051 to 1.443)
	Constant	0.077
Intellectual disabilities	Age	
	25–34 (reference)	–
	35–44	1.163 (1.008 to 1.343)
	45–54	1.363 (1.172 to 1.584)
	55–64	1.656 (1.384 to 1.981)
	65+	1.505 (1.254 to 1.807)
	Gender	
	Male (reference)	–
	Female	1.354 (1.209 to 1.516)
	Constant	0.309
Mental health condition	Age	
	25–34 (reference)	–
	35–44	1.455 (1.269 to 1.668)
	45–54	1.485 (1.284 to 1.719)
	55–64	1.548 (1.297 to 1.849)
	65+	1.531 (1.280 to 1.832)
	Gender	
	Male (reference)	–
	Female	1.155 (1.034 to 1.291)
	Constant	0.362
Physical disability	Age	
	25–34 (reference)	–
	35–44	1.22 (1.038 to 1.434)
	45–54	1.487 (1.258 to 1.758)
	55–64	2.312 (1.913 to 2.795)

Continued

Table 4 Continued

Condition	Variable	OR (95% CI)
	65+	3.634 (3.022 to 4.370)
	Gender	
	Male (reference)	–
	Female	1.504 (1.333 to 1.697)
	Constant	0.187
Other condition	Age	
	25–34 (reference)	–
	35–44	1.276 (1.109 to 1.467)
	45–54	1.419 (1.224 to 1.645)
	55–64	2.134 (1.792 to 2.542)
	65+	2.901 (2.433 to 3.459)
	Gender	
	Male (reference)	–
	Female	1.563 (1.400 to 1.745)
	Constant	0.321

six comorbidities than did the men with autism; OR of female gender predicting each of the six comorbidities was greater for all conditions (except mental health) in the population with autism compared with the whole population, and indeed reversed for deafness and intellectual disabilities which were more common in men in the whole population. Alternatively, these findings could support the view that in some cases it is the concept of 'Autism Plus' (ie, autism co-occurring with any other major neurodevelopmental disorder),[17] which ultimately results in people receiving a diagnosis of autism. While this Plus element of co-occurring conditions is currently often neglected in assessment, diagnosis and intervention, in some populations, possibly including women, it may be the initial or primary reason for considering a diagnosis of autism.

Strengths and limitations

We believe this study to be unique in including the whole population of a country, with a high response rate, and systematic enquiry of everyone regarding autism and selected long-term conditions. The results of this study are generalisable to other adult populations in high-income countries. The concept of autistic spectrum disorder has broadened in recent years; hence, our findings relate to the narrower definition that was used to diagnose autism in the past, as the study is one of adults who most likely were originally diagnosed in childhood. This accounts for the 0.2% identified prevalence; more recent studies conducting autism assessments have reported higher prevalence.[18] It is important to note that undiagnosed adults with milder forms of autism may have lower levels of comorbidity than those with more severe autism. Limitations may include the use of the term developmental disorder in the Census, although the clarification of this term provided on the Census form included only autistic spectrum disorder and Asperger's syndrome, and the phrasing of the question was carefully selected specifically to capture autism, from results of the cognitive question testing procedure. Furthermore, this category was distinguished from intellectual disabilities, specific learning disability and mental health conditions, and tested with people with all these conditions. Hence, we consider that respondents will have replied accordingly, that is, responded regarding autism. However, we have no further means to check this on the whole population. Furthermore, respondents reported whether or not each person was known to have autism rather than each person having an assessment for autism, so some reporting error is possible. Given the large number of households, we are unable to state how each household reference person approached completing the Census form, although cognitive question testing was completed with a broad range of 70 respondents on the whole questionnaire in advance of the Census (in addition to the 102 respondents who completed cognitive question testing interviews specifically on the health questions). The Census form was also broad-brush in its questioning rather than including detailed subquestions on each of the six categories of health conditions. Finally, while we describe the imputation process, we cannot state with certainty whether or not the imputed 6% of records contained the same, higher or lower proportion of adults with autism, but note that this missing 6% is a small proportion overall. Despite this, we believe the results of this study are generalisable to other high-income countries, as well as filling a significant gap in existing research on the prevalence of long-term health conditions in adults with autism.

Implications for clinicians

This study advances our knowledge of comorbidities in adults with autism, which is otherwise somewhat invisible in previous studies. Adults with reported autism have very high rates of comorbid physical disabilities as well as mental health conditions. Hearing and visual impairments are also very common, and their impact on reciprocal communication, especially if undiagnosed/unattended, may compound core features of autism. Clinicians require a heightened awareness of this, especially given the greater complexity of health assessments in adults with autism compared with other people. It is essential to have accurate information on the prevalence of comorbid conditions in adults with autism in order to accurately plan for service provision and to tackle health inequalities. Our study is large scale and robust in design, but requires replication given the relative lack of previous studies on this topic.

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Contributors ER analysed the data, jointly interpreted it and wrote the first draft of the manuscript. LAH-M, CG and AH jointly interpreted the data and contributed to the manuscript. CM and JR worked on the Census, jointly interpreted the data and contributed to the manuscript. S-AC conceived the project, interpreted the data and contributed to the manuscript. All authors approved the final version of the manuscript. S-AC is the study guarantor.

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Corrections: *Prevalence of long-term health conditions in adults with autism: observational study of a whole country population*

Rydzewska E, Hughes-McCormack LA, Gillberg C, *et al*. Prevalence of long-term health conditions in adults with autism: observational study of a whole country population *BMJ Open* 2018;**8**:e023945. doi: 10.1136/bmjopen-2018-023945.

This article was previously published with some errors.

In result section of the abstract, the percentage of deafness/hearing loss, physical disability and the OR for mental health conditions are incorrect.

deafness/hearing loss should be 14.1% instead of 17.5%
physical disability should be 24.0% instead of 30.7%
OR for mental health conditions should be OR 8.6 (95% CI 8.2 to 9.1) instead of OR 8.6 (95% CI 8.2 to 9.0).

On the prevalence of reported comorbidities section (page 3) it reads:

Table 3 shows the OR (95% CI) of autism predicting each of the six conditions: OR 3.3 (3.1 to 3.6) for deafness or partial hearing loss, OR 8.5 (7.9 to 9.2) for blindness or partial sight loss, OR 94.6 (89.4 to 100.0) for intellectual disabilities, OR 8.6 (8.2 to 9.0) for mental health conditions, OR 6.2 (5.8 to 6.6) for physical disability and OR 2.6 (2.5 to 2.8) for other condition.

It should read:

Table 3 shows the OR (95% CI) of autism predicting each of the six conditions: OR 3.3 (3.1 to 3.6) for deafness or partial hearing loss, OR 8.5 (7.9 to 9.2) for blindness or partial sight loss, OR 94.6 (89.4 to 100.0) for intellectual disabilities, OR 8.6 (8.2 to 9.1) for mental health conditions, OR 6.2 (5.8 to 6.6) for physical disability and OR 2.6 (2.5 to 2.8) for other condition.

On the discussion section, first sentence of the first paragraph reads:

Comorbidity is substantially greater in adults with reported autism than in other people; with ORs of 95 for intellectual disabilities, 9 for mental health conditions, 9 for deafness or partial hearing loss, 6 for physical disability, 3 for blindness or partial sight loss and 3 for other condition.

It should read:

Comorbidity is substantially greater in adults with reported autism than in other people; with ORs of 95 for intellectual disabilities, 9 for mental health conditions, 9 for blindness or partial sight loss, 6 for physical disability, 3 for deafness or partial hearing loss and 3 for other condition.

On the discussion section, third sentence of the second paragraph reads:

We found 14% with hearing impairment (range 7%–46% depending on age group; 5%–44% for men and 11%–47% for women), and 12% with visual impairments (range 7%–30% depending on age group; 7%–27% for men and 10%–35% for women), notably higher than the rates recorded in medical records reported in the North California study (4.7% and 1.1% respectively, though ORs were not dissimilar)9 likely reflecting the different study methodologies.

It should read:

We found 14% with hearing impairment (range 7%–46% depending on age group; 5%–44% for men and 9%–47% for women), and 12% with visual impairments (range 7%–30% depending on age group; 7%–27% for men and 10%–35% for women), notably higher than the rates recorded in medical records reported in the North California study (4.7% and 1.1% respectively, though ORs were not dissimilar)9 likely reflecting the different study methodologies.

On the discussion section, fifth sentence of the second paragraph reads:

A high rate of intellectual disabilities in children with autism has been described previously; we have now quantified the extent of this—29% (25%–32% depending on age group; 22%–35% for men and 31%–42% for women)—in a much larger study of adults.

It should read:

A high rate of intellectual disabilities in children with autism has been described previously; we have now quantified the extent of this—29% (23%–35% depending on age group; 22%–35% for men and 31%–42% for women)—in a much larger study of adults.

On the discussion section, sixth sentence of the second paragraph reads:

There are few other studies with which we can draw comparisons, and we identified none on physical disability in adults with autism with which we could compare the high rate of 24% for all adults aged 25+ (range 15%–45% depending on age group; 14%–42% for men and 24%–45% for women).

It should read:

There are few other studies with which we can draw comparisons, and we identified none on physical disability in adults with autism with which we could compare the high rate of 24% for all adults aged 25+ (range 15%–45% depending on age group; 14%–42% for men and 24%–48% for women).

In table 1, the total number for the gender of 25-34 years is 667 327 instead of 632 488.

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Paper VI (Full reference and link to the manuscript below)

Rydzewska E, Hughes-McCormack LA, Gillberg C, Henderson A, MacIntyre C, Rintoul J, Cooper SA. Prevalence of sensory impairments, physical and intellectual disabilities, and mental health in children and young people with self/proxy-reported autism: Observational study of a whole country population. Autism. 2019 Jul;23(5):1201-1209. doi: 10.1177/1362361318791279. Epub 2018 Oct 17. PMID: 30328695.

Prevalence of sensory impairments, physical and intellectual disabilities, and mental health in children and young people with self/proxy-reported autism: Observational study of a whole country population - PubMed (nih.gov)

Paper VII

Open access Research

BMJ Open

Relative influence of intellectual disabilities and autism on mental and general health in Scotland: a cross-sectional study of a whole country of 5.3 million children and adults

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ABSTRACT

Objectives To determine the relative extent that autism and intellectual disabilities are independently associated with poor mental and general health, in children and adults.

Design Cross-sectional study. For Scotland's population, logistic regressions investigated odds of intellectual disabilities and autism predicting mental health conditions, and poor general health, adjusted for age and gender.

Participants 1 548 819 children/youth aged 0-24 years, and 3 746 584 adults aged more than 25 years, of whom 9396/1 548 819 children/youth had intellectual disabilities (0.6%), 25 063/1 548 819 children/youth had autism (1.6%); and 16 953/3 746 584 adults had intellectual disabilities (0.5%), 6649/3 746 584 adults had autism (0.2%). These figures are based on self-report.

Main outcome measures Self-reported general health status and mental health.

Results In children/youth, intellectual disabilities (OR 7.04, 95% CI 6.30 to 7.87) and autism (OR 25.08, 95% CI 23.08 to 27.32) both independently predicted mental health conditions. In adults, intellectual disabilities (OR 3.50, 95% CI 3.20 to 3.84) and autism (OR 5.30, 95% CI 4.80 to 5.85) both independently predicted mental health conditions. In children/youth, intellectual disabilities (OR 18.34, 95% CI 17.17 to 19.58) and autism (OR 8.40, 95% CI 8.02 to 8.80) both independently predicted poor general health. In adults, intellectual disabilities (OR 7.54, 95% CI 7.02 to 8.10) and autism (OR 4.46, 95% CI 4.06 to 4.89) both independently predicted poor general health.

Conclusions Both intellectual disabilities and autism independently predict poor health, intellectual disabilities more so for general health and autism more so for mental health. Intellectual disabilities and autism are not uncommon, and due to their associated poor health, sufficient services/supports are needed. This is not just due to coexistence of these conditions or just to having intellectual disabilities, as the population with autism is independently associated with substantial health inequalities compared with the general population, across the entire life course.

Strengths and limitations of this study

- This large study is the first study to have reported on the extent to which autism and intellectual disabilities are independently associated with poor mental and general health, in children and adults.
- The study comprises a whole country population, with high participation rate (94%), and the conditions were systematically enquired about on everyone.
- A limitation is that conditions were self/proxy reports rather than in-depth diagnostic assessments.

INTRODUCTION

Both intellectual disabilities and autism occur not uncommonly in children and adults, and can co-occur. Children and adults with intellectual disabilities have notably poorer mental and general health than other people.[1–4] This has also been reported for autistic children and adults,[5–9] although the quantity of research is limited, particularly with regard to adults. The extent of co-occurrence of intellectual disabilities in autistic people used to be considered to be as high as 50%–70%,[10] although more recent reports suggest that it may be lower, though still considerable, at about 20%.[9 11] This may in part relate to the broadening of criteria for the autism spectrum to include 'milder' autism and greater awareness about autism in children and young people in recent years, as it is well established that autism prevalence is higher in people with more severe intellectual disabilities and vice versa.[12] Autism is generally considered to be associated with poor mental health. However, the largest study to examine this in adults in a general community population found no difference in rates of mental ill health in adults with co-occurring autism and

intellectual disabilities, compared with age-gender-Down syndrome level of ability-matched adults with intellectual disabilities but no autism.[13] As intellectual disabilities and autism have tended to be studied separately, the relative extent to which being autistic, or having intellectual disabilities, accounts for their poor population health is not clear. This is important to understand, given the frequent co-occurrence of these conditions, and is important to understand in both child and adult populations, given the more recent change in co-occurrence due to higher frequency of diagnosis of autism.

The aim of this paper is to study the extent to which autism and intellectual disabilities are independently associated with poor mental and general health, in children and adults.

METHOD

Strengthening the Reporting of Observational Studies in Epidemiology guidelines

The Strengthening the Reporting of Observational Studies in Epidemiology checklist for cross-sectional studies was adhered to.

Census process and variables

Scotland has performed a national Census every 10 years since 1841, the most recent being Scotland's Census, 2011. Information was collected on every resident in Scotland on the Census date, 27 March 2011. This included people in private households and also people in community residences (such as care homes, prisons and student halls of residence). In private households (typically family households), one person was responsible for completing the Census details for all the household's residents; for communal establishments, the manager was responsible for providing the information. It is a legal requirement in the UK to complete the Census. Failure to provide information or for providing false information attracted a fine of up to £1000. Non-responses were followed up by the Census team and help provided. These factors accounted for the high response rate; Scotland's Census 2011 achieved a 94% response rate.[14] The Census team adjusted for the 6% non-response rate using a Census Coverage Survey to estimate numbers and characteristics. The Census Coverage Survey included around 40 000 households; the records from it were matched with Census records, with all individuals deterministically matched to check for duplicates. Individuals estimated to be missing from the Census were then imputed, using a subset of characteristics from real individuals, including health information. This edit and imputation methodology was adapted from the Office for National Statistics rigorous and systematic guidelines, available at: http://webarchive.nationalarchives.gov.uk/20160108193745/http://www.ons.gov.uk/ons/guide-method/method-quality/survey-methodology-bulletin/smb-69/index.html and further details on the Census population estimates are available at: http://www.scotlandscensus.gov.uk/documents/censusresults/release1b/rel1bmethodology.pdf

Full details of the methodology and other background information on Scotland's Census, 2011 are available at: http://www.scotlandscensus.gov.uk/supporting-information.

The Census included questions on demography, long-term conditions and on general health.

The question on long-term conditions enquired:

'Do you have any of the following conditions, which have lasted, or are expected to last, at least 12 months? Tick all that apply:

- Deafness or partial hearing loss.
- Blindness or partial sight loss.
- Learning disability (eg, Down's syndrome).
- Learning difficulty (eg, dyslexia)
- Developmental disorder (eg, autistic spectrum disorder or Asperger's syndrome).
- Physical disability.
- Mental health condition.
- Long-term illness, disease or condition.
- Other condition, please write in
  - free-text space was then provided for conditions to be listed.
- No condition'.

The question on general health enquired:

'How is your health in general?

- Very good.
- Good.
- Fair.
- Bad.
- Very bad'.

The terminology used in both these questions was specifically investigated prior to implementation of data collection. The General Register Office for Scotland commissioned Ipsos MORI Scotland to undertake cognitive question testing, to determine whether the questions were answered accurately and willingly by respondents, and what changes if any might be required to improve data quality and/or the acceptability of the response options. Cognitive interviewing is a widely used approach to critically evaluate survey questionnaires.[15] It tests the way respondents understand, mentally process and respond to survey materials. It enables researchers to modify survey material to enhance clarity. Retrospective probing was deemed to be the most appropriate of the different techniques available. It involved the interviewer presenting the question, the respondent answering it, and the interviewer then probing for specific information relevant to the question or to the specific answer given (eg, What does this question mean in your own words?). This research was undertaken with 102 participants with a mix of gender and age, both with and without the health conditions and disabilities (including people with more than one of the conditions). This included people with autism, intellectual disabilities, dyslexia, dyspraxia, speech impairment, mental health conditions (both milder and more serious) and other long-term conditions. The

results found that the question on general health status functioned well and did not need amendment, as did the questions on long-term conditions, including intellectual disabilities and mental health condition, while the question on autism was redesigned to that listed above in order to more accurately capture the data specifically on autism. Additionally, the response 'no' was amended to 'no condition'. The other questions did not require any modification. Further information can be found at: http://www.scotlandscensus.gov.uk/documents/research/2011-census-health-disability-questions.pdf http://www.scotlandscensus.gov.uk/documents/legislation/changes-to-gov-statement-report.pdf

In Scotland, the term 'learning disability' is synonymous with the international term 'intellectual disabilities'.[16 17]

For 2.6% of the Census returns, information on long-term conditions was not completed. The Census team assumed the most plausible explanation was that the person had no long-term condition but did not see the 'No condition' check box at the end of the question. They, thus, recorded them to have none of the long-term conditions.

Data analysis

First, frequency data were generated. Next, we used logistic regressions to calculate the ORs with 95% CIs of autism, intellectual disabilities, age and gender in predicting (1) having a mental health condition and (2) poor general health. We dichotomised the general health status variable to good health (very good or good health) or poor health (fair, bad, or very bad health). The gender variable was binary, the reference group was male. We conducted the analyses separately for children and young people (aged 0–24 years) and adults (aged 25+ years). This was because in Scotland's Census, 2011, the prevalence of autism is higher in the children and young people than in the adults, most likely due to widening out of the diagnostic criteria and greater awareness of autism in recent decades. Hence the adults with autism are more likely to be on the more severely affected range of the autism spectrum. For the children and young people, the reference group was aged 0–15 years (childhood), given the physiological changes and changing life experiences that occur in adolescence/transition compared with younger children, which may have a bearing on general and mental health. The adults were grouped into 10-year age bands, with the reference group being aged 25–34 years. We then conducted a second round of the regressions, including the interaction terms age x intellectual disabilities and age x autism. This was because the influence of age on mental health and general health is likely to differ in people with intellectual disabilities and possibly in people with autism to that seen in other people. All analyses were conducted with SPSS software V.22.

Patient and public involvement

The question on intellectual disabilities and autism was included in Scotland's Census, 2011 at the behest of third sector organisations for people with intellectual disabilities and autism. This study was undertaken by the Scottish Learning Disabilities Observatory, which has a specific remit for people with intellectual disabilities and autism; its steering group includes partners from the third sector organisations. Results from this study will be disseminated for people with intellectual disabilities and autism in easy-read version via the Scottish Learning Disabilities Observatory website and newsletters.

RESULTS

Scotland's Census, 2011, includes records on 5 295 403 people aged more than 0–75 years, of whom 1 548 819 (29.2%) were children and young people, and 3 746 584 (70.8%) were adults aged 25 years and over. Of the children and young people, 9396 (0.6%) reported having intellectual disabilities and 25 063 (1.6%) reported having autism. Of the adults aged 25 years and over, 16 953 (0.5%) reported having intellectual disabilities and 6649 (0.2%) reported having autism. Of the children and young people with intellectual disabilities, 3756/9396 (40.0%) additionally had autism, and of the adults aged 25 years and over with intellectual disabilities, 1953/16 953 (11.5%) additionally had autism. Of the children and young people with autism, 3756/25 063 (15.0%) additionally had intellectual disabilities, and of the adults aged 25 years and over with autism, 1953/6649 (29.4%) additionally had intellectual disabilities.

538/5640 (9.5%) of the children and young people with intellectual disabilities but no autism had a mental health condition, and 3383/15 000 (22.6%) of the adults with intellectual disabilities but no autism had a mental health condition. A total of 1601/21 307 (7.5%) of the children and young people with autism but no intellectual disabilities had a mental health condition, and 1314/4696 (28.0%) of adults with autism but no intellectual disabilities had a mental health condition. A total of 15 829/1 518 116 (1.0%) of the children and young people with neither condition had a mental health condition, and 208 493/3 724 935 (5.6%) of the adults with neither condition had a mental health condition.

Table 1 presents the OR (95% CI) of intellectual disabilities, autism, age and gender in predicting a mental health condition in the children and young people. It presents the results of two regressions, the second one including the interaction terms. Both intellectual disabilities (OR 7.0, 95% CI 6.3 to 7.9)and autism (OR 25.1, 95% CI 23.0 to 27.3) independently increased the odds of having a mental health condition, more so for autism. Mental health conditions were also predicted by female gender (OR 1.5, 95% CI 1.4 to 1.5) and being a young person rather than a child (OR 10.5, 95% CI 10.1 to 11.0).

In adults (table 2), a similar pattern was seen with both intellectual disabilities (OR 3.5, 95% CI 3.2 to 3.8) and autism (OR 5.3, 95% CI 4.8 to 5.9) independently predicting a mental health condition, as did female gender (OR 1.3, 95% CI 1.2 to 1.3). All age groups had

Table 1 Predictors of mental health conditions in the whole population of children and young people

Variable		Regression 1 OR	Regression 1 95% CI	Regression 2 (including interaction terms) OR	Regression 2 (including interaction terms) 95% CI
Age	0–15 (ref)	–		–	
	16–24	7.65	7.36 to 7.95	10.54	10.06 to 11.05
Gender	Male (ref)	–		–	
	Female	1.49	1.45 to 1.54	1.48	1.44 to 1.53
Autism	No autism (ref)	–		–	
	Autism	10.21	9.67 to 10.78	25.08	23.02 to 27.32
Intellectual disabilities	No intellectual disabilities (ref)	–		–	
	Intellectual disabilities	5.85	5.44 to 6.29	7.04	6.30 to 7.87
Age x intellectual disabilities	0–15 (ref)	–		–	
	16–24	–		0.66	0.57 to 0.76
Age x autism	0–15 (ref)	–		–	
	16–24	–		0.24	0.21 to 0.26
Constant		0.00		0.00	

higher odds ratios than the 25–34 years of predicting having a mental health condition, except for the oldest age group, aged 65+ years who had a lower rate.

A total of 2453/5640 (43.5%) of children and young people with intellectual disabilities but no autism, and 7834/15 000 (52.2%) of adults with intellectual disabilities

Table 2 Predictors of mental health conditions in the whole population of adults

Variable		Regression 1 OR	Regression 1 95% CI	Regression 2 (including interaction terms) OR	Regression 2 (including interaction terms) 95% CI
Age	25–34 (ref)	–		–	
	35–44	1.40	1.38 to 1.42	1.40	1.38 to 1.42
	45–54	1.38	1.36 to 1.40	1.38	1.36 to 1.40
	55–64	1.08	1.06 to 1.09	1.07	1.05 to 1.08
	65+	0.92	0.90 to 0.93	0.91	0.90 to 0.92
Gender	Male (ref)	–		–	
	Female	1.25	1.24 to 1.26	1.25	1.24 to 1.26
Autism	No autism (ref)	–		–	
	Autism (ref)	5.29	5.00 to 5.59	5.30	4.80 to 5.85
Intellectual disabilities	No intellectual disabilities (ref)			–	
	Intellectual disabilities	4.42	4.26 to 4.59	3.50	3.20 to 3.84
Age x intellectual disabilities	25–34 (ref)	–		–	
	35–44	–		0.99	0.87 to 1.11
	45–54	–		1.24	1.10 to 1.39
	55–64	–		1.71	1.51 to 1.94
	65+	–		1.82	1.59 to 2.08
Age x autism	25–34 (ref)	–		–	
	35–44	–		1.03	0.89 to 1.19
	45–54	–		0.94	0.80 to 1.10
	55–64	–		1.02	0.84 to 1.25
	65+	–		1.18	0.97 to 1.44
Constant		0.46		0.46	

Table 3 Predictors of poor general health in the whole population of children and young people

Variable		Regression 1 OR	Regression 1 95% CI	Regression 2 (including interaction terms) OR	Regression 2 (including interaction terms) 95% CI
Age	0–15 (ref)	–		–	
	16–24	2.14	2.10 to 2.18	2.28	2.24 to 2.33
Gender	Male (ref)	–		–	
	Female	1.11	1.09 to 1.14	1.11	1.09 to 1.13
Autism	No autism (ref)	–		–	
	Autism	6.70	6.46 to 6.95	8.40	8.02 to 8.80
Intellectual disabilities	No intellectual disabilities (ref)	–		–	
	Intellectual disabilities	14.05	13.39 to 14.73	18.34	17.17 to 19.58
Age x intellectual disabilities	0–15 (ref)	–		–	
	16–24	–		0.57	0.52 to 0.63
Age x autism	0–15 (ref)	–		–	
	16–24	–		0.54	0.50 to 0.58
Constant		0.02		0.02	

but no autism had poor general health. A total of 3898/21 307 (18.3%) of children and young people with autism but no intellectual disabilities, and 2 134/4 696 (45.4%) of adults with autism but no intellectual disabilities had poor general health. A total of 42 713/1 518 116 (2.8%) of the children and young people with neither condition, and 880 044/3 724 935 (23.6%) of the adults with neither condition had poor general health.

Table 3 presents the OR (95% CI) of intellectual disabilities, autism, age and gender in predicting poor general health in the children and young people. It presents the results of two regressions, the second one including the interaction terms. Both intellectual disabilities (OR 18.3, 95% CI 17.2 to 19.6) and autism (OR 8.4, 95% CI 8.0 to 8.8) independently increased the odds of having poor general health, more so for intellectual disabilities. Poor general health was also predicted by female gender (OR 1.1, 955 CI 1.1 to 1.1) and being a young person rather than a child (OR 2.3, 95% CI 2.2 to 2.3.

In adults (table 4), a similar pattern was seen with both intellectual disabilities (OR 7.5, 95% CI 7.0 to 8.1 and autism OR 4.5, 95% CI 4.1 to 4.9) independently predicting poor general health, as did female gender (OR 1.1, 95% CI 1.1 to 1.1). A gradient is seen, with older age groups progressively predicting having poor general health.

DISCUSSION

Principal findings and interpretation

This is the largest study to date on this topic, comprising a whole country population. Our findings have demonstrated that both intellectual disabilities and autism are associated with having a mental health condition and with poor general health. This is so in both children/young people and in adults, after the overlap between these two conditions (intellectual disabilities and autism) is accounted for. For mental health conditions, this is particularly so for autism (OR 25.1, 95% CI 23.0 to 27.3 for children/young people; OR 5.3, 95% CI 4.8 to 5.9 for adults). For poor general health, this is particularly so for intellectual disabilities (OR18.3, 95% CI 17.2 to 19.6 for children/young people; OR 7.5, 95% CI 7.0 to 8.1 for adults). Previous literature on this is limited, and has not taken account of the overlap between autism and intellectual disabilities. It is of particular note that autism contributes to poor general health and especially to having a mental health condition even after taking account of the contribution of intellectual disabilities. The mental health conditions did not include transient common mental disorders, as the question referred to mental health conditions lasting or expected to last at least 12 months, that is, severe mental health conditions.

The extent of mental and general health inequality experienced by the population with intellectual disabilities and the population with autism, in comparison with the general population, is greatest in children/young people than it is for adults, though is substantial at all ages. This reflects that both mental health conditions and poor general health are much more common in adults than children and young people in the general population, while they are common at all ages in people with autism and in people with intellectual disabilities. Indeed, in people with intellectual disabilities, those with more severe intellectual disabilities have more comorbidity[18] and die at an earlier age[19] including in childhood. Hence, with increasing age, although acquiring age-related conditions, the population with intellectual disabilities has less disability-related comorbidity and perversely may be healthier than the younger population with intellectual disabilities.

Table 4 Predictors of poor general health in the whole population of adults

Variable		Regression 1 OR	Regression 1 95% CI	Regression 2 (including interaction terms) OR	Regression 2 (including interaction terms) 95% CI
Age	25–34 (ref)	–		–	
	35–44	1.78	1.76 to 1.80	1.79	1.77 to 1.81
	45–54	2.86	2.83 to 2.89	2.90	2.87 to 2.93
	55–64	4.81	4.76 to 4.86	4.88	4.82 to 4.93
	65+	10.25	10.15 to 10.36	10.39	10.29 to 10.50
Gender	Male (ref)	–		–	
	Female	1.05	1.05 to 1.06	1.05	1.05 to 1.06
Autism	No autism (ref)	–		–	
	Autism (ref)	3.39	3.21 to 3.58	4.46	4.06 to 4.89
Intellectual disabilities	No intellectual disabilities (ref)			–	
	Intellectual disabilities	4.39	4.25 to 4.53	7.54	7.02 to 8.10
Age x intellectual disabilities	25–34 (ref)	–		–	
	35–44	–		0.72	0.65 to 0.79
	45–54	–		0.60	0.54 to 0.65
	55–64	–		0.45	0.40 to 0.50
	65+	–		0.24	0.21 to 0.26
Age x autism	25–34 (ref)	–		–	
	35–44	–		0.83	0.72 to 0.96
	45–54	–		0.59	0.50 to 0.68
	55–64	–		0.49	0.41 to 0.59
	65+	–		0.44	0.36 to 0.53
Constant		0.08		0.08	

The aetiology of mental and general ill health in people with intellectual disabilities or people with autism includes genetic predisposition[18]; indeed the term, Early Symptomatic Syndromes Eliciting Neurodevelopmental Clinical Examination, has been coined for the association of problems in one or more of 10 health domains in young children.[19] It is clear though that aetiology is multifactorial, and social and environmental factors such as life events, which occur more commonly in people with intellectual disabilities, have been shown to precede onset of mental health conditions in adults with intellectual disabilities.[20] Therefore, underpinning mechanisms appear to include the interplay between genes, environment and lifestyle[21] including differences in health related behaviours, such as diet and exercise,[22] and inequalities in access to services.[23] This is important, as understanding these factors provides pathways to the development of interventions to improve health. Examples tailored to these populations, in addition to drug treatments, include interventions developed to address lifestyle,[24] general health[25] and psychological interventions for mental health conditions.[26]

Comparison with previous literature

No previous studies have been identified which investigated the extent to which autism and intellectual disabilities are independently associated with poor mental and general health, in children and adults. We believe that this is, therefore, the first study to do so and subsequently we cannot compare these results.

Strengths and limitations

The main strengths of the study are the 94% whole population response, rather than biassed sampling; the large population size of 5.3 million; that the conditions (intellectual disabilities, autism, mental health condition and general health) were systematically enquired about for each person; and that the phrasing of the questions underwent cognitive question testing in advance of the Census to ensure they captured the intended meaning. Consequently, we believe that these results are generalisable to other high-income countries.

Limitations include the use of the term of 'developmental disorders' in the Census. However, the Census form prompted responses only for autistic spectrum disorder or Asperger's syndrome. Furthermore, the developmental disorders category was distinguished from intellectual disabilities, learning difficulties and mental health conditions, which are important distinctions. Hence, we consider that respondents will have replied accordingly, that is, responded regarding autism.

However, we have no means to check this. In addition, conditions were self/proxy reports rather than in-depth diagnostic assessments (which would not be possible on such a large scale). Respondents reported whether or not each person was known to have autism and/or intellectual disabilities, rather than each person having an assessment, so some reporting error is possible. However, intellectual disabilities and autism are conditions that are typically diagnosed during infant/primary school age, if not before. In Scotland these diagnoses attract additional educational support, which is to the child's advantage; once diagnosed these are lifetime diagnoses. Consequently, there may be an undercount in the early years of childhood, whereas reporting of these conditions should be accurate in later childhood, youth and in adults, within the diagnostic criteria prevailing at the time of diagnosis. The proportion of people in the population reported to have autism was lower after age 25. This reflects the broadening of diagnostic criteria and greater awareness of autism in recent years; hence, the older people with autism might have more severe autism than the children/youth reported to have autism. The children/youth with autism are likely to include some who function well. We do not know the extent to which reporting of mental health conditions and general health status would reflect that found in in-depth diagnostic assessments, although subjective general health status is commonly used in population studies, and it is well established as an extremely valid measure of health. There is a strongly predictive linear gradient across subjective health status and subsequent number of medical appointments, hospital admissions and mortality.[27–29] We do not know the proportion who self-reported or for whom the report was by another household reference person (eg, parent). However, the latter is likely to be more common for the people with intellectual disabilities in view of their intellectual disabilities, and for the children and young people. Six per cent of Census entries were imputed. The Census team assumed the 2.6% who did not provide information on long-term conditions did not have any of them, but we are unable to confirm the accuracy of this assumption.

Future research investigating narrower age bands of children/youth may be useful, and next steps must importantly include study of the aetiology of poor health in these populations, to inform the development of further effective interventions.

IMPLICATIONS

Intellectual disabilities and autism are not uncommon, and due to their associated poor mental and general health, services and supports need to be available in sufficient quantity and quality. Our findings demonstrate that this is not just related to the coexistence of these conditions, or just to having intellectual disabilities, as the population with autism is also independently associated with substantial health inequalities compared with the general population, across the entire life course.

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Open access Original research

BMJ Open

Birth incidence, deaths and hospitalisations of children and young people with Down syndrome, 1990–2015: birth cohort study

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ABSTRACT

Objective To investigate current Down syndrome live birth and death rates, and childhood hospitalisations, compared with peers.

Setting General community.

Participants All live births with Down syndrome, 1990–2015, identified via Scottish regional cytogenetic laboratories, each age–sex–neighbourhood deprivation matched with five non-Down syndrome controls. Record linkage to Scotland's hospital admissions and death data.

Primary outcome HRs comparing risk of first hospitalisation (any and emergency), readmission for children with Down syndrome and matched controls were calculated using stratified Cox proportional hazards (PH) model, and length of hospital stay was calculated using a conditional log-linear regression model.

Results 689/1479 (46.6%) female and 769/1479 (51.9%) male children/young people with Down syndrome were identified (1.0/1000 births, with no reduction over time); 1235 were matched. 92/1235 (7.4%) died during the period, 18.5 times more than controls. More of the Down syndrome group had at least one admission (incidence rate ratio(IRR) 72.89 (68.72–77.32) vs 40.51 (39.15–41.92); adjusted HR=1.84 (1.68, 2.01)) and readmissions (IRR 54.85 (51.46–58.46) vs 15.06 (14.36–15.80); adjusted HR=2.56 (2.08, 3.14)). More of their admissions were emergencies (IRR 56.78 (53.13–60.72) vs 28.88 (27.73–30.07); first emergency admission adjusted HR=2.87 (2.61, 3.15)). Children with Down syndrome had 28% longer first admission after birth. Admission rate increased from 1990–2003 to 2004–2014 for the Down syndrome group (from 90.7% to 92.2%) and decreased for controls (from 63.3% to 44.8%).

Conclusions We provide contemporaneous statistics on the live birth rate of babies with Down syndrome, and their childhood death rate. They require more hospital admissions, readmissions emergency admissions and longer lengths of stays than their peers, which has received scant research attention in the past. This demonstrates the importance of statutory planning as well as informal support to families to avoid added problems in child development and family bonding over and above that brought by the intellectual disabilities associated with Down syndrome.

Strengths and limitations of this study

- A whole country population-based study on Down syndrome, with comprehensive identification via all NHS Scottish Regional Genetics Centres.
- 25-Year period of linked data.
- Account taken of the potential confounders of age, sex and extent of neighbourhood deprivation; underlying conditions were not investigated.
- Some of the Down syndrome individuals could not be matched, mostly due to missing data on postcode, and so were omitted from analyses.
- The use of routinely collected hospital and death statistics has many advantages including large-scale coverage, but there is likely to be a degree of coding inaccuracy which is not quantified.

INTRODUCTION

Improvements in health and social care and attitudes to disability have increased the survival of people with Down syndrome in recent decades.[1–6] In England and Wales, it was estimated that there were 37090 people with Down syndrome in 2011,[7] but exact numbers are not reported. The National Down Syndrome Cytogenetic Register for England and Wales produces data on the estimated live birth rate for Down syndrome, using assumptions on termination rates for those receiving a prenatal diagnosis. In 2013, it reported an estimated live birth rate of 1.0/1000.[8] In the USA, de Graaf *et al*[9] estimated live birth and population prevalence for Down syndrome for all nine states in which data were available. They reported a live birth prevalence rate of 12.1/10 000, and an increase in the number of people living with Down syndrome from 1950 to 2010. In Canada, the live birth rate for Down syndrome was reported to have remained stable between 2005 and 2013, and higher than in the UK and USA rates, at approximately 13.5/10 000.[10] Similar data are

not available for Scotland. Given the changing population demographic for people with Down syndrome, and limited evidence base, we require a better understanding of their live birth rate and future health service needs.[4]

Down syndrome is associated with a range of congenital anomalies, particularly cardiac, and a higher risk for respiratory, immunological, endocrine and gastrointestinal conditions.[4 11] This phenotype puts people with Down syndrome at risk of hospitalisation. This may impact on their development and family relationships, and so is an added disadvantage on top of their intellectual disabilities. Despite this, few previous studies have investigated hospitalisation of children and young people with Down syndrome, compared with the general population. Fitzgerald *et al*[12] studied hospital admissions of 405 children with Down syndrome born in Western Australia between 1983 and 2004. They found that the children with Down syndrome were hospitalised five times more often when compared with previously published general population data for the single year of 1995. They had no general population comparison data for length of stay. Zhu *et al*[13] investigated hospitalisations of 3212 children and adults with Down syndrome compared with 67 204 children and adults without Down syndrome in Denmark, between 1977 and 2008. The people with Down syndrome had more than twice the rate of hospital admissions, and nearly three times as many bed days. Both studies found that length of admissions of the people with Down syndrome had reduced over the periods studied, and Zhu *et al*[13] reported more bed days for males, children under 5 years, and those with congenital heart anomalies. Other studies have lacked general population comparison data or been limited to single conditions. We are not aware of any large-scale studies specifically of children and young people with Down syndrome investigating these issues compared with their peers. There has been very little other investigation of the factors that influence length of hospital stay of children and young people with Down syndrome, and we located no studies on risk of readmission or on emergency admissions of children and young people.

Without accurate information on live birth and death rates, and hospital admission needs, it is not possible to plan for the healthcare support that children with Down syndrome and their families need.

This study's aims were to investigate the (1) incidence of live births of people with Down syndrome over a 25-year period, (2) frequency of deaths of children and young people with Down syndrome compared with matched controls and (3) hospital admission frequency and duration, emergency admissions and readmissions of children and young people with Down syndrome compared with matched controls.

METHODS

Written informed consent was not obtained from participants for their clinical records to be used in this study, but patient records were anonymised and de-identified prior to analysis. Safehaven approval was granted to approved researchers to analyse the data.

Patient and public involvement

This study was undertaken by the Scottish Learning Disabilities Observatory, which has a specific remit for people with intellectual disabilities, including people with Down syndrome; its steering group includes partners from third sector organisations, including Down Syndrome Scotland. Results from this study will be disseminated for people with Down syndrome and their families/carers in easy-read version via the Scottish Learning Disabilities Observatory website and newsletters.

Study sample, setting and process

All NHS Scottish Regional Genetics Centres (the east, north, south-east and west Scotland centres) identified all live birth infants screened positive for Down syndrome postnatally (live births with trisomy 21, mosaic trisomy 21 or unbalanced translocation resulting in trisomy 21) between 1 January 1990 and 1 December 2015. In Scotland, everyone is given a unique Community Health Index (CHI) at birth which is used in their health records. The CHI database is held centrally by National Services Scotland (NSS), and was used to identify five controls without Down syndrome for each person with Down syndrome, matched on sex, age (by month and year of birth) and neighbourhood deprivation (using the Scottish Index of Multiple Deprivation (SIMD)).[14 15] Down syndrome infants who could not be matched due to missing data or had less than 1 year of potential follow-up time (ie, births in 2015) were excluded from statisitcal analyses. The CHI also provides a means to record link each person identified with Down syndrome and their matched controls to routinely collected hospital admissions data (Scottish Morbidity Records 01: SMR01),[16] and National Records of Scotland death certificate data.[17] The end of the period of follow-up was 1 December 2015. However, children born in 2015 were excluded from the statistical analyses to allow a minimum potential follow-up time of 1 year for all children.

Data sources and definitions

Live births

Live births of babies with Down syndrome from all NHS Scottish Regional Genetics Centres, 1990–2015; Scottish live births in the whole population, from the National Records of Scotland births time series, 1990–2015 (NRS, 2019).[17]

Death

Deaths and causes of death by the International Classification of Diseases codes[18] according to death certificates registered at National Records of Scotland.

Hospital admissions

SMR01 contains episode-based records for all non-psychiatric, non-obstetric acute hospital admissions in

Scotland. The information collected includes the date of admission to hospital and discharge from it and whether the admission was routine or emergency. In SMR01, continuous periods of care are accounted for with a continuous inpatient stay (CIS) marker. This CIS marker ensures that a series of individual episodes over an unbroken period of care (eg, transfers between wards or hospitals) can be identified and treated as one admission rather than several admissions. Transfer of a baby to a neonatal intensive care unit after birth is counted as a first admission. Data quality assurance assessments for SMR01 are performed periodically.[19 20]

Admission type

Admission type is coded as emergency or routine. 'Emergency' admissions are those which were unplanned in advance; planned admissions are 'routine' admissions.

Birth year group

Grouped into the most recent cohort (2004–2014) and the least recent cohort (1990–2003).

Scottish Index of Multiple Deprivation

SIMD matching with controls and reporting is by quintiles, where SIMD 1 is the most deprived neighbourhoods and SIMD 5 is the most affluent neighbourhoods. SIMD is calculated at datazone level, identified from postcodes.

Discharge type

Discharge type is coded as a regular discharge or irregular discharge. Irregular discharges include, for example, a patient discharging himself/herself against medical advice, or death.

All follow-up/censoring

Children were followed up from birth, and all models were censored on death or 1 December 2015 (whichever came first) unless stated otherwise.

Analyses

Incidence of Down syndrome

Incidence of Down syndrome births was calculated for each calendar year from 1990 to 2015 inclusive.

Death

Incidence rates for mortality per person time (per 1000) were described, split by age group (for all ages, 0–1 month of life, from 1 to 12 months, from 13 to 60 months, from 61 to 120 months, from 121 to 180 months and from 181 to 240 months), as were underlying causes of death, for people with Down syndrome and their matched controls.

First hospital admissions

Incidence rates for first admissions per person time (per 1000) for people with Down syndrome compared with controls were described, split by age group (for all ages, 0–1 month, from 1 to 12 months and from 13+ months). Descriptive data for first admissions were presented including by sex, SIMD, year of birth and duration of first admissions in days, and this was compared using dependent t-tests for continuous variables and χ^2 tests for categorical variables or Mann-Whitney U tests for length of hospital stay. The relative risk of first hospital admission was compared for people with Down syndrome and controls using stratified (by sex, birth year group, SIMD) Cox regression models (log–log plots confirmed the proportional hazards assumption was met). The follow-up period was defined as from date of birth until date of first admission, and admission type was entered as a potential confounder.

Risk of emergency hospital admission

The relative risk of emergency first hospital admission was compared for people with Down syndrome and controls, analysed using stratified (by sex, birth year group, SIMD) Cox regression models. The follow-up period was defined as from date of birth until date of first admission.

Duration of hospital admission

Conditional (stratified by sex, birth year group, SIMD) linear regression was used to model the duration of first hospital admission (log transformed to help negate potential skewing of results) as the outcome, comparing Down syndrome and control groups. Admission type was entered as a potential confounder.

Risk of readmission

The relative risk of readmission (the next admission after the first admission) was compared for people with Down syndrome and controls, analysed using stratified (by sex, birth year group, SIMD) Cox regression models. The follow-up period was defined as date of discharge from the first admission until date of the next admission. First admission type and discharge type from first admission were entered as potential confounders, and the interaction of group (Down syndrome vs controls)*admission type (emergency vs routine) was included.

Reference groups for all regression analyses were: not having Down syndrome, females, most affluent neighbourhoods, routine admissions and regular type of hospital discharges. All analyses were conducted with IBM SPSS V.22.

RESULTS

Between 1 January 1990 and 1 December 2015, the Scottish Regional Genetics Centres identified 1479 live births with Down syndrome. One thousand two hundred and thirty-five people were matched with 6175 controls. One hundred eighty-seven (12.6%) infants with Down syndrome were excluded from statistical analyses, as they could not be matched due to missing data on SIMD quintile (n=160) in view of missing postcode information, sex (n=21), month and year of birth (n=<5) and CHI for example, due to migration out of Scotland (n=<5). A further 57 children were excluded as they were born in 2015 (and had less than 1 year of potential follow-up time). Of the 1479 infants born with Down syndrome, 689

Table 1 Incidence of Down syndrome by year of birth

Year of birth	Number of births registered in Scotland (all people) n	Down syndrome n (%)
1990	65973	60 (0.09)
1991	67024	66 (0.10)
1992	65789	38 (0.06)
1993	63337	35 (0.06)
1994	61656	43 (0.07)
1995	60051	46 (0.08)
1996	59296	40 (0.07)
1997	59440	72 (0.12)
1998	57319	54 (0.09)
1999	55147	52 (0.09)
2000	53076	60 (0.11)
2001	52527	61 (0.12)
2002	51270	44 (0.09)
2003	52432	58 (0.11)
2004	53957	79 (0.15)
2005	54386	63 (0.12)
2006	55690	44 (0.08)
2007	57781	70 (0.12)
2008	60041	63 (0.10)
2009	59046	73 (0.12)
2010	58791	59 (0.10)
2011	58590	63 (0.11)
2012	58027	59 (0.10)
2013	56014	58 (0.10)
2014	56725	62 (0.11)
2015	55098	57 (0.11)*
Total	1508483	1479 (0.10)

*In 2015, the Down syndrome total is for 11 months only, so the % in 2015 is adjusted to 11/12 months in the general population.

(46.6%) were girls, 769 (51.9%) were boys and sex was not recorded for 21 (1.4%). The proportion of births for the Down syndrome population by the five SIMD quintiles were: 1=21.3%, 2=20.4%, 3=18.5%, 4=20.1% and 5=19.7%, and SIMD data were missing for 160 (10.8%). Of the 1235 infants with Down syndrome with matched controls, 591 (47.9%) were girls and 644 (52.1%) were boys. The proportion of births for the Down syndrome population, by the five SIMD quintiles were: 1=21.0%, 2=20.0%, 3=18.2%, 4=19.8% and 5=19.9%. Hence they appear to be representative of all the Down syndrome infants that were born.

Incidence of Down syndrome live births over time

Given the total number of infants born was 1 508 483, the incidence of Down syndrome live births was 1479/1 508 483 (1.0/1000 births). Table 1 and figure 1 show the incidence of Down syndrome live births for each year of the study period (with a moving average smoothed trend line to account for noise from year on year variation). The birth rate in Scotland has fallen since the early 1990s, while the incidence of Down syndrome shows some year-to-year variation and appears to have remained the same or risen since the early 1990s.

Deaths

Of the 1235 people with Down syndrome with matched controls, 92 (7.4%) died during the study period (165 744 person months of follow-up for the Down syndrome group), of whom 47 (51.0%) were women and 45 (48.9%) were men. Of the 6175 matched controls, 23 (0.4%) died during the period (895 776 person months follow-up for the controls), of whom 9 (39.1%) were women and 14 (60.9%) were men. Death was therefore 18.5 times more common in the Down syndrome group. Most of the deaths of the people with Down syndrome occurred in infancy. Death incidence rates per person time (per 1000) by age groups for the Down syndrome population versus controls were 0.56 (95% CI 0.45 to 0.68) (n=92) vs 0.03 (95% CI 0.02 to 0.04) (n=23) for all ages, 17.81 (95% CI 9.87 to 32.17) (n=11) vs 0 for the first month of life, 3.87 (95% CI 2.96 to 5.07) (n=53) vs 0.17 (95% CI 0.09 to 0.29) (n=12) from 1 to 12 months, 0.36 (95% CI 2.96 to 5.07) (n=18) vs 0.02 (95% CI 0.01 to 0.05) (n=6) from 13 to 60 months, 0.15 (95% CI 0.07 to 0.32) (n=7) vs 0.01 (95% CI 0.00 to 0.04) (n=<5) from 61 to 120 months, 0.09 (95% CI 0.03 to 0.31) (n=<5) vs 0 from 121 to 180 months and 0 vs 0.02 (95% CI 0.00 to 0.08) (n=<5) from 181 to 240 months. The most common certified underlying causes of death for the Down syndrome group were congenital heart anomalies (n=33, 34.4%), Down syndrome (n=26, 27.1%) and infection (excludes respiratory infections) (n=11, 11.5%). This pattern was different compared with controls in whom causes of deaths were due to a range of other factors not found for people with Down syndrome (n=8, 34.8%), prematurity/perinatal causes (n=5, 21.7%) and congenital heart anomalies (n=5, 21.7%). Although less common than some other causes, leukaemia was a cause of death among people with Down syndrome (n=<5) compared with controls where no deaths were from leukaemia.

Admissions

Table 2 presents details on the first all-cause hospital admissions for the two groups. One thousand one hundred and five people out of 1235 (89.5%) of the Down syndrome group had at least one hospital admission compared with 3305/6175 (53.5%) of the control group. Admission incidence rates per person time (per 1000) by age groups for the Down syndrome population versus controls were 72.89 (95% CI 68.72 to 77.32) (n=1105) vs 40.51 (95% CI 39.15 to 41.92) (n=3305) for all ages, 38.87 (95% CI 26.05 to 57.99) (n=24) vs 13.29 (95% CI 9.79 to 18.06) (n=41) for the first month of life, 56.44 (95% CI 52.03 to

Figure 1 Incidence of Down syndrome births by year in Scotland from 1990 to 2015.

61.23) (n=580) vs 24.94 (95% CI 23.62 to 26.33) (n=1298) from 1 to 12 months, 117.47 (95% CI 107.62 to 128.22) (n=501) vs 74.55 (95% CI 71.32 to 77.92) (n=1966) from 13+ months. Compared with the control group, the Down syndrome group were younger at the time of first hospital admission (median=0 months (IQR 0–300 months) vs 24 months (IQR 0–288)), had less admissions among females, were more equally spread across neighbourhoods while the control group had a gradient across SIMD groups with those in the most deprived areas having more hospital admissions, and had longer duration of admissions (5.03 days (median=2.00; IQR=1.00–4.00), vs 1.78 days (median=1; IQR=1.00–1.00)) by 3.25 days. The rate of admissions slightly increased over time for people with Down syndrome (from 528, 90.7% (in 1990–2003) to 577, 92.2% (in 2004–2014)) and decreased for controls (from 1837, 63.3% (in 1990–2003) to 1468, 44.8% (in 2004–2014)). The gap between the higher rate of admissions has widened over time for people with Down syndrome compared with controls.

Emergency admission incidence rates per person time (per 1000) by age groups for the Down syndrome population versus controls were 56.78 (95% CI 53.13 to 60.72) (n=861) vs 28.88 (95% CI 27.73 to 30.07) (n=2352) for all ages, 27.53 (95% CI 17.12 to 44.23) (n=17) vs 11.03 (95% CI 7.88 to 15.43) (n=34) for the first month of life, 48.66 (95% CI 44.57 to 53.11) (n=500) vs 20.42 (95% CI 19.23 to 21.69) (n=1063) from 1 to 12 months, 80.66 (95% CI 72.57 to 89.65) (n=344) vs 47.71 (95% CI 45.14 to 50.42) (n=1255) from 13+ months. Table 3 displays the results

Table 2 First all-cause hospital admissions characteristics

	Down syndrome (n=1105)	Control group (n=3305)	
	n(%), Median (IQR)	n(%), Median (IQR)	P value
Age at admission (months)	0 (IQR=0–300)	24 (IQR=0–288)	<0.001
Sex			
Male	572 (51.8%)	1840 (55.7%)	<0.001
Female	533 (48.2%)	1465 (44.3%)	
Birth year group			
Most recent	577 (92.2%)	1468 (44.8%)	<0.001
Less recent	528 (90.7%)	1837 (63.3%)	
SIMD			
Most affluent (5)	217 (19.6%)	570 (17.2%)	<0.001
4	226 (20.4%)	609 (18.4%)	
3	203 (18.4%)	591 (17.9%)	
2	230 (20.8%)	720 (21.8%)	
Most deprived (1)	229 (20.7%)	814 (24.6%)	
Duration of admission (days)	2.00 (IQR=1.00–4.00)	1.00 (IQR=1.00–1.00)	<0.001

Denominator for birth year group most recent/less recent are for Down syndrome n=653/582/control group n=3275/2900.
SIMD, Scottish Index of Multiple Deprivation.

Table 3 Risk for first all cause hospital admission

	Separately entered		Simultaneously entered	
	HR value	95% CI	HR value	95% CI
Group				
No Down syndrome	ref	ref	ref	ref
Down syndrome	2.74†	2.52 to 2.98	1.84†	1.68 to 2.01
First admission type				
Routine	ref	ref	ref	ref
Emergency	1.10*	1.00 to 1.20	1.19†	1.08 to 1.30

Stratified models by sex, birth year group, SIMD.
*P<0.05.
†P<0.001.
SIMD, Scottish Index of Multiple Deprivation.

of the Cox regressions for time to first all-cause hospital admission. The risk of admission was higher among people with Down syndrome compared with controls, with an unadjusted HR=2.74 (95% CI 2.52 to 2.98). This remained after adjusting for type of first admission: HR=1.84 (95% CI 1.68 to 2.01).

Table 4 displays the results of the Cox regressions for time to first emergency hospital admission. The risk of emergency admission was higher among people with Down syndrome compared with controls, with a HR=2.87 (95% CI 2.61 to 3.15).

Duration of hospitalisation

The duration of first hospital admission was significantly longer for the Down syndrome group at 5.03 days (median=2 days) versus 1.78 days (median=1 day) by 3.25 days. After adjustment (with a log transformed time variable, to help account for any data skew) for sex, birth year group, SIMD and type of admission, the Coefficient=0.25 (95% CI 0.22 to 0.28) (table 5). This log transformed coefficient can be interpreted as a ratio of difference for the Down syndrome population compared with the control group (exponentiate of the log transformed coefficient, exp(0.248)=1.28145993219–1×100=28, with a baseline length of stay (intercept) exp(0.12)=1.12749685158). This means the average length of stay was 28% higher in the Down syndrome population, compared with the controls. All exponentiated values are presented as percentages in table 5.

Table 4 Risk for first emergency hospital admission

	Separately entered	
	HR value	95% CI
Group		
No Down syndrome	ref	ref
Down syndrome	2.87*	2.61 to 3.15

Stratified models by sex, birth year group, SIMD.
*P<0.001.

Risk of readmission

Of the Down syndrome group, 945/1105 (85.5%) had at least one readmission, compared with 1685/3305 (51.0%) of the control group. Readmission incidence rates per person time (per 1000) for the Down syndrome population versus controls were 54.85 (95% CI 51.46 to 58.46) (n=945) vs 15.06 (95% CI 14.36 to 15.80) (n=1695). The baseline age at readmission was lower for the Down syndrome group with a median age of 6 months (IQR 1–17) compared with the controls with a median age of 11 months (IQR 5–20; p<0.001). The unadjusted HR of Down syndrome for time to readmission following discharge from first admission was 2.44 (95% CI 2.13 to 2.79). The HR when adjusted for type of admission, type of first discharge and group*emergency interaction was 2.56 (95% CI 2.08 to 3.14). Results are displayed in table 6.

DISCUSSION

Principal findings and interpretation

We found the Scottish incidence of Down syndrome live births to be 1.0/1000 births over the last 25 years, with it being possibly higher now than in the early 1990s. The Scottish birth rate has fallen overall, but incidence of Down syndrome has not. Death rate was 18.5 times higher for the children/young people with Down syndrome than for their controls, and their causes of death differed, including causes that might be expected to have been amenable to good healthcare such as infections.

Over a 25-year period, we found that children/young people with Down syndrome compared with matched controls were considerably more likely to have had at least one hospital admission (IRR of 72.89 compared with 40.51; adjusted HR=1.84) and readmissions (IRR of 54.85 compared with 15.06; adjusted HR=2.56). Their admissions were more likely to have been emergency admissions (IRR of 56.78 compared with 28.88; first emergency admission HR=2.87) and of longer duration 28% longer first admission after birth. Given the higher rate of first emergency admissions in the Down syndrome group, we

Table 5 Duration of first all-cause hospital admission (log transformed time variable)

	Separately entered			Simultaneously entered		
	Coef	95% CI	Exp %	Coef	95% CI	Exp %
Group						
No Down syndrome	ref	ref	27	ref	ref	28
Down syndrome	0.24*	0.21 to 0.27		0.25*	0.22 to 0.28	
First admission type						
Routine	ref	ref	2	ref	ref	4
Emergency	0.02	0.00 to 0.04		0.04*	0.02 to 0.06	

Stratified models by sex, birth year group, SIMD.
*P<0.001.
Exp %, exponentiated values as percentages; Coef, Coefficient.

included the interaction of group×type of first admission (emergency or routine) in the analysis of readmissions, but they still remained more likely to be readmitted. We are not aware of any previous study of readmissions and emergency admissions in this population. The rate of admissions slightly increased over time (from 1990–2003 to 2004–2014) for the children/young people with Down syndrome (from 90.7% to 92.2%), while it decreased for the control group (from 63.3% to 44.8%), perhaps reflecting changes in medical practice over time, to keep children out of hospital as far as possible, but admitting for serious problems which the Down syndrome children may be more likely to experience. These findings are important, as they demonstrate the impact of additional health needs to both children and young people with Down syndrome, and to their families. This demonstrates the importance of statutory as well as informal support to families to avoid added problems in child development over and above the intellectual disabilities that the children with Down syndrome experience, and the impact caring can have on families. Additionally, the high rates of emergency admissions and readmissions may indicate some poorer quality management of healthcare at the primary care level (to avoid admissions), and have not to our knowledge been reported before, though other factors, including underlying health conditions may contribute.

The Down syndrome group were younger at the time of first hospital admission, had a more equal sex distribution for first admission than the control group and an equal distribution across extent of neighbourhood deprivation, while the control group had a gradient with those in the most deprived areas more likely to have had a hospital admission. This suggests that at present, the children with Down syndrome appear to receive comparable care in terms of first admission, regardless of their neighbourhood, and highlights that findings in the general population cannot be relied on when planning services

Table 6 Risk of readmission following discharge from first admission

	Separately entered		Simultaneously entered	
	HR value	95% CI	HR value	95% CI
Group				
No Down syndrome	ref	ref	ref	ref
Down syndrome	2.44†	2.13 to 2.79	2.56†	2.08 to 3.14
First admission type				
Routine	ref	ref	ref	ref
Emergency	0.97*	0.83 to 1.13	1.39	1.05 to 1.84
Discharge type				
Routine	ref	ref	ref	ref
Irregular	0.86	0.83 to 0.89	0.85	0.82 to 0.88
Group* first admission type				
–	–	–	1.06	0.78 to 1.44

Stratified models by sex, birth year group, SIMD.
*P<.05.
†P<0.001.

for people with Down syndrome/intellectual disabilities, such as focussing services in areas of greatest deprivation. Families of children and young people with Down syndrome need focused support regardless of the areas they reside in.

Comparison with previous literature

As well as being 18.5 times more likely to die than their controls, we found that the children/young people with Down syndrome were more likely to be hospitalised, emergency hospitalised, readmitted and for longer durations. We have not identified any previous studies on readmission with which to compare our findings, nor on emergency admissions. For the earlier period of 1977–2008 in Denmark, Zhu *et al*[13] reported that children and adults with Down syndrome had more than twice the rate of hospital admissions (all admissions, not separately reported for first admissions or repeat admissions), similar to our adjusted HRs for first and repeat admissions, but we specifically investigated children and young people unlike that study. The smaller Australian study for the earlier period of 1983–2004 suggested that the hospitalisation rate was five times higher, but relied on previously reported general population data from 1995.[12]

As our study shows, and as previously reported in a study of children with Down syndrome identified from population-based sources in 1997 (n=210) and 2004 (n=208) in Western Australia,[21] it is highly likely that admission rates change over time, so these two studies are not strictly comparable to ours, and we have found no studies contemporaneous with ours with which to draw comparisons. Previously, small-scale studies reported length of admissions of children with Down syndrome to be 4.7 days in a Swedish study of 211 children followed up for at least 17 years,[22] and 7 days specifically for bronchiolitis in a small retrospective case–control study in Singapore,[23] compared with the 5.0 days we found. A further small study of 213 children with Down syndrome aged 0–3 years in America reported high admission rates.[24]

The live birth incidence for Down syndrome we report is similar to that recently reported in England, USA and Canada.[8–10] Antenatal screening changes may underlie the variability in the birth rates found over time. Previous research in Scotland showed that decisions to terminate pregnancy following antenatal diagnosis of aneuploidy had fallen over time,[1] which our results are also possibly suggestive of from the early 1990s to more recently. In contrast, in some other countries (eg, Denmark), following the introduction of antenatal combined screening for Down syndrome in 2004, there was a concurrent increase in prenatal diagnoses and the number of Down syndrome live births decreased suddenly and significantly before then stabilising.[25] A higher proportion of the Scottish population have catholic faith compared with Denmark which might contribute in part to differences between countries: a range of complex factors influence women's decision making regarding prenatal screening and diagnosis of Down syndrome, and it is possible that more societal acceptance of diversity and people with disabilities, or better availability of family support may influence decisions in Scotland over time,[1 26] although this requires further investigation.

Strengths and limitations

This is the first population-based study examining incidence of Down syndrome and healthcare utilisation over time in Scotland compared with the general population, and the first worldwide to investigate emergency hospital admissions and readmissions. It controlled for the potential confounders of age, sex and SIMD. It was not subject to selection bias, as the population identification was based on routine data sources from all genetic service laboratories in Scotland (ie, the whole population). The Cox regression analysis for risk of admissions and readmissions provides advantages over logistic or linear techniques as this includes consideration of time to event (person time at risk). However, there were some missing control data, mostly due to missing postcodes, and this is a limitation of the study. There were no data on ethnicity which is a limitation; the Scottish population is 96% white. The use of routinely collected hospital and death statistics has many advantages including large-scale coverage, but there is likely to be a degree of coding inaccuracy which is not quantified. Children with Down syndrome have higher risk of comorbidities, so may be more likely to be transferred to neonatal intensive care units which might explain the increased risk of first hospital admission rates, but we could not distinguish such admissions in our dataset. We did not aim to investigate underlying conditions which are associated with admissions and death in the two groups.

CONCLUSIONS

As people with Down syndrome are living longer than in the past, we have provided contemporaneous statistics for the live birth rate of babies with Down syndrome and their childhood death rate compared with peers. We have found that their increasing survival is not without health consequences for the child, their family and health services—we report the majority require hospital admissions, readmissions and longer lengths of stays than their peers, which has received scant research attention in the past. Some factors may even suggest poorer healthcare than their peers, such as higher rates of emergency admissions, although we did not aim to investigate the conditions associated with admission. Together this brings important new findings necessary to address the challenge of planning support that children with Down syndrome and their families need. They require adequate healthcare, and also an awareness of the impact that hospitalisation can have on child development and family bonding. There may be a need to develop tailored packages of support for families with children/young people with Down syndrome to cope with the higher healthcare needs of their offspring.

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Contributors LAH-M contributed to the study design, the analysis and interpretation of data, drafted the first article and revised it critically; DM, AH and LO contributed to the study design, interpretation of data and revised the article critically; S-AC, JPP and RM conceived and designed the study, contributed to study analysis, interpretation of data and revised the article critically. All authors approved the final version.

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Competing interests None declared.

Patient and public involvement Patients and/or the public were not involved in the design, or conduct, or reporting, or dissemination plans of this research.

Patient consent for publication Not required.

Ethics approval Permission to access, link and analyse these data was granted by the Privacy Advisory Committee (PAC) to the National Health Service (NHS), National Services Scotland (NSS) and the Registrar General.

Provenance and peer review Not commissioned; externally peer reviewed.

Data availability statement No data are available. This study linked patient information held across administrative health datasets within ISD Scotland, with externally held data held by the Scottish Government and National Records of Scotland. Linkage and de-identification of data was performed by the Information Services Division (ISD) of NHS National Services Scotland (NSS). A data processing agreement between NHS NSS and University of Glasgow was drafted. The University of Glasgow were authorised to receive record linked data controlled and held by ISD within NSS, via access through the national safe haven. The ISD Statistical Disclosure Control Protocol was followed. It is therefore not possible to share data with other parties.

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Appendix 2: List of scientific meetings where the work in the thesis was presented

- **Hughes-McCormack, L-A.,** McGowan.R, Pell, J.,Mackay, D., Henderson, A., O'Leary, L., Cooper S-A. (2019). Live-births, survival, and hospitalisation rates of children and young people with Down syndrome 1990-2015: a birth cohort study. *Journal of Intellectual Disability Research*. 2019 IASSIDD World Congress, Glasgow, Scotland, August 2019. (doi.org/10.1111/jir.12660)
- **Hughes-McCormack, L-A**., Evans, A., Hurt, L., McGowan, R., Pell, J., Daniel, R., Demmler, J., Bandyopadhyay, A., Mackay, D., Henderson, A., O'Leary, L., Paranjothy, A., Cooper, S.A. (2019). Comparing mortality and hospital admissions to five years of age in children with and without Down syndrome within and across counties 2003-2012: an electronic record matched cohort study in Scotland and Wales. *Journal of Intellectual Disability Research.* 2019 IASSIDD World Congress, Glasgow, Scotland, August 2019. (doi.org/10.1111/jir.12660)
- **Hughes-McCormack, L-A.,** Henderson, A., Kinnear, D., Wiseman, P., Watson, N., Cooper S-A. Gender differences in the health of people with intellectual disabilities. *Journal of Intellectual Disability Research.* 2019 IASSIDD World Congress, Glasgow, Scotland, August 2019. (doi.org/10.1111/jir.12660)
- **Hughes-McCormack, L-A.,** McGowan.R, Pell, J.,Mackay, D., Henderson, A., O'Leary, L., Cooper S-A. (2018). Birth Prevalence and Hospitalisation Rates of Children and Young People with Down Syndrome in Scotland 1990-2015: A Birth Cohort Study. Gatlinburg Conference, Texas, USA, 3-5 April 2019.
- **Hughes-McCormack, L-A.,** McGowan.R, Pell, J.,Mackay, D., Henderson, A., O'Leary, L., Cooper S-A. (2018). Survival rates and hospitalisation rates of people with Down syndrome born in Scotland 1990-2015: a population based cohort study. *Journal of Applied Research in Intellectual Disabilities.* Fifth International IASSIDD Europe Congress, Athens, Greece, 17-20 July 2018. (doi:10.1111/jar.12485)
- **Hughes-McCormack, L-A.,** Rydzewska, E., Henderson, A., MacIntyre, C., Rintoul, J., Cooper S-A. (2018). Prevalence of mental health conditions and relationship with general health in a whole country population of 26,349 people with intellectual disabilities, compared with the general population. *Journal of Applied Research in Intellectual Disabilities.* Fifth International IASSIDD Europe Congress, Athens, Greece, 17-20 July 2018. (doi:10.1111/jar.12485)
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- Rydzewska, E., **Hughes-Mccormack, L.,** Henderson, A. and Cooper, S.-A. (2018) Adults with Autism – Observational Study of Comorbidity in a Whole Country Population. Journal of Applied Research in Intellectual Disabilities, Fifth International IASSIDD Europe Congress, Athens, Greece, 17-20 July 2018. (doi:10.1111/jar.12485)
- Rydzewska, E., **Hughes-Mccormack, L.,** Henderson, A. and Cooper, S.-A. (2018) Adults with Autism – Observational Study of General Health Status in a Whole Country Population. *Journal of Applied Research in Intellectual Disabilities*. Fifth International IASSIDD Europe Congress, Athens, Greece, 17-20 July 2018. (doi:10.1111/jar.12485)
- Rydzewska, E., **Hughes-Mccormack, L.,** Henderson, A. and Cooper, S.-A. (2018) Children and Young People with Autism–Observational Study of Comorbidity in a Whole Country Population. *Journal of Applied Research in*

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- Rydzewska, E., **Hughes-Mccormack, L.,** Henderson, A. and Cooper, S.-A. (2018) Children and Young People with Autism–Observational Study of General Health Status in a Whole Country Population. *Journal of Applied Research in Intellectual Disabilities.* Fifth International IASSIDD Europe Congress, Athens, Greece, 17-20 July 2018. (doi:10.1111/jar.12485)
- **Hughes-McCormack, L-A.,** Cooper, S-A., Greenlaw, N., McConnachie, A., Allan, L., Baltzer, M., McArthur, L., Henderson, A., Melville, C., Morrison, J., Ross, K (2016). Management of long term conditions in primary health care for adults with intellectual disabilities compared with the general population: a comparison over time. Proceedings of the Seattle Club Conference (page 6), 12-13 December, 2016, Glasgow, Scotland.
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