An investigation of healthcare and health inequalities experienced by people with intellectual disabilities, and improving their inclusion in health research

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Abstract

Background

People with intellectual disabilities experience health care and health inequalities across the life course. Compared to the general population, they are more likely to experience multimorbidity, poor mental and physical health, premature mortality and poorer quality care. Despite increasing recognition of these inequalities, much of the existing research has been methodologically limited, often using small unrepresentative samples that are not generalisable. Also, the voices of people with intellectual disabilities are typically excluded from research and research processes that seek to understand and address these inequalities.

The objectives of this thesis are to:

1. Investigate the health care experienced by people with intellectual disabilities, and health care inequalities compared to the general population.
2. Investigate the health outcomes experienced by people with intellectual disabilities, and health inequalities compared with the general population.
3. Develop and implement a novel methodological approach to inclusive health research for people with intellectual disabilities.

Methods

This is a mixed methods thesis which includes 11 studies (9 quantitative and 2 qualitative). Seven of the quantitative studies use Scottish administrative datasets linked using the unique Community Health Index number to construct large, population-level cohorts of children, young people and adults with and without intellectual disabilities. Data sources include Scotland's 2011 Census, the annual Scottish Pupil Census, Prescribing Information System, health and social care expenditure, and death registrations. These data enabled analysis of health and care expenditure, medication use, multimorbidity and mortality.

In the qualitative studies an adapted Citizens' Jury was developed and implemented to ascertain its effectiveness in engaging people with intellectual disabilities in deliberations on inclusive research and academic peer review. These juries incorporated capacity-

building workshops, accessible materials and communications support to enable meaningful participation and coproduction in research.

Results

Health and social care expenditure for adults with intellectual disabilities in Scotland fell in real terms by 3.41% over three years (2012/13, 2013/14 and 2014/15), with disproportionate reductions in health funding and wide regional variation. Psychotropic prescribing, a marker of health care, was consistently high, 50.7% of adults were prescribed psychotropic medications in 2002-04 (T1) and 48.2% in 2014 (T2); antipsychotics: 24.5% (292/1190) in T1 and 16.7% (653/3906) in T2; antidepressants: 11.2% (133/1190) in T1 and 19.1% (746/3906) in T2. 21.2% (62/292) prescribed antipsychotics in T1 had psychosis or bipolar disorder, 33.2% (97/292) had no mental ill health or problem behaviours, 20.6% (60/292) had problem behaviours but no psychosis or bipolar disorder. A longitudinal, linked cohort, which included 545 adults for whom data was available at both T1 and T2, was established. In this group psychotropic prescribing increased over time and 77.3% of those prescribed antipsychotics in T1 remained on them 10 years later. Those with problem behaviours at T1 were over six times more likely to be prescribed antipsychotics (OR 6.45, 4.41-9.45) than those with no problem behaviours. Children and young people with intellectual disabilities were also more likely than their peers to be prescribed antipsychotics (OR 16.85, 15.29-18.56) and antidepressants (OR 2.28, 2.03-2.56). However, the magnitude of the difference between those with and without intellectual disabilities fell over time. Adults with intellectual disabilities were at greater risk of higher anticholinergic burden than their peers at all ages. Anticholinergic polypharmacy was more common, 61.3% vs, 54.7%, (OR 1.61, 1.41-1.83) as was psychotropic polypharmacy, 33.7% vs, 14.3%, (OR 2.79, 2.41-3.23).

A national e-cohort study linking >92% of the Scottish population to other administrative datasets, was established to create a resource for future academic research, including cancer and mortality studies. Of the whole population 0.5% (n=22,538) had intellectual disabilities, 12,837 males and 9,701 females. Analysis of data from Scotland's largest health board reported that almost all adults (98.7%) with intellectual disabilities had multimorbidity, with an average of 11 cooccurring conditions. The pattern of

multimorbidity observed was different to that of the general population. Multimorbidity was present across the life course. Adults with intellectual disabilities also had a higher mortality risk (Standardised Mortality Ratio (SMR) 2.24, 1.99-2.50), particularly those with Down syndrome (SMR 5.28, 3.98-6.57). Those with intellectual disabilities were twice as likely to experience avoidable deaths (29.8% vs, 14%). Children had an even higher mortality risk (SMR 11.6, 9.6-14.0) and were 3.6 (2.2-5.8) times more likely to experience avoidable deaths. During the COVID-19 pandemic, adults with intellectual disabilities had higher rates of infection (SIR 2.61, 1.81-3.40) and severe disease and/or death (SMR 3.26, 2.19-4.32), especially under the age of 65.

Inclusive research methods using adapted citizens' juries enabled people with intellectual disabilities to deliberate on inclusive research practices and peer review. In the Research Voices citizens' jury members produced a consensus report of 10 recommendations to improve the inclusion of people with intellectual disabilities in research. The peer review citizens' jury proposed adaptations to the peer review process to enable inclusion of people with intellectual disabilities in reviewing research, including proposing the establishment of group-based reviews and improvements to the accessibility of academic publishing.

Impact

The research has already influenced national policy and data systems in Scotland, including the decision by Scottish Government to prioritise people with intellectual disabilities for COVID-19 vaccination and the inclusion of intellectual disabilities as a specific data field in Scotland's National Hub for reviewing child deaths. It has advanced methodological innovation through the creation of a national e-cohort, enabling robust population-based research that continues to inform health and care policy.

Conclusions

This thesis provides strong evidence of the persistent health care and health inequalities experienced by people with intellectual disabilities and highlights the value of inclusive methodologies in amplifying their voices within complex research processes. The thesis also highlights the need for ongoing investment in high quality research to address the

evidence gaps, inform policy and ensure responsive, equitable health and care for people with intellectual disabilities.

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List of Publications and abstracts

The full text of the articles is 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.

Output-1:

Henderson A., Fleming M., Cooper S.-A., Pell, J.P., Melville, C., MacKay, D.F., Hatton, C., Kinnear, D. (2022) COVID-19 infection and outcomes in a population-based cohort of 17 203 adults with intellectual disabilities compared with the general population Journal of Epidemiology and Community Health. Doi: 10.1136/jech-2021-218192

Abstract

Background: Adults with intellectual disabilities may be at higher risk of COVID-19 death than other people. We compared COVID-19 infection, severe infection, mortality, case fatality and excess deaths, among adults with, and without, intellectual disabilities.

Methods: Adults with intellectual disabilities identified from Scotland's Census, 2011, and a 5% sample of other adults, were linked to COVID-19 test results, hospitalisation data and deaths (24 January 2020–15 August 2020). We report crude rates of COVID-19 infection, severe infection (hospitalisation/death), mortality, case fatality; age-standardised, sex-standardised and deprivation-standardised severe infection and mortality ratios; and annual all-cause mortality for 2020 and 2015–2019.

Findings: Successful linkage of 94.9% provided data on 17 203 adults with, and 188 634 without, intellectual disabilities. Adults with intellectual disabilities had more infection (905/100 000 vs 521/100 000); severe infection (538/100 000 vs 242/100 000); mortality (258/100 000 vs 116/100 000) and case fatality (30% vs 24%). Poorer outcomes remained after standardisation: standardised severe infection ratio 2.61 (95% CI 1.81 to 3.40) and mortality ratio 3.26 (95% CI 2.19 to 4.32). Standardised Severe Infection Ratio and Standardised Mortality Ratios were higher at ages 55–64: 7.39 (95% CI 3.88 to 10.91) and 19.05 (95% CI 9.07 to 29.02), respectively, and in men, and less deprived neighbourhoods. All-cause mortality was slightly higher in 2020 than 2015–2019 for people with intellectual disabilities: standardised mortality ratio 2.50 (95% CI 2.18 to 2.82) and 2.39 (95% CI 2.28 to 2.51), respectively.

Conclusion: Adults with intellectual disabilities had more COVID-19 infections, and worse outcomes once infected, particularly adults under 65 years. Non-pharmaceutical

interventions directed at formal and informal carers are essential to reduce transmission. All adults with intellectual disabilities should be prioritised for vaccination and boosters regardless of age.

Output-2:

Henderson, A., Kinnear, D., Fleming, M., Stanley, B., Greenlaw, N., Young-Southward, G., Pell, J. P., Cooper, S.-A. (2021) Antipsychotic and antidepressant prescribing for 704 297 children and young people with and without intellectual disabilities: record linkage study. British Journal of Psychiatry, 218(1), 58-62. (doi: 10.1192/bjp.2020.232)

Abstract

Background: Psychotropics are overprescribed for adults with intellectual disabilities; there are few studies in children and young people. This study investigated antipsychotic and antidepressant prescribing in children and young people with and without intellectual disabilities, and prescribing trends.

Method: Scotland's annual Pupil Census, which identifies pupils with and without intellectual disabilities, was record-linked to the Prescribing Information System. Antidepressant and antipsychotic data were extracted. Logistic regression was used to analyse prescribing between 2010 and 2013.

Results: Of the 704 297 pupils, 16 142 (2.29%) had a record of intellectual disabilities. Antipsychotic and antidepressant use increased over time and was higher in older pupils; antipsychotic use was higher in boys, and antidepressant use was higher in girls. Overall, antipsychotics were prescribed to 281 (1.74%) pupils with intellectual disabilities and 802 (0.12%) without (adjusted odds ratio 16.85, 95% CI 15.29–18.56). The higher use among those with intellectual disabilities fell each year (adjusted odds ratio 20.19 in 2010 v. 14.24 in 2013). Overall, 191 (1.18%) pupils with intellectual disabilities and 4561 (0.66%) without were prescribed antidepressants (adjusted odds ratio 2.28, 95% CI 2.03–2.56). The difference decreased each year (adjusted odds ratio 3.10 in 2010 v. 2.02 in 2013).

Conclusions: Significantly more pupils with intellectual disabilities are prescribed antipsychotics and antidepressants than are other pupils. Prescribing overall increased over time, but less so for pupils with intellectual disabilities; either they are not receiving the same treatment advances as other pupils, or possible overprescribing in the past is changing. More longitudinal data are required.

Output-3:

Henderson, A., McSkimming, P., Kinnear, D., McCowan, C., McIntosh, A., Allan, L., Cooper, S.-A. (2020) Changes over a decade in psychotropic prescribing for people with intellectual disabilities: prospective cohort study. British Medical Journal Open, 10(9), e036862. (doi: 10.1136/bmjopen-2020-036862) (PMID:32912946)

Abstract

Background: Psychotropic prescribing rates are higher than reported rates of mental ill-health in adults with intellectual disabilities. This study investigated psychotropic prescribing in the intellectual disabilities population over 10 years and associated mental ill health diagnoses.

Methods: Comparison of cross-sectional data in 2002–2004 (T1) and 2014 (T2). Longitudinal cohort study with detailed health assessments at T1 and record linkage to T2 prescribing data using encashed regular and as-required psychotropic prescriptions.

Results: 1190 adults with intellectual disabilities in T1 compared with 3906 adults with intellectual disabilities in T2. 545/1190 adults with intellectual disabilities in T1 were alive and their records linked to T2 prescribing data. 50.7% (603/1190) of adults in T1 and 48.2% (1881/3906) in T2 were prescribed at least one psychotropic; antipsychotics: 24.5% (292/1190) in T1 and 16.7% (653/3906) in T2; antidepressants: 11.2% (133/1190) in T1 and 19.1% (746/3906) in T2. 21.2% (62/292) prescribed antipsychotics in T1 had psychosis or bipolar disorder, 33.2% (97/292) had no mental ill health or problem behaviours, 20.6% (60/292) had problem behaviours but no psychosis or bipolar disorder. Psychotropics increased from 47.0% (256/545) in T1 to 57.8% (315/545) in T2 ($p<0.001$): antipsychotics did not change (OR 1.18; 95% CI 0.87 to 1.60; $p=0.280$), there was an increase for antidepressants (OR 2.80; 95% CI 1.96 to 4.00; $p<0.001$), hypnotics/anxiolytics (OR 2.19; 95% CI 1.34 to 3.61; $p=0.002$), and antiepileptics (OR 1.40; 95% CI 1.06 to 1.84; $p=0.017$). Antipsychotic prescribing increased for people with problem behaviours in T1 (OR 6.45; 95% CI 4.41 to 9.45; $p<0.001$), more so than for people with other mental ill health in T1 (OR 4.11; 95% CI 2.76 to 6.11; $p<0.001$).

Conclusions: Despite concerns about antipsychotic prescribing and guidelines recommending their withdrawal, it appears that while fewer antipsychotic prescriptions were initiated by T2 than in T1, people were not withdrawn from them once commenced. People with problem behaviours had increased prescribing. There was also a striking increase in antidepressant prescriptions. Adults with intellectual disabilities need frequent and careful medication reviews

Output-4

Henderson, A., Cassidy, J., Croydon, A., Nind, M. (2024) Inclusive peer review: reflections on an adapted citizens' jury with people with learning disabilities. British Journal of Learning Disabilities, (doi: 10.1111/bld.12603)

Abstract

Background: Inclusive research is widely accepted as an essential part of the process to democratise knowledge creation and dissemination. However, while peer review is an important part of academic publishing, the potential to include people with intellectual disabilities in this element of the research process has not previously been explored using a deliberative approach.

Methods: Accessibility adaptations were made to the citizens' jury approach enabling people with learning disabilities to participate. Sixteen adults with mild to moderate intellectual disabilities were recruited to participate in the adapted citizens' jury. Jury members took part in capacity-building workshops to develop their knowledge of research and research processes. Six expert witnesses presented evidence to the citizens' jury and were questioned on aspects of inclusive research, representation, peer review and academic publishing processes. Facilitators supported citizens' jury members to reflect on the evidence presented and to develop recommendations for inclusive peer review.

Results: The citizens' jury was an effective inclusive research approach in this case. Jurors made recommendations related to the question of inclusive peer review: inclusive reviews should be done by groups rather than individuals; the research under review must be in accessible formats and on relevant topics; reviewers need sufficient time to conduct reviews; and diverse groups of people with intellectual disabilities should be involved.

Conclusions: People with intellectual disabilities appreciate the importance of peer review. This jury suggested creative approaches to disseminating, reviewing and engaging with research, including building more opportunities for dialogue between researchers and self-advocates. The adapted citizens' jury was a novel and effective method of supporting deliberation on this topic but other approaches to including the views and experiences of those with more severe intellectual disabilities should be explored.

Output-5

Henderson, A., McLean, R., Kinnear, D. (2022) Including the voices of people with learning disabilities in health research. In: Volkmer, A., and Broomfield, K. eds. Seldom Heard Voices in Service User Involvement: The how and why of meaningful collaboration. Havant. J&R Press. Pp 87-105.

Abstract

Background: People with intellectual disabilities experience significant health inequalities, including higher rates of poor health, multimorbidity, and premature mortality. There is growing consensus on the need to include people with intellectual disabilities in health research and policy development. Traditional research methods generally exclude this population. This project aimed to address this gap by developing and adapting a citizens' jury model to consider the inclusion of people with learning disabilities in health research.

Methods: A demographically representative group of people with intellectual disabilities were recruited to participate in an adapted citizens' jury. Adaptations included preparatory workshops, development of communication supports and accessible expert witness presentations. The jury met over five days.

Findings: The adapted Citizens' Jury was effective in enabling in-depth discussion on complex issues in the field of health research. The jurors produced a consensus report with 10 recommendations for the health research community. The project demonstrated that, with the right support, people with intellectual disabilities can engage in informed discussions on areas of health research. Jurors reported increase confidence and valued the opportunity to be heard.

Conclusions: The project demonstrated the viability of adapting the Citizens' Jury method to include people with intellectual disabilities in health research. Investment in capacity building, accessible communication tools and tailored support is crucial.

Output-6

Cooper, S.-A., **Henderson, A.,** Kinnear, D., MacKay, D., Fleming, M., Smith, G., Hughes-McCormack, L., Rydzewska, E., Dunn, K., Clark, D., Pell, J.P., Melville, C. (2022) Cohort profile: Scotland's record-linkage e-cohorts of people with intellectual disabilities, and autistic people (SCIDA). British Medical Journal Open, 12, e057230. (doi: 10.1136/bmjopen-2021-057230) (PMID:35568493) (PMCID:PMC9109103)

Abstract

Background: There is a lack of population level data available to investigate health, mortality and healthcare inequalities experienced by people with intellectual disabilities, and autistic people, and their determinants. This linkage study is an important step towards identifying and implementing solutions to reduce inequalities. This paper describes the cohorts, record-linkages and variables that will be used.

Methods: Scotland's Census, 2011 was used to identify Scotland's citizens with intellectual disabilities, and autistic citizens, and representative general population samples with neither. Using Scotland's community health index, the Census data (demography, household, employment, long-term conditions) were linked with routinely collected health, death and healthcare data: Scotland's register of deaths, Scottish morbidity data 06 (SMR06: cancer incidence, mortality, treatments), Prescribing Information System (identifying asthma/chronic obstructive pulmonary disease; angina/congestive heart failure/hypertension; peptic ulcer/reflux; constipation; diabetes; thyroid disorder; depression; bipolar disorders; anxiety/sleep; psychosis; attention deficit hyperactivity disorder; epilepsy; glaucoma), SMR01 (general/acute hospital admissions and causes, ambulatory care sensitive admissions), SMR04 (mental health admissions and causes), Scottish Care Information–Diabetes Collaboration (diabetic care quality, diabetic outcomes), national bowel screening programme and cervical screening.

Results: Of the whole population, 0.5% had intellectual disabilities, and 0.6% were autistic. Linkage was successful for >92%. The resultant e-cohorts include: (1) 22 538 people with intellectual disabilities (12 837 men and 9701 women), 4509 of whom are children <16 years, (2) 27 741 autistic people (21 390 men and 6351 women), 15 387 of whom are children <16 years and (3) representative general population samples with neither condition. Very good general health was reported for only 3389 (15.0%) people with intellectual disabilities, 10 510 (38.0%) autistic people, compared with 52.4% general population. Mental health conditions were reported for 4755 (21.1%) people with intellectual disabilities, 3998 (14.4%) autistic people, compared with 4.2% general population.

Future plans: Analyses will determine the extent of premature mortality, causes of death, and avoidable deaths, profile of health conditions and cancers, healthcare quality and screening and determinants of mortality and healthcare.

Output-7

Okon, M., **Henderson, A.,** Kinnear, D., Cooper, S.-A. (2018) Trends and variations in per capita expenditure on adult intellectual disabilities health and social care across Scotland, and by urban/rural class. Journal of Applied Research in Intellectual Disabilities, 32(1), pp. 121-130. (doi: 10.1111/jar.12514) (PMID:29993166)

Abstract

Background: Following the global financial crisis in 2007/08, the UK implemented an austerity programme which may impact on services. Scotland comprises both densely populated urban conurbations and highly dispersed remote rural and island communities.

Methods: Expenditure data were extracted from Scottish Government statistics. Per capita expenditure was calculated using adjusted Scotland's 2011 census data.

Results: There was a 3.41% decrease in real term expenditure on adult intellectual disabilities services between 2012/13 and 2014/15 (>£32 million). In 2014/15, per capita expenditure on adult intellectual disabilities health care ranged from £1,211 to £17,595; social care from £21,147 to £83,831; and combined health and social care expenditures from £37,703 to £85,929. Per capita expenditure on combined health and social care was greater in rural areas, with more on intellectual disabilities social care, though less on health care.

Conclusions: Scottish expenditure on adult intellectual disabilities services has not kept abreast of rising living costs. It varies considerably across the country: a postcode lottery.

Output-8

McKernan, L.W., Cooper, S.-A., **Henderson, A.,** Stanley, B., Greenlaw, N., Pacitti, C., Cairns, D. (2022) A study on prescriptions contributing to the risk of high anticholinergic burden in adults with intellectual disabilities: retrospective record linkage study. Annals of General Psychiatry 21, 41. https://doi.org/10.1186/s12991-022-00418-x

Abstract

Background: People with intellectual disabilities may face a disproportionate risk of experiencing high anticholinergic burden, and its negative sequalae, from a range of medications, and at younger ages than the general population, but there has been little previous study. Our aim was to determine the source of anticholinergic burden from prescribed medication.

Methods: Retrospective matched observational study using record linkage. Adults with (n = 4,305), and without (n = 12,915), intellectual disabilities matched by age-, sex- and

neighbourhood deprivation. The main outcome measure was the prescription of long-term (approximately 12 months use) anticholinergic medications overall (classified according to the Anticholinergic Risk Scale [ARS]), by drug class, individual drugs, and polypharmacy.

Results: Adults with n = 1,654 (38.4%), and without n = 3,047 (23.6%), intellectual disabilities were prescribed medications long-term with anticholinergic effects. Of those on such drugs, adults with intellectual disabilities were most likely to be on central nervous system (62.6%), gastrointestinal (46.7%), and cardiovascular (28.4%) medications. They were prescribed more central nervous system, gynaecological/urinary tract, musculoskeletal, and respiratory medications, and less cardiovascular, infection, and endocrine medications than their matched comparators. Regardless of age, sex, or neighbourhood deprivation, adults with intellectual disabilities had greater odds of being prescribed antipsychotics (OR = 5.37 [4.40–6.57], $p < 0.001$), antiepileptics (OR = 2.57 [2.22–2.99], $p < 0.001$), and anxiolytics/hypnotics (OR = 1.28 [1.06–1.56], $p = 0.012$). Compared to the general population, adults with intellectual disabilities were more likely to be exposed to overall anticholinergic polypharmacy (OR = 1.48 [1.33–1.66], $p < 0.001$), and to psychotropic polypharmacy (OR = 2.79 [2.41–3.23], $p < 0.001$).

Conclusions: Adults with intellectual disabilities are exposed to a greater risk of having very high anticholinergic burden through polypharmacy from several classes of medications, which may be prescribed by several different prescribers. There is a need for evidence-based recommendations specifically about people with intellectual disabilities with multiple physical and mental ill-health conditions to optimise medication use, reduce inappropriate prescribing and adverse anticholinergic effects.

Output-9

Smith, G. S., Fleming, M. , Kinnear, D. , **Henderson, A.** , Pell, J. P. , Melville, C., Cooper, S.-A. (2020) Rates and causes of mortality among children and young people with and without intellectual disabilities in Scotland: a record linkage cohort study of 796,190 schoolchildren. British Medical Journal Open, 10, e034077. (doi: 10.1136/bmjopen-2019-034077) (PMID:32773385)

Abstract

Background: Children and young people with intellectual disabilities experience high rates of physical and mental ill-health than their peers. There is significant variability in the reported risks of mortality within this group and few studies investigate causes of

mortality. This study investigates mortality rates and causes in children and young people with intellectual disabilities.

Methods: Retrospective cohort; individual record linkage between Scotland's annual pupil census and National Records of Scotland death register. Data on pupils receiving local authority-funded schooling in Scotland, 2008 to 2013, with an Additional Support Need due to intellectual disabilities, were compared with other pupils. Deaths up to 2015: age of death, age-standardised mortality ratios (age-SMRs); causes of death including cause-specific age-SMRs; avoidable deaths as defined by the UK Office of National Statistics were analysed.

Results: 18 278/947 922 (1.9%) pupils had intellectual disabilities. 106 died over 67 342 person-years (crude mortality rate=157/100 000 person-years), compared with 458 controls over 3 672 224 person-years (crude mortality rate=12/100 000 person-years). Age-SMR was 11.6 (95% CI 9.6 to 14.0); 16.6 (95% CI 12.2 to 22.6) for female pupils and 9.8 (95% CI 7.7 to 12.5) for male pupils. Most common main underlying causes were diseases of the nervous system, followed by congenital anomalies; most common all-contributing causes were diseases of the nervous system, followed by respiratory system; most common specific contributing causes were cerebral palsy, pneumonia, respiratory failure and epilepsy. For all contributing causes, SMR was 98.8 (95% CI 69.9 to 139.7) for congenital anomalies, 76.5 (95% CI 58.9 to 99.4) for nervous system, 63.7 (95% CI 37.0 to 109.7) for digestive system, 55.3 (95% CI 42.5 to 72.1) for respiratory system, 32.1 (95% CI 17.8 to 57.9) for endocrine and 14.8 (95% CI 8.9 to 24.5) for circulatory system. External causes accounted for 46% of control deaths, but the SMR for external-related deaths was still higher (3.6 (95% CI 2.2 to 5.8)) for pupils with intellectual disabilities. Deaths amenable to good care were common.

Conclusion: Pupils with intellectual disabilities were much more likely to die than their peers, and had a different pattern of causes, including amenable deaths across a wide range of disease categories. Improvements are needed to reduce amenable deaths, for example, epilepsy-related and dysphagia, and to support families of children with life-limiting conditions.

Output-10

Cooper, S.-A. , Allan, L., Greenlaw, N., McSkimming, P., Jasilek, A., **Henderson, A.** , McCowan, C. , Kinnear, D., Melville, C. (2020) Rates, causes, place, and predictors of mortality in adults with intellectual disabilities with and without Down syndrome: cohort

study with record linkage. British Medical Journal Open, 10, e03646. (doi: 10.1136/bmjopen-2019-036465) (PMID:32423940) (PMCID:PMC7239521)

Abstract

Background: People with intellectual disabilities die prematurely, however there are gaps in our knowledge of the risks factors associated with higher rates of mortality in the intellectual disabilities population with and without Down syndrome and the causes of mortality. This study investigates mortality in adults with intellectual disabilities: rates, causes, place, demographic and clinical predictors.

Methods: Cohort study with record linkage to death data. 961/1023 (94%) adults (16–83 years; mean=44.1 years; 54.6% male) with intellectual disabilities, clinically examined in 2001–2004; subsequently record-linked to their National Health Service number, allowing linkage to death certificate data, 2018. Standardised mortality ratios (SMRs), underlying and all contributing causes of death, avoidable deaths, place, and demographic and clinical predictors of death.

Results: 294/961 (30.6%) had died; 64/179 (35.8%) with Down syndrome, 230/783 (29.4%) without Down syndrome. SMR overall=2.24 (1.98, 2.49); Down syndrome adults=5.28 (3.98, 6.57), adults without Down syndrome=1.93 (1.68, 2.18); male=1.69 (1.42, 1.95), female=3.48 (2.90, 4.06). SMRs decreased as age increased. More severe intellectual disabilities increased SMR, but ability was not retained in the multivariable model. SMRs were higher for most International Statistical Classification of Diseases and Related Health Problems, 10th Revision chapters. For adults without Down syndrome, aspiration/reflux/choking and respiratory infection were the the most common underlying causes of mortality; for Down syndrome adults 'Down syndrome', and dementia were most common. Amenable deaths (29.8%) were double that in the general population (14%); 60.3% died in hospital. Mortality risk related to percutaneous endoscopic gastrostomy/tube fed, Down syndrome, diabetes, lower respiratory tract infection at cohort-entry, smoking, epilepsy, hearing impairment, increasing number of prescribed drugs, increasing age. Bowel incontinence reduced mortality risk.

Conclusions: Adults with intellectual disabilities with and without Down syndrome have different SMRs and causes of death which should be separately reported. Both die younger, from different causes than other people. Some mortality risks are similar to other people, with earlier mortality reflecting more multimorbidity; amenable deaths are also common. This should inform actions to reduce early mortality, for example, training to avoid aspiration/choking, pain identification to address problems before they are advanced, and reasonable adjustments to improve healthcare quality.

Output-11

Kinnear, D., Morrison, J., Allan, L., **Henderson, A.** , Smiley, E., Cooper, S.-A. (2018) Prevalence of physical conditions and multimorbidity in a cohort of adults with intellectual disabilities with and without Down syndrome: cross-sectional study. British Medical Journal Open, 8(2), e018292. (doi: 10.1136/bmjopen-2017-018292) (PMID:29431619) (PMCID:PMC5829598)

Abstract

Background: People with intellectual disabilities have different health needs and a shorter life expectancy than other people. This study investigates the prevalence of multimorbidity in adults with intellectual disabilities with and without Down syndrome.

Methods: Large, population-based cross-sectional study in the geographical area of one Health Board, Scotland. All adults (aged 16+ years) known to general practitioners to have intellectual disabilities and adults receiving services provided or paid by intellectual disabilities health or social work services. 1023/1562 potential participants took part (65.5%); 562 (54.9%) men and 461 (45.1%) women, aged 43.9 years (16–83 years). 186 had Down syndrome and 837 did not. The prevalence of International Statistical Classification of Diseases, 10th revision, physical health conditions and multimorbidity detected at a comprehensive health assessment are reported.

Results: The mean number of physical health conditions/participant was 11.04, and 98.7% had multimorbidity. The most prevalent conditions are painful and/or disabling and, in some cases, life threatening. The five most prevalent were visual impairment, obesity, epilepsy, constipation and ataxic/gait disorders. The pattern of multimorbidity differs from that seen in the general population and is spread across the entire adult life course. The extent of multimorbidity in the adults with Down syndrome was similar to that of the adults without Down syndrome, while the prevalence of individual conditions differed.

Conclusions: This robustly designed study with a large population found an extremely high prevalence of multimorbidity in adults with intellectual disabilities across the entire adult life course. This increases complexity of medical management that secondary healthcare services and medical education are not yet geared towards, as these tend to focus on single conditions. This is in addition to complexity due to limitations in communication and understanding. As the physical conditions within their

multimorbidity also differ from that seen in the older general population, urgent attention is needed to develop the care pathways and guidelines that are required to inform and so improve their healthcare.

All submitted outputs, except 4, 5 and 7 include a published contributors statement which was approved by all authors prior to publication. On request, the available co-authors of outputs 4, 5, and 7 have now approved in writing my contribution to those outputs, and submission for this PhD by publication. Rhiann McLean (output 5) and Marion Okon (output 7) were not contactable.

Acknowledgements

I am grateful to Professor Sally-Ann Cooper (OBE), my supervisor, whose guidance, challenge, and inspiration have helped shaped this work. Her leadership in intellectual disabilities research has led to real change in Scotland, and working under her supervision over a number of years has encouraged me to aim high and produce research that makes a difference.

I would also like to thank the many people with intellectual disabilities I have had the privilege to work alongside. Their generosity in sharing time, insights, and their experiences has been invaluable, and many of these collaborations have grown into lasting friendships, continually reminding me of the human impact of this research.

This thesis has been strengthened by the support and collaboration of many colleagues across research, health, policy, and the third sector. These multi-author papers reflect the expertise, insights, and constructive engagement of all involved, and I am grateful for their shared commitment to advancing knowledge, improving practice, and translating research into meaningful policy change.

I also owe thanks to my family. My husband has been my unwavering supporter and my children, a constant motivation to be the best person I can be. I am proud that they are kind, compassionate people who are committed to justice and equality — their example reinforces the purpose of this work and its broader contribution to a fairer, more inclusive world.

Personal statement

Throughout my personal, academic and professional life, I have sought to understand and challenge inequalities. My postgraduate studies in social policy and research methods helped me to develop a more nuanced understanding of the structural and biosocial nature of inequality and reinforced my motivation to work in an area where I could make a meaningful contribution to addressing it. A formative turning point was securing a role in a third sector organisation with a mission to advocate for the rights of people with intellectual disabilities across all domains of life, including the right to live independent and healthy lives. My roles in this organisation provided the opportunity to work alongside people with intellectual disabilities themselves, supporting them to have a voice and to influence policy and practice. This way of working became central to my approach and enabled me to learn directly from people with intellectual disabilities, shaping how I understood both inequality and evidence. Alongside this, I was able to apply my theoretical understanding of inequality across a broad range of qualitative and quantitative research and policy-focused activities. This work exposed me to the practical consequences of public policy decisions provided insight into how decisions across health and social care interact to shape outcomes over the life course. Through this experience, I developed an understanding that the inequalities experienced by people with intellectual disabilities are fundamentally multifactorial, arising not from individual characteristics alone, but from the cumulative effects of historical and contemporary policy design, service organisation and structural exclusion. Crucially, this third-sector context also embedded inclusive ways of working as a core part of my professional practice. Through this work I established enduring relationships and a practical appreciation of the value of lived experience in shaping research priorities, evidence and impact.

Following publication of Scotland's national learning disabilities strategy, which included a commitment to identifying and addressing health inequalities through the establishment of a specialist public health observatory, I secured an opportunity to develop my knowledge and experience within academia by contributing to the establishment of this new body dedicated to building the infrastructure for undertaking population level research primarily using administrative data. In the early stages of this work my focus was on finding opportunities to identify people with intellectual disabilities within these datasets, building relationships with data providers and working to secure approvals for large scale linkage projects with the potential to address gaps in the research on the health of people

with intellectual disabilities across the lifespan, uncovering previously hidden trajectories of inequality thus increasing their visibility within population health data. A key part of my role in building this function was in strategically positioning the organisation as a reliable source of policy relevant evidence to support policy and practice development. Another critical aspect of this role was to address the question of how to meaningfully engage people with intellectual disabilities in this complex, quantitative and tightly regulated research context. This challenge led to the establishment of the adapted deliberative model of engagement designed to include people with intellectual disabilities in discussions on inclusive research that are presented in this thesis.

Overall, these experiences reflect my progression from third-sector policy research to a role focused on building the data and analytical capacity necessary to inform policy, enhance accountability, and support sustained action on health and healthcare inequalities. These experiences provide the foundations for the programme of research presented in this thesis.

Explanatory essay

Introduction

Intellectual disabilities include a diverse set of conditions with onset in the developmental phase. They significantly affect a person's intellectual functioning across various domains, including cognitive, language, motor, and social skills, and adaptive behaviours across conceptual, social, and practical areas (1) People with intellectual disabilities of all ages, experience significant inequalities in many areas of life. (2, 3) These inequalities are reflected in their high rates of multi-morbidity, diverse and complex health needs, (4) premature mortality (5) and high rates of psychotropic prescribing. (6) Existing studies on these issues were previously limited, and few were large-scale.

Increasing awareness of the health and healthcare inequalities experienced by people with intellectual disabilities has led to calls for improvements in the methods of health studies for this population group, (7) as there are significant gaps in this research (8) and variable results reported due to sampling and methodological limitations. (9) Research specifically on the health of children and young people with intellectual disabilities is even less well developed than for adults with intellectual disabilities. (3, 10) It is therefore clear that good quality evidence that is generalisable at a population level is needed as the first step towards informing action to address the poor health, high prescribing rates and premature mortality of people with intellectual disabilities. Despite Scotland possessing a rich set of routinely collected administrative datasets providing information on population health, routine surveillance and reporting of the health and healthcare utilisation of the population with intellectual disabilities is lacking. (11) While a burgeoning number of studies in the last two decades have linked multiple data sources to enable the investigation of health and healthcare outcomes for the general population in Scotland (12) significant barriers remain in reporting on the specific health and healthcare outcomes of people with intellectual disabilities, as they are not routinely identifiable in Scotland's health datasets. Ascertainment of this population within Scotland's national censuses and administrative datasets, coupled with robust record linkage is crucial. This would enable research to better understand health inequalities, to identify associated health and care support usage, and ultimately to enable effective public service planning and resource allocation. Nine of the papers which are included in this thesis utilise population level, administrative datasets to

investigate health and healthcare patterns and outcomes for this under-researched population.

Understanding of the allocation and utilisation of resources is similarly important in effectively addressing the identified systemic health care inequalities. No longitudinal studies have comprehensively investigated the allocation of funding to both health and social care specialist intellectual disabilities services. One English study, (13) explored resource allocation in intellectual disabilities community services across 151 English councils, reporting inequities in the distribution of funding for intellectual disabilities services. Understanding the allocation of resources at a national level for intellectual disabilities services is essential in identifying potential inequities in geographic distribution. Such studies are essential for improving understanding of system level inequities in specialist health and care provision, and for informing actions to address them thereby supporting evidenced informed policy making. (14) Whilst patterns of resource allocation provide important insights into the structural aspects of service provision, assessing how these resources are utilised in practice offers an additional perspective on healthcare quality. Prescribing patterns, for example, serve as a useful proxy for evaluating the nature and quality of care delivered, highlighting variations in clinical decision-making, alignment with best practice guidelines, and access to personalised, non-pharmacological interventions. Studies have reported that adults with intellectual disabilities are more likely to be prescribed psychotropic medications (15, 16) than other people. One large UK-based study (16) which used primary care records to investigate mental ill-health, challenging behaviour and psychotropic prescribing rates in adults with intellectual disabilities reported high rates of psychotropic prescribing, higher than general practitioner (GP) recorded mental ill-health, a significant association between challenging behaviours and antipsychotic prescribing and a moderate decrease in new antipsychotic prescriptions over time. However, no studies have incorporated detailed psychiatric assessments, by specialist intellectual disabilities psychiatrists, alongside longitudinal analysis of prescribing patterns in a community-based cohort of adults with intellectual disabilities. There are no studies investigating longitudinal patterns of psychotropic prescribing in children and young people with intellectual disabilities for a whole country. This is a significant gap in the literature particularly given the lack of good quality evidence on the safety and efficacy of psychotropic prescribing in children and young people. (17)

In addition to understanding comparative rates of prescribing, it is also important to consider the potential health effects of prescribed medications. Medications with anticholinergic properties are commonly prescribed for a wide range of conditions. These medications can cause a wide range of adverse effects. Studies investigating the cumulative effect of anticholinergic polypharmacy have shown that older people are at high risk of anticholinergic burden due to higher rates of multimorbidity and prescribing. (18) However, few studies have investigated the risk of anticholinergic burden and risk in people with intellectual disabilities. One Irish study (19) investigated the cumulative impact of anticholinergic burden in a representative cohort of 736 older adults with intellectual disabilities (aged 41-90 years). This study reported high rates of cumulative anticholinergic medication exposure, more so in those aged 65 and over with co-occurring mental ill-health. Daytime drowsiness and constipation associated with high cholinergic burden were also observed. Psychotropic medications were identified as the most common drugs contributing to anticholinergic burden. No studies have investigated anticholinergic burden in a population-based sample compared to a matched general population cohort.

Access to good quality health care services is an important determinant of health outcomes for people with intellectual disabilities of all age groups. However, the planning, delivery and resourcing of these services should be informed by epidemiological research to understand the patterns of health, disease burden and mortality that underpin and interact with these service level disparities. Previous research has reported that people with intellectual disabilities experience significantly poorer health outcomes than the general population, but significant gaps remain regarding the prevalence and nature of physical and mental health conditions, multimorbidity and mortality across the lifespan in this population. International research has shown that people with intellectual disabilities have significantly higher rates of morbidity across a range of conditions, including obesity, (20) epilepsy, (21) gastrointestinal disorders, (22) sensory impairments (23) and mental health disorders, (24) compared to the general population. Crucially, the pattern of multimorbidity also differs from that observed in the general population with people with intellectual disabilities experiencing multiple co-occurring conditions from a much younger age. (4) Multimorbidity represents a significant challenge to the organisation and delivery of high-quality healthcare services which tend to be organised around management of single conditions. (25) Mortality studies have shown that those with intellectual disabilities are at increased risk of death, with reported standardised mortality ratios (SMRs) two to three

times higher than those observed in the general population (5) and even higher in some population subgroups, such as those with Down Syndrome. (26) Mortality studies have also reported that people with intellectual disabilities are more likely to die from preventable or amenable causes, which could have been avoided through the effective healthcare interventions. (27) However, much of the existing literature is methodologically flawed, including small, unrepresentative samples that are not generalisable.

The studies presented in this section of the thesis directly address these gaps by drawing on robust, nationally representative datasets to investigate the inequalities in health care and health of people with intellectual disabilities. They include investigation of the prevalence of physical and mental health conditions, the nature and extent of multimorbidity, and patterns of mortality in both adults and children, including those with and without Down Syndrome. These novel studies also include evidence on the impact of the COVID-19 pandemic on the population with intellectual disabilities, at a point in the pandemic when there was a lack of evidence to inform public health policy. Collectively these studies provide comprehensive, generalisable evidence with direct implications for policy and practice, demonstrating the need for public health strategies, preventative interventions and clinical pathways that are designed to meet the complex needs of people with intellectual disabilities, instead of relying on approaches developed to address general population health needs.

Alongside such quantitative research, it is essential to develop robust methods for inclusive research practices. There is consensus that people with intellectual disabilities should be included in research that aims to better understand and address their health and wellbeing inequalities; however, there are significant barriers to their engagement in such research. These barriers occur at different levels (28) and result in the underrepresentation of people with intellectual disabilities as both participants and subjects of research. (29) The inclusion of people with intellectual disabilities in research enhances the quality and relevance of research and can lead to targeted health and care interventions that are better aligned to the needs of people. (28) However, methodological adaptations and creative techniques are required to support the inclusion of people with intellectual disabilities in research. (30, 31) There are currently few studies that have systematically investigated the views of people with intellectual disabilities on how, and at what stages in the research

process, people with intellectual disabilities should be included. Thus, there is a need for robust engagement with people with intellectual disabilities to investigate the specific contribution and benefits of inclusive health research, across different stages of the research cycle. This is particularly relevant when considering how to involve people with intellectual disabilities in large scale quantitative research studies or in essential parts of the research process, such as peer review.

Objectives

The outputs included in this thesis were published from research that was undertaken to:

1. Investigate the health care experienced by people with intellectual disabilities, and health care inequalities compared to the general population.
2. Investigate the health outcomes experienced by people with intellectual disabilities, and health inequalities compared with the general population.
3. Develop and implement a novel methodological approach to inclusive health research for people with intellectual disabilities

Methods

Administrative data sources

Nine of the 11 submitted studies used administrative data from different public bodies including primary care, secondary health, finance and education data to investigate specific health and care outcomes for people with intellectual disabilities; three of which draw on national level census data (table 1).

Table 1: Overview of administrative data sources

Dataset	**Details**	**Dates**	**Associated outputs**
Scotland's Census, 2011(32)	The 2011 Scottish census, conducted on 27 March 2011, collected data on people living in both private households and communal establishments. This census included questions relating to general health, the presence of long-term conditions and the extent to which daily activities were limited by such conditions. It is estimated that the census captured 94% of Scotland's population. Notably, the 2011 census included a specific question on intellectual disabilities, explicitly distinguishing these from specific learning difficulties such as dyslexia, thus enabling reliable ascertainment of the population with intellectual disabilities for the whole of Scotland.	27/03/11	1, 6
Pupil Census (33)	The annual pupil census collects information on all children and young people attending publicly funded schools, which is 95% of all children and young people in Scotland. This census includes information on additional support needs and their reason. The types of additional support needs that can be recorded include intellectual disabilities and are differentiated from specific learning disabilities.	2010-2013 2008-2013	2 9
General Practice data, NHS Greater	Data from participants with intellectual disabilities, living in Scotland's largest health board area were collected following comprehensive physical and mental health assessments by specialist	2002-2004 & 2014	3

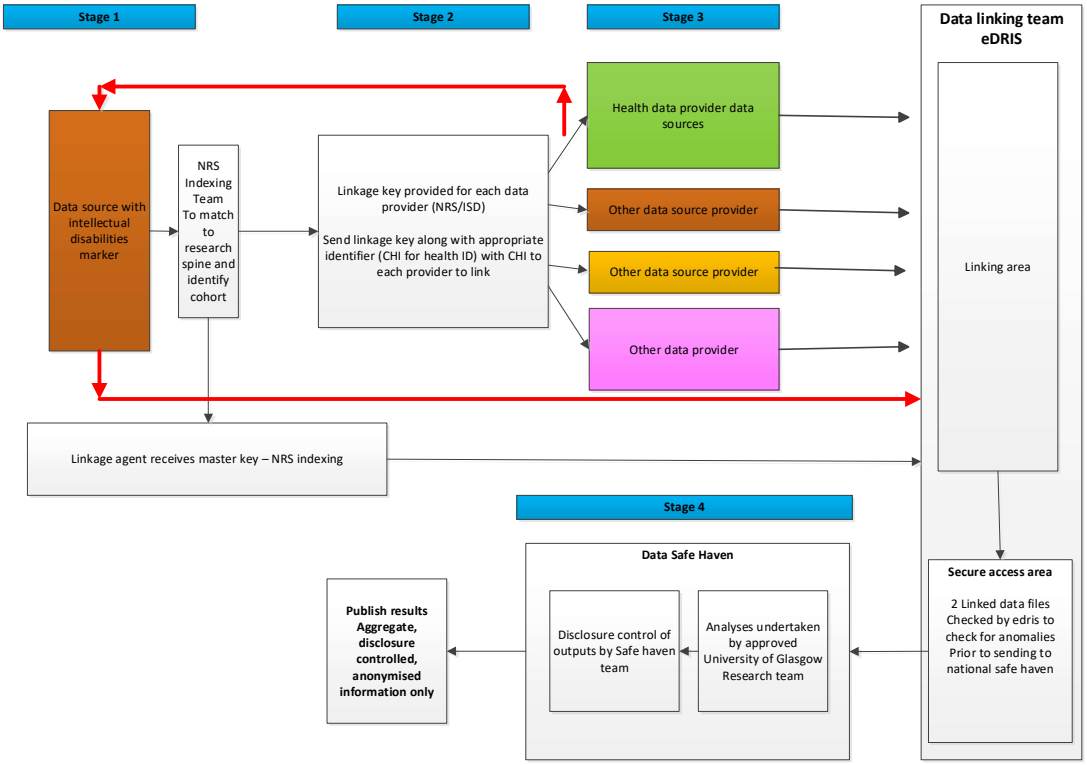
Glasgow and Clyde (34)	intellectual disabilities nurses and doctors. Clinical diagnoses were recorded using International Classification of Diseases -10 (Diagnostic Criteria for Research), (35) Diagnostic and Statistical Manual of Mental Disorders-IV-TR (36) and Diagnostic Criteria for Psychiatric Disorders for use with Adults with Intellectual Disabilities, (37) along with demographic criteria	2002-2004	10, 11
		2014	8
Prescribing Information System (PIS) (38)	PIS is a record of all medicines that are prescribed and dispensed in the community, including prescriptions written in Scotland but encashed elsewhere in the UK. Prescribing data is coded according to British National Formulary (BNF) codes. (39)	2010-2013	2
		2014	3
		09/16 – 08/17	8
Local Government Financial Statistics (40)	Annual collection of social care expenditure for all Scottish local authorities. Data on employee and operating costs, transfer payments, support service costs and revenue contribution to capital from all 32 local authorities.	2012/13	7
Scottish Health Service Costs dataset (41)	Annual collection of health service costs from all Scottish territorial health boards. Expenditure data on adult intellectual disabilities in-patient, day-patient, and community intellectual disabilities teams from 13/14 Scottish health boards.	2012/13, 2013/2014, 2014/2015	7
Electronic Communication of Surveillance in	ECOSS includes a record of laboratory-tested COVID-19 results.	24/01/20 – 15/08/20	1

Scotland (ECOSS) (42)			
Death Registrations (43)	The Scottish Death Registration dataset contains records of all deaths in Scotland, collected by National Records of Scotland. It includes information on the date and cause of death and is coded using the ICD.	24/01/15 – 15/08/19 & 24/01/20 – 15/08/20	1
		2008 - 2015	9
		2018	10

Data linkage

Seven of the 11 studies presented have used data linked across multiple sources of data, using the Community Health Index (CHI) (44) as a linkage key. The CHI database of all patients in NHS Scotland facilitates the unique identification of individual records and covers between 96.5% and 99.9% of Scotland's population (45). The CHI number enables linkage between different data sets at an individual level to allow investigation of health, education and care outcomes. Diagram 1 provides an overview of this linkage process.

Figure 1: Linkage process diagram



Inclusive methods

Inclusive deliberative approaches, such as Citizens' Juries, have been used frequently in diverse, global contexts to engage communities in complex deliberations on important areas. (46, 47) Citizens' juries involve the recruitment of a representative sample of citizens to deliberate on a complex area of public concern. Jury members are presented with detailed evidence, from expert 'witnesses' related to the issue being considered. Following presentation and 'cross-examination' of the witnesses jurors deliberate on the issue and produce a consensus set of recommendations, that address the issue under

consideration, in the form of a report. To support the engagement of people with intellectual disabilities in complex discussions on inclusive research practice an adapted Citizens' Jury model was developed. This novel approach adhered to the core principles of the 'Jefferson Center' model for Citizens' Juries, (48) with adaptations to support accessibility of participants with intellectual disabilities. These adaptations included the use of visual communication tools, accessible information and other inclusive methods to support the specific communication needs of the participants. Jury members were recruited to reflect the demographic profile of adults with learning disabilities in terms of age and sex, and informed consent was sought from participants.

Project ethical approvals

Approvals were gained, as required via the NHS Scotland Public Benefit and Privacy Panel (PBPP), the Statistics PBPP, NHS REC, NHS Greater Glasgow and Clyde Local Privacy Advisory Committee and the University of Glasgow's College of Medical, Veterinary and Life Sciences Ethics Committee.

Summary of results

1. Health care experienced by people with intellectual disabilities, and health care inequalities compared to the general population

People with intellectual disabilities have significant health needs and thus require equal access to well-resourced, evidence-informed health and care services that are designed to meet their specific health needs. The significant inequalities experienced by people with intellectual disabilities are reflected in the ways that health and care is funded, delivered and utilised.

Longitudinal trends and variation in health and care expenditure

Routine statistical reports of spending on health and care services, over 3 years, were analysed to investigate the resourcing of health and care services for adults with intellectual disabilities across Scotland. Output 7, (49) found that Scotland's combined health and social care expenditure on adult intellectual disabilities services was £975,904,242 in 2012/13; £950,759,710 in 2013/14; and £943,692,001 in 2014/15. This is a real term decrease of 3.41% across the 3 years, equating to a reduction of over £32 million. Whilst combined per capita expenditure on adult intellectual disabilities services may appear a lot (on average £44,265 in 2014/15), the cost to each adult in the general population is not (on average £213 in 2014/15). Within the context of whole population expenditure on other services, the 4.29% real term decrease in intellectual disabilities health care expenditure and 3.02% real term decrease in intellectual disabilities social care expenditure demonstrated that intellectual disabilities health expenditure was being disproportionally reduced, whereas intellectual disabilities social care expenditure (whilst reduced) was not disproportionately so. Significant regional disparities in health and social care per capita expenditure were also observed. In 2014/15, per capita intellectual disabilities health expenditure ranged from £1,211 to £17,595, and social care expenditure from £21,147 to £83,831. Differences in expenditure between urban and rural areas were also reported, where remote and rural areas saw increased spending on social care services but reduced levels of spending on specialist intellectual disabilities health services.

Psychotropic prescribing

Prescribing practices are an effective proxy for health service quality and use as they reflect key elements of specialist and routine health care delivery, including diagnostic accuracy, access to appropriate treatments and health reviews. Output 3 (50) was a longitudinal study of psychotropic prescribing at two timepoints, 2002-2004 (T1) and 2014 (T2). Data on a cross-sectional cohort of adults with intellectual disabilities was linked to prescribing data. All participants included in the T1 cohort (n=1,190) underwent comprehensive health assessments, including assessment of mental ill-health by intellectual disabilities nurses or a GP with special interest in intellectual disabilities. 54% of participants who were identified as having possible, probable or definite mental ill health were then separately assessed by specialist intellectual disabilities psychiatrists, and through consensus, mental ill-health was classified using ICD-10-DCR, DSM-IV-TR, DC-LD and clinical criteria. At T2, primary care records, in the same health board were linked to the Prescribing Information System (PIS) to create an additional cohort of people with intellectual disabilities aged ≥18 years (n=3,906). Overall, high rates of psychotropic prescribing (antipsychotic, antidepressant, hypnotic and anxiolytic, antiepileptic and lithium) were observed across both timepoints (50.7% in T1 and 48.2% in T2). 24.5% were prescribed antipsychotics in T1 and 16.7% in T2. For antidepressants the rate was 11.2% in T1 and 19.1% in T2. Of those prescribed antipsychotic medications in T1 only 21.2% had psychosis or bipolar disorder (severe mental ill-health) and 33.2% had no mental health problem.

A linked cohort, of participants for whom data was available in both T1 and T2 was created (n=545). Analysis of data from the same group of people over 10 years showed that overall psychotropic prescribing significantly increased from 47.0% to 57.8% ($p<0.001$); antipsychotics stayed the same and antidepressants increased. However, of those prescribed antipsychotics in T1, 77.3% were still prescribed 10 years later in T2. This highlights the long-term nature of antipsychotic prescribing in this cohort and once commenced it is unlikely that antipsychotic prescriptions will be reduced or removed. Those who had problem behaviours in T1 were more than 6 times more likely to be prescribed an antipsychotic at T2 (OR 6.45; 95% CI 4.41 to 9.45; $p<0.001$).

Output 2 (51) is one of few studies of psychotropic prescribing in children and young people with intellectual disabilities in comparison with other children and young people, and the first longitudinal study of a country's whole population. Using data from Scotland's annual pupil census linked with the PIS, the study analysed prescribing outcomes for 704,297 pupils, of whom 16,142 (2.29%) had a record of intellectual disabilities. This study revealed that those with intellectual disabilities were significantly more likely to be prescribed both antipsychotics (OR 16.85 95% CI 15.29 to 18.56) and antidepressants (OR 2.28 95% CI 2.03 to 2.56) when compared to other children. The relative difference between them and their peers reduced over four years, but this was because of increased use of antipsychotics and antidepressants by their peers, not decreased use among children with intellectual disabilities.

High rates of psychotropic prescribing over long periods can have significant detrimental health impacts and the increased risk of prescribing of psychotropic medications observed in the previous 2 studies indicate that people with intellectual disabilities could be more likely to experience such negative health outcomes, including increased anticholinergic burden. Output 8 (52) investigated the prevalence and sources of anticholinergic burden in adults with intellectual disabilities. This retrospective cohort study compared medication use for adults with and without intellectual disabilities, matched by age, sex and socioeconomic factors. The study reports high rates of prescriptions leading to high anticholinergic burden in the intellectual disabilities population. Adults with intellectual disabilities were more likely to be prescribed anticholinergic medicines, than those with no intellectual disabilities (38.4% vs, 23.6%). Gastrointestinal, cardiovascular and central nervous system drugs, and in particular psychotropic medications were a major contributor to anticholinergic burden. Those with intellectual disabilities had higher odds of being prescribed antipsychotics OR=5.37 (4.40-6.57), antiepileptics OR=2.57 (2.22-2.99) and anxiolytics/hypnotics OR=1.28 (1.06-1.56). Both anticholinergic and psychotropic polypharmacy (the use of 2 or more anticholinergic or psychotropic medications at one time) were also significantly higher in the intellectual disabilities group, OR=1.48 (1.33-1.66) and OR=2.79 (2.41-3.23) respectively. Prescriptions of anticholinergic drugs from an early age increase the likelihood of burden across the lifespan. There is robust evidence correlating anticholinergic burden to poor health outcomes, with recommendations for avoiding strong anticholinergic medication use long-term. These findings indicate a role

for medication reviews to additionally consider anticholinergic side effects and cumulative burden across concomitant medications.

Output 6 (53) reports the establishment of a national e-cohort of people with intellectual disabilities compared to a random sample of the general population. Scotland's Census, 2011 was used to identify Scotland's citizens with intellectual disabilities, and representative general population samples. Their Census data was linked with data from routinely collected health, death and health care data. Of the whole population, 0.5% had intellectual disabilities. Linkage was successful for >92% of records. The resultant intellectual disabilities e-cohort includes: 22,538 people with intellectual disabilities (12,837 men and 9,701 women), 4,509 of whom are children <16 years. Very good general health was reported for only 3389 (15.0%) people with intellectual disabilities, compared with 52.4% of the general population. Mental health conditions were reported for 4,755 (21.1%) people with intellectual disabilities, compared with 4.2% of the general population. The e-cohort provides data for future analyses on health care and health care inequalities to identify health care barriers and provide evidence to inform the future development of policy and health care interventions.

2. Health outcomes experienced by people with intellectual disabilities, and health inequalities compared with the general population.

The outputs addressing objective 2 investigated the prevalence of physical and mental ill health, multimorbidity, and mortality across the life span for people with intellectual disabilities, including the impact and outcomes of COVID-19. There were significant gaps in the evidence and few high-quality, generalisable studies that drew on population-level data, hence the importance of these novel studies.

Output 11 (54) reported prevalence of physical conditions and multimorbidity in a large cohort of adults with intellectual disabilities with, and without Down syndrome, following detailed health assessments. The mean number of physical health conditions was 11.04 and 98.7% had multimorbidity. The most prevalent conditions were visual impairment, obesity, epilepsy, constipation and ataxic gait disorders. The extent of multimorbidity was similar for both the adults with, and without, Down syndrome, though, as expected, there were

some differences in the pattern of conditions. The pattern of multimorbidity also differed from the general population, hence findings from the general population are not transferrable; multimorbidity among people with intellectual disabilities requires specific study. Multimorbidity was prevalent across the entire adult life course, unlike the general population in whom it increases over the age of 50 years, thus health care availability is equally essential at all ages for those with intellectual disabilities. In the UK, secondary healthcare, and medical education is organised around single conditions, yet management of multimorbidity is far more complex.

Output 10 (55) investigated standardised mortality ratios (SMRs), causes of death and avoidable deaths in adults with intellectual disabilities, with and without Down syndrome. This study reported a standardised mortality ratio (SMR) of 2.24 (1.98, 2.49) for adults with intellectual disabilities; for those with Down syndrome it was 5.28 (3.98, 6.57), and for adults without Down syndrome it was 1.93 (1.68, 2.18); male=1.69 (1.42, 1.95), female=3.48 (2.90, 4.06). SMRs decreased as age increased. SMRs were higher for most ICD chapter groupings of conditions. Aspiration/reflux/choking was the most common underlying cause of death in adults with intellectual disabilities, followed by respiratory infection. Amenable deaths (29.8%) were double that in the general population (14%). Mortality risk was related to percutaneous endoscopic gastrostomy/tube fed, Down syndrome, diabetes, lower respiratory tract infection at cohort-entry, smoking, epilepsy, hearing impairment, increasing number of prescribed drugs, and increasing age. In view of the different profile of deaths among people with intellectual disabilities, it is not adequate to solely rely on the public health interventions informed by general population studies, even when they are accessible.

Output 9, (56) is one of very few studies that have reported mortality rates and causes among children and young people with intellectual disabilities. Children and young people in Scotland with intellectual disabilities have a SMR of 11.6 (95% CI 9.6 to 14), rising to 22-fold on excluding external causes. SMR was 16.6 (95% CI 12.2 to 22.6) for female pupils and 9.8 (95% CI 7.7 to 12.5) for male pupils. Results were more striking for children aged 5 to 14 years (SMR 21.6) than the young people aged ≥15 years (SMR 7.7). Among the children and young people with intellectual disabilities, deaths were higher across several disease categories, with nervous system and respiratory causes of death

being the most common. Pupils with intellectual disabilities were also approximately 3.6 times more likely to experience an avoidable death. These are deaths from causes that would have been amenable to quality health care, such as epilepsy, pneumonia and pneumonitis due to food and gastric contents. It is highly important to identify amenable deaths so that actions can be devised and taken.

Given that this research demonstrated that people with intellectual disabilities have higher rates of death from respiratory conditions compared with the general population, the onset of the COVID 19 pandemic raised concerns that people with intellectual disabilities may be particularly at risk. The studies already undertaken ensured capability within Scotland's datasets to investigate this robustly. Output 1, (57) reported that adults with intellectual disabilities had higher rates of COVID 19 infection, severe infection (resulting in hospitalisation and/or death) (SMR 2.61, 95% CI 1.81 to 3.40) and higher rates of mortality once infected (SMR 3.26, 95% CI 2.19 to 4.32). So, adults with intellectual disabilities had more COVID 19 infections, and worse outcomes once infected, particularly adults under 65 years. This showed that non-pharmaceutical interventions are even more important for carers and care-provider organisations supporting people with intellectual disabilities, in minimising the transmission of COVID 19, than for the rest of the population; and that age cut-offs in use at the time for prioritising COVID 19 vaccination and booster jabs should not be applied to adults with intellectual disabilities who are at higher risk even at younger ages.

As described under the results addressing objective 1, Output 6 reports the establishment of a national e-cohort of people with intellectual disabilities and a comparison group of other people. The e-cohort provides data for future analyses on health and health inequalities to enhance understanding of health profiles and determinants.

3. Development and implementation of a novel methodological approach to inclusive health research practice for people with intellectual disabilities

People with intellectual disabilities experience multiple barriers to inclusion in research, including attitudinal concerns, viability and ethical concerns, communication issues and

systemic challenges in research processes. The outputs from 2 studies aimed to develop and implement a novel, adapted Citizens' jury methodology to gain insights into the views of people with intellectual disabilities on research, challenge existing barriers and generate actionable recommendations for inclusive research practices. The Research Voices project, output 5, (58) explored the views of people with intellectual disabilities on inclusive health research practice, while the second, output 4, (59) considered their participation in academic peer review processes. Both studies were designed to develop and implement strategies to support inclusion in discussions on research priorities, practices and processes.

The first study (58) successfully developed and delivered an accessible Citizens' jury through implementation of a series of adaptations to the conventional method, accommodating the diverse communication and support needs of the intellectual disabilities community. During the developmental phase, each element of the citizens' jury approach was considered to identify adaptations that would facilitate engagement with people with intellectual disabilities. Key adaptations were identified, including reducing the duration of meetings and incorporating frequent breaks, using specialised communication and recall aids such as Talking Mats, 'I want to speak' cards and graphic facilitation, providing accessible presentations from expert witnesses, and allowing support staff to attend. Another crucial adaptation was in empowering jury members to collaboratively set their own research question within the broad field of health research, to confer greater autonomy and control over the project's agenda. A 5-day preparatory programme was developed to build participants' confidence, knowledge, and communication skills. This was followed by implementation of the citizens' jury sessions, which involved structured evidence presentation, questioning, and deliberation. A further 4 workshops were held to support the co-production of project outputs. This included the production of a consensus report (60) of 10 recommendations designed to guide the health research community on how people with intellectual disabilities can influence research development, delivery, and dissemination. The recommendations included the provision of more accessible health information, increased opportunities to participate in research, improved representativeness of the whole intellectual disabilities community, the establishment of an organisation dedicated to supporting inclusion in research, greater influence of people with intellectual disabilities in research funding decision making, and involvement in research ethics committees. Additionally, participants reported that they valued the opportunity to deliberate on research practices, and that participation had

increased their confidence and enabled them to build new social connections. They also said that they valued the opportunity to contribute to improving inclusive research practices. Expert witnesses also reported positive experiences and highlighted the quality and insightful nature of the questions posed by the jury, acknowledging their personal perspectives.

The second, related study, specifically investigated the feasibility and value of involving people with intellectual disabilities in the academic peer review process for the British Journal of Learning Disabilities. Participants were recruited from support and advocacy organisations and groups and invited to participate in the Citizens' jury. Jurors in this project demonstrated a clear understanding of inclusive research, with some drawing on extensive prior experience of working with research teams. Following the jury several systemic challenges within academic publishing were identified and proposed. First, the accessibility of information was highlighted as an ongoing issue and even 'easy read' formats were cited as lacking in utility. This led to a strong preference for video and audio summaries of research outputs. Second, jurors expressed frustration with the slowness of academic research processes in translating findings into social change, which was a particular concern for those engaged in activism. Third, concerns were raised regarding the balance between the effort required for peer review and the perceived rewards, whether financial, intrinsic or in terms of impact. Finally, jury members expressed a rejection of the exclusive nature, priorities and timescales of traditional academic writing and publishing.

A set of consensus recommendations was agreed upon by the jury, which challenge traditional academic norms and present a roadmap for engaging people with intellectual disabilities in research dissemination. They agreed that research reviews should be conducted by groups, rather than individuals, in dialogue with authors, rather than anonymously; that research under review should be genuinely accessible and engaging; they expressed a strong preference for engaging in reviews on topics that concerned them and were relevant to their own lives; and underlined the importance of being given sufficient time to process information and engage in discussion. They also argued that it was vital that inclusive reviews included diverse representation, including those with more severe intellectual disabilities. In addition to the recommendations relating to peer review the jurors proposed broader suggestions for improving engagement with research outputs. They called for the development of an accessible research journal targeted at the wider

public, beyond academia and the establishment of accessible forums for learning and discussion about research and its implications.

Outputs 4 and 5 described the development and implementation of an adapted Citizens' jury model to provide insight into the views of people with intellectual disabilities on inclusive health research practices, reflecting on the utility and effectiveness of this adapted model of engagement. Output 5 demonstrates that early investment in planning and development of a structured framework is an important component of successful inclusive practice along with the role of experienced facilitators. However, there is a need to provide consistent and dedicated resources to support inclusive research at all stages of research from setting research agendas to dissemination of results. Output 4 developed this work further to investigate the inclusion of people with intellectual disabilities in the peer review process. This study also demonstrated that the adapted citizens' jury method was effective, and participants valued the experience. However, further adaptations would be needed to ensure representation of a more diverse group of people with intellectual disabilities, including those with more severe intellectual disabilities. Both projects demonstrate that people with intellectual disabilities can engage in informed discussions about the concept of inclusive health research, when given time, expert facilitation and tailored communication support.

Discussion

This thesis presents a comprehensive and cohesive body of work examining the health care and health inequalities experienced by people with intellectual disabilities across the lifespan in Scotland and an exploration of approaches to inclusive research methods. Through a combination of population-level quantitative analyses and methodological innovation the thesis addresses three core objectives: 1. To investigate the health care experienced by people with intellectual disabilities, 2. To investigate the health of people with intellectual disabilities and 3. To develop and implement a novel, inclusive methodological approach to support the active involvement of people with intellectual disabilities in research.

This body of work provides an integrated account of systemic inequalities in health care, service resourcing and utilisation; significant inequalities in health, with high rates of morbidity and avoidable mortality compared to the general population; and important insights into inclusive research method development and conceptualisation that can support addressing the systematic exclusion of people with intellectual disabilities from research. This discussion synthesises these findings, critically evaluates their implications and reviews them in the context of the broader literature. Finally, it will reflect on the strengths and limitations of the included studies, their impact to date, and indicate future directions for research, policy, and practice.

Health care inequalities

Resourcing of intellectual disabilities services

An important dimension of equality is access to high quality, well-funded health and care services at the point of need. The longitudinal study on health and care expenditure demonstrated that the despite high levels of health and care needs of people with intellectual disabilities, a real term decrease of 3.14% in health and social care expenditure on targeted services were observed across the study period. Whilst per-capita expenditure on intellectual disabilities services appeared substantial, the overall proportion of health care funding allocated to these services declined relative to that allocated to general population services. Furthermore, substantial regional variation in expenditure was also observed, with remote and rural areas experiencing disproportionate reductions in

intellectual disabilities health care funding, exacerbating geographical inequities in service access. These findings raise questions about the capacity of these services to meet the complex health and care needs of this population as well as they did in the past. Evidence from a recent scoping review (61) highlights the critical role of targeted funding initiatives in improving health care access, through the provision of a specialist workforce that can facilitate reasonable adjustments, improve engagement of people with intellectual disabilities in decision-making, and support personalised care. Recent qualitative studies have investigated the impact of budget reductions in health and social care spending in England following the introduction of austerity measures on the lives of people with intellectual disabilities reporting negative impacts on the health and well-being (62) and the loss of care. (63) Another study investigating expenditure on intellectual disabilities health services across English local authorities (64) reported significant spatial variation in expenditure with higher rates of spending concentrated in urban areas. However, current studies on resource allocation are limited and out of date. The systemic inequalities in resource allocation reported in this study are significant and indicate the likely impact of budget cuts on the quality of specialist health and care provision, including limited access to comprehensive care and gaps in service provision. The spatial inconsistencies in funding allocation underscore the structural nature of health inequalities, where geographical location intersects with systemic underinvestment and fragmented service planning, potentially leading to unequal experiences and outcomes for people with intellectual disabilities. This study now provides crucial baseline expenditure data for the whole country against which subsequent trends/changes can be measured. Such studies can also provide vital context to understanding the persistent inequalities across all domains of life for people with intellectual disabilities. For example, highlighting how spatial and temporal variation in the resourcing, quality, and accessibility of these specialist services can compound health inequalities and potentially result in delayed diagnoses, limited access to appropriate treatments and preventative care, social isolation, and poorer quality of life and health outcomes for those living in under-resourced areas.

Psychotropic prescribing practices and anticholinergic burden

Access to appropriate, high-quality and equitable health care is vital to addressing inequalities in health. The studies on prescribing patterns provide additional evidence that good practice guidelines are not being followed for people with intellectual disabilities. Outputs 2 (51) and 3 (50) reported persistently high rates of psychotropic prescribing for

both adults and children with intellectual disabilities. Output 3 showed that over half of the adult intellectual disabilities population were prescribed psychotropic medications, with rates of antipsychotic prescribing higher than the clinically assessed prevalence of severe mental ill-health. At time 1 42.2% of those who were prescribed antipsychotics had no mental ill-health or problem behaviours. For the longitudinal cohort, those with problem behaviours assessed at time 1 were over 6 times more likely to experience antipsychotic prescribing and more than 3 times more likely to be prescribed antidepressants at time 2. These results suggest that people with intellectual disabilities were routinely prescribed antipsychotics, and other psychotropic medications off-licence, in the absence of clear clinical indication. Analysis of data, at two time points, over 10 years showed no significant decrease in antipsychotic prescribing for those that were prescribed antipsychotics at time 1, indicating that once prescribed antipsychotics these were unlikely to be reduced or removed. However, overall antipsychotic prescribing rates did reduce at time 2, indicating a reduction in newly initiated antipsychotic prescriptions. These findings indicate that further action is required to support deprescribing of antipsychotic medications and rigorous monitoring of the potential adverse effects of these medications, (65) given the evidence on the potential risks of long-term prescribing. (66) This study also reported that there was a significant increase in the prescription of antidepressant medications, more so for women and people with mild intellectual disabilities. The continued high levels of use of antipsychotics, despite guidelines recommending their withdrawal in the absence of severe mental ill-health, indicate that these medications are being used to manage problem behaviours. The increases in antidepressant prescribing that were observed may reflect better recognition of mental ill-health in the population with intellectual disabilities; however, it also underscores the need for comprehensive, non-pharmacological approaches to health care delivery, such as suitably modified Cognitive Behavioural Therapy (CBT) or Positive Behavioural Support (PBS).

A recent study of psychotropic prescribing in adults with intellectual disabilities in Ireland aged ≥ 40 also reported a significant increase in antidepressant prescribing, from (28.6-35.8%) and no change in antipsychotic prescribing rates. (67) Significant increases in antidepressant prescribing in the general population have been widely reported internationally. A recent retrospective, longitudinal study of antidepressant prescribing trends in England reported that prescriptions increased from 377 to 1266 items per 1000 population between 1998 and 2018. (68) Another, Italian study observed a similar rate of

increase of 36.7% between 2008 and 2022. (69) These increases have been attributed to diverse factors including increased awareness of mental health conditions and diagnostics (70) and longer term prescribing. (71) However, the drivers and implications of the increase in antidepressant prescribing observed in our study on prescribing in the intellectual disabilities population are unclear and require further research (72).

A further recent (2025) study from the Czech Republic on prescribing in the intellectual disabilities population have replicated the trends reported in output 3, observing significant associations between psychotropic prescribing and problem behaviours, long-term use of psychotropic medications and increases in psychotropic prescribing overall (55% in 2010 to 59% in 2022), including significant increases in antipsychotics (35% to 37%), antidepressants (17% to 25%) and mood stabilisers (23% to 28%) over the same period. (73) A recent systematic review and meta-analysis (74) of psychotropic prescribing in adults with intellectual disabilities up to 31 December 2021, reported a pooled prevalence of 41% (95% CI, 35%-46%) of psychotropic, 31% (27%-35%) for antipsychotic and 14% (9-19%) for antidepressant prescribing in this group. However, significant heterogeneity across the 24 included studies was reported, reflecting potentially significant methodological and cohort variation.

The literature on longitudinal psychotropic prescribing rates in children and young people with intellectual disabilities is even less well developed. Output 2 contributes to addressing this gap, reporting elevated prescribing rates among children and young people with intellectual disabilities who were more likely to be prescribed antipsychotics (OR 16.85) and antidepressants (OR 2.28) compared to their peers with no intellectual disabilities. Whilst, prescribing of antipsychotics and antidepressants increased for children and young people with and without intellectual disabilities between 2010 and 2013 the magnitude of the increase was greater in those with no intellectual disabilities. We do not know if these results suggest that children and young people with intellectual disabilities are receiving the same treatment advances (i.e. better identification of mental ill-health leading to increased levels of clinically indicated prescribing) over time as children and young people without intellectual disabilities, or if children and young people with intellectual disabilities were being overprescribed psychotropics in the past and this was addressed to

an extent in this time period, or if overprescribing is increasing in children and young people without intellectual disabilities.

Recent studies have also reported significant increases in antidepressant prescribing in children and young people in the general population. One Irish retrospective, cross-sectional study of psychotropic medications reported a 32% increase in psychotropic prescribing between 2017 and 2021. (75) Another retrospective cohort study of Australian primary care prescribing of psychotropic medications also reported significant increases in antidepressant (42.8%) and antipsychotic (62.8%) prescribing rates for children and young people between 2011 and 2018. (76) Only one other study was identified that compared prescribing rates for those with and without intellectual disabilities. This retrospective cohort study of prescription use in young people aged 15-24 years between 2010-2019 in British Columbia reported higher odds of psychotropic prescribing for all psychotropic medications in the intellectual disabilities group. Those with intellectual disabilities were almost 11 times more likely to be prescribed antipsychotics (OR 10.932, 10.576-11.300) and more than 4 times more likely to be prescribed antidepressants (OR 4.316, 3.912 – 4.186) (77) To date no studies, other than the one I report on have investigated longitudinal psychotropic prescribing rates in all school age children and young people with intellectual disabilities, nor for a whole country population.

These findings are important because they highlight a significant and under-researched area of health care inequality with potentially long-term consequences for the health of children and young people with intellectual disabilities. The high rates of psychotropic prescribing in this population, combined with the limited evidence on their efficacy and safety in younger age groups should be further investigated. One recent systematic review and meta-analysis investigating the use of second-generation antipsychotics in children and young people in the general population reported weight gain, increased BMI and metabolic alterations which may predispose those on these medications in childhood to adult obesity. (78) Another systematic review and meta-analysis of metabolic adverse effects of antipsychotic use in people with intellectual and developmental disabilities reported that antipsychotic prescribing is highly associated with clinically significant weight gain with the majority of included studies focusing on children and young people. (79) Further research is needed to understand the potential impact of early exposure to psychotropic

medications and to support assessment of their associated risks for adverse health outcomes. This evidence is critical for enabling prescribers to make evidence-informed decisions that appropriately balance potential benefits against associated risks. Further research is needed to investigate prescribing practices and to uncover the reasons why people with intellectual disabilities are not being withdrawn from certain medications, this could include qualitative studies involving prescribing clinicians.

In the case of adult antipsychotic prescribing these results reflect limited adherence to best practice guidelines recommending the reduction or cessation of antipsychotic prescribing in the absence of severe mental ill-health. Guidelines on prescribing of psychotropic drugs (80) which include medications with high anticholinergic burden for the population with intellectual disabilities indicate the need for improved access to high quality health and care services that facilitate comprehensive, person-centred assessments which prioritise non-pharmacological, therapeutic interventions and regular medication reviews. The high rates of psychotropic prescribing, in both adults and children, and prescribing of medications with high levels of anticholinergic burden observed in the included studies indicate that people with learning disabilities are not receiving the recommended standards of care. These patterns of prescribing should be further investigated and monitored given the correlation of psychotropic prescribing with poor health outcomes in both adults (74) and children and young people (17, 53, 78). Output 8 directly addresses this by investigating the prevalence and sources of high anticholinergic burden, where psychotropic medications were a significant contributor, in the intellectual disabilities population. Irrespective of age, adults with intellectual disabilities had a higher risk than their peers with no intellectual disabilities of being prescribed antipsychotics, antiepileptics and hypnotics/anxiolytics, as well as exposure to higher rates of psychotropic polypharmacy. This finding further reinforces the need for recommended medication reviews, which should also consider anticholinergic side effects and cumulative burden. However, current research on the adverse effects of anticholinergic burden is lacking, and most studies have investigated their effects in older people and those with dementia but no intellectual disabilities. (81)

The studies on health care inequalities in this thesis report persistent and systemic inequalities affecting people with intellectual disabilities across Scotland. Despite the

substantial and frequently, complex health and care needs of people with intellectual disabilities there has been a real term decline in overall expenditure and a proportional reduction in the allocation of resources to specialist intellectual disabilities services. The potential impact of this disinvestment is exacerbated by the regional disparities in service funding. Reductions in the funding of specialist intellectual disabilities health and care services are likely to decrease the quality of care and support services and exacerbate health inequalities further. In addition to the investigation of patterns of resource allocation this thesis also explores patterns of prescribing as a proxy indicator of access to health care provision for people with intellectual disabilities. The prescribing studies demonstrate that people with intellectual disabilities are potentially experiencing poor quality of health care. Persistently high rates of psychotropic prescribing have been associated with increased risk of adverse health outcomes. These findings reflect underlying deficiencies in service provision, particularly in relation to the limited availability of specialist services and non-pharmacological interventions. The need for regular medication reviews, metabolic monitoring and action to reduce long-term prescribing to avoid further exacerbating the mental and physical health inequalities experienced by people with intellectual disabilities across the lifespan is also indicated.

Health inequalities

The studies addressing the second objective of this thesis investigated the prevalence of physical and mental ill-health, multimorbidity, and mortality across the lifespan for people with intellectual disabilities, as well as the impact and outcomes of COVID-19. These studies draw on large, nationally representative datasets, addressing previous limitations in the existing literature. Output 6 reports on the successful establishment of a national e-cohort of people with intellectual disabilities from Scotland's census 2011, providing a unique platform for future analyses of health and health care inequalities. This resource has already been used to investigate health and mortality outcomes, (82, 83) with other research in progress. Output 11 investigated the prevalence of physical health conditions and multimorbidity in adults with intellectual disabilities. It found that 98.7% of participants had multimorbidity, with a mean of 11.04 physical health conditions. This pattern differed from that of the general population, being prevalent across the life course, unlike in the general population, where it increases after the age of 50. This reinforces the need for tailored models of care that provide continuity of care and are responsive to the

specific health needs of the intellectual disabilities population across the lifespan. It also highlights the need for restructuring of health and social care systems to enhance coordination and support the development of integrated, multidisciplinary care pathways for people with intellectual disabilities and complex care needs. The studies on mortality further address significant methodological gaps in the research. Previous studies indicated that people with intellectual disabilities were at increased risk of death, with most reporting SMRs between two and five times higher than the general population. (27) Output 10 provides robust data for adults with intellectual disabilities in Scotland, reporting an SMR of 2.24, which was even higher for those with Down syndrome (5.28). Other factors associated with increased mortality risk included diabetes, respiratory infection, epilepsy, and the total number of different medications prescribed. Neighbourhood deprivation was not found to be a predictive factor. This study also found that amenable deaths were double that of the general population (29.8% vs. 14%), highlighting that deaths could have been avoided through the provision of effective support and health care interventions. More recent studies have replicated these results. A large, English observational study (84) of linked primary care data reported SMRs for males and females with intellectual disabilities of between 2.91 and 3.51, with no relationship between lower neighbourhood deprivation and mortality risk. SMRs for epilepsy and aspiration pneumonia were highest. A systematic narrative review (85) of mortality, predictors and causes in people with intellectual disabilities reported that the risk of mortality was between 3 and 4 times higher than in the general population, with deaths from respiratory causes and epilepsy being particularly common. However, these studies did not disaggregate results for children and young people from those of adults, despite differences in their rate and causes of mortality. Output 9 is one of the few studies investigating mortality rates in children and young people with intellectual disabilities, and reported an even higher SMR of 11.6, finding that children and young people with intellectual disabilities were 3.6 times more likely to die from potentially avoidable causes than their peers with no intellectual disabilities. These concur with those of another, more recent population-level study (86) on the rates and causes of mortality in children and young people (aged 5-24 years), which reported an overall SMR of 10.7 (9.47-12.1) for all causes and 5.17 (4.19-6.37) for avoidable causes, with the most common causes of death being respiratory or epilepsy.

The finding that respiratory conditions were one of the most significant causes of death for people with intellectual disabilities informed concerns raised by the COVID-19 pandemic.

Output 1 robustly demonstrated that adults with intellectual disabilities had higher rates of COVID-19 infection, severe infection, and mortality once infected, particularly those under the age of 65. These findings provided timely evidence for policy, leading to the early prioritisation of people with intellectual disabilities for COVID-19 vaccination in Scotland.

The health inequalities reported in this thesis are not inevitable, rather they arise from a complex interplay of social, structural and systemic factors. Contributing influences include barriers to reciprocal communication, lack of awareness of the health needs of people with intellectual disabilities among health and care professionals, diagnostic overshadowing, lack of reasonable adjustments, reliance on carers to mediate access to care and inadequacies in the provision and accessibility of health information. These are all modifiable and reflect broader equity issues in relation to service design and delivery.

This research is significant because it addresses previous gaps in the literature with few generalisable studies that drew on population-level data. These studies demonstrate that people with intellectual disabilities experience differential health outcomes compared to the general population, very high rates of multimorbidity, mental ill-health and premature/avoidable mortality across all ages. The pattern of morbidities and the main causes of death also differed for the intellectual disabilities population compared to other people. Respiratory conditions were a major cause of death, and a high proportion of deaths of those with intellectual disabilities were potentially avoidable. Additionally, people with intellectual disabilities were more likely to experience severe outcomes during the COVID-19 pandemic. The successful establishment of a national e-cohort provides a unique and valuable platform to further investigate the health inequalities experienced by this group.

Inclusive research methods

The selected inclusive research studies demonstrate the value of adapting existing deliberative public engagement methods, such as citizens' juries, to support including people with intellectual disabilities in deliberations on inclusion in research. Both studies,

make a significant contribution to inclusive research by demonstrating the viability and wisdom of involving people with intellectual disabilities in discourse around academic processes, particularly within the context of health research. The overall message from these novel studies was that, whilst people with intellectual disabilities understand the value of research, they do not want inclusive research practices to simply replicate existing academic structures. Instead, they wish to contribute in ways that fundamentally challenge and transform these systems to better serve their interests and reflect their lived experiences. There was a strong emphasis on the need for inclusive research content to be personally relevant to participants in both projects. Other recent studies have also identified this preference for participation in research that has a personal connection to the lives of participants with intellectual disabilities. (87) Their contributions, via these projects, were to recommend greater dialogue between researchers and the researched community, and more relevant and accessible engagement with the outputs of research. This is closely aligned with the work of proponents of 'epistemic justice' in knowledge production, who argue that value is placed on propositional knowledge over that gained through experience, to the inevitable exclusion of people with intellectual disabilities, who may not be able to express their knowledge verbally. (88)

In their deliberations on the potential role of inclusive peer review, participants expressed a strong preference for group-based reviews conducted in dialogue with authors that would signify a move towards more collaborative, supportive interactions, if adopted. This is in direct contrast to the anonymous and solitary nature of traditional academic peer review. This approach is consistent with the British Journal of Learning Disability 'In Response' method, which empowers people with intellectual disabilities to 'speak back to' academic research directly, prioritising personal relevance and community benefit over conventional academic metrics.

The importance of accessibility, through the use of diverse formats, was consistently highlighted as a significant gap in traditional academic dissemination practices. The Jurors' scepticism of complex academic language and their desire to engage with topics directly impacting their lives is reflective of a pragmatic and activist-oriented view of research.

Whilst the adapted citizens' jury method proved effective in facilitating deep engagement with complex issues for participants with mild and moderate intellectual disabilities, there

was an acknowledged lack of representation from those with more severe intellectual disabilities. The juror's concerns about the exclusion of people with more severe intellectual disabilities and the overrepresentation of established self-advocacy groups also align with broader calls for epistemic justice in inclusive research practices. This has led other researchers to argue that modifications to existing inclusive research methods are insufficient to include those with more severe intellectual disabilities. (89) and that partial representation is harmful and should be addressed through the development of approaches tailored to diverse subgroups. (90)

These projects provide a robust foundation for democratising research and working towards greater epistemic justice by demonstrating that people with intellectual disabilities can engage with and offer insightful critiques of academic processes. Their recommendations present actionable solutions to support moving from tokenistic involvement towards fostering genuine dialogue and ensuring that research serves, and is informed by the experiences of, people with intellectual disabilities. The emphasis on capacity building, well-being and social change as motivating factors for participants also calls for a re-evaluation of academic incentives and dissemination strategies and moving away from producing answers to research questions towards fostering empowering, life-improving discussions on the outputs of research.

Strengths and limitations: quantitative studies

A major strength across the body of work presented in this thesis is the use of large, nationally representative datasets, often including whole-country populations of people with intellectual disabilities. The comprehensive coverage of administrative data sources, such as Scotland's Census 2011, education records, health records and expenditure data allowed for robust sample sizes, minimisation of selection bias and enhanced generalisability of findings to the broader population with intellectual disabilities. The novel use of Scotland's Census 2011 as a means of population ascertainment for the population with intellectual disabilities represents a particular methodological strength, uniquely enabling the identification of this otherwise hard to identify population at a national level. Record linkage success rates were high (typically exceeding 90%), and the inclusion of longitudinal data, for some studies, provided valuable insights into temporal trends and outcomes over time. The inclusion of a wide range of administrative data

enabled the examination of diverse outcomes including mortality, morbidity, health service use and medication exposure. Additionally, a subset of the included studies benefitted from comprehensive baseline health assessment conducted by intellectual disabilities specialists, contributing rich, clinically verified information on participants.

Several limitations that are inherent to administrative data research must also be acknowledged. Whilst administrative records provide breadth and depth of coverage, they often lack clinical precision, for example, data on the severity or cause of intellectual disabilities are often missing. The reliance on routinely collected information, such as death certificate data or general practice records, raises potential questions about misclassification, under-ascertainment or inaccuracies in clinical coding, which may affect the reliability of some outcomes. In some analyses, small cell sizes limited the reliability of subgroup outcomes, particularly when stratifying outcomes by age. Furthermore, variations in data completeness, quality and recording practices across different health boards, local authorities or time periods may have introduced unmeasured bias. Collectively these strengths and limitations highlight both the value and constraints of using administrative data in research about the population with intellectual disabilities when undertaking large-scale, population-based investigations with minimal burden on data providers.

Strengths and limitations: inclusive studies

The 2 studies included which focus on the development of inclusive research methodologies demonstrate a strong commitment to working collaboratively with people with intellectual disabilities to understand their views in relation to involvement in different stages of the research process. The adapted citizens' jury method facilitated meaningful dialogue and generated practical, participant driven recommendations aligned to wider inclusive research frameworks and theoretical perspectives, contributing valuable insights into how inclusive research should be reconceptualised to better reflect the perspectives of people with intellectual disabilities. This approach also highlighted the participants' desire to extend inclusion beyond the jury itself, emphasising the importance of representing diverse voices within the intellectual disabilities community.

Several limitations were also observed, including the method's reliance on support staff to facilitate access to meetings which introduced complexity in distinguishing authentic voices of participants from those of their advocates, potentially obscuring power dynamics that are critical in any inclusive research. A clearer framework, delineating the role and influence of support workers would enhance future methodological rigour.

Impact

This thesis makes a significant contribution to the knowledge base on health care, health inequalities, and inclusion in research of people with intellectual disabilities, contributing knowledge to support clinical practice, public health policy, and system change. It addresses longstanding methodological gaps in the existing literature through the use of robust, population-based administrative data, novel methodological approaches to inclusive research, and a translational focus, bridging the gap between evidence and policy. The studies included in this thesis were conducted with the explicit aim of improving the health and care of people with intellectual disabilities through the provision of high-quality research that can inform national and local policy and practice. Thus, impact and knowledge exchange have been central to the design, dissemination, and application of the research included in this thesis.

Specific impact examples demonstrating how the studies included in the thesis have already contributed to advancements in the field of intellectual disabilities, across the lifespan, are described below.

At the beginning of the COVID-19 pandemic, there was no information on the potential impact and risks of the virus to people with intellectual disabilities. Emerging data on the heightened risks to older people and other vulnerable groups rapidly informed public health responses, including vaccine prioritisation. However, early calls to include all people with intellectual disabilities in the prioritisation schedule were initially rejected despite respiratory causes contributing to their early and avoidable deaths, due to a lack of COVID-19-specific data.

In May 2020, following receipt of approval for the national e-cohort study, I was advised by Public Health Scotland of potential delays in accessing data due to prioritisation of COVID-19 studies. Recognising a critical gap in the knowledge, I proposed the incorporation of COVID-19 laboratory data into the e-cohort to enable investigation of the infection and mortality risks to those with intellectual disabilities. To progress this, I convened rapid discussions with the intellectual disabilities lead in Scottish Government and the Director of Statistical Services at National Records of Scotland, focusing on the hypothesis that people with intellectual disabilities, across all age groups, were likely to experience increased risk of severe outcomes due to their complex morbidity profile. Approval for this amendment was secured, analysis commenced, and preliminary findings were published (91) on 9 February 2021 to expedite dissemination of the results. Following publication, the co-conveners of the Scottish Parliamentary Cross Party Group on Learning Disabilities formally cited the findings in correspondence to the Cabinet Secretary for Health & Sport, advocating for the inclusion of all people with intellectual disabilities in vaccine priority group 6. On 22 February 2021, Scotland's First Minister publicly announced that all people with intellectual disabilities would be added to priority group 6 for COVID-19 vaccination. This sequence of events demonstrates the influence of my timely data-driven research on national health policy, contributing to the improved protection of people with intellectual disabilities during a public health emergency.

Findings from the study investigating mortality in children and young people with intellectual disabilities were disseminated across a range of policy and practice forums, including via a presentation to the team responsible for the planning and implementation of the National Hub for Reviewing and Learning from the Deaths of Children and Young People. The primary objective of the National Hub was to establish a standardised, high-quality and consistent methodology for the review of all child and young person deaths in Scotland. As part of this process, a core review dataset was developed for use by NHS health boards and local authorities to ensure standardisation in the collection of data when reviewing deaths. During these engagements with the presentation of research findings I was able to highlight the potential value of incorporating minor adaptations to the core dataset template that would enable disaggregation of intellectual disabilities as a potential contributing factor in the death of a child or young person. This direct engagement and dissemination of research with the National Hub implementation team resulted in a change to include intellectual disabilities as a specific data field within the review dataset when the

hub was officially launched by the Scottish Government in 2021. As a result, intellectual disabilities are now systematically recorded and reported during routine reviews of child deaths. The first national data overview report, (92), which summarised national child death data between 1 October 2021 and 31 March 2023, identified that over 25% of the deaths of children and young people that were reviewed were recorded as having a developmental impairment or intellectual disability as a contributory factor. This development represents an important advancement in national surveillance, providing greater visibility of the potential contribution of intellectual disabilities to child mortality and enabling more targeted policy and practice responses to address preventable deaths in this population.

The establishment of Scotland's national e-cohort of people with intellectual disabilities linked to administrative health and care data represents a significant methodological innovation with academic and policy impact. This e-cohort provides the academic infrastructure to investigate health, mortality, and health care experiences of people with intellectual disabilities at a whole population level, facilitating the generation of robust, generalisable evidence, directly relevant to policy and practice. Since its establishment, the e-cohort has supported a range of studies investigating mortality across the lifespan, cancer incidence and mortality rates in adults, and hospitalisations and deaths due to ambulatory care sensitive conditions. The creation and application of this e-cohort has thus established a sustainable research resource that has the potential to continue to produce research that can directly inform policy decisions, guide service improvements, and contribute to reducing inequalities experienced by people with intellectual disabilities.

Implications for research, policy, and practice

The research presented in this thesis has important implications for future research, health policy, and clinical practice. Collectively, these studies reinforce the need for systemic reform to improve the coordination and integration of health and social care services for people with intellectual disabilities, ensuring more seamless, person-centred and equitable support across services. They highlight persistent inequalities in service access and quality, resource allocation across the lifespan, and health outcomes, indicating clear areas for evidence-informed action.

The thesis highlighted a need for further research that addresses the complex and intersecting health challenges experienced by people with intellectual disabilities. Further studies should build on the methodological advances described here, particularly those involving population-based cohorts and longitudinal data linkage to better understand the trajectories of health, health care use, and mortality.

Further research is needed to understand the clustering and trajectories of multimorbidity in the population with intellectual disabilities. While general population studies have increasingly recognised the importance and pattern of multimorbidity, few studies have investigated the distinct clustering of multimorbidity among people with intellectual disabilities. Similarly, the health effects of long-term exposure to psychotropic medications are under-researched in people with intellectual disabilities. The evidence on prescribing practices raises questions about the quality and safety of health care. The long-term use of antipsychotics without clear clinical indication and the rising use of antidepressants reinforce the need for regular medication reviews. The significant risk of anticholinergic burden identified highlight the need for regular reviews that also take account of the cumulative effects of medications in this group. There are currently no clinical trials evaluating the potential benefits or harms of antidepressant prescribing in this group. The high rates of psychotropic prescribing, polypharmacy, and increased anticholinergic burden observed in the included studies highlight an important gap in the evidence base around the cumulative health risks associated with long-term prescribing in this group. Most studies on anticholinergic burden to date have focused on older adults in the general population, leaving a significant gap in understanding the impact on adults, of all ages, with intellectual disabilities.

The thesis also contributes important knowledge to the field of inclusive research, demonstrating that people with intellectual disabilities can meaningfully engage in, and deliberate on, complex research processes when skilled facilitation is provided. However, further work is needed to ensure that people with more severe intellectual disabilities are not excluded. This will require further methodological innovation as well as sustained resourcing of coproduced research practice. Future research is needed to further investigate inclusion of people with intellectual disabilities in research agenda setting,

conceptualisation of the concept of inclusive research and inclusive knowledge production, study design, and dissemination and communication of research outputs.

The evidence presented in this thesis has important implications for health and social care policy and practice. The findings underscore deficits in current approaches to health care management, which are based predominantly on single condition pathways. The evidence clearly shows that people with intellectual disabilities have complex and often overlapping health needs. This thesis reinforces calls for more integrated, multidisciplinary care pathways that are responsive to and informed by the specific health needs of people with intellectual disabilities. This approach will also require better digital infrastructure to support the sharing of electronic patient records and collaborative health care planning.

Conclusions

This thesis provides robust evidence of substantial and persistent disparities in the provision, utilisation and accessibility of health and care services for people with intellectual disabilities compared to the general population across the lifespan. These systemic inequalities are potentially both a reflection of and a contributing factor to the significant physical and mental health inequalities that are documented in the included studies. Importantly, this body of work also demonstrates that the application of novel and inclusive research methodologies can facilitate the meaningful involvement of people with intellectual disabilities in complex research, ensuring that their perspectives and priorities inform both evidence generation and communication of research. The research has had a tangible policy impact, contributing to immediate, quantifiable change: influencing the Scottish Government's COVID-19 vaccination prioritisation policy, securing the inclusion of an intellectual disabilities identifier in a key Scottish child health dataset, and delivering a dedicated research resource to enable future national-level Scottish studies on intellectual disabilities health and health care.

Sustained and meaningful improvement in the health and care of people with intellectual disabilities will require continued investment in high-quality and inclusive research across these and related areas. Such research is essential not only for addressing the persistent gaps in evidence but also for informing policy, guiding service development and ensuring

that future interventions are responsive to the needs and rights of all people with intellectual disabilities.

Personal reflections

My research journey has deepened my appreciation of both the potential and the limitations of routinely collected administrative and population level data for understanding the lives of people with intellectual disabilities. Although I began this work motivated by the power of large-scale datasets to address evidence gaps and to support equitable, evidence informed policy, sustained engagement with these sources has also highlighted important limitations. Working with these datasets has highlighted issues of population ascertainment, service and diagnostic access, recording and data completeness, and limited granularity, demonstrating that no single administrative or census source can provide a full or unbiased account of this heterogenous population. These experiences have shifted my focus toward understanding how data systems themselves shape what is visible, measurable and actionable, prompting more critical reflection on the structural and methodological foundations of this kind of research.

This growing awareness has in turn shaped my future aspirations as both a researcher and a policy advocate. Recognising the limitations and inequities embedded within existing data infrastructures has strengthened my commitment to more inclusive, participatory and person-centred approaches to data generation, governance and use. I am increasingly motivated to contribute to digital and data innovations that actively reduce bias and exclusion, with a focus on data quality improvement. This is grounded in the principle that systems designed to work well for people with intellectual disabilities tend also to be more robust, accessible and beneficial for wider populations. These reflections have not only shaped the analyses presented in this thesis but have also crystallised a long-term commitment to improving the quality, integrity and justice of the data on which research, policy and practice depend.

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Appendices

COVID-19 infection and outcomes in a population-based cohort of 17 203 adults with intellectual disabilities compared with the general population

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ABSTRACT

Background Adults with intellectual disabilities (ID) may be at higher risk of COVID-19 death. We compared COVID-19 infection, severe infection, mortality, case fatality and excess deaths, among adults with, and without, ID.

Methods Adults with ID in Scotland's Census, 2011, and a 5% sample of other adults, were linked to COVID-19 test results, hospitalisation data and deaths (24 January 2020–15 August 2020). We report crude rates of COVID-19 infection, severe infection (hospitalisation/death), mortality, case fatality; age-standardised, sex-standardised and deprivation-standardised severe infection and mortality ratios; and annual all-cause mortality for 2020 and 2015–2019.

Findings Successful linkage of 94.9% provided data on 17 203 adults with, and 188 634 without, ID. Adults with ID had more infection (905/100 000 vs 521/100 000); severe infection (538/100 000 vs 242/100 000); mortality (258/100 000 vs 116/100 000) and case fatality (30% vs 24%). Poorer outcomes remained after standardisation: standardised severe infection ratio 2.61 (95% CI 1.81 to 3.40) and mortality ratio 3.26 (95% CI 2.19 to 4.32). These were higher at ages 55–64: 7.39 (95% CI 3.88 to 10.91) and 19.05 (95% CI 9.07 to 29.02), respectively, and in men, and less deprived neighbourhoods. All-cause mortality was slightly higher in 2020 than 2015–2019 for people with ID: standardised mortality ratio 2.50 (95% CI 2.18 to 2.82) and 2.39 (95% CI 2.28 to 2.51), respectively.

Conclusion Adults with ID had more COVID-19 infections, and worse outcomes once infected, particularly adults under 65 years. Non-pharmaceutical interventions directed at formal and informal carers are essential to reduce transmission. All adults with ID should be prioritised for vaccination and boosters regardless of age.

INTRODUCTION

The first case of COVID-19 was confirmed in the UK on 24 January 2020 and a pandemic declared by WHO on 12 March 2020. There is global concern that adults with intellectual disabilities may be at higher risk of death from COVID-19, but there are gaps in the evidence.[1] The WHO defines intellectual disabilities as impairments in adaptive, social, and intellectual functioning (IQ<70), requiring daily support, with the onset in the developmental phase (<18 years).[2] People with intellectual disabilities account for <1% of the global population,[3 4] and about 0.5% of adults.[4] They experience substantial health inequalities, including multimorbidity[5] and premature mortality,[6 7] often from respiratory conditions.[8 9] They are more likely to live in congregate settings or be in receipt of social care[10]; recent studies have reported high rates of COVID-19 mortality within multioccupancy residences.[11] However, questions remain as to whether people with intellectual disabilities are more likely to contract COVID-19, and whether they have more severe infections, and higher COVID-19 mortality compared with others.

Existing evidence is inconclusive and has limitations. Five studies reported COVID-19 mortality compared with the general population. One cohort study linked data for over 17 million people on general practitioner registers in England across two waves (1 March 2020–31 August 2020 and 1 September 2020–8 February 2021) investigating COVID-19 hospital admissions and deaths in children and adults.[12] Adults with intellectual disabilities were over five times more likely to have a COVID-19 hospital admission (HR 5.3, 95% CI 4.9 to 5.8) and eight times more likely to die from COVID-19 (HR 8.2, 95% CI 7.2 to 9.4). Results were similar in wave 2 ((4.3, 95% CI 4.1 to 4.6 COVID-19 hospital admission; 7.2, 95% CI 6.4 to 8.1 COVID-19 deaths). They acknowledged their methods of population ascertainment may have led to possible overestimates of HRs. A Canadian cohort study used health records for the province of Ontario to investigate COVID-19 infection rates, hospital admissions and deaths between 15 January 2020 and 10 January 21 for adults with intellectual and developmental disabilities, and with Down syndrome compared with the general population.[13] Infection rates for people with intellectual and developmental disabilities were 1.28 times higher, and 1.42 times higher for those with Down syndrome. For adults with intellectual and developmental disabilities risk of hospitalisation (RR 2.21, 95% CI 1.93 to 2.5) and death (RR 2.23, 95% CI 1.86 to 2.67)) was also higher compared with the general population. Population ascertainment was via hospital health records and therefore excludes those with intellectual and developmental disabilities who may have not been correctly classified or had contact with hospitals. A large US study of data from 547 healthcare organisations investigated risk of COVID-19 infection, hospitalisation and death in people with intellectual disabilities compared with

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the general population, between January 2019 and November 2020.[14] People with intellectual disabilities were significantly more likely to become infected with COVID-19 (3.1% vs 0.9%, p<0.001) and be admitted to hospital (63.1% vs 29.1%, p<0.001). The authors recognised ascertainment of intellectual disabilities in these data was much lower than expected. One non-peer-reviewed study, used three data sources from England to identify adults with intellectual disabilities who definitely or possibly died from COVID-19 from 1 February 2020 to 5 June 2020.[15] Underestimates and uncertainty around figures were acknowledged in the report, due to limitations in data sources. Analysis of two of these data sources resulted in crude COVID-19 mortality rates of 240/100 000 (2.3 times the general population) and 192/100 000 (3.1 times the general population). Welsh general practice records, 1 March 2020–26 May 2020, recorded 31 deaths from COVID-19 among people with intellectual disabilities, equating to a higher age-standardised COVID-19 mortality than observed in the general population.[16]

Five studies reported COVID-19 case-fatality rates, though with biased samples and conflicting results. Electronic medical records from 42 healthcare organisations, across 30 countries, up to 14 May 2020, ascertained >30 000 patients with COVID-19 infections.[17] No difference was found in overall case-fatality rates between the 150 people with intellectual disabilities and those without, but possibly higher case-fatality rates among younger ages.[17] A large US study using private insurance claims, between 1 April 2020 and 31 August 2020, reported higher COVID-19 case-fatality among people with intellectual disabilities compared with those without (OR 2.75, 95% CI 1.66 to 4.56), especially at <70 years of age (OR 3.61, 95% CI 1.89 to 6.93).[18] These results may not be generalisable as they did not include people with public insurance or no insurance. A large self-selected sample of English general practices covering >4 million patients reported higher COVID-19 case-fatality among people with intellectual disabilities over weeks 2–20 of 2020 (OR 1.97, 95% CI 1.22 to 3.18).[11] Another large study of people with intellectual and developmental disabilities living in New York state's residential settings were indirectly compared with the general population, reporting COVID-19 infection rate to be about four times higher, case-fatality almost double, and mortality rate 7.8 times higher.[19] Gleason *et al*[14] reported that people with intellectual disabilities were more likely to die than those in the general population following COVID-19 infection (8.2% vs 3.8%, p<0.001).

A prediction algorithm of COVID-19 mortality risk following infection, was derived (24 January 2020–30 April 2020) and validated (1 May 2020–30 June 20) using a large English primary care database of >8 million patients.[20] It reported higher fatality among adults with intellectual disabilities without Down syndrome (men: HR 1.36, 95% CI 1.14 to 1.60), women: HR 1.36, 95% CI 1.11 to 1.65); and a further increase risk in the small sample of adults with Down syndrome (men: HR 9.80, 95% CI 4.62 to 20.78), women: HR 32.55, 95% CI 18.13 to 58.42). This led to the inclusion of Down syndrome, but not intellectual disabilities, onto the clinically extremely vulnerable list used in the UK.[21]

Adults with intellectual disabilities may be at greater risk of contracting COVID-19 and at greater risk of case fatality, though evidence for both is currently limited. This study investigated in a whole-country adult population with intellectual disabilities, COVID-19 infection, severe infection, mortality, case fatality and excess deaths, compared with adults without intellectual disabilities, during the first wave of the COVID-19 pandemic (24 January 2020–15 August 2020).

METHODS

Population, data sources and record linkage

We used Scotland's Census 2011 to ascertain all adults recorded with intellectual disabilities[4] and a random 5% sample of the general population (without intellectual disabilities or autism) linked to COVID-19 laboratory tests, hospital admissions, and death registrations. Scotland's Census, 2011 provides information on Scotland's population, recorded on 27 March 2011. The Scottish Morbidity Record (SMR) 01 records acute hospital admissions including International Classification of Diseases 10th Revision (ICD-10) diagnoses. Laboratory results from COVID-19 tests are stored electronically within the Electronic Communication of Surveillance in Scotland (ECOSS) database. Personal identifiers from Census 2011 have previously been linked to allow Census 2011 data to be further linked to Public Health Scotland health data for research purposes.[22]

We presented demographic characteristics for adults with and without intellectual disabilities; sex, age and neighbourhood deprivation recorded at the time of the Census in 2011. To reduce the risk of disclosing personally identifiable information age was categorised as adults 18–54 years, 55–64 years and ≥65 years, and Scottish Index of Multiple Deprivation (SIMD) was categorised into two groups: more deprived (deciles 1–5) and less deprived (deciles 6–10). People <18 years in 2011 were excluded from analyses. Analysis was undertaken for the period 24 January (first UK confirmed COVID-19 case)—15 August for each year of the study. When analysing all outcomes, age was calculated at time of event for those who had an event of interest, or age at 24 January in the respective year of interest for people within the denominator population who did not have any events of interest.

Outcomes

Outcomes included: COVID-19 infection (positive COVID-19 test, hospitalisation for COVID-19, or death due to COVID-19); severe COVID-19 infection (hospitalisation for COVID-19 or death due to COVID-19); COVID-19 mortality; COVID-19 case fatality (death from any cause among those who had COVID-19 infection); and excess deaths (difference between average annual all-cause mortality rates 24 January 2015–15 August 2019 and all-cause death rate in 24 January 2020–15 August 2020).

Hospitalisation or death due to COVID-19 was defined by ICD-10 code of U07.1 (confirmed COVID-19 19) or U07.2 (suspected COVID-19 19) in any primary or secondary diagnostic or cause of death position, no timescale was applied.

Analyses

Complete (National Records of Scotland) NRS death data up to 15 August 2020 was available and results from 24 January 2020–15 August 2020 were investigated. Crude rates (per 100 000 people) were compared for those with and without intellectual disabilities using the number of people still alive within each group on 24 January 2020 as respective denominators. Crude outcomes included rates of COVID-19 infection, severe infection, mortality and case fatality. To take into account demographic differences between groups with and without intellectual disabilities, we performed indirect standardisation using sex, age and deprivation. We produced COVID-19-specific standardised mortality ratios (SMRs) and COVID-19-specific Standardised hospitalisation/mortality ratios for 2020. We then produced all-cause SMRs for deaths in 2020 and separately for deaths over the previous 5 years. For each standardisation we used the 5% sample of the census population without intellectual disabilities or autism as the standard population and compared relevant age-sex-deprivation specific rates to the population with intellectual

disabilities to ascertain expected and observed counts. Analyses were repeated within sex, age and deprivation subgroups, standardising each time for the other two variables.

When calculating standardised ratios for deaths and admissions in 2020, we used the respective denominator populations including everyone in the original linked census cohort minus those who died before 24 January 2020. When calculating standardised ratios for deaths between 2015 and 2019 we counted deaths between 24 January and 15 August in each of the respective years to enable an accurate comparison with 2020. People who died before the 24th January in each of the respective years from 2015 to 2019 were removed from the respective denominator populations. For all outcomes age was calculated at time of event (not age in 2011) for those with an event of interest (positive COVID-19 test, hospital admission, death) or age at 24 January in the respective year of interest for those people within the denominator population who did not have any events of interest.

RESULTS

Patient characteristics

Of the 269 771 people (24 264 with, and 245 507 without, intellectual disabilities) included in our Census 2011 cohort, 255 916 (94.9%) were linked to the NRS Population Spine. The linkage rate was 92.9% (n=22 538) among people with intellectual disabilities and 95.1% (n=233 378) of the original 5% comparison sample with no intellectual disabilities or autism. People <18 years old were excluded (figure 1) leaving a final cohort of 213 062 adults (17 203 with intellectual disabilities and 188 634 without intellectual disabilities or autism).

As expected, there were more men than women with intellectual disabilities, who were younger and more likely to live in deprived areas (table 1).

Crude COVID-19 infection rates and outcomes

Adults with intellectual disabilities were almost twice as likely as those without to become infected with COVID-19 (905/100 000 vs 521/100 000) and 2.2 times as likely to have severe infection resulting in hospitalisation or death (538/100 000 vs 242/100 000) or fatal infection (258/100 000 vs 116/100 000) (table 2). Following COVID-19 infection, people with intellectual disabilities were more likely to die 28.5% (95% CI 23.3% to 32.3%) vs 22.3% (95% CI 20.5% to 23.9%).

Age-standardised, sex-standardised, deprivation-standardised COVID-19 outcomes

In 2020, the age-standardised, sex-standardised, deprivation-standardised ratio for severe COVID-19 infection among adults with intellectual disabilities compared with those without was

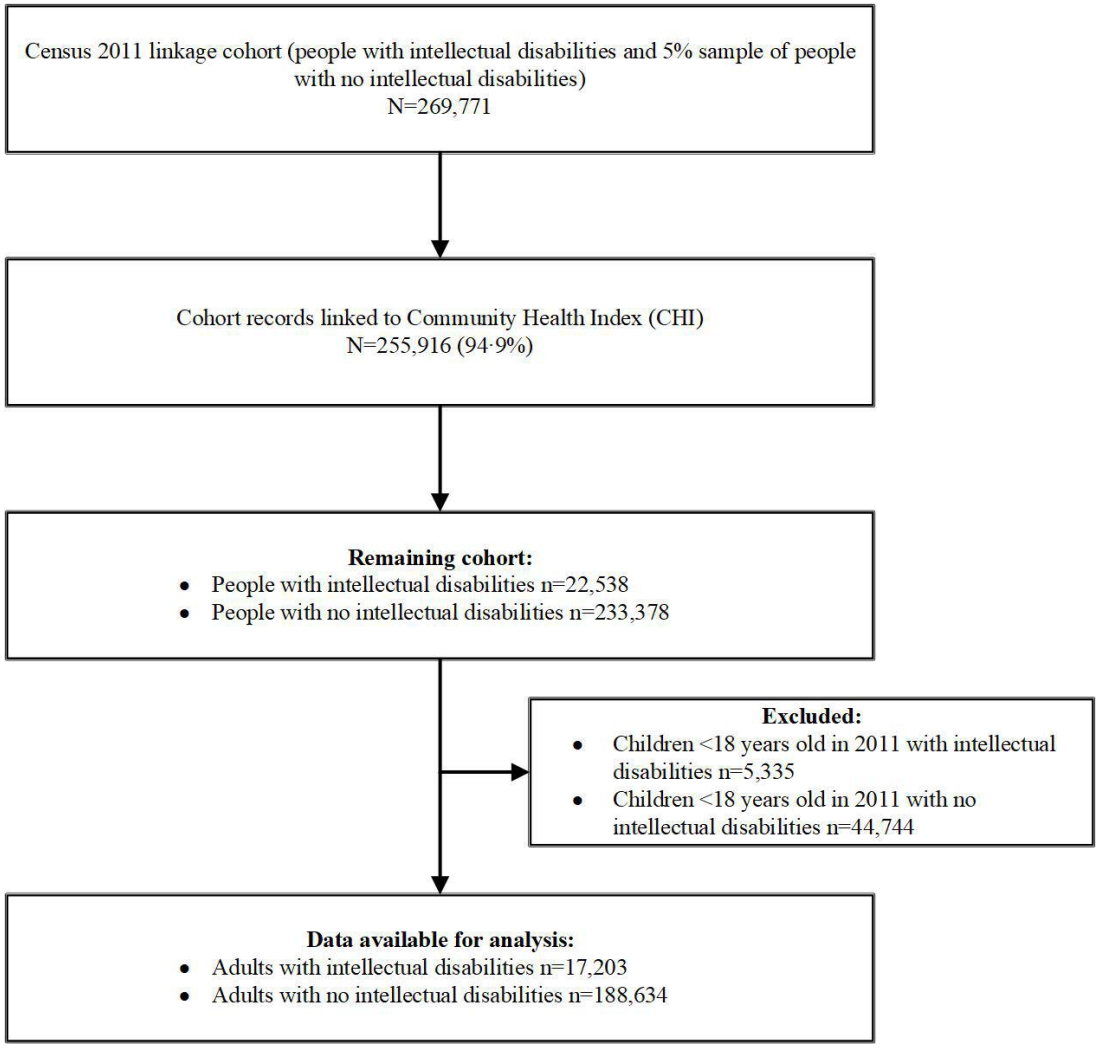

Figure 1 Participant flow diagram.

Table 1 Characteristics of the study population in 2011

	Adults with intellectual disabilities		5% sample of adults with no intellectual disabilities or autism	
	N=17 203		N=188 634	
	n	%	n	%
Sex				
Male	9565	55.6	88 863	47.1
Female	7638	44.4	99 771	52.9
SIMD decile				
1–5 (more deprived)	11 099	64.5	90 406	47.9
6–10 (less deprived)	6104	35.5	98 228	52.1
Age (years) at 2011 census				
Adults ≤54	12 637	73.5	116 534	61.8
55–64	2494	14.5	31 022	16.4
≥65	2072	12.0	41 078	21.8

SIMD, Scottish Index of Multiple Deprivation.

2.61 (95% CI 1.81, 3.40) and for COVID-19 mortality was 3.26 (95% CI 2.19, 4.32) (table 3). The standardised ratios were slightly higher in men than women and in less deprived areas. They were higher in people under 65 years of age and particularly high in the 55–64 years age group where the risk of severe infection, resulting in hospitalisation or death, was more than 7 times higher and the risk of death was over 19 times higher (table 3).

Excess overall mortality

Overall, age-standardised, sex-standardised, deprivation-standardised all-cause mortality ratios for adults with intellectual disabilities were 2.39 (95% CI 2.28 to 2.51) over the 5 years prior to COVID-19, and only slightly higher at 2.50 (95% CI 2.17 to 2.81) in 2020 (table 4). In the subgroup analyses, the largest increase occurred in the 55–64 years age group where the standardised all-cause mortality ratio increased from 4.27 (95% CI 3.87 to 4.67) between 2015 and 2019 to 5.12 (95% CI 3.95 to 6.29) in 2020. However, the CIs still overlapped.

DISCUSSION

Principal findings and interpretation

This is the first comprehensive study investigating COVID-19 infection, severe infection, mortality, case fatality and excess mortality among a whole country's population of adults with intellectual disabilities compared with the general population.

Table 2 Crude outcomes of study populations

	Adults with intellectual disabilities			5% sample of adults without intellectual disabilities or autism		
	N	Crude rate per 100 000	95% CI*	N	Crude rate per 100 000	95% CI*
COVID-19						
Infection†	126	905	747 to 1061	871	521	487 to 556
Severe infection‡	75	538	417 to 660	404	242	218 to 265
Mortality§	36	258	174 to 343	194	116	100 to 133

*95% CIs calculated based on normal approximation.
†Positive COVID-19 test, hospitalisation for COVID-19 or death from COVID-19.
‡Hospitalisation for COVID-19 or death from COVID-19.
§Death from COVID-19 in the population.

Table 3 Age-standardised, sex-standardised, deprivation-standardised COVID-19 outcomes, overall and by subgroup

		Standardised hospitalisation /mortality ratio (95% CI)*	Standardised mortality ratio (95% CI)*
Overall		2.61 (1.81 to 3.40)	3.26 (2.19 to 4.32)
Sex	Male	2.93 (1.85 to 4.02)	3.70 (2.22 to 5.18)
	Female	2.10 (0.96 to 3.25)†	2.63 (1.14 to 4.12)†
Deprivation	SIMD 1–5 (more deprived)	2.27 (1.43 to 3.11)	3.09 (1.92 to 4.25)
	SIMD 6–10 (less deprived)	3.81 (1.74 to 5.88)†	3.90 (1.35 to 6.45)‡
Age (years)§	Adults ≤54	2.83 (1.08 to 4.59)†	7.47 (2.29 to 12.65)¶
	55–64	7.39 (3.88 to 10.91)†	19.05 (9.07 to 29.02)**
	≥65	1.41 (0.67 to 2.15)†	1.51 (0.72 to 2.31) †

*95% CIs calculated based on normal approximation.
†Ratios are based on less than 20 observed events in the intellectual disabilities group.
‡Ratios are based on less than 10 observed events in the intellectual disabilities group.
§Age at time of event.
¶Ratios are based on less than 10 observed events in the intellectual disabilities group and less than 10 observed events in the 5% sample of the general population.
**Ratios are based on less than 20 observed events in the intellectual disabilities group and less than 10 observed events in the 5% sample of the general population.
SIMD, Scottish Index of Multiple Deprivation.

Adults with intellectual disabilities were twice as likely to become infected with COVID-19, 2.2 times as likely to have severe infection resulting in hospitalisation or death, and following COVID-19 infection, had a case fatality of 30% compared with 24% in the general population. The risk of severe or fatal

Table 4 Age-standardised, sex-standardised, deprivation-standardised all-cause mortality, overall and by subgroup

		Standardised mortality ratio*	95% CI†
2015–2019			
Overall		2.39	2.28 to 2.51
Sex	Male	2.23	2.08 to 2.38
	Female	2.60	2.42 to 2.78
Deprivation	SIMD 1–5 (more deprived)	2.08	1.96 to 2.20
	SIMD 6–10 (less deprived)	3.38	3.11 to 3.66
Age (years)‡	Adults≤54	4.55	4.09 to 5.00
	55–64	4.27	3.87 to 4.67
	≥65	1.68	1.57 to 1.79
2020			
Overall		2.50	2.17 to 2.81
Sex	Male	2.50	2.08 to 2.92
	Female	2.51	2.01 to 3.00
Deprivation	SIMD 1–5 (more deprived)	2.18	1.84 to 2.52
	SIMD 6–10 (less deprived)	3.65	2.82 to 4.47
Age (years)‡	Adults ≤54	4.79	3.52 to 6.04
	55–64	5.12	3.95 to 6.29
	≥65	1.55	1.25 to 1.85

*Standardised for age, sex, SIMD (whichever not used to define subgroup); referent to the 5% comparison group.
†95% CIs calculated based on normal approximation.
‡Age at time of event.
SIMD, Scottish Index of Multiple Deprivation.

COVID-19 infection, relative to the general population, was higher among non-elderly age groups and particularly high in those aged 55–64 years. Compared with the general population, risk was greater in less deprived areas due to the association of more deprived neighbourhoods with poorer outcomes in the general population, and perhaps as congregate housing for people with intellectual disabilities tends to be in more affluent areas (larger houses). Risk was also greater for men. This highlights the importance of action to reduce COVID-19 infection and mortality risks for adults with intellectual disabilities across all ages, and in all neighbourhoods. The overall risk of dying from any cause was already higher among adults with intellectual disabilities prior to the COVID-19 pandemic. The net effect of COVID-19 is a slight, non-significant increase in excess deaths from any cause.

Many people with intellectual disabilities require support with daily activities, meaning that public health advice on shielding has limited effectiveness as regular contact with other people is often unavoidable. They are also more likely to be residents in communal establishments and have higher rates of multimorbidity resulting in more frequent contact with healthcare workers. This highlights the importance of non-pharmaceutical interventions, such as social distancing, face coverings and hand hygiene in minimising risk of infection. It is also critical that public health policy and messaging increases awareness among caregivers and people with intellectual disabilities of COVID-19 symptoms, infection risk, the need for carers and people with intellectual disabilities to isolate when symptomatic, and to consider hospital early to improve outcomes given the higher risk to this population, as well as the high importance of vaccination. It is critical that further comparative research is conducted to investigate any disparities or inequalities in hospital treatment following admission for COVID-19 between people with intellectual disabilities and those without.

Comparison with previous studies

Previous studies have suggested variable COVID-19 outcomes for people with intellectual disabilities including: COVID-19 mortality rates 2–8 fold higher than the general population,[12–15 23] increased risk of infection,[13 14] increased risk of hospital admissions,[12–14] no difference in case-fatality,[17] overall case-fatality with OR=2.75 or OR=3.61 in the <70 years old,[18] and all-cause mortality in those with known COVID-19 with OR=1.97.[11] A study on adults with intellectual disabilities in residential settings (compared with whole-community general population) reported COVID-19 infection rates to be about four times higher, case fatality almost double and mortality rates 7.8 times higher.[19] Our study of a whole nation's adult population with intellectual disabilities found they were twice as likely to become infected with COVID-19, with a case fatality 26% higher and COVID-19 mortality 2.2 times higher than the general population. Poorer COVID-19 outcomes in younger adults compared with the general population were expected and have also been suggested.[13 15 17] This is in view of premature deaths resulting in people with milder intellectual disabilities and fewer comorbidities surviving into old age, and therefore, more closely resembling the elderly general population. This difference in the age profile of COVID-19 mortality is important given the current prioritisation of ongoing vaccination boosters in the UK, and initial vaccinations internationally in countries with lesser current coverage and on those in older age groups, which will potentially lead to increased levels of potentially preventable COVID-19 mortality in unvaccinated younger people with intellectual disabilities.

What is already known

⇒ To date, five studies have reported mortality rates for people with intellectual disabilities compared with the general population. All reported higher rates of COVID-19 mortality, ranging between 2.23 and 8 times higher for people with intellectual disabilities than those in the general population. Studies that report case fatality rates in the intellectual disabilities population have been based on biased samples and have resulted in conflicting findings, with some studies reporting no difference between rates or as much as 2.75 times higher rates in adults with intellectual disabilities compared with other adults.

⇒ In summary studies investigating the impact of COVID-19 on adults with intellectual disabilities may indicate a higher risk of COVID-19 mortality and case fatality than other adults, but the evidence is limited and inconclusive.

What this study adds

⇒ This study analysed COVID-19 mortality, case-fatality, severe infection and infection rates for all adults with intellectual disabilities compared with a 5% sample of the general population. Adults with intellectual disabilities were almost twice as likely to become infected with COVID-19, 2.2 times as likely to have severe infection, 2.2 times as likely to have COVID-19 mortality and had 26% higher COVID-19 case fatality compared with those with no intellectual disabilities. After standardising for age, sex and deprivation, people with intellectual disabilities were 3.2 times more at risk of COVID-19 mortality and 2.6 times more at risk of severe infection relative to those with no intellectual disabilities. We also report that adults with intellectual disabilities had poorer outcomes among non-elderly age-groups particularly those aged 55–65 years, men and those living in less-deprived neighbourhoods compared with people with no intellectual disabilities. Our data indicate that people with intellectual disabilities under the age of 65 are at significantly greater risk of COVID-19 mortality than those in the general population.

Study implications

⇒ The increased risk of COVID-19 infection and mortality suggests that non-pharmaceutical initiatives should be treated as vital interventions to enable carers and care-provider organisations to ameliorate the risk of infection. All adults with intellectual disabilities should be prioritised in the national roll-outs of COVID-19 vaccination programmes, regardless of age, sex or neighbourhood deprivation.

Strengths and limitations

The study is large, including the entire country's adult population with intellectual disabilities, as well as a proportion of adults in the general population. There was a 94%, response rate. Record linkage was successful on 94.9% providing data on a wide range of outcomes.

COVID-19 testing data are likely to be an underestimate of true community incidence of COVID-19 infection rates

due to limited COVID-19 testing during the first wave of the pandemic. However, this approach is preferable to the inclusion of suspected cases that may not be COVID-19. Case fatality rates were high in both groups and are likely to be an overestimate due to lack of testing of less severe infections, particularly in wave one. We did not have information on ability level, Down syndrome or living circumstances, or data on other risk factors for COVID-19 such as comorbidities which are more common in adults with intellectual disabilities than other people. As 97.3% of the intellectual disabilities population in Scotland is white, we were unable to analyse ethnicity due to small cell sizes. In the subgroup calculations of standardised ratios of severe and fatal COVID-19 infections some cells contained less than twenty events. The Office for National Statistics advises that, in such situations derived rates should be interpreted as having low reliability.[24] It is important that COVID-19 outcomes in the second and third waves are investigated as this data becomes available.

Implications for clinicians and policymakers

Non-pharmaceutical interventions are critical in minimising the transmission of COVID-19. Our findings are important for policy-makers, clinicians and public health physicians to make evidence-based decisions about targeting preventive measures such as shielding, surveillance strategies, criteria for testing and prioritisation for vaccination, including those providing care and support to people with intellectual disabilities. These results are relevant for all adults with intellectual disabilities, regardless of age, sex or extent of neighbourhood deprivation. The age cut-offs used in the general population for prioritising COVID-19 vaccination and boosters should not be applied to adults with intellectual disabilities who are a higher risk even at younger ages.

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Contributors AH and DK conceived the study. AH and S-AC led on the acquisition of data for the study. AH, DK, MF, JPP, S-AC, DFM, CM and CH were involved in the design of the study and interpretation of the data. MF led the analysis of the data and CM verified data. AH and DK prepared the first draft of the manuscript. MF, JPP, S-AC, DFM, CM and CH also contributed to the manuscript writing. All authors approved the final manuscript. AH is the study guarantor.

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Competing interests None declared.

Patient consent for publication Not applicable.

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Antipsychotic and antidepressant prescribing for 704 297 children and young people with and without intellectual disabilities: record linkage study

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Background

Psychotropics are overprescribed for adults with intellectual disabilities; there are few studies in children and young people.

Aims

To investigate antipsychotic and antidepressant prescribing in children and young people with and without intellectual disabilities, and prescribing trends.

Method

Scotland's annual Pupil Census, which identifies pupils with and without intellectual disabilities, was record-linked to the Prescribing Information System. Antidepressant and antipsychotic data were extracted. Logistic regression was used to analyse prescribing between 2010 and 2013.

Results

Of the 704 297 pupils, 16 142 (2.29%) had a record of intellectual disabilities. Antipsychotic and antidepressant use increased over time, and was higher in older pupils; antipsychotic use was higher in boys, and antidepressant use was higher in girls. Overall, antipsychotics were prescribed to 281 (1.74%) pupils with intellectual disabilities and 802 (0.12%) without (adjusted odds ratio 16.85, 95% CI 15.29–18.56). The higher use among those with intellectual disabilities fell each year (adjusted odds ratio 20.19 in 2010 *v*. 14.24 in 2013). Overall, 191 (1.18%) pupils with intellectual disabilities and 4561 (0.66%) without were prescribed antidepressants (adjusted odds ratio 2.28, 95% CI 2.03–2.56). The difference decreased each year (adjusted odds ratio 3.10 in 2010 *v*. 2.02 in 2013).

Conclusions

Significantly more pupils with intellectual disabilities are prescribed antipsychotics and antidepressants than are other pupils. Prescribing overall increased over time, but less so for pupils with intellectual disabilities; either they are not receiving the same treatment advances as other pupils, or possible overprescribing in the past is changing. More longitudinal data are required.

Keywords

Intellectual disabilities; antipsychotics; antidepressants; children; young people.

Adults with intellectual disabilities are overprescribed psychotropic prescriptions,[1,2] but surprisingly few studies have focussed on children and young people. Children and young people with intellectual disabilities are more likely to experience psychiatric disorders than those without intellectual disabilities.[3–5] However, few children or young people experience psychosis, as the peak onset in males is at age 20–24 years and, for females, a less prominent peak occurs at 20–24 years, followed by another at >35 years.[6] However, antipsychotics are also prescribed 'off-label' in children and young people with intellectual disabilities with problem behaviours, such as aggression, disruptive behaviour or hyperactivity.[7] Although there is some evidence of effectiveness, more so for risperidone, the quality of evidence is low, and side-effects are common, including for atypical antipsychotics.[8–11] Indeed, it has been suggested that antipsychotic side-effects occur more commonly in children with intellectual disabilities than in other children, and they have difficulty reporting them;[3] it has also been suggested that prescribing antipsychotics to children with problem behaviours is maltreatment.[12] The UK National Institute for Health and Care Excellence and other guidelines recommend that antipsychotic drugs should be reduced or discontinued in people with intellectual disabilities who are not experiencing psychosis,[13,14] and that use is considered only when psychological or other interventions alone do not produce change within an agreed time, or risk is very severe.[15] Similarly, systematic reviews of antidepressant prescribing in this population have also deemed studies to be low quality, with small sample sizes,[16] and most antidepressants are contraindicated in children and youth, with suicidal ideation being a side-effect. Despite these cautions, there has been little study of the rates of prescription of antipsychotics and antidepressants in children with intellectual disabilities, or any changes over time. A review of general practice records in England reported that 2.4% with intellectual disabilities aged ≤18 years were prescribed antipsychotics and 1.2% were prescribed antidepressants between 2009 and 2012, but the authors did not provide longitudinal data.[1] A Taiwanese study reported an increase in out-patient clinic psychotropic prescribing between 1997 and 2007 of 16.67% for children with intellectual disabilities, and 14.69% for youth with intellectual disabilities.[17] For antidepressant prescribing, the increases were 46.05% for children with intellectual disabilities and 124.19% for youth with intellectual disabilities, but the sample was not population-based.[17]

Aims

This study aimed to investigate the use of antipsychotic and antidepressant prescribing to the whole population of children and young people with intellectual disabilities in Scotland, compared with those without intellectual disabilities, and to investigate trends in prescribing over a 4-year period.

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Method

Approvals

This study was approval by the National Health Service (NHS) National Services Scotland Public Benefit and Privacy Panel (Application number 1617-0259). Individual participant consent was not required.

Data sources and linkage

In Scotland, a Pupil Census is conducted in September each year. It collects information on all children and young people attending local authority primary, secondary and special schools in Scotland, or children and young people with local authority-funded placements in alternative schools. This includes 95% of all children and young people in Scotland. We can identify no reason to suspect that children with intellectual disabilities would be more likely than other children to be home-educated or have parents paying school fees for their education at a public school. In 2010, 35.8% of pupils with intellectual disabilities attended special schools, and 64.2% attended mainstream schools; in 2011, these figures were 29.1% and 70.9%; in 2012, they were 27.7% and 72.4% and in 2013, they were 29.0% and 71.0%, respectively. The information collected in the Pupil Census includes whether the child/young person has a record of additional support needs, and the type of additional support needs including intellectual disabilities. The types of additional support needs recorded differentiate between intellectual disabilities and specific learning disabilities such as dyslexia and dyscalculia. Under the Education (Additional Support for Learning) (Scotland) Act 2004 (as amended), education authorities have a statutory requirement to identify and provide for the additional support needs of children and young people. The assessment includes informal observations by teachers, and more formal assessments from education, medical or psychological professionals (educational psychology, NHS community child health teams and clinical genetics services are available throughout Scotland, to provide these and other assessments). In the Pupil Census, teachers record the additional support needs identified from these assessments. It is held by the Scottish Exchange of Education Data. We used data from the four censuses conducted in 2010–2013, to identify children and young people with and without intellectual disabilities, aged up to 19 years. Records for each year were only included in the analysis if the child was aged 4 years or older in that particular year. We then used individual record linkage based on probabilistic record matching (on date of birth, gender and postcode) to the Community Health Index (CHI; Scotland's list of all unique patient identifiers). We excluded non-singleton births (identified from maternity records), as names were not used to link the pupil records to the CHI, and therefore we could not decipher whether the correct child had linked. We excluded any records with duplicate pupil records or where the linkage was tied with another pupil. CHI was used to link the Pupil Census data at an individual level to prescribing data for each child; it did not provide any additional information for the study. Using the CHI, we then linked the Pupil Census data to Scotland's Prescribing Information System (PIS) to investigate prescriptions encashed over 2010–2013. PIS records all medicines that are prescribed and dispensed in the community in Scotland, or issued in hospitals and dispensed in the community, and prescriptions written in Scotland that were dispensed elsewhere in the UK, and includes coding of the prescriptions by British National Formulary codes. Only pupils with intellectual disabilities recorded in at least two different school years were included in the intellectual disabilities group, to reduce potential bias as a result of miscoding. Pupils who were included in at least two pupil censuses over the study period and had no record of intellectual disabilities were used as the comparison group. Antidepressant and antipsychotic prescribing data were extracted from the PIS for the two groups, for each of the 4 years (2010–2013). Prescription codes for each year were determined using the British National Formulary chapter 4, section code 402 for antipsychotics and 403 for antidepressants.[18]

Statistical analyses

Participant characteristics were summarised for pupils with and without intellectual disabilities, using counts and percentages for categorical data and mean and s.d. for age. Prescribing of antipsychotics and antidepressants at any time during 2010–2013 was summarised for pupils with and without intellectual disabilities by age group and gender, using counts and percentages. Prescribing was then compared between pupils with and without intellectual disabilities, using univariate logistic regression models to produce odds ratios and corresponding 95% confidence intervals. The models were run univariately and then adjusted for age, gender and census year. These models were then extended again to include an interaction term between census year and population (intellectual disabilities or without intellectual disabilities), to assess whether there was a statistically significant difference in prescribing between census years for the different populations. If a statistically significant interaction was found, then subgroup analysis was performed by individual census years 2010 to 2013, and by population group (intellectual disabilities or without intellectual disabilities).

All analyses were performed with R version 3.5.0 for Windows (R Core team, Vienna, Austria; see https://www.R-project.org/).

Results

Participant characteristics

The final linked data-set contained records for 704 297 pupils aged between 4 and 19 years, who had been included in at least two censuses. Of these, 16 142 (2.29%) had at least two records of additional support needs owing to intellectual disabilities, and 688 155 (97.71%) had no record of intellectual disabilities. The mean age of the pupils with intellectual disabilities was 9.53 years (s.d. 3.55), and the mean age of the pupils without intellectual disabilities was 9.22 years (s.d. 3.96). As expected, there were more male than female pupils with intellectual disabilities,[19] and an almost even split in gender for the population without intellectual disabilities (Table 1).

Antipsychotic prescribing

Antipsychotics were prescribed at some point from 2010 to 2013 for 281 (1.74%) of the pupils with intellectual disabilities and 802 (0.12%) of the pupils without intellectual disabilities (Table 2). More male than female pupils were prescribed antipsychotics in

Table 1 Characteristics of pupils at the year of first inclusion in a census

	Intellectual disabilities		No intellectual disabilities	
	n (*n* = 16 142)	%	*n* (*n* = 688 155)	%
Gender				
Male	10 424	64.6%	347 539	50.5%
Female	5718	35.4%	340 616	49.5%
Age in 2010 (years)				
<4	554	3.4%	56 797	8.3%
4–11	10 292	63.8%	398 532	57.9%
12–16	5201	32.2%	231 912	33.7%
17–19	95	0.6%	914	0.1%

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Table 2 Antipsychotic and antidepressant prescribing in the period 2010–2013 in pupils with and without intellectual disabilities

	Antipsychotics		Antidepressants	
	Intellectual disabilities, *n* (%)	Without intellectual disabilities, *n* (%)	Intellectual disabilities, *n* (%)	Without intellectual disabilities, *n* (%)
All	281/16 142 (1.74%)	802/688 155 (0.12%)	191/16 142 (1.18%)	4561/688 155 (0.66%)
Gender				
Male	204/10 424 (1.96%)	499/347 539 (0.14%)	111/10 424 (1.06%)	1424/347 539 (0.41%)
Female	77/5718 (1.35%)	303/340 616 (0.09%)	80/5718 (1.4%)	3137/340 616 (0.92%)
Age in 2010 (years)				
4–11	120/10 292 (1.17%)	282/398 532 (0.07%)	55/10 292 (0.53%)	819/398 532 (0.21%)
12–16	150/5201 (2.88%)	505/231 912 (0.22%)	131/5201 (2.52%)	3693/231 912 (1.59%)
17–19	8/95 (8.42%)	5/914 (0.55%)	<5/95 (<5.22%)	30/94 (3.28%)

both populations. The unadjusted odds ratio of the association of intellectual disabilities with antipsychotic prescribing was 18.21 (95% CI 16.54–20.04).

Antipsychotics were prescribed to pupils with intellectual disabilities at rates of 0.85% in 2010, 1.02% in 2011, 0.90% in 2012 and 0.99% in 2013; compared with rates in pupils without intellectual disabilities of 0.04% in 2010, 0.05% in 2011, 0.05% in 2012 and 0.06% in 2013. Table 3 shows the logistic regression results, which indicated an odds ratio of 16.85 (95% CI 15.29–18.56) of intellectual disabilities being associated with antipsychotic prescribing, when adjusted for census year, age and gender. Older age, male gender and later census year were also associated with an increase in antipsychotic prescribing (Table 3). The interaction term between population (intellectual disabilities or without intellectual disabilities) and census year was also significant ($P = 0.002$). Hence, subgroup analyses were conducted, and we found that the adjusted association of intellectual disabilities with being prescribed antipsychotics showed a gradient of falling over time (odds ratios of 20.19 (95% CI 16.34–24.84) in 2010, 18.84 (95% CI 15.57–22.70) in 2011, 15.55 (95% CI 12.75–18.86) in 2012 and 14.24 (95% CI 11.80–17.10) in 2013). In the subgroup analyses for the association of census year on antipsychotic prescribing in the population with intellectual disabilities, referenced to 2010, there was also a gradient showing increase over time (adjusted odds ratios of 1.18 (95% CI 0.94–1.49) for 2011, 1.27 (95% CI 1.00–1.61) for 2012 and 1.80 (95% CI 1.42–2.28) for 2013). For the population without intellectual disabilities, there was an increase over time; corresponding adjusted odds ratios were 1.29 (95% CI 1.10–1.52) for 2011, 1.68 (95% CI 1.43–1.98) for 2012 and 2.62 (95% CI 2.24–3.08) for 2013. These subgroup analyses show that the increase in prescribing previously observed for year remains in both populations, most clearly for those without intellectual disabilities who have a greater increase with year.

Antidepressant prescribing

Antidepressants were prescribed at some point in 2010–2013 to 191 (1.18%) of the pupils with intellectual disabilities, and 4561 (0.66%) of the pupils without intellectual disabilities (Table 2). More female than male pupils were prescribed antidepressants in both populations. The unadjusted odds ratio of the association of intellectual disabilities with antidepressant prescribing was 2.05 (95% CI 1.82–2.29).

Antidepressants were prescribed to pupils with intellectual disabilities at rates of 0.29% in 2010, 0.47% in 2011, 0.51% in 2012 and 0.60% in 2013; compared with rates in the pupils without intellectual disabilities of 0.10% in 2010, 0.22% in 2011, 0.25% in 2012 and 0.31% in 2013. The logistic regression (Table 4), to determine the association between intellectual disabilities and antidepressant prescribing, found that pupils with intellectual disabilities were over twice as likely as other pupils to be prescribed antidepressants when adjusted for census year, age and gender. Older age, female gender and later census years were also associated with higher odds of antidepressant prescribing (Table 4). The interaction term between population (intellectual disabilities or without intellectual disabilities) and census year was significant ($P = 0.007$). Hence, subgroup analyses were conducted, and we found that the adjusted association of intellectual disabilities with being prescribed antidepressants showed a gradient of falling over time (odds ratios of 3.10 (95% CI 2.27–4.12) in 2010, 2.48 (95% CI 1.95–3.11) in 2011, 2.12 (95% CI 1.69–2.64) in 2012 and 2.02 (95% CI 1.64–2.47) in 2013). Similar to the antipsychotic results, this indicates that the increase in prescribing for the intellectual disabilities population overall remained across all years of the census, although the scale of the increased odds reduced by year. In the subgroup analyses exploring the association of census year on antidepressant prescribing in the intellectual disabilities population, referenced to census year 2010, there was also a gradient showing increase over time (adjusted odds ratios of 1.61 (95% CI 1.12–2.33) for 2011, 2.27 (95% CI 1.59–3.29) for 2012 and 3.82 (95% CI 2.68–5.51) for 2013). For the population without intellectual disabilities, there was an increase over time; corresponding adjusted odds ratios were 2.14 (95% CI 1.96–2.34) for 2011, 3.95 (95% CI 3.61–4.31) for 2012 and 7.76 (95% CI 7.10–8.49) for 2013. These subgroup analyses show that the increase in prescribing previously observed for year also remains in both populations, and is greater for those without intellectual disabilities.

Table 3 Logistic regression to determine the association of intellectual disabilities with antipsychotic prescribing

	Unadjusted model			Adjusted model[a]			
	Odds ratio	95% confidence interval	*P*-value	Odds ratio	95% confidence interval	*P*-value	Interaction *P*-value[b]
Intellectual disabilities	18.21	16.54–20.04	<0.001	16.85	15.29–18.56	<0.001	
Age (per 1-yr increase)				1.29	1.27–1.31	<0.001	
Female gender				0.53	0.48–0.58	<0.001	
Census year (per 1-yr increase)				1.32	1.27–1.38	<0.001	
Interaction of census year and population (with or without intellectual disabilities)							0.002

a. Model for population effect, adjusted for age, gender and census year.
b. Interaction between census year and population added to above adjusted model.

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Table 4 Logistic regression to determine the association of intellectual disabilities with antidepressant prescribing

	Unadjusted model			Adjusted model[a]			
	Odds ratio	95% confidence interval	*P*-value	Odds ratio	95% confidence interval	*P*-value	Interaction *P*-value[b]
Intellectual disabilities	2.05	1.82–2.29	<0.001	2.28	2.03–2.56	<0.001	
Age (per 1-yr increase)				1.59	1.57–1.61	<0.001	
Female gender				1.99	1.89–2.10	<0.001	
Year (per 1-yr increase)				1.93	1.88–1.98	<0.001	
Interaction of census year and population (with or without intellectual disabilities)							0.007

a. Model for population effect, adjusted for age, gender and census year.
b. Interaction between census year and population added to above adjusted model.

Discussion

Principal findings and interpretation

As far as we are aware, this is the first study of antipsychotic and antidepressant prescribing, and trends in prescribing, in a country's whole population of children and young people with intellectual disabilities, in comparison with those without intellectual disabilities. Over the study period, 1.74% of pupils with intellectual disabilities received antipsychotics and 1.18% received antidepressants. Compared with their peers, their odds of receiving the drugs were 16 and 2, respectively, after adjusting for potential confounders. Psychotropic prescribing in children and young people must be undertaken cautiously after other options have been exhausted, but it does play a role in certain situations. The relative difference between them and their peers fell over time, but this was because of increased use of antipsychotics and antidepressants by their peers, not decreased use among children with intellectual disabilities. These apparent changes in antipsychotic, and particularly antidepressant use over a 4-year period highlight the need for further longitudinal studies, especially as there have been no other population-based longitudinal studies of psychotropic drug use conducted with children with intellectual disabilities.

Comparison with previous literature

Our finding of 1.74% of pupils with intellectual disabilities being prescribed antipsychotics in 2010–2013 compares with the higher rate reported in England of 2.4% for the similar period of 2009–2012; however, our finding of 1.18% for antidepressants is similar to the 1.2% reported in England.[1] We found that antipsychotic prescribing had increased, which was also reported in a study in Wales, but for a non-comparable population as it included children and youth with autism together with those with intellectual disabilities.[20] The Taiwanese study reported considerable increase in prescribing of psychotropics, particularly antidepressants, but included only attenders at out-patient clinics, and has a different healthcare system to that in the UK, so is not comparable with our study.[17] Additionally, in a study comparing 16 countries, Taiwan was shown to have the highest antipsychotic prescribing rates for general population children and young people of all the countries, so prescribing practices may differ from the UK.[21] A small USA study is also not directly comparable with ours as they reported on children with comorbid intellectual disabilities and mental disorders; unlike our study, they found no change in antipsychotic use between 2007–2011.[22] Psychotropic prescribing for children and young people has increased over time in the general population, particularly in Europe, the USA and Canada.[23] In our study, we have shown that the extent of increase is less in pupils with intellectual disabilities than in pupils without intellectual disabilities.

Higher prescribing rates in youth rather than childhood mirrors the pattern of emerging mental disorders with age, and may be related to the greater challenges for families in managing aggressive problem behaviour in adolescents rather than smaller children. Higher prescribing rates of antipsychotics in males and antidepressants in females follow the same pattern as that seen in the general population,[23] and in the English study of children and young people with intellectual disabilities.[1]

Strengths and limitations

Our study included the whole country, and the pupil census has approximately 95% coverage of school pupils. The data were longitudinal. Scotland is a high-income country, provides additional support in school to children with intellectual disabilities (which is advantageous to children with this diagnosis) and has comprehensive multidisciplinary community paediatric and genetic services, so few children with intellectual disabilities would not be identified. However, there may still be some recording errors in the census, hence we required the children to have a record of intellectual disabilities on at least two occasions to be included in the intellectual disabilities population. We did not have data on the severity of pupil's intellectual disabilities. We did not have data on the mental health of children and young people. Additionally, we were unable to access data on the 5% of children and young people not in school. Further, the pupil census is focussed on information about the pupils, not individual data about their parents or households, and so we have no contextual information that might have been relevant on any associations with, for example, parental mental health, parental income, parental occupation or parental educational attainments.

Implications

Children and young people with intellectual disabilities are significantly more likely to be prescribed antipsychotics and antidepressants than children and young people without intellectual disabilities. Psychotropic prescribing for children and young people is increasing, but at a lesser rate for those with intellectual disabilities compared with those without. We do not know if that suggests that children and young people with intellectual disabilities are not receiving the same treatment advances (i.e. better identification of mental ill-health and more prescribing, appropriately) over time as children and young people without intellectual disabilities, or if children and young people with intellectual disabilities were being overprescribed psychotropics in the past and this was addressed to an extent in this time period, or if overprescribing is increasing in children and young people without intellectual disabilities. It is important that prescribers in psychiatric services are vigilant to these possibilities, and prescribe when beneficial to the child, using the lowest therapeutic dose that is effective for each child, and withdrawing psychotropic drugs when there is no clear indication or benefit. Longitudinal observation is clearly important to quantify and understand these prescribing trends, and future research should

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investigate longitudinal prescribing in children and adolescents with intellectual disabilities in relation to mental health diagnoses, and adverse health outcomes.

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Supplementary material

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Data availability

The researchers were authorised to receive data controlled and held by the Information Services Division, NHS National Services Scotland (NHS NSS), via access through the national safe haven. Data processing and sharing agreements were drafted with NHS NSS and ScotXed education. The Information Services Division Statistical Disclosure Control Protocol was followed. Therefore, data cannot be shared with other parties, but can be requested from the data controllers subject to the approval processes.

Author contributions

A.H., D.K. and S.-A.C. conceived the study. G.Y.-S. conducted the initial literature review. M.F. and J.J.P. developed the record linkage. B.S. and N.G. analysed the data. A.H. drafted the first version of the manuscript. All authors contributed to interpreting the data and writing the manuscript, and approved the final version of the manuscript.

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Declaration of interest

None.

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Changes over a decade in psychotropic prescribing for people with intellectual disabilities: prospective cohort study

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ABSTRACT

Objectives To investigate psychotropic prescribing in the intellectual disabilities population over 10 years, and associated mental ill health diagnoses.

Design Comparison of cross-sectional data in 2002–2004 (T1) and 2014 (T2). Longitudinal cohort study with detailed health assessments at T1 and record linkage to T2 prescribing data.

Setting General community.

Participants 1190 adults with intellectual disabilities in T1 compared with 3906 adults with intellectual disabilities in T2. 545/1190 adults with intellectual disabilities in T1 were alive and their records linked to T2 prescribing data.

Main outcome measures Encashed regular and as-required psychotropic prescriptions.

Results 50.7% (603/1190) of adults in T1 and 48.2% (1881/3906) in T2 were prescribed at least one psychotropic; antipsychotics: 24.5% (292/1190) in T1 and 16.7% (653/3906) in T2; antidepressants: 11.2% (133/1190) in T1 and 19.1% (746/3906) in T2. 21.2% (62/292) prescribed antipsychotics in T1 had psychosis or bipolar disorder, 33.2% (97/292) had no mental ill health or problem behaviours, 20.6% (60/292) had problem behaviours but no psychosis or bipolar disorder. Psychotropics increased from 47.0% (256/545) in T1 to 57.8% (315/545) in T2 (p<0.001): antipsychotics did not change (OR 1.18; 95% CI 0.87 to 1.60; p=0.280), there was an increase for antidepressants (OR 2.80; 95% CI 1.96 to 4.00; p<0.001), hypnotics/anxiolytics (OR 2.19; 95% CI 1.34 to 3.61; p=0.002), and antiepileptics (OR 1.40; 95% CI 1.06 to 1.84; p=0.017). Antipsychotic prescribing increased for people with problem behaviours in T1 (OR 6.45; 95% CI 4.41 to 9.45; p<0.001), more so than for people with other mental ill health in T1 (OR 4.11; 95% CI 2.76 to 6.11; p<0.001).

Conclusions Despite concerns about antipsychotic prescribing and guidelines recommending their withdrawal, it appears that while fewer antipsychotic prescriptions were initiated by T2 than in T1, people were not withdrawn from them once commenced. People with problem behaviours had increased prescribing. There was also a striking increase in antidepressant prescriptions. Adults with intellectual disabilities need frequent and careful medication reviews.

Strengths and limitations of this study

- The large cohort size, longitudinal design, detailed ascertainment of the population with intellectual disabilities, and the in-depth health assessments at T1.
- The cross-sectional cohorts were population based at T1 and T2, and representative of the population with intellectual disabilities; the linked cohort had similar characteristics to the cross-sectional cohort at T1, suggesting this cohort is also representative and therefore that the results are generalisable.
- Only 73% of general practices agreed to data extraction, and this combined with deaths are likely to be the main reasons for 545/1190 of the participants being linked in the T2 data 10 years later.
- The different methods of data collection, with specialist individual assessments at T1 and electronic data extraction at T2; in particular, a large proportion of information is missing and inaccuracies might exist relating to recorded level of intellectual disabilities in the general practitioner data at T2, limiting comparability of this variable between the T1 and T2 cohorts.
- The study did not investigate changes in dosages, polypharmacy or duration of use, and mental ill health data at T2 are lacking.

INTRODUCTION

Mental ill health is common in people with intellectual disabilities.[1] The prevalence of psychosis in this population is reported to be around 4% based on cross-sectional data, and the rate of people with a first psychotic episode is about 10 times that of the general population.[2] While the rates of psychosis are relatively high, antipsychotics are often prescribed for adults with intellectual disabilities who do not have a record of severe mental ill health,[3 4] often for problem behaviours,[5–9] and despite limited evidence to support their use beyond short-term sedation.[7] Indeed, 71% of people with intellectual disabilities who are prescribed antipsychotics have been reported to have no record of serious mental ill health.[10] This is important because antipsychotics have numerous disabling, painful and disfiguring side effects, some of which are life

threatening, such as tardive dyskinesia, cardiac arrhythmias and sudden cardiac death.[11–13] Antipsychotics are also frequently prescribed for children and young people with a range of developmental disabilities and problem behaviours,[14 15] and in the young general population, rates increase during adolescence.[16]

Concerns have repeatedly been raised about the overuse of antipsychotics, and the need for more proportionate prescribing for people with intellectual disabilities.[7 17–19] In 2016 a national campaign was launched by NHS England in partnership with the Royal Colleges of General Practitioners, Psychiatrists, and Nursing, the Royal Pharmaceutical Society, and the British Psychological Society to address these concerns in England: 'Stopping over medication of people with a learning disability, autism or both (STOMP)'. Guidelines from STOMP, the National Institute for Health and Care Excellence, and the Royal College of Psychiatrists highlight that prescribers, where appropriate, should reduce or withdraw antipsychotics for people with intellectual disabilities who do not have psychosis.[7 20 21] However, there is very little empirical evidence from the UK on any changes in antipsychotic prescribing patterns over time. An exception is a study by Sheehan and colleagues that extracted data from general practice records on 33 016 adults with a record of intellectual disabilities, with a median follow-up of 5.5 years.[10] These authors reported the incidence of new psychotropic prescription to be 518/10 000 person years. Prescriptions of antipsychotics fell by 4% per year over the study period, as did mood stabilisers, while there was no consistent trend for antidepressants or anxiolytics/hypnotics. Sheehan and colleagues reported that 47% of those with 'challenging behaviour' had received antipsychotic drugs, but only 12% had a record of severe mental ill health, and that 26% of those prescribed antipsychotics did not have a record of severe mental ill health or 'challenging behaviour'. A limitation of this study is in the identification of 'challenging behaviour' through a heterogeneous list of 45 Read codes (the system used in general practices in the UK to code diagnoses). Read codes do not provide a robust method for ascertaining problem behaviours. Additionally, incomplete and variable recording practices do not always accurately reflect a person's health.[10]

Another study from Australia investigated psychotropic medication use between 1999 and 2015 in a cohort of 138 participants[22] and also found a strong association between problem behaviours and psychotropic medication. In this cohort the study reported that once psychotropic medications were prescribed they were unlikely to be removed, and little change was observed in prescribing of antipsychotics between 1999 and 2015 (24/138 (24%) to 23/92 (23%)). A sharp increase in the prescribing of antidepressants from 16.7% to 36.1% across the same period was also observed. However, while this was a longitudinal cohort, not all participants took part in all waves of data collection, therefore it is not possible to ascertain within group changes in prescribing.

Aim

The aim of this study was to investigate changes over a decade in psychotropic prescribing for adults with intellectual disabilities, and the diagnoses associated with antipsychotics by using detailed psychiatric assessments.

METHODS

Ethical approval

Between 2002 and 2004 (T1), individual consent to participate was taken in line with Scottish law. In 2014 (T2), 191/263 (73%) general practices in NHS Greater Glasgow and Clyde area participated, and the NHS Greater Glasgow and Clyde Local Privacy Advisory Committee approved electronic extraction and linkage of primary care records.

Participants

In 2000–2001, a primary care intellectual disabilities register was established of adults with intellectual disabilities, aged 16 years and older, living in the NHS Greater Glasgow area. This initiative was delivered through partnership between the intellectual disabilities clinical service and all general practitioners in the area. People with intellectual disabilities were identified through social work services for people with intellectual disabilities; local authority funding arrangements for people receiving paid support of any kind, including day opportunities; local specialist health services for people with intellectual disabilities; the Health Board; and general practices who were financially incentivised to identify their registered patients with intellectual disabilities (100% of general practices participated). Intellectual disabilities nurses reviewed all cases on the register to determine if intellectual disabilities were present; those that did not have intellectual disabilities were removed from the register. The register was then updated annually by the general practices and the intellectual disabilities clinical service.

Between 2002 and 2004, the register was used to invite people living in a representative part of the Health Board area to participate in the study; 67% agreed to take part. These participants were recruited to a longitudinal cohort between 2002 and 2004 (T1), and had detailed health assessments at that time; 1190 were aged 18 years and older and comprise the study population reported here. In 2014 (T2), for people on the register and living in the Health Board area, data were extracted from primary care records; 73% of general practices in the Health Board area agreed to the data extraction. Data were extracted on 3906 patients with intellectual disabilities aged 18 years and older, who comprise the study population reported here.

Process and measures

Semi-structured individual health assessments, including medication review, assessment of level of intellectual disabilities (via structured questions on abilities, and the Vineland Scale[23]), mental ill health symptoms including

problem behaviours and autism, were conducted at T1 by one of six intellectual disabilities nurses and one of three general practitioners with a special interest in intellectual disabilities. These assessments were preceded by data collection from the person's general practitioner medical records, and then a review conducted with the person with intellectual disabilities and their carer(s). This included a review of drug charts for participants in supported care. The 54% of individuals identified with possible, probable or definite mental ill health (including problem behaviours and autism) were then assessed by the study psychiatrists who were specialists in intellectual disabilities psychiatry. Information from each person's psychiatric assessment was reviewed by two psychiatrists who had a case conference and agreed the classification of the mental ill health using ICD-10-DCR,[24] DSM-IV-TR,[25] DC-LD[26] and clinical criteria. Details have been previously reported.[1] Given that ICD-10 and DSM criteria function poorly for adults with moderate to severe intellectual disabilities (particularly with regards to problem behaviours), in this study we report the clinical diagnoses agreed by the study psychiatrists. Data collection was over a 2-year period. Drugs were coded using British National Formulary (BNF) codes.

At T2, the 3906 adults with intellectual disabilities identified from primary care records were record linked to Prescribing Information System (PIS) data by using the Community Health Index (CHI), which is the NHS patient identification number, unique to each person. PIS is Scotland's electronic record of all encashed prescriptions (ie, not prescriptions issued, or drugs administered, but those that the carers/person with intellectual disabilities actually took to a pharmacist and exchanged for the drugs), and includes a record of the BNF code of each prescribed drug.[27] Prescribing information was then extracted using BNF codes for the 3906 adults with intellectual disabilities to identify all prescriptions of antipsychotics, antidepressants, antiepileptics, lithium, and hypnotics/anxiolytics across a specific 12-week prescribing window in 2014, including both regular prescriptions and as-required medication. To establish the longitudinal cohort the CHI number was used to identify T1 participants in the T2 dataset, enabling comparison of encashed medications across the decade. Only participants with complete data who were aged 18 years and older were included in the analyses (figure 1).

Statistical analysis

Subject characteristics and prescribing information were summarised descriptively with mean and SD for continuous outcomes and number and percentage for categorical outcomes at each time point (T1 and T2). Prescribing information at each time point was summarised using binary variables for each class of medications of interest (yes/no), allowing prescribing patterns to be investigated between the two time points in the study using McNemar's tests on the subset of the linked cohort, for whom there were prescribing records at both T1 and

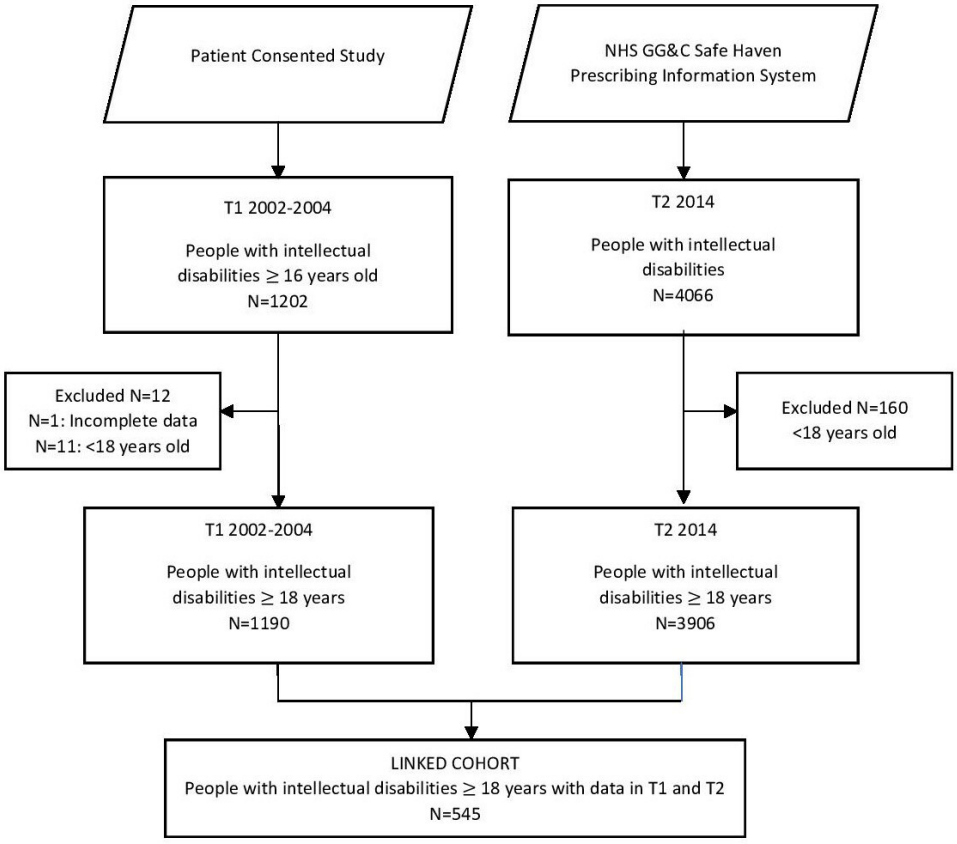

Figure 1 Participant flow diagram. GG&C, Greater Glasgow and Clyde.

T2. This analysis was extended to explore whether there were associations between time or the subject characteristics at T1, with each prescription category using binary logistic regression models. Each model included multiple explanatory variables; specifically, time as a binary variable to indicate each time point T1 and T2; sex; age as a continuous measure; level of intellectual disabilities as four-level categorical variable; presence of mental ill health (yes/no, excluding problem behaviours); having problem behaviours (yes/no); and a binary dependant variable for each class of medication (yes/no). Logistic regression models were also fitted with the above T1 subject characteristics to explore their association with each prescribing category specifically at T2. Odds ratios are reported for all logistic regression models with corresponding 95% confidence intervals (CIs) and p values. A p value less than 0.05 is considered statistically significant. Statistical analyses were conducted using SAS version 9.3.

Patient and public involvement

The Scottish Learning Disabilities Observatory has a steering committee which meets twice a year and provides strategic direction and oversight of all of the Observatory's research, including this project. The steering committee includes people with intellectual disabilities from 'People First', a national group of self-advocates with intellectual disabilities.

RESULTS

Participant characteristics of the cross-sectional cohorts

Data for those who had incomplete data at T1 and for those who were aged under 18 years at either time point were excluded from further analyses. Table 1 shows participant characteristics: age, sex, level of intellectual disabilities at T1 (n=1190) and T2 (n=3906), and mental ill health and epilepsy diagnoses at T1. No mental health or epilepsy data were available at T2.

Table 1 Participant characteristics for the cross-sectional cohorts at T1 and T2

Characteristic	T1 aged ≥18 years (n=1190)	T2 aged ≥18 years (n=3906)
Age, mean (SD)	44.6 (14.3)	45.4 (15.5)
Sex, No (%)		
Male	671 (56.4)	2260 (57.9)
Female	519 (43.6)	1646 (42.1)
Level of intellectual disabilities, No (%)		
Mild	451 (37.9)	1047 (26.8)
Moderate	319 (26.8)	859 (22.0)
Severe	233 (19.6)	595 (15.2)
Profound	187 (15.7)	197 (5.0)
Unknown	0	1208 (30.9)
Epilepsy, No (%)	419 (35.2%)	Not collected
Type of mental ill health, No (%)		
Psychosis, including psychosis in remission	52 (4.4)	Not collected
Problem behaviours	244 (20.5)	
Autism	80 (6.7)	
Attention deficit hyperactivity disorder	15 (1.3)	
Unipolar depression	51 (4.3)	
Bipolar disorder	21 (1.8)	
Anxiety disorders	32 (2.7)	
Organic disorder	20 (1.7)	
Personality disorder	9 (0.8)	
Obsessive compulsive disorder	7 (0.6)	
Psychosexual disorder	<5	
Other	15 (1.3)	
Mental ill health (including problem behaviours)	438 (36.8)	
Mental ill health (excluding problem behaviours)	194 (16.3)	

Prescribing for the cross-sectional cohorts

At least one psychotropic was prescribed at T1 for 50.7% (603/1190) and at T2 for 48.2% (1881/3906) (table 2) of participants. Antipsychotics were prescribed to 24.5% (292/1190) of participants at T1 and 16.7% (653/3906) at T2. At T1, antidepressants were prescribed for 11.2% (133/1190) and at T2 for 19.1% (746/3906) of participants. Hypnotic/anxiolytic, lithium and anti-epileptic prescribing was similar at T1 and T2.

The types of mental ill health experienced by the 292 participants at T1 who were taking antipsychotics are shown in table 3. The most common diagnosis within this group was problem behaviours at 40.8% (119/292). Of note, 33.2% (97/292) of the people taking antipsychotics did not have any identified mental ill health or problem behaviours. Figure 2 demonstrates the overlap between groups of the people who were taking antipsychotics at T1 and selected diagnoses.

Table 3 also shows the types of mental ill health experienced by the 230 participants at T1 who were taking antipsychotics, after excluding people with psychosis (or psychosis in remission) or bipolar disorder (given that they would be expected to be prescribed antipsychotics, and given the considerable overlap between disorders shown in figure 2). Most strikingly, 97/230 (42.2%) of those prescribed antipsychotics had no mental ill health or problem behaviours. The proportion of people in each diagnostic category, without co-occurring psychosis or bipolar disorder who were taking antipsychotics was considerable for all types of mental ill health: 11.7% (27/230) for autism, 7.0% (16/230) for unipolar depression, 2.6% (6/230) for anxiety disorders and 2.2% (5/230) or less for all other diagnoses.

Table 2 Psychotropics prescribed for the cross-sectional cohorts at T1 and T2

Prescriptions	T1 aged ≥18 years (n=1190)	T2 aged ≥18 years (n=3906)
Any psychotropic drug	603 (50.7)	1881 (48.2)
Antipsychotics	292 (24.5)	653 (16.7)
Antidepressants	133 (11.2)	746 (19.1)
Antiepileptics	333 (28.0)	1028 (26.3)
Lithium	14 (1.2)	31 (0.8)
Hypnotics/anxiolytics	81 (6.8)	305 (7.8)
Missing data	0	3 (0.1)

Participant characteristics of the longitudinal, linked cohort

The longitudinal, linked cohort included the 545 adults who were in the T1 cohort and who were also identified within the GP records at T2. Table 4 shows their age, sex, level of intellectual disabilities, epilepsy diagnosis and mental ill health at T1. They appear to be broadly representative of the whole cohort at T1 based on these characteristics.

Table 3 Types of mental ill health at T1 experienced by people prescribed antipsychotics at T1, and after excluding people with psychosis and bipolar disorder

Mental ill health at T1		Adults (≥18 years) taking antipsychotics at T1	
Diagnosis	All (n=1190)	All (n=292)	Excluding people with psychosis and bipolar disorder (n=230)
Psychosis, including psychosis in remission	52	45 (15.4)	–
Problem behaviours	244	119 (40.8)	100 (43.5)
Autism	80	30 (10.3)	27 (11.7)
Attention deficit hyperactivity disorder	15	12 (4.1)	11 (4.8)
Unipolar depression	51	20 (6.9)	16 (7.0)
Bipolar disorder	21	17 (5.8)	–
Anxiety disorders	32	7 (2.4)	6 (2.6)
Organic disorder	20	5 (1.7)	<5
Personality disorder	9	5 (1.7)	<5
Obsessive compulsive disorder	7	3 (1.0)	<5
Psychosexual disorder	<5	<5	<5
Other	15	6 (2.1)	5 (2.2)
Mental ill health (including problem behaviours)	438	195 (66.8)	133 (57.8)
Mental ill health (excluding problem behaviours)	194	76 (26.3)	33 (14.4)
No mental ill health or problem behaviours	752	97 (33.2)	97 (42.2)

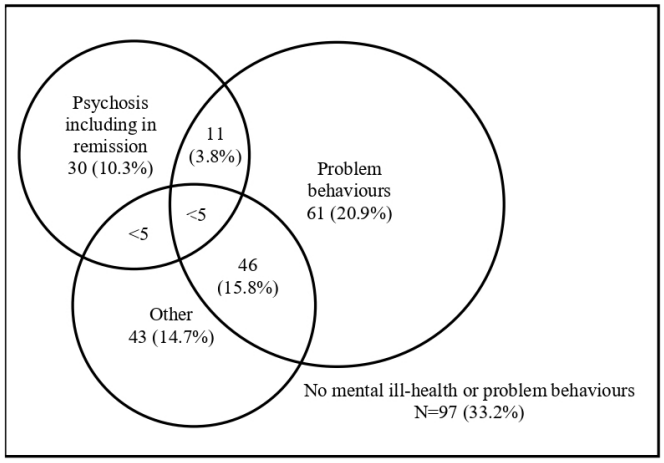

Figure 2 Types of mental ill health experienced by people prescribed antipsychotics at T1 (n=292)

Prescribing for the longitudinal, linked cohort

At least one psychotropic medication was prescribed for 47.0% (256/545) at T1 and for 57.8% (315/545) at

Table 4 Participant characteristics at T1 for people in the longitudinal cohort

Characteristic	T1 (aged ≥18 years) n=545 (%)
Age, mean (SD)	41.8 (13.2)
Sex, No (%)	
Male	322 (59.1)
Female	223 (40.9)
Level of intellectual disabilities, No (%)	
Mild	237 (43.5)
Moderate	154 (28.3)
Severe	89 (16.3)
Profound	65 (11.9)
Epilepsy, No (%)	173 (31.7)
Type of mental ill health, No (%)	
Psychosis, including psychosis in remission	32 (5.9)
Problem behaviours	109 (20.0)
Autism	38 (7.0)
Attention deficit hyperactivity disorder	8 (1.5)
Unipolar depression	23 (4.2)
Bipolar disorder	10 (1.8)
Anxiety disorders	14 (2.6)
Organic disorder	<5
Personality disorder	7 (1.3)
Obsessive compulsive disorder	<5
Psychosexual disorder	<5
Other	6 (1.1)
Mental ill health (including problem behaviours)	190 (34.9)
Mental ill health (excluding problem behaviours)	81 (14.9)

Table 5 Psychotropic medications prescribed for the longitudinal, linked cohort at T1 and T2

Medication group	T1 (n=545)	T2 (n=545)	p-value
Any psychotropic medication	256 (47.0%)	315 (57.8%)	p<0.001
Antipsychotics	128 (23.5%)	142 (26.1%)	p=0.099
Antidepressants	54 (9.9%)	120 (22.0%)	p<0.001
Hypnotics/anxiolytics	25 (4.6%)	51 (9.4%)	p<0.001
Antiepileptics	135 (24.8%)	169 (31.0%)	p<0.001
Lithium	7 (1.3%)	10 (1.8%)	p=0.180

T2 (table 5), which is a significant increase over time (p<0.001). Antidepressants were prescribed for 9.9% (54/545) of participants at T1 and for 22.0% (120/545) at T2, showing a significant increase (p<0.001). At T1, hypnotics/anxiolytics were prescribed for 4.6% (25/545) of participants, and at T2 for 9.4% (51/545), a significant increase (p<0.001). At T1, antiepileptics were prescribed for 24.8% (135/545) of participants, and at T2 for 31.0% (169/545), a significant increase (p<0.001). Prescribing patterns at T1 and T2 were similar for both antipsychotics and lithium. Of the 128 people prescribed antipsychotics at T1, 77.3% (99/128) were prescribed antipsychotics at T2; only 29 (22.7%) had been withdrawn, and 43/545 (7.9%) had started on an antipsychotic between the two timepoints.

The logistic regression analyses, taking account of sex, age, level of intellectual disabilities, having mental ill health (excluding problem behaviours) and having problem behaviours at T1 (table 6) show no significant difference in antipsychotic prescribing rate in the linked cohort over the decade (OR=1.18; 95% CI 0.87 to 1.60; p=0.280), an increase in antidepressants (OR 2.80; 95% CI 1.96 to 4.00; p<0.001), hypnotics/anxiolytics (OR 2.19; 95% CI 1.34 to 3.6; p=0.002), and antiepileptic prescribing (OR 1.40; 95% CI 1.06 to 1.84; p=0.017). Sex was not independently associated with prescribing, except that women were more likely to have an increase in antidepressants than men after adjusting for time (OR 0.53; 95% CI 0.37 to 0.78; p<0.001). Older age had a small effect on prescribing for antipsychotics and antidepressants.

Effects are also observed for level of intellectual disabilities. There was a gradient for antiepileptics (increased prescribing with increasing severity of intellectual disabilities) and a gradient for antidepressants (reduced prescribing with increasing severity of intellectual disabilities). However, there was no gradient across different ability levels for antipsychotic prescribing. As expected, participants with a diagnosed mental health problem (excluding problem behaviours) at T1 were more likely to be prescribed antipsychotics (OR 4.11; 95% CI 2.76 to 6.11; p<0.001), antidepressants (OR 3.90; 95% CI 2.53 to 6.02; p<0.001), and hypnotics/anxiolytics (OR 3.25; 95% CI 1.78 to 5.94; p<0.001). Strikingly though, people with problem behaviours identified at T1 were over six

Table 6 Multivariable analysis of exploratory T1 factors and time with psychotropic prescriptions for the linked cohort (n=545)

	Antipsychotics		Antidepressants		Hypnotics/anxiolytics		Antiepileptics		Lithium*	
	OR (95% CI)	p-value	OR (95% CI)	p-value	OR (95% CI)	p-value	OR (95% CI)	p-value	OR (95% CI)	p-value
Time	1.18 (0.87 to 1.60)	0.280	2.80 (1.96 to 4.00)	<0.001	2.19 (1.34 to 3.60)	0.002	1.40 (1.06 to 1.84)	0.017	0.69 (0.26 to 1.84)	0.4612
Male sex	0.99 (0.73 to 1.34)	0.954	0.53 (0.37 to 0.76)	<0.001	0.83 (0.52 to 1.35)	0.456	1.02 (0.77 to 1.35)	0.896	0.93 (0.34 to 2.59)	0.890
Age at T1	1.04 (1.03 to 1.05)	<0.001	1.02 (1.00 to 1.03)	0.010	1.01 (0.99 to 1.03)	0.260	0.99 (0.98 to 1.00)	0.057	0.96 (0.93 to 0.99)	0.016
Level of intellectual disabilities (compared with mild intellectual disabilities)	–	<0.001	–	0.001	–	0.444	–	<0.001	–	0.094
Moderate	1.88 (1.30 to 2.74)	<0.001	0.82 (0.54 to 1.24)	0.346	1.64 (0.92 to 2.92)	0.093	1.78 (1.26 to 2.51)	<0.001	0.23 (0.07 to 0.71)	0.011
Severe	2.49 (1.61 to 3.85)	<0.001	0.62 (0.37 to 1.03)	0.063	1.12 (0.55 to 2.31)	0.750	2.30 (1.55 to 3.41)	<0.001	1.49 (0.17 to 13.36)	0.721
Profound	1.61 (0.97 to 2.69)	0.067	0.22 (0.11 to 0.46)	<0.001	1.24 (0.58 to 2.62)	0.579	4.73 (3.07 to 7.31)	<0.001	0.46 (0.08 to 2.64)	0.386
Mental ill health†	4.11 (2.76 to 6.11)	<0.001	3.90 (2.53 to 6.02)	<0.001	3.25 (1.78 to 5.94)	<0.001	1.13 (0.76 to 1.70)	0.547	–	–
Problem behaviours	6.45 (4.41 to 9.45)	<0.001	3.44 (2.22 to 5.35)	<0.001	3.06 (1.72 to 5.44)	<0.001	1.27 (0.90 to 1.81)	0.174	–	–

*Mental illness and problem behaviours excluded from lithium model due to small numbers .
†Not including problem behaviours.

times more likely to have increased prescribing of an antipsychotic (OR 6.45; 95% CI 4.41 to 9.45; p<0.001), over three times more likely for antidepressants (OR 3.44; 95% CI 2.22 to 5.35; p<0.001) and three times more likely for hypnotics/anxiolytics (OR 3.06; 95% CI 1.72 to 5.44; p<0.001).

The further regression (online supplementary table 1) investigating factors at T1 which are associated with prescribing at T2 (as opposed to change in prescribing reported in the paragraph above) shows that women were more likely to be prescribed antidepressants at T2, that older age had a small effect for antipsychotics and antidepressants at T2, a gradient across ability level for antiepileptics, a relationship with moderate and severe (but not profound) intellectual disabilities for antipsychotics at T2, and fewer antidepressants for people with profound intellectual disabilities. Mental ill health and problem behaviours at T1 predicted prescribing of all classes.

DISCUSSION

Principal findings

Despite numerous calls and guidelines in the UK for the withdrawal of antipsychotic drugs from people with intellectual disabilities who do not have psychosis/bipolar disorders,[7 20 21] our longitudinal, linked cohort analysis shows no progress over a decade. The comparison of the two cross-sectional cohorts does show a lower rate of antipsychotic prescribing in T2 than was observed in T1, but the rate is still high in T2 at 16.7% of the population. It appears that while few people are being withdrawn from antipsychotics once they start them, new antipsychotic prescriptions are less commonly initiated than in the past. Over the decade, comparison of both the cross-sectional cohorts, and of the longitudinal, linked cohort, reveal a striking increase in the prescription of antidepressants (11.2% to 19.1%, and 9.9% to 22.0%). This was particularly so for women and for people with mild intellectual disabilities. To a lesser extent, there were also increases in prescribing of hypnotics/anxiolytics and antiepileptics in the linked cohort, but not in the comparison of the cross-sectional cohorts. This difference may be accounted for by the known increase in these prescriptions with age,[5] as the linked cohort is of course 10 years older in T2, whereas age and sex are similar in the whole cohorts in T2 and in T1. The age-related change in antiepileptic prescribing in the longitudinal linked cohort, but not in the comparison of the similarly aged cross-sectional cohorts, contextualises the antipsychotic and antidepressant findings (prescribing trends in general) because antiepileptics were almost all prescribed for the highly prevalent condition of epilepsy in this population. While previous studies have reported high rates of antipsychotic prescribing, we are not aware of any that have investigated prescribing over this length of time along with related fluctuations in assessed mental ill health.

Comparison with previous literature

To our knowledge only two studies have investigated longitudinal psychotropic prescribing patterns in community-based samples of people with intellectual disabilities in the UK. Both studies were large and relied on data extracted from primary care records. One reported antipsychotic prescribing for 17.1%, and antidepressants for 16.9% of adults with intellectual disabilities, with age being associated with both, and sex with antidepressants, similar to our T2 results.[5] The results of the other study differed, reporting antipsychotic prescribing in 27.7% of participants at the end of their study period, but also reporting a decrease of 4% per year over the whole study period, and no consistent trend in antidepressant prescriptions was reported.[5] Neither study conducted psychiatric assessments on the population, limiting the precision of findings related to clinical diagnosis and GP-recorded symptoms.

This study reaffirms the strong association between antipsychotic prescribing and problem behaviours reported in a number of other studies.[28–31] However, few studies have separately reported associations between problem behaviours and antidepressants or hynotics/anxiolytics. An Irish study which investigated rates of prescribing of psychotropics in older adults with intellectual disabilities reported no increased risk of antidepressant prescribing or any association with problem behaviours.[32]

Several studies have reported the increase in rates of antidepressant prescribing in the general population across the UK, which our findings mirror.[33–35] In Scotland the number of antidepressant prescriptions rose from 1.16 to 3.53 million per year between 1992 and 2006,[36] and women were prescribed antidepressants more frequently than men.[33] The reasons for the increase are unclear and have been attributed to multiple factors such as the availability of newer classes of drugs with fewer side effects, improved management of depression, lack of availability of alternative interventions,[36] a widening of clinical uses[33] and patient expectations. Earlier studies have cited concerns that depression may have been underdiagnosed in the population with intellectual disabilities.[37] One American study which retrospectively analysed outpatient psychiatric charts reported a higher than expected rate of antidepressant prescribing for the subgroup with intellectual disabilities and suggested this was indicative of increasing diagnosis of depressive disorders in adults with intellectual disabilities.[38] Another US study analysed data from adults with intellectual disabilities living in community settings in New York State between 2006 and 2007 and also reported a higher than expected rate of antidepressant prescribing in this group.[39] The substantial increase in antidepressant prescribing observed in the current study may indicate improved diagnosis in primary care for this population.[24] This study has also observed that problem behaviours were independently associated with antidepressant prescribing in adults with intellectual disabilities. However a systematic review of antidepressants and problem behaviour management in people

with intellectual disabilities concluded that evidence of their effectiveness in this context is lacking.[40] Longitudinal patterns of antidepressant prescribing require further investigation.

Strengths and limitations

Strengths of the study include its large size, the longitudinal design, the detailed ascertainment of the population with intellectual disabilities, and the detailed health assessments at T1. The cross-sectional cohorts were population based at T1 and T2, and so were more widely representative of the population with intellectual disabilities. Additionally, the linked cohort was similar in characteristics to the whole cohort at T1, suggesting it is representative and hence that the results are generalisable. The 12-week extraction period of PIS data was selected to account for the frequency of prescriptions being issued; it included both regular and as-required drugs. Given the 12-week prescribing period it is likely that the as-required drugs were being actively used (as a fresh prescription had been issued and was encashed by the person with intellectual disabilities/their carer during this period). The time period for encashment was identical at both time points for the longitudinal, linked cohort. As a matter of caution in interpreting the data, the case-conferenced clinical mental ill health diagnoses agreed by the study psychiatrists were used rather than ICD-10 or DSM-IV-TR diagnoses, in view of the under-recording of mental ill health that these two classification systems produce with this population; had we used either of these classifications, our results would have been even more striking in terms of the discrepancy between mental ill health and prescription of antipsychotics.

Only 73% of general practices agreed to data extraction, and this combined with deaths are likely to be the main reasons for 545/1190 of the participants being linked in the T2 data 10 years later. Limitations are the different methods of data collection, with specialist individual assessments at T1 and electronic data extraction at T2. In particular,a large proportion of information is missing and there may be inaccuracies on the recorded level of intellectual disabilities in the general practitioner data at T2, so comparison of this variable between the T1 and T2 cohorts is limited. Additionally, mental ill health data at T2 are lacking. The study did not investigate changes in dosages, polypharmacy or duration of use. Some antipsychotic drugs are licenced for indications other than psychosis, and it is possible that other conditions accounted for their use, for example promazine. Antidepressants and antiepileptics have also seen increased use in the general population over this time period for neuralgic pain. We do not know how relevant this is to people with intellectual disabilities who may have difficulties in communicating pain, and note that encashed antiepileptics did not increase between the two cohorts.

Implications for research and practice

This study has shown that fewer new antipsychotic prescriptions are being initiated, but patients prescribed antipsychotics in T1 were unlikely to have these drugs withdrawn over the next decade. This implies possible reluctance of carers, families and individuals to stop medications, combined with a lack of evidence available to prescribers about direct cessation interventions.[22 41] The issue therefore remains far from addressed, and the risks of long-term health problems, death and impact on quality of life associated with long-term antipsychotic prescriptions still need further highlighting.[42] This study reinforces the need for frequent medication reviews for people with intellectual disabilities, alongside further research to investigate the long-term effects of antipsychotic medications on this population.[8] Further research to examine the barriers to antipsychotic drug reduction and to evaluate approaches to promoting reduction and withdrawal of antipsychotics for people with intellectual disabilities is needed. There is a dearth of evidence on antidepressant prescribing in the population with intellectual disabilities. The sharp increase in antidepressant prescribing observed in this study demands further research to understand the drivers for this practice. The association between increasing age and prescribing of antipsychotics and antidepressants also supports calls for research to investigate the implications of long-term psychotropic prescribing for older people with intellectual disabilities.[43]

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Contributors AH and PM analysed the data, jointly interpreted it. AH wrote the first draft of the manuscript. AH, S-AC, DK, CM, and LA jointly conceived the project, interpreted the data and contributed to the manuscript. AM contributed to additional analyses in response to reviewer comments. All authors approved the final version of the manuscript. S-AC is the study guarantor.

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Competing interests All authors have completed the ICMJE uniform disclosure form at www.icmje.org/coi_disclosure.pdf and declare: no support from any organisation for the submitted work; no financial relationships with any organisations that might have an interest in the submitted work in the previous 3 years; no other relationships or activities that could appear to have influenced the submitted work .

Patient consent for publication Not required.

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ORIGINAL ARTICLE

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Inclusive peer review: Reflections on an adapted citizens' jury with people with learning disabilities

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Abstract

Background: Inclusive research is widely accepted as an essential part of the process to democratise knowledge creation and dissemination. However, while peer review is an important part of academic publishing, the potential to include people with learning disabilities in this element of the research process has not previously been explored using a deliberative approach.

Methods: Accessibility adaptations were made to the citizens' jury approach enabling people with learning disabilities to participate. Sixteen adults with mild to moderate learning disabilities were recruited to participate in the adapted citizens' jury. Jury members took part in capacity-building workshops to develop their knowledge of research and research processes. Six expert witnesses presented evidence to the citizens' jury and were questioned on aspects of inclusive research, representation, peer review and academic publishing processes. Facilitators supported citizens' jury members to reflect on the evidence presented and to develop recommendations for inclusive peer review.

Findings: The citizens' jury was an effective inclusive research approach in this case. Jurors made recommendations related to the question of inclusive peer review: inclusive reviews should be done by groups rather than individuals; the research under review must be in accessible formats and on relevant topics; reviewers need sufficient time to conduct reviews; and diverse groups of people with learning disabilities should be involved.

Conclusions: People with learning disabilities appreciate the importance of peer review but do not necessarily want to participate in it. This jury suggested creative approaches to disseminating, reviewing and engaging with research, including building more opportunities for dialogue between researchers and self-advocates. The adapted citizens' jury was a novel and effective method of supporting deliberation on this topic but other approaches to including the views and experiences of those with more severe learning disabilities should be explored.

KEYWORDS
citizens' jury, deliberative democracy, inclusive research, intellectual disabilities, learning disabilities, peer review

Accessible Summaries

- Peer review is an important part of making sure that research is done properly.
- The project was set up to make recommendations about how research reviews could be more inclusive of people with learning disabilities.
- We used an accessible citizens' jury to talk about research reviews with a group of people with learning disabilities.
- People with learning disabilities understand that reviewing research is important. The group suggested different ways that people with learning disabilities could be involved in publishing and sharing research.
- We found out that an accessible citizens' jury is a good way to talk about complicated issues with people with mild to moderate learning disabilities.

1 | INTRODUCTION

The objective of this paper is to critically evaluate the adapted citizens' jury method that we adopted in a novel project concerned with developing recommendations for the largely unchartered territory of including people with learning disabilities in the peer review process for the British Journal of Learning Disabilities (BJLD). In evaluating this innovative approach, we discuss the jury's priorities in engaging with research that concerns them and discuss the outcome. We begin by outlining the key ideas in the paper.

BJLD is committed to supporting the rights of people with learning disabilities and working to improve their lives (Nind, 2020). Inclusive research is a core part of democratising the research process and opening it up to people with learning disabilities. It enables people with learning disabilities to take active roles in the research process and not just be the object of the academic research gaze. However, as we show in our literature review below, the peer review process has tended to remain the purview of academics and the involvement of people with learning disabilities and other researchers outside the academy happens rarely.

Peer review is an essential part of the research cycle and involves other researchers (peers) commenting on the quality of a paper. Most authors seek feedback from their peers even before submitting a paper for consideration by a journal for publication. After submission, the peer review process becomes a formal part of the process of deciding whether to accept or reject the paper and giving feedback on how it might be improved. As Wiley (2023), the publishers of this journal, clarify, peer review is designed 'to assess the validity, quality and often the originality of articles for publication' maintaining academic integrity by 'filtering out invalid or poor-quality articles'; it has to establish that the research was done responsibly, with respect, and following agreed rules. The nature of peer review is that the feedback comments are made by experts in the same field as the author/s.

We created a citizens' jury of people with learning disabilities to deliberate on what the inclusion of people with learning disabilities might add to the research process (the wisdom of doing this) and what kind of model of inclusive peer review might work (the feasibility). We wanted to explore these wisdom and viability dimensions of inclusive peer review for a journal that values dialogue with people with learning disabilities *and* the usefulness of citizen jury as a method for doing this. We describe citizens' juries in more detail in the methods section.

2 | LITERATURE REVIEW

Inclusive research, which has become gradually more established in learning disabilities research since the 1980s, asserts that disabled people can be active participants in research rather than mere objects of research. It has been named, characterised, advocated for and experimented with as a phenomenon of importance to people with learning disabilities and their allies. Walmsley and Johnson (2003) originally characterised inclusive research as beyond participation; it was research in which disabled people owned the research problem, collaborated in the research process and exerted some control over it, such that the research represented their views and experiences, and became for their benefit and improved lives, and was respectful and accessible. Writing recently with Strnadová, they have expanded their defining characteristics to stress that the research must be aimed at social change and be research 'in which those involved in it are "standing with" those whose issues are being explored or investigated' (Walmsley et al., 2017, p. 758). Inclusive research, therefore, is unashamedly political (Woelders et al., 2015), becoming more so if anything. Exploring the rise of inclusive research across fields and disciplines, Nind (2014) argues for its position within a wider movement to democratise research.

For this paper, what matters most is what people with learning disabilities say about inclusive research and why it—and aspects of it—matter to people. García Iriarte et al. (2023) show a level of maturity emerging in which established inclusive research teams can evolve and become self-critical. Nind and Vinha (2014) and Armstrong et al. (2022), using dialogue to take stock of progress in inclusive research note that we ask it to do a lot, including producing important, impactful findings via egalitarian processes that are empowering for self-advocates and self-advocacy. Hence, as Woelders et al. (2015) argue, idealisation can take root, expecting inclusive research to be some kind of panacea. In 'a world where people with learning disabilities are routinely excluded' (Armstrong et al., 2022, p. 314), inclusive research has become a symbol of change as well as a route to change; it represents the rights of people with learning disabilities to 'interpret their own lives' (p. 317).

The political and ethical arguments for the democratisation of research urge us all to be inclusive and to support the building of capacity for undertaking research among people with learning disabilities interested in generating their own knowledge. One of the conclusions of Armstrong et al. (2022, p. 326) is that such investment in building capacity 'is not a matter of teaching them research skills but supporting them to understand the academic world so that they can contribute'. Through being involved in all stages of research, various groups of people with learning disabilities have come to understand research bidding, ethics applications, data generation, data analysis and dissemination, including in academic journals (O'Brien et al., 2022; Tilley et al., 2021). Much of this learning has been through researching alongside academic researchers, doing research collaboratively, asking questions, posing challenges, and solving problems (Bigby & Frawley, 2010; Nind, 2016). BJLD has supported inclusive research (see O'Brien et al., 2022) and fostered the involvement of people with learning disabilities in academic roles as collaborating authors (e.g., Mikulak et al., 2022; Townson et al., 2004), guest editors (e.g., Blunt et al., 2012; Chadwick et al., 2023) incorporating an element of reviewing, and since 2020 with the *In Response* initiative, as partners in academic dialogue (see e.g., Lewis et al., 2020).

In this paper we focus on the largely unexplored role of people with learning disabilities as peer reviewers. Reflecting on managing a whole special issue of BJLD, Blunt et al. (2012, p. 83) comment:

> it is time that people with learning disabilities became involved in *more than just* co-writing and research. We wanted to see a partnership approach to the whole process of peer review and publishing research.

They add:

> The process of peer reviewing and making decisions about the articles submitted was not an easy one, and we think we have learnt some important lessons that we would like to share with you.

These lessons include how to make difficult decisions on which papers to accept and reject, how to deal with the emotional impact of papers addressing distressing issues, how to handle difficult and inaccessible language, and how to respond when authors write as if people with learning disabilities are all the same. Most recently, Cameron Richards, self-advocate, co-guest editor of the special issue on digital inclusion, echoed the accessibility challenge point and reflected: 'I got a lot of new knowledge and work experience out of being an editor' (Chadwick et al., 2023, p. 122). Similarly, Lewis et al. (2020, p. 271) ended their inaugural *In Response* paper with, 'Thank you for giving us the opportunity to respond. It is rare to be asked what we think about academic research'.

There is a nascent sense then, that inclusion in the world of academic publishing of research is appreciated by people with learning disabilities who have taken up opportunities to participate. Giving people recognition and a 'bridge' into new worlds constitute some of what we ask of inclusive research (Nind & Vinha, 2014). There is richness and purpose in having diverse perspectives informing research outputs, and epistemic justice in hearing from people with learning disabilities when it comes to research about their lives (Armstrong et al., 2022). However, there are strong warnings in the literature about bringing difference into dialogue rather than eradicating it in 'striving for normalisation', which 'can be paralysing' (Woelders et al., 2015, p. 528): Inclusive researchers may need to resist pressures for people with learning disabilities to do research roles in the same way as academic researchers, while still achieving the same levels of value for their distinctive part in the research process as well as their empowerment.

In the next section, we report on the methods we adopted to further explore, alongside people with learning disabilities, their role in the peer review process using the specific approach of an (adapted) citizens' jury. Citizens' juries have developed as an approach to public engagement that is underpinned by deliberative democratic theories (Street et al., 2014). In common with other methods of deliberative democracy, the citizens' jury approach is based on the principles of informed deliberation between a group of citizens on a specific topic. Citizens' juries have been widely used internationally to enhance public engagement with public policy-making processes, particularly around complex and contested issues of concern to communities (Roberts & Escobar, 2015; Tully et al., 2019). Wakeford and Walcon (2015) describe the process as bringing together:

> Twelve or more members of the general public (the "jurors") [to] participate in a process of dialogue under the guidance of a chair or "facilitator". They interrogate specialist commentators (sometimes called witnesses) chosen because of their knowledge of a particular subject … Jurors then draw up and publish their conclusions.

A key part of the process, which was developed by the Jefferson Center (now the Center for New Democratic Processes) in the United States, is the deliberation, usually over 2–7 days (Involve, n.d.). As the

popularity of this method of deliberative and active citizenship has increased so has the development of adaptations to the original model. Street et al. (2014, p. 8) argue for the importance of these adaptations in furthering our understanding of 'how various methodological decisions can shape jury processes and outcomes'.

Henderson et al. (2022) developed and evaluated an adapted citizens' jury model to enable a group of people (aged over 16 years) with mild to moderate learning disabilities to deliberate on the broad area of inclusive health research. The jurors on this project made 10 recommendations for inclusive health research and their work informed the development of the peer review citizens' jury project we report on here. The adapted citizens' jury model developed by Henderson et al. (2022) prioritised four components of citizens' juries: recruitment of a demographically *representative* group of people with learning disabilities, who were then *informed* through the provision of high-quality accessible information relevant to the project; witness testimonies were *impartial*, and the process was facilitated to enable a *deliberative* approach. In their review of public engagement in a research priority exercise, Gooberman-Hill et al. (2008) concluded that the structured processes of a citizens' jury (evidence sessions, facilitation and deliberation) facilitated informed and deep engagement with the topic. The adaptations to the Citizens' jury model described in detail by Henderson et al. (2022) had previously demonstrated that people with learning disabilities, when effectively supported, can deliberate and form consensus recommendations on complex issues. This provided an empirically validated methodological foundation for an approach well-suited to the aims of this study on inclusive peer review.

3 | METHODS

The current study investigated:

- What can a citizens' jury tell us about the viability and wisdom of inclusive peer review in the quest to democratise the research process and include dialogue with people with learning disabilities?
- To what extent is the adapted citizens' jury approach an effective method for delivering rapid research evidence in keeping with the ethos of doing research inclusively?

Ethics approval was given by University of Southampton Faculty of Social Science Research Ethics Committee [no. 70302]. Participants, who were people with mild to moderate learning disabilities, were given accessible information sheets and consent forms and the opportunity to discuss the research with people who support them and the research team before giving informed consent.

The citizens' jury had to adapt to the constraints of a short project timescale and the COVID-19 pandemic which impacted on demographic representativeness of the jury and mitigated against in-person meetings. Therefore, one major adaptation made to the method for this project was in the selection of participants. In essence, these were a convenience sample of self-advocates known to the researchers. Some of the participants attended as independent individuals who had collaborated with the researchers in previous projects and others as members of self-advocacy organisations. In keeping with wider uses of citizens' juries, however, the findings were to be fed back to policymakers (in this case the editorial board and publishers) to inform policy decision-making.

There were 16 individual participants (9 male, 7 female), including five organisations and three countries—England, Scotland, and Wales. They had previous experience of inclusive research and in most cases of self-advocacy. Four additional people from the participating advocacy organisations were present in the meetings to provide practical support for participation to the self-advocates (the jury members). The role and remit of the jurors was to hear evidence from expert witnesses on inclusive peer review, to deliberate as a group on the evidence presented and to develop a set of evidence informed recommendations on which all jurors agreed. The language of juror comes from the original citizen jury method. Expert witnesses were recruited to present their perspectives on issues relevant to deliberation on inclusive peer review.

The jury process, adapted from the Center for New Democratic Processes (Figure 1) took the jurors through the stages of capacity-building, considering evidence, and finally collaborating on guidelines and recommendations for inclusive peer review. Two preparatory 2-hour workshops, using the Zoom platform were delivered to introduce the jurors to each other, develop their knowledge of research and research processes including the role of peer review, and to agree ground rules. These workshops were followed by four online sessions where the jury members heard evidence from six expert witnesses on separate topics. The topics were selected to enable jurors to hear and deliberate on evidence relating to inclusive research and peer review processes. Expert witnesses were drawn from diverse backgrounds and were selected on the basis of their knowledge and experience of self-advocacy in research, academic authorship, as experts by experience and in academic publishing. All had knowledge of peer and academic review processes, with the exception of the expert by experience. These evidence sessions addressed the topics of: (i) inclusive research, focusing specifically on defining this concept and reflecting on inclusive research practice; (ii) speaking for others (e.g., people with profound and multiple learning disabilities), which considered issues of representativeness within self-advocacy groups; (iii) giving and receiving a research review, examining the concept and practice of academic peer review; and (iv) the editor's and publisher's perspectives on the role of peer review in the editorial and publishing process. Jurors heard from each expert witness who explained relevant concepts or issues in a 20-minute presentation. Following each presentation, the jurors prepared questions in discussion in small groups. Each group was facilitated by a member of the research team who supported jurors to reflect on the presentation, and using prompts to ensure key issues were discussed. However, it was the jury members who agreed the final questions for the expert witnesses. Expert witnesses were given guidance on accessible presenting and asked to share their presentation with researchers in advance to support this aim. In a

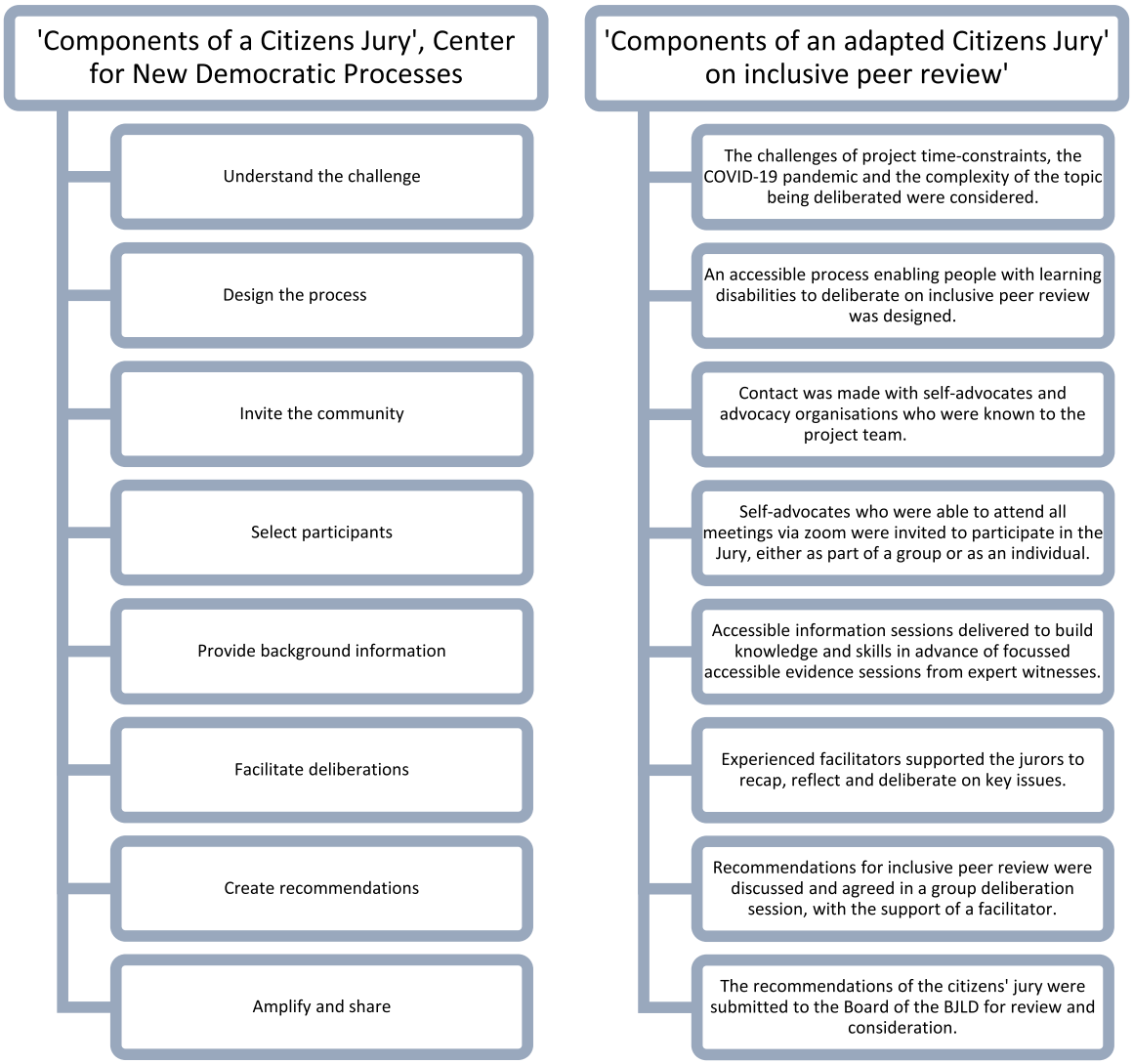

FIGURE 1 Components of an adapted Citizens' Jury.

final 2-hour session dedicated to review, deliberation and consensus forming, jurors worked together in groups, with facilitation from the research team or support staff, to find consensus and make recommendations. This session drew on analysis of recordings and summaries from all jury sessions (including data from Facebook summaries of questions and responses). A total of seven online sessions were held across the project, each session was video-recorded via Zoom. The jurors also had a group Facebook page that hosted a video of the presentations to jurors and accessible summaries of questions and discussions that followed expert witness sessions. These were available for reference and for communication and community-building between sessions. The preparatory, evidence and deliberation sessions were delivered and facilitated by academic researchers Abigail Croydon, Rhiann McLean, Angela Henderson and Angeliki Katramadou.

In addressing the wisdom of inclusive peer review, we sought to investigate the extent to which the inclusion of people with learning disabilities in peer review could add value to this part of the research process. If worth doing we needed to investigate how feasible it was and what the challenges were that we would have to negotiate. Findings in relation to these questions were drawn from the recommendations reached through the jury process. In the final meeting, jurors reviewed expert witness presentations and points made during deliberations to develop guidelines for inclusive peer review. Their conclusions were summarised in accessible language with visual supports by the research team and returned to jurors for comment and amendment. In this sense, the findings emerged from a thorough participatory process of analysis, accessible presentation, and review/amendment by jury members. For the present paper, Abigail Croydon reviewed the workshop recordings to identify potential omissions regarding coverage of the issues and to select quotations that aligned with findings. The findings with respect to the usefulness of the jury method are presented thematically and we drew these from the juror's and moderators' comments on the jury experience captured in the session recordings, field notes and reflections from the academic team.

A member of the citizens' jury, John Cassidy, agreed to collaborate in the development of this manuscript and was supported by Angela Henderson to reflect on the initial objectives of the project and the extent to which these were achieved. Through six meetings, John and Angela met (via a combination of virtual and in-person meetings) to discuss, agree and amend sections of the manuscript and to reflect on specific themes from the perspective of a jury member. All contributing authors met via Zoom at different stages in the development of the manuscript to support this collaboration.

4 | FINDINGS

The findings address first the jurors' conclusions about the viability and wisdom of inclusive peer review before addressing the effectiveness of the citizen's jury method. We summarise the recommendations made by the Jury to BJLD, which were reached following the final review session where jury members were supported to reflect on all previous jury sessions, undergo further deliberation and reach a group consensus.

4.1 | Inclusive peer review

Jurors were confident in their understanding of the logic of inclusive research. Some referred to extensive experience within research teams. Three themes emerged early and recurred regularly in later sessions. First was the perennial issue of accessibility of information about research ('a journal needs to be easy read and accessible and photo symbols' said one juror). The second was the frustrating slowness of academic research processes, especially in terms of delivering social change. This was a particular concern for jurors engaged in activism. Third, and most positively, jurors talked about the value of learning collectively about research, summed up by one juror as 'the more we get together and talk, the smarter we get, and knowledge is power'.

Bearing in mind the priority for diverse perspectives to inform research outputs, and our recruitment of people involved in self-advocacy and with prior research experience, we explored with jurors their role in 'representing others', especially those experiencing more severe disability. The expert witness for this session presented on the challenge of participation and 'voice' for people with profound and multiple learning disabilities. She introduced the example of her daughter and highlighted the challenge—'we want to give them their voice and not give them our voice'. This session was effective in extending thinking about the means of inclusion and representation at the intersection of learning disability and academic research. A juror reflected on the difference between his aspirations for himself and what the witness's daughter's preferences might have been:

> My voice is like trying to find the right sort of questions to ask, to try not to say something which might feel like the wrong thing to say … I want to be independent, but she couldn't be … not everyone can or even wants to do this…

His reflection gave rise to discussion of whether the witness and her daughter together might be able to take part in peer review and question to the witness, 'is there a way in which teams can get involved rather than individuals?' The idea of joint peer review developed through subsequent sessions as jurors thought about the 'know-how' challenges involved in peer reviewing and the social, well-being and learning priorities of people with learning disabilities. Working on reviewing in teams might reduce the investment of time and effort involved in addressing peer review, while providing meaningful social and learning opportunities important for the wellbeing and agency of people with learning disabilities.

Jurors considered who might be invited to do peer review. A juror with experience of Patient and Public Involvement in research made the case to widen recruitment beyond self-advocacy groups as 'more people with disabilities are not in self-advocacy'. In their recommendations, jurors emphasised the importance of diverse representation of groups of people with learning disabilities in inclusive reviews.

The BJLD *In Response* initiative provided the expert witnesses for the issues of receiving and giving feedback on a journal paper. Jurors heard from the lead researcher of a project on fathers with learning disabilities (Symonds et al., 2021) and from members of the group that had read and responded to the paper (Williams et al., 2021). This session produced intense discussion, with focus on questions about recruitment and the findings of the research as well as the reviewing process. Jurors with experience of parenting saw this as an outstanding example of research with personal relevance. The associated video featuring reconstruction of vignettes and debates from the research contributed significantly to communicating the aims and findings and this feature was a focus of deliberation.

The expert witnesses who had given feedback on the journal paper made powerful points regarding the process of choosing what to engage with. The 'accessible' summaries they selected included some that they regarded as wholly inaccessible, meaning that 'people aren't going to want to engage with that research'. Following this session, in particular, jurors discussed the limitations of easy read formats ('I'm no bothered about easy read') and moved towards seeking video and audio research summaries.

Jurors developed the theme of investing time and effort in producing inclusive peer reviews. There was some scepticism about the balance between effort and reward, in terms of pay, intrinsic reward and achieving social change. Developing reviews in in-person meetings and in dialogue with academic authors (the process followed in the *In response* initiative) appeared to improve the balance. The witnesses on giving and receiving feedback gave positive accounts of the benefits of the experience, with the reviewers saying, 'we all felt - what's the word - boosted by being part of this conversation'. This contrasted with the academic witness' account of anonymous peer review processes as sometimes 'nerve-wracking'.

The research topic of the session (fathers with learning disabilities) prompted discussion about the purposes of learning about research for jury members. One juror was 'spellbound' by the topic, 'it's so close to my heart, I got a lump in my throat thinking about this'. He returned to the theme raised earlier of the benefits of meeting and talking in groups about research, 'what I think is missing is the encouragement to show people with disabilities that you can help improve your life or somebody else's life because you learn something … researchers don't make enough of that right, they're just "let's get your answer"'. This was a key reason for seeking group debate about research—its function in terms of learning and well-being: 'if a guy finds out that it's no just peer review, but it could help their mental health—that would encourage a man [otherwise it's] "can't be bothered mate"'. This juror argued for structured and supported debate among peers with learning disabilities about personally relevant research, whether producing a peer review or another form of engagement with research. Others endorsed this emphasis on learning, though there was also contrast with activists who saw research more in terms of serving the purposes of activism. This debate arose in response to some jurors' doubts about the benefits of peer review to people with pressing practical difficulties: 'people might ask themselves: What's the point in joining the peer review groups?'

The discussion highlighted that jurors' interest in reviewing research was selective, even conditional. One juror argued that if research 'said something about your life', it might be worth the time and effort involved in getting to grips with it. This became a key point in the guidance for inclusive peer review. Jurors who saw themselves as primarily activists sought better focus by researchers on orienting research towards support for self-advocacy campaigns. Other aspects of research, setting research priorities and taking part in dissemination/impact activities, in particular, might have a higher priority for people with learning disabilities. An advocacy group leader felt that people might need a clear understanding that contributing to peer review might deliver change in academia rather than wider social change. Implicit in these strands of thought was a resistance to the exclusive nature of academic writing and publishing, its priorities, timescales, and accessibility ('you can't buy [journals] in WH Smiths or on Amazon or anything'). Ultimately, the set of consensus recommendations on inclusive peer review were that:

- Reviewing should be done by groups not individuals, in dialogue with authors, not anonymously.
- Research for review must be genuinely accessible and engaging, for example, using video and audio formats.
- We would like to review research concerning topics that we know about and are relevant to our lives.
- We need time for processing information and engaging in debate about the research reviewed, which means proper funding.
- We want to include as many different people as possible, including people with more severe disabilities, people who are not members of self-advocacy groups and people from black and minority ethnic groups.

The jurors also had concerns and practical suggestions for improving research outputs:

- We need an accessible research journal for all people not just people in universities.
- We need forums for learning and discussion about research besides peer review.
- We would like to be paid, but some of us think that taking part is more important than being paid.

4.2 | The citizen's jury method

Street et al. (2014) recommend that in evaluating citizens' juries, special attention should be paid to recruitment, moderation, and jury duration. In the case of people with learning disabilities, there are intrinsic difficulties in attempting to represent the population concerned. In our research, short timescales and limited funding also ruled out seeking to represent the demographic profile of the population with learning disabilities. Knowledge of inclusive research was determined to be a requirement to progress efficiently to deliberating the case for peer review. This meant that juror recruitment focused on people with stronger verbal skills and milder learning disabilities. However, we sought to address the question of representation as part of the jury process itself. Citizens' juries though lend themselves to populations who can converse with ease.

With respect to moderation, the research team, who all work in learning disabilities research, selected and briefed witnesses, moderated all sessions and summarised group questions and discussions in accessible formats for jurors' reference between sessions. Two team members provided support to breakout groups of jurors to facilitate discussion and develop questions for witnesses. We balanced the priority to make progress on the topic with the wider remit of enabling participation and deliberation. Facilitators experienced some limitations in providing remote support to jury members, including technical support. This was particularly the case where members attended as a group sharing one screen, preventing them from joining breakout rooms as individuals. This meant that the research team could not always distinguish individual voices, including the voice of group support staff from that of jurors.

The duration of our jury was 14 hours, over a 7-week period, a comparatively long participation compared with others reviewed by Street et al. (2014). Nevertheless, some participants felt there was not enough time to process information and to deliberate. This may have reflected, in part, the conceptual difficulty of the topic of peer review, but also jurors' interests in debating issues of inclusion, accessibility, and research dissemination beyond peer review.

The Facebook group was designed to extend the possibilities for participation, by giving jurors access to presentations and questions, and the facility to engage in debate between sessions. There was little active participation in this, though participants referred to the resources. A greater sense of community and participation might have been achieved through attendance in person, though this would have reduced geographical spread. Jurors' concerns at the outset

(e.g., regarding the accessibility of language) developed as they deliberated. The idea of group peer review, a key recommendation, gained traction across the sessions as jurors became confident that the academic format could be reformulated.

Jurors reported that the jury process had provided an enriching, collaborative experience, which was valuable in terms of learning, opportunities to collaborate with others with learning disabilities and to be heard in a supportive environment. The jury process successfully established the jurors' views on academic review and provided a strong foundation for developing a more inclusive approach to academic publishing. In summary, the citizen's jury method adapted successfully to provide guidance on inclusive peer review from a particular learning disability perspective at short notice. It was experienced as democratic and deliberative in line with the wider principles the citizens' jury concept.

5 | DISCUSSION

This novel and inclusive approach to exploring the potential role of people with learning disabilities in the specific area of peer review makes a significant contribution to the conceptualisation and practice of inclusive research. In this project we investigated the viability and wisdom of inclusive peer review through a structured dialogue with people with learning disabilities, using an adapted citizens' jury method. A key driver was to provide recommendations to the editor and publishers of BJLD, on how to further democratise the academic publishing process, and specifically on how to further include the perspectives of people with learning disabilities. Thus, following a process of deliberation and consensus forming this citizens' jury produced a series of clear recommendations on the question of the viability and wisdom of inclusive peer review.

The jurors welcomed the inclusive potential of peer review by people with learning disabilities. Their consensus recommendations, however, did not suggest methods of replicating the peer review system that is already an established part of the academic process. In exploring the potential for people with learning disabilities to become meaningfully engaged in peer review we were reminded of the argument of a researcher with learning disabilities that 'it was not important to do exactly the same thing as an academic researcher did, but to do what was within her capacity' (Woelders et al., 2015, p. 538) and of the 'taken-for-granted academic frameworks and demands' (p. 539). Instead, the jury recommendations prioritised the need to foster dialogue between authors and reviewers, community representation, participation in reviewing research of direct relevance to their experiences and interests and improving accessibility of research outputs. The citizens' jury on inclusive peer review thus reinforces the conclusions made by Armstrong et al. (2022, p. 326) that self-advocates want support to contribute to the research process in ways that are defined by them and designed to further benefit their community.

Instead of suggesting ways to engage in the academic peer review system, the citizens' jury recommended establishing a platform for dialogue between people with learning disabilities and authors as a prerequisite for democratising research. If, like Jones et al. (2020) and Milner et al. (2020) who cite them, we draw on the lens of relational equity, we also need to appreciate that many of the ontologies and epistemologies at work are contestable. It may be that in seeking to admit people with learning disabilities to another academic process, there are dangers of assimilation too, in requiring them 'to approximate ourselves' (Milner & Frawley, 2019, cited by Milner et al. (2020, p. 128). These points are further reinforced by the jurors' recommendation that reviewers should be able to choose topics of personal interest, otherwise, as one member said, 'people might ask themselves what's the point?'

We see a real resonance in our findings from this project with the liberating effect found by Milner and colleagues of people with learning disabilities refusing traditional methods or roles, preferring to steer things according to their motivations. The power of the *In Response* initiative is that people with learning disabilities can 'speak back to' academic research about their lives, rather than following an academic agenda about the quality of papers which matters more to people who have not taken on an academic researcher identity. In reflecting on his own experiences while we were writing this paper John Cassidy said that some research topics would be too troubling for him to be a part of.

Jurors also agreed that representation of diverse voices within the learning disabilities community was important. They expressed their concerns on representativeness in two ways. First, concern that people with more severe learning disabilities might not have the chance to contribute to inclusive research and second that inclusive research groups often tend be drawn from established self-advocacy groups, which they argued could lead to exclusion of more diverse voices and experiences. This is resonant with the systematic review of inclusive health and social care research in which Hewitt et al. (2023, p. 698) reflect on representativeness and highlight the potential risk of creating a 'subsection of researchers with [learning disabilities], who no longer hold the position of "outsider"' and are therefore less likely to challenge the established research paradigm. The jury's concern that people with more severe learning disabilities were generally excluded from inclusive research and their assertion that efforts should be made to ensure their voices are heard echoes the argument of de Haas et al. (2022, p. 2) that 'inclusive research has not been able to stretch its parameters sufficiently to enable people with profound [learning disabilities] to belong' and that simple modification of existing methods of inclusive research will not achieve 'epistemic justice' for people with more profound learning disabilities. McCoy et al. (2020) also identify harms in partial representation. They acknowledge the challenge in achieving comprehensive representation and suggest that claims of representation should reflect the specific subgroup engaged with (e.g., self-advocates with learning disabilities). Alternatively, they suggest that comprehensive representation might be attempted using deliberative and consultative methods to understand the interests of all subgroups.

Throughout the deliberative process, jury members raised practical barriers to engagement in inclusive peer review. These included challenges related to the accessibility of research processes and outputs, dissemination methods, timescales and funding of research. Jury members reflected on their positive experiences of self-advocacy in research, where they felt rewarded by making a difference to the lives of other people with learning disabilities while experiencing personal growth and extending knowledge and skills. For John, as a coauthor on this paper and member of the citizens' jury these aspects made him feel empowered ('it has improved the quality of my life in all aspects').

Our reflections on the effectiveness of the adapted citizens' jury method of delivering rapid research evidence in keeping with the ethos of doing research inclusively were generally positive. In their systematic review of the application of the citizens' jury method in health policy decision-making, Street et al. (2014) argue that pragmatic adaptations to the 'ideal' Jefferson Centre [now the Center for New Democratic Processes] citizens' jury method are necessary to the development and application of effective, influential, and inclusive community engagement. However, they also assert that it is important to reflect on and record the impact of 'methodological decisions' on the essential component of providing an 'unbiased inclusive deliberative process'.

At an early stage in this project, we opted for pragmatic recruitment at the expense of demographic representativeness, which was a significant adaptation. This adaptation, Wise (2017) would argue, risks introducing bias to the project. However, for John and others, their prior experiences of inclusive research were important to the deliberative process.

> In my first experience of inclusive research, I was shy and didn't have as much confidence and I wasn't as outgoing. I think if I didn't have that experience, I might not have been able to participate in this remote group so well. Having the opportunity to be involved in the remote group during COVID really helped me to get through that time - being involved [has] been so important to my mental health.

In thinking about the limitations of our method in relation to inclusion of a broader spectrum of people with learning disabilities, we are reminded by de Haas et al. (2022) that a more radical rethinking of inclusive research practices is necessary to include those with profound learning disabilities.

A further limitation of our adapted method relates to the role of support staff who facilitated access to online meetings. Teasing apart people's voices can be important for understanding power dynamics (Chapman, 2014; Woelders et al., 2015) and this was sometimes difficult in our sessions. A clearer set of guidelines on the role of advocacy support workers, and whether their own views were to be included in the project or not would have been useful and could be incorporated in future citizens' jury work of this kind.

6 | CONCLUSIONS

The citizens' jury was tasked with considering the viability and wisdom of inclusive peer review in the quest to democratise the research process and presented five well-considered and practical recommendations on this. The recommendations indicate that people with learning disabilities perceive inclusive peer review as more than an opportunity to replicate traditional academic roles. Instead, they seek to challenge and contribute to changing the established system, which has historically positioned them only as research subjects. The jury's recommendations are aligned with the BJLD's *In Response* approach and provide a route-map to furthering inclusive approaches to research generation, communication, and impact.

The adapted citizens' jury model was an effective method for investigating a complex conceptual aspect of the inclusive research paradigm. However, further adaptations would be needed to facilitate the representation of a wider range of people with learning disabilities.

The aim of this citizens' jury was to focus on the question of inclusive peer review. However, the group demonstrated that their involvement was driven by a commitment to addressing the routine exclusion that people learning disabilities experience in all domains of life. They advocated for more accessible information on the outcomes of research, particularly research perceived to be of relevance to them. They sought opportunities to collaborate for the benefit of other people with learning disabilities, including those whose voices were not represented in the jury. The resulting recommendations do not align neatly with academic journals' definition of peer review but offer an approach to inclusion in research publishing that could complement the existing peer review process. Their guidelines could facilitate the dissemination of research knowledge, promote dialogue between researchers and people with learning disabilities, and bring new perspectives to the validity and quality of research. The attention of the editorial board of BJLD is now on progressing this agenda.

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CONFLICTS OF INTEREST STATEMENT

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Research data are not shared.

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Chapter 7:

Including the voices of people with learning disabilities in health research

by Angela Henderson, Rhiann McLean & Deborah Kinnear.

Learning disabilities occur before adulthood and people with learning disabilities will require different levels of support for learning and independent living. People with learning disabilities can experience difficulties in understanding complex information, learning new skills and living independently. However, learning disabilities vary greatly and with support people with learning disabilities can live independent lives. People with learning disabilities experience higher rates of poor health and multimorbidity (Kinnear et al., 2018) and die, on average 20 years earlier than other people (O'Leary, Cooper, & Hughes-McCormack, 2018) . There is consensus that people with learning disabilities should be included in research that aims to better understand and address these inequalities. This is reflected in the growing number of academic studies that seek to describe and identify inclusive health research (Frankena, Naaldenberg, Cardol, Linehan, & van Schrojenstein Lantman-de Valk, 2015; Schwartz, Kramer, Cohn, & McDonald, 2020). There is also a greater focus in public policy emphasising the need to include patients in health research and service development (Bigby, Frawley, & Ramcharan, 2014; Crook, Tomlins, Bancroft, & Ogi, 2016).

In recent years there has been a growth in the development of frameworks that conceptualise and describe the essential elements and principles of inclusive research (Bigby et al., 2014; Frankena et al., 2019; Walmsley, 2004). Despite this, examples of research that systematically engage people with learning disabilities are rare and people with learning disabilities face multiple barriers to engagement in research that seeks to identify, influence and address their health needs. In a review of clinical trials, Feldman et al (2013) found that only 2% of randomly selected trials explicitly included people with learning disabilities (Feldman, Bosett, Collet, & Burnham-Riosa, 2014). In this chapter we provide a detailed account of the planning and

delivery of a project to include people with learning difficulties in research and to work with them to establish a set of research priorities.

Background

There are multiple barriers to inclusive health research with people with learning disabilities, including: attitudinal barriers; viability and ethical concerns; communication barriers and systemic barriers. With regards to attitudinal barriers, 'gatekeepers' often block access to people with learning disabilities due to perceived vulnerabilities/lack of capacity to give consent (Lennox et al., 2005). A further barrier is concern by various professionals around the viability and ethics of including people with learning disabilities in research. Communication can be a barrier for some people with learning disabilities who have difficulty with language comprehension, abstract thought and memory and are often acquiescent in their views with those they see as professionals. Finally, systemic barriers can include research commissioning processes which can mitigate against robust engagement. Overcoming these, and other barriers, to inclusive research demands concerted action to develop and share good inclusive practice and resources.

The Scottish Learning Disabilities Observatory was set up in 2015 to address the gap in evidence about the health of people with learning disabilities by conducting research, using routinely collected health data, to help inform the development of public policy and clinical practice in Scotland. From the start, we were clear that we wanted to address some of the big challenges of including people with learning disabilities in different stages of the health research process – challenges we felt had led to tokenistic involvement or even worse, to the total exclusion of people with learning disabilities from research. This was the beginning of our journey towards the development and adaptation of a model of deliberative democratic engagement. We recognised that if we were to adopt a truly inclusive approach to involving people with learning disabilities in our research programme then we would need to be innovative as well as ambitious. It was also clear from the start that whatever method we adopted would require a significant, dedicated resource. In 2018 we secured funding for the 'Research Voices' project from The Wellcome Trust Public Engagement Fund with the aim of developing and testing an adapted Citizens' Jury on health research. The project aimed to:

- Provide insight into the views of people with learning disabilities on health research
- Challenge attitudinal barriers to involvement of people with learning disabilities in research as participants, subjects and collaborators
- Challenge structural barriers that limit opportunities for research that includes people with learning disabilities
- Develop, evaluate and share project resources
- Generate recommendations supporting inclusive health research

What is a Citizens' Jury and why did we choose this method?

The Citizens' Jury method engages members of the public or a community in important issues, enabling them to make informed decisions and to produce recommendations through a process of deliberation. Although there is consensus that the Citizens' Jury method has evolved considerably to encompass a range of different models, the Research Voices project best fits within the Wakeford, et al. (2015) definition of:

> *Twelve or more members of the general public (the 'jurors') participate in a process of dialogue under the guidance of a chair or 'facilitator'. They interrogate specialist commentators (sometimes called witnesses) chosen because of their knowledge of a particular subject… Jurors then draw up and publish their conclusions. (Wakeford T., 2015)*

The method focuses on presenting good quality evidence and supporting jury members to debate, deliberate, challenge and find consensus. Citizens' Juries are often effective when exploring complex issues with social or moral implications and have been applied across a wide range of policy areas across the globe. However, there are few examples of this approach being used to support deliberation with people with learning disabilities.

People with learning disabilities experience multiple barriers to engagement in research and our aim was to implement a method that would enable deliberation on health research at a conceptual, ethical and practical level. These core elements of the Citizens' Jury model provided a useful framework to allow people with learning disabilities to consider complex

ideas around inclusive health research. However, a number of significant adaptations to this method were required to enable deliberative engagement with a demographically representative group of people with learning disabilities.

The Research Voices project team had considerable experience of working on a range of engagement projects with people with learning disabilities but had not planned or delivered a Citizens' Jury before. The design of this project relied on evidence of best practice in other Citizens' Juries, including the Scottish Health Council 'Our Voice Citizens' Jury on Shared Decision-making (Scottish Health Council, 2020) and Roberts and Escobar's in-depth evaluation (Roberts J., 2015). However, our review of other Citizen Jury practices was limited by the lack of inclusion of people with learning disabilities. To address this gap, the project team liaised with People First Scotland, a national self-advocacy organisation that held a Citizens' Grand Jury in 2011. This meeting helped the team to consider necessary adaptations in the early design and development stages.

A Citizens' Jury is a useful method for projects who want to facilitate in-depth debate about challenging social issues. A Citizens' Jury may be the right choice if the aim of your work is to:

- Explore evidence and present well-reasoned recommendations that move beyond consultation
- Provide the opportunity of active citizenship to groups that are typically excluded from public decision making
- Ask communities to make concise decisions or set priorities

We collaborated with Talking Mats (https://www.talkingmats.com/), a social enterprise, who have developed and pioneered innovative techniques to enable effective communication for people of all abilities in order to develop our vision for an adapted Citizen's Jury. Our approach was designed to recognise the heterogeneity of the population with learning disabilities by supporting the specific communication skills and support needs of a range of people with learning disabilities whose communication skills were likely to vary significantly.

An inclusive approach to project planning

The project team took an inclusive approach to project planning as early as possible in the project, starting with the interview for the project engagement lead, which was designed and conducted with input from a member of the National Involvement Network (NIN) with learning disabilities, who said following the interview:

> *"I really liked being involved in making the [Talking] mat. At the interview I knew what I had to do and I felt the other interviewers listened to me and my opinion." (Juror)*

The involvement of a person with learning disabilities was possible from the start of the project because we were able to harness the skills and experience of a person from the NIN who was trained in Talking Mats. This was a clear independent role which was central to the interview process and improved the quality of the information gained by panel members and provided interview candidates with the opportunity to demonstrate their accessible communication skills.

Delivering an accessible Citizens' Jury

In its planning phase, the project team examined the core elements of the Citizens' Jury model and decided to prioritise four critical elements in our approach (see Figure 7.1).

Figure 7.1. Four core principles of Citizen's Juries

Adaptations to the Citizens' Jury model

As previously mentioned, in developing a Citizens' Jury which was accessible for people with learning disabilities a number of adaptations and supports were necessary to enable the participation of a group of adults with diverse support and communication needs. We identified the core components of jury planning for accessibility as shown in Figure 7.2. To achieve these we placed a strong focus on capacity building for engagement and held five preparatory workshops to build confidence, knowledge, communication skills and trust within the group. Other adaptations to the standard Citizens' Jury method included:

- Reducing the duration of meetings and incorporating plenty of breaks
- Using additional communication supports such as Talking Mats and 'I want to speak' cards
- Working with expert witness to make their presentations accessible
- Allowing support staff to attend meetings (this was usually a support worker and a transcriber)

One of the most significant adaptations of the Citizens' Jury method was in our decision to work with the group to set their own jury question within the broad field of health research. This decision was intended to confer as much autonomy as possible onto the group by giving them greater control of the agenda for the Research Voices project.

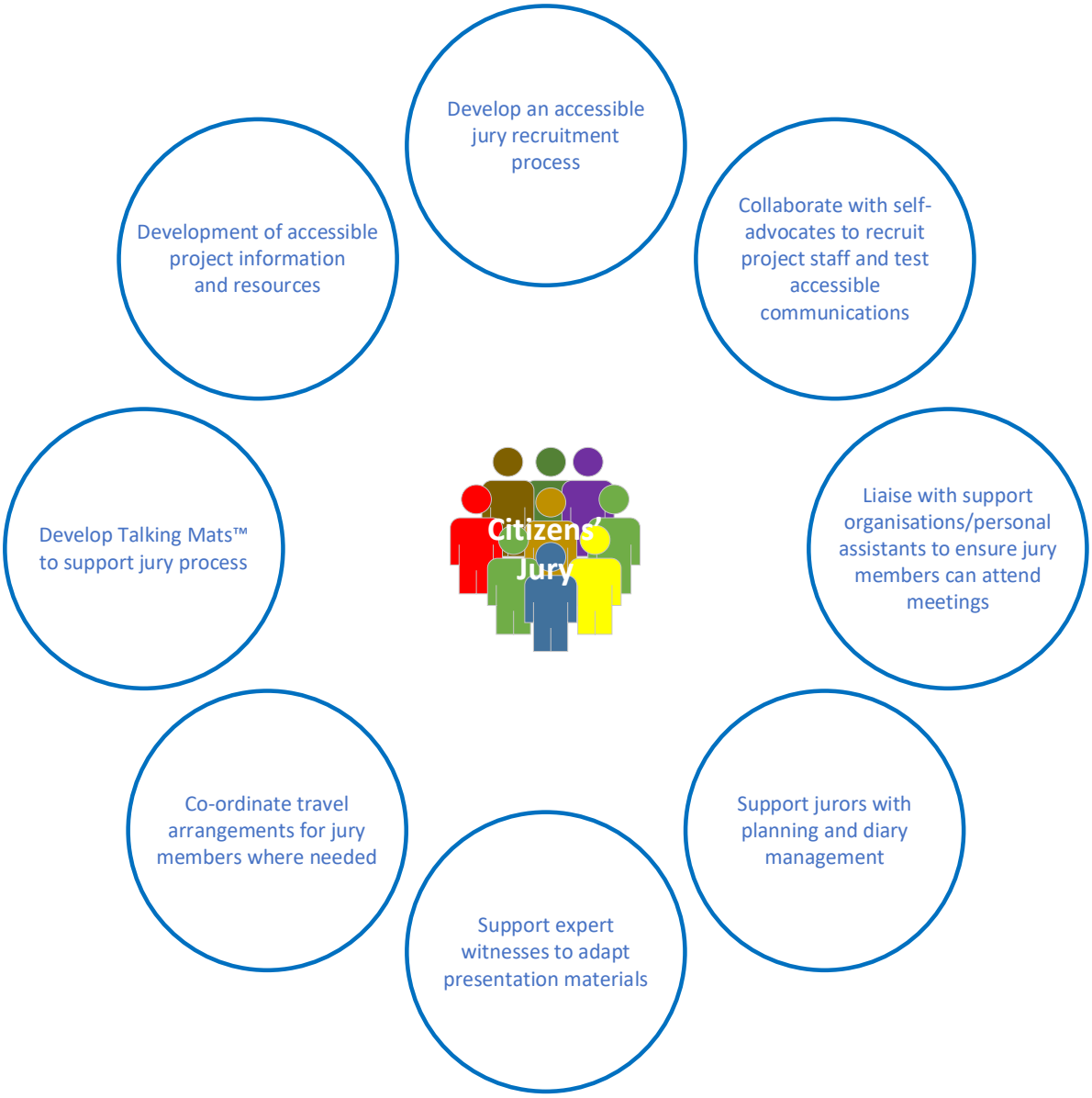

Figure 7.2. Core components of a jury planning for accessibility

Recruiting jury members

Traditionally, Citizens' Juries are recruited using stratified random sampling, often drawing on voting registries, or using third party market research companies to recruit a demographically

representative group. However, this was a challenge for our key audience, who are harder to reach and often democratically excluded. Therefore, this project used purposeful recruitment, engaging with community leaders and 'Gatekeepers' to cascade information about the project throughout the learning disabilities 'community'. The recruitment approach for the project was designed to be demographically representative. This was based on the learning disabilities population demographics observed in Scotland's 2011 census. Twelve people were initially recruited, the final group of nine included five men and four women aged 16 to 66 years old with different experiences of health and inequality. Recruitment of people with learning disabilities to research is a known challenge (Cleaver, Ouellette-Kuntz, & Sakar, 2010) and this project was no exception. The project struggled to find representation in the 45-54 age group (projected n=2, achieved n=0). It was important to us that this group looked beyond 'the usual suspects' - people with learning disabilities who are already very involved and active in other self-advocacy projects. With this in mind, we met with candidates who had no background of participating in co-production, project work or group advocacy, and only half of jurors had previous experience of self-advocacy or research. See the appendix 1 and 2 for the accessible invitations and information sheet. Our accessible juror recruitment process consisted of one informal meeting or phone call with potential jurors, followed by an interview using the Talking Mats tool that was developed for the project.

Following recruitment, the project team delivered 5 preparatory workshops on these core topics:

1. What is a Citizens' Jury?
2. What is health research?
3. Developing communication and questioning skills
4. Choosing a Jury question
5. Preparing for the Citizens' Jury

These workshops were followed by the Citizens' Jury itself.

Image 7.1: The Jury in action

Following the jury, the team delivered 4 additional workshops focused on evaluation and co-producing outputs for the project including a final recommendations report and an accessible video report.

Jury session design

The Citizens' Jury met for 5 days (10am – 3pm) and followed a set structure for each day (see Table 7.1). Four of these days focused on presenting evidence, asking questions and identifying key learning and the final day focused on deliberation and proposing recommendations.

Day 1- Morning Session	Day 2 – Morning Session	Day 3 – Morning Session	Day 4 – Morning Session	Day 5: Deliberation day
Introduction to the Citizens' Jury Discussion of expectations and schedule Development of or summary of working together agreement Pre-jury survey **Day 1 – Afternoon Session** **WITNESS 1** Prep for witness: who are they? What are their motivations? What expert knowledge might they have? Witness 1 Presentation Questions for Witness 1 Summary of key learning points from Witness Evaluation Day 1	Warm up Reminder of working together agreement **WITNESS 2** Prep for witness: who are they? What are their motivations? What expert knowledge might they have? Witness 2 Presentation Questions for Witness 2 Summary of key learning points from Witness **Day 2- Afternoon Session** **WITNESS 3** Prep for witness: who are they? What are their motivations? What expert knowledge might they have? Witness 3 Presentation Questions for Witness 3 Summary of key learning points from Witness Evaluation day 2	Warm up Reminder of working together agreement **WITNESS 4** Prep for witness: who are they? What are their motivations? What expert knowledge might they have? Witness 4 Presentation Questions for Witness 4 Summary of key learning points from Witness **Day 3 – Afternoon Session** **WITNESS 5** Prep for witness: who are they? What are their motivations? What expert knowledge might they have? Witness 5 Presentation Questions for Witness 5 Summary of key learning points from Witness Evaluation day 3	Warm up Reminder of working together agreement **WITNESS 6** Prep for witness: who are they? What are their motivations? What expert knowledge might they have? Witness 6 Presentation Questions for Witness 6 Summary of key learning points from Witness **Day 4 – Afternoon Session** **WITNESS 7** Prep for witness: who are they? What are their motivations? What expert knowledge might they have? Witness 7 Presentation Questions for Witness 7 Summary of key learning points from Witness Evaluation day 4	**Morning session** Warm up Reminder of working together agreement Outline goals for the day Review evidence from presentations in small groups, highlighting gaps or key learning points Develop initial recommendations in small groups to present to wider group for comment and addition Discuss any gaps in learning or questions remaining **Day 5 – Afternoon Session** Vote on recommendations seeking consensus and noting any areas where there was less consensus/any concerns raised Post jury survey Discussion of next steps/writeup of recommendations Final evaluation and celebration

Table 7.1. Jury session design

Photo 1: Deliberation following Citizens' Jury

Photo 2: Talking Mats in practice

Photo 4: Graphic representation of expert testimony

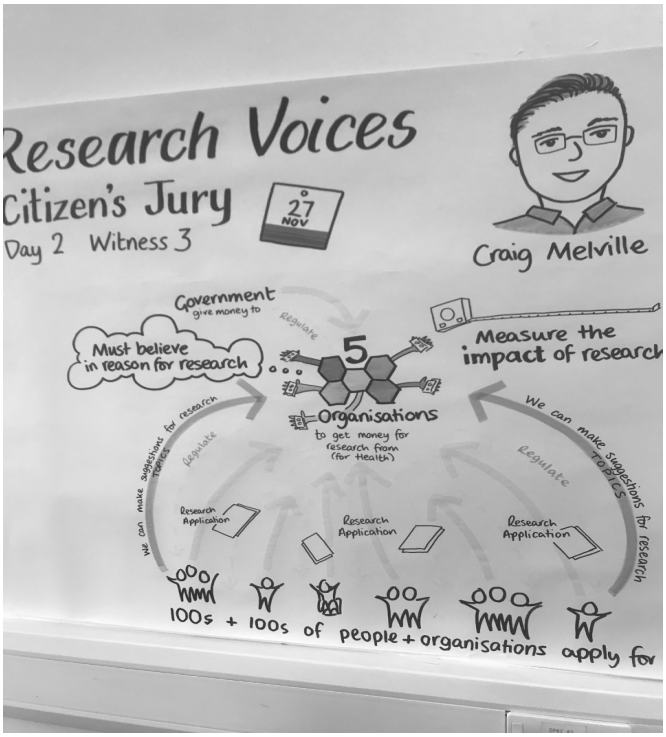

Photo 4: Graphic representation of expert testimony

The project team reflected that there was benefit in compressing meetings into two weeks, but the final deliberation took longer than expected, and consensus voting was extended into an additional workshop to ensure that all jurors were satisfied with the final recommendations.

The pre-Jury workshop and jury sessions all shared similar approaches. Each workshop had clear stated aims, a mix of activities and discussions and an opportunity for group members to reflect on what had worked well, what learning materials had been most effective and on their experience of facilitation.

Workshops were a mix of small group discussions, wider discussion and individual contribution. Smaller groups and sub-groups were always allocated in advance rather than free-formed. In part, this was to support socialisation and relationship building as some group members had met before and had a tendency to gravitate towards each other at the expense of collaborating with new people. However, it was primarily to allow each juror the space to make their contribution to the group. For example, the facilitator might place an individual into a group because they had a strength (such as reading or questions development) that the group, as a whole, would benefit from. These smaller group discussions often lent themselves to more in-depth engagement and ideas sharing, a benefit recorded in other Citizens' Juries. It is important to note that each sub-group needed an individual skilled facilitator to support discussion.

Following the jury, the team delivered 4 additional workshops focused on evaluation and co-producing outputs for the project including a final recommendations report and an accessible video report.

Tools used in the jury

We used a range of facilitation tools to support engagement in the Research Voices Citizens' Jury. These are explored in Table 7.2 below:

Tool	Description	Use	Limitations/Considerations
The 'car park'	This is a large piece of poster paper placed in a visible position with space to add post-it notes. The aim of the car park is to 'park' topics for later discussion, by adding post-it notes with discussion points.	This tool is used to redirect tangential discussions and keep the group on track. The tool acknowledges the importance of other topics, and makes people feel heard whilst allowing facilitators to focus on the current task.	Facilitators must set aside time to properly address issues in the car park 1-1 or in a group. This way, they are honouring the system and ensuring that it is not used to dismiss talking points Facilitators must also be open to exploring what is 'on topic' or 'off topic' when working with a group with lived experience and expertise. For example, if one topic emerges time and time again – they must consider if this is related to the topic in ways they haven't considered before
'Stop' and 'I want to speak' cards	These are red and green cards that are given to each individual that give them the opportunity to stop the conversation at any point (to ask a question or say they don't understand) or to ask to speak.	This tool is useful for building flow in group discussions by limiting interruptions and allowing facilitators to notice who wants to speak, which can be challenging in groups of people with different communication needs. This tool also works for those who struggle to find appropriate moments to	Some group members may still struggle with not interrupting despite the use of the tool, and may still need support to navigate this This tool does not negate the need for facilitators to actively support balanced discussion, which may involve engaging with people not using their cards.

		interrupt, or fear seeming rude	
Graphic facilitation	Graphic facilitation is a form of visual facilitation that takes complex information and transforms it into an easier to understand narrative using key words and drawings	This tool visualises and distils learning from expert testimony in an accessible way. It can be also be a visual record of the process and can be used during deliberation or reflection.	Graphic facilitation is a specialist skill that requires a trained professional Graphic facilitators may need materials in advance of the Jury to be able to anticipate the flow of their work Alternative recording may also be needed if any group members have sensory impairments.
Accessible jury presentations	In this project, jury presentations were made accessible through the use of easy read language and clear images that corresponded to ideas Jury members received hard copies of presentations in advance, which allowed them to access the materials at their own pace For other groups, accessibility may mean availability of evidence in different languages, or in audio format or braille	This allows individuals who learn at different paces and individuals who may struggle with memory to refresh their learning between jury sessions	Expert witnesses may need support to ensure that they can produce evidence in an accessible format In some cases, Jury organisers may need to take on responsibility for making these resources more accessible

Talking Mats	A form of augmentative and alternative communication (AAC) that facilitates conversations using visual symbols Talking Mats can be used digitally or using physical symbols	This tool can deconstruct complex ideas and concepts into smaller, more focused ones Talking Mats can be used on an individual basis to understand views and preferences or in a group for discussions or evaluation (see appendix 3 for an example)	Talking Mats require trained listeners to be an effective tool Project teams may need to design bespoke Talking Mats to support conversation in their processes Talking Mats require forward planning, and may not be responsive to emerging conversation topics
Visual voting tools	Visual voting tools might include coloured stickers for dot voting or 1st, 2nd and 3rd visuals for nominal voting processes.	This tool supports active decision making and priority setting. This tool also visualises votes to allow the group to understand how a consensus has been reached	Jurors may need individual support to cast a vote and understand how their vote fits in with wider decision making Facilitators will need to present clear visual choices for the group for jurors to understand what they are voting for (an idea, a process, a recommendation)

Table 7.2. Tools and resources developed to support the jury

Expert Witness Selection

One of the most important elements of Citizens' Jury is the role of Expert Witnesses. Expert witnesses are experts who provide testimony to the jury, providing good quality evidence that the jury can critically engage with through questions. Expertise can mean a person is an expert in their professional field but can also include experts by experience. When organising a Citizens' Jury, it is important to present the group with experts with a balance of opinions who can offer different perspectives for jurors to consider. It is even more important when

working with marginalised groups to ensure that people have as much choice and autonomy as possible and recognise that they may value and recognise different types of expertise.

When planning an adapted Citizens' Jury, it is vital that expert witnesses understand that they must adapt their evidence to be accessible to the group they are speaking to. For this project, we supplied clear communications guidelines in advance as well as sample accessible presentations. We also offered in-depth support to witnesses who wanted help to develop their communication style. Serving as an Expert Witness was a positive experience for most who took part in our project, but it is important to recognise the time commitment involved, as well as the opportunity for skills development.

Impact

In 2020 the Research Voices Citizens' Jury published their consensus report with 10 recommendations for the health research community outlining how people with learning disabilities can influence research development, delivery and dissemination. A key aim of this project was to evaluate the viability of an adapted Citizens' Jury method for engagement and to share the learning from this approach in order to influence:

- The health research community, including those who focus on learning disabilities health research
- Community and civic engagement teams who want to know how to adapt Citizens assemblies, Citizens juries and wider deliberative democracy to involve people with learning disabilities
- Public and patient involvement and engagement practitioners who work with marginalised groups

Jurors reflected positively on their participation in the Research Voices Citizens' Jury, and valued the opportunity to debate and deliberate on health research. Many jurors said that their confidence had increased, that they had made important social connections and crucially that they had been listened to.

> "*It was good. Felt part a team or a group, because I don't go out to work.. I felt really part of a team - all are together. It did good for my self-confidence.*" - Juror

Jury members also gained a personal investment in the impact of the work and hoped it could support the delivery of more inclusive future research.

> "*It may improve on how the research has done*" - Juror

In a blog post about his experience, one group member wrote:

> *"we feel left out speechless, voiceless never feeling part of or important, feeling alone is never good for anyone but with a disability it's scary harder to put your trust in people to do right things by you and be let down now I personally am now a better person better speaker, talker never feeling alone"*

The Research Voices Citizen's Jury also had an important impact on the expert witnesses. All of the expert witnesses were positive about their experience and felt jurors were able to engage with their presentation. Expert witnesses consistently praised the quality and depth of questions presented by the Jury, indicating positive engagement in the process as well as the value of the jurors' personal perspective:

> *"They were insightful questions, ones with emotion and personal experience reflected in the wording and thoughts. I spent the entire journey home considering some of their reflections… They made me consider areas that I hadn't really focused on …*" - Expert Witness

Final reflections

For the project team, early investment in detailed planning enabled the delivery of a structured framework for engagement that encouraged commitment and a sense of ownership of the jury members from the outset. The group quickly developed a supportive and collaborative environment, which built confidence in discussing complex ideas. Investing in planning meant we understood the individual communication/support needs of the group as well as the skills each individual could offer the process. As facilitators we decided early on that the process needed to minimise our voices and opinions, and by establishing ground rules for engagement the jury members defined their preferences for the role of facilitators. Personal stories of jurors made a vital contribution to the process and were often the lens through which people were able to share their views on the subject. It was important for the team to tune into these stories rather than dismiss them as tangential.

Conclusions

The Research Voices Citizens' Jury was a rewarding and successful project for everyone who was involved, above all it showed that people with learning disabilities can engage in informed discussions about complex and contentious issues, when given time, expert facilitation and tailored communication support.

Top tips:

- People with learning disabilities may need more time and more support at the outset of an engagement/participation project but investing in capacity-building and peer support work early in the project leads to better outcomes.
- When inviting expert witnesses to present information, it is important that they adapt the information to their audiences' needs.
- When debating complex social and ethical issues, personal experience is a rich source of information.
- A Citizens' Jury is one of many potential models of public engagement, but an accessibility-first approach is what made this project successful.

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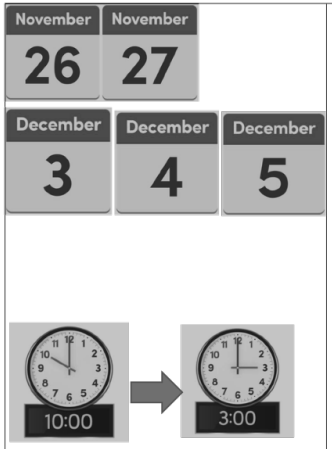
Appendices:

Appendix 1: Invitation to citizens Jury

You are invited to the Research Voices Citizens' Jury



When is the Citizens' Jury?

The Citizens' Jury is five days long

We will meet on the 26th and 27th November

We will meet again on the 3rd, 4th and 5th December

All of our meetings are 10am to 3pm

Where is the Citizens' Jury?

On the 26th and 27th of November we will be meeting at our usual place, The Albany

On the 3rd and 4th of December we will be meeting at our usual place, The Albany

On **5th December** we will meet at Kelvin Hall

This is a new meeting place that we will only use for one meeting

Rhiann will remind you about the new meeting place

Rhiann will make sure your taxi knows about the new meeting place

If you want to take a bus, Rhiann will help you make a plan for the new meeting place

Please tell us if you can come to Citizens' Jury

Please phone Rhiann to tell her if you are coming to the meeting

- We will pay for your travel costs
- Please keep your bus ticket, train ticket or receipt from your taxi
- We will give you the money back on the day

If Rhiann usually books you a taxi, she will do that for you

Food and drink

We will have tea and coffee

We will have lunch together

Tell Rhiann if there is any food you do not like or cannot eat

More information about the Citizens' Jury

We have included more information about the Citizens' Jury and what will happen in this letter

Scottish
Learning Disabilities
Observatory

Information about the Research Voices Project

We are looking for 12 people with learning disabilities to take part in a project called 'Research Voices'

Research Voices is a project about helping more people with Learning Disabilities in Scotland to talk about Health Research

Scottish
Learning Disabilities
Observatory
TalkingMats

The Research voices Project is being run by the Scottish Learning Disabilities Observatory and Talking Mats

Research can help us find out about lots of different things

Health Research helps us to find information out about peoples' health

The Scottish Government
TalkingMats
wellcome

Appendix 2: Information pack

Health research is important

It is important because health research helps professionals to give the best care to people with learning disabilities

It is important because people with learning disabilities have worse health than other people in Scotland

This is not fair

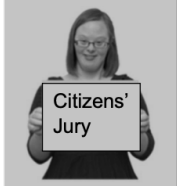
Researchers want to make health care better for people with learning disabilities

They want to find out more about why health is so bad

They want to find out how to makes people's health better

Not many people with learning disabilities get the chance to be involved in health research

We want to bring people with learning disabilities together to talk about health research



We are starting something called a Citizens' Jury

A Citizens' Jury is a group of people who want to talk about an important issue

This Citizens' Jury will be talking about Health Research

The Citizens' Jury will meet people who know about health research and ask them questions

The Citizens' Jury will learn new things

The Citizens' Jury will

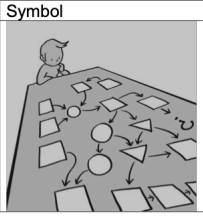
- Listen to what is said
- Think about what is said
- Talk about it what is said
- Tell people what they think should change

Appendix 3: Feedback using Talking Mats

Opinion statements

After the Jury

I am going to read you some opinions. Place each card on the mat where you think it needs to go. The cards can go under I think this is true **most of the time**, true **some of the time** or true **not very often**. You can also talk a bit about what you think.

Statement	Symbol	Placement / Comments
People with learning disabilities are able to have a say in health research		
Health Research already looks at what matters to people with learning disabilities		
It is easy for people with learning disabilities to find helpful information about health research that makes our lives better		
It is easy for people with learning disabilities to be part of health research		
Everyone with learning disabilities is given opportunities be involved in health research		

BMJ Open

Cohort profile: Scotland's record-linkage e-cohorts of people with intellectual disabilities, and autistic people (SCIDA)

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ABSTRACT

Purpose To investigate health, mortality and healthcare inequalities experienced by people with intellectual disabilities, and autistic people, and their determinants; an important step towards identifying and implementing solutions to reduce inequalities. This paper describes the cohorts, record-linkages and variables that will be used.

Participants Scotland's Census, 2011 was used to identify Scotland's citizens with intellectual disabilities, and autistic citizens, and representative general population samples with neither. Using Scotland's community health index, the Census data (demography, household, employment, long-term conditions) were linked with routinely collected health, death and healthcare data: Scotland's register of deaths, Scottish morbidity data 06 (SMR06: cancer incidence, mortality, treatments), Prescribing Information System (identifying asthma/chronic obstructive pulmonary disease; angina/congestive heart failure/hypertension; peptic ulcer/reflux; constipation; diabetes; thyroid disorder; depression; bipolar disorders; anxiety/sleep; psychosis; attention deficit hyperactivity disorder; epilepsy; glaucoma), SMR01 (general/acute hospital admissions and causes, ambulatory care sensitive admissions), SMR04 (mental health admissions and causes), Scottish Care Information–Diabetes Collaboration (diabetic care quality, diabetic outcomes), national bowel screening programme and cervical screening.

Findings to date Of the whole population, 0.5% had intellectual disabilities, and 0.6% were autistic. Linkage was successful for >92%. The resultant e-cohorts include: (1) 22 538 people with intellectual disabilities (12 837 men and 9701 women), 4509 of whom are children <16 years, (2) 27 741 autistic people (21 390 men and 6351 women), 15 387 of whom are children <16 years and (3) representative general population samples with neither condition. Very good general health was reported for only 3389 (15.0%) people with intellectual disabilities, 10 510 (38.0%) autistic people, compared with 52.4% general population. Mental health conditions were reported for 4755 (21.1%) people with intellectual disabilities, 3998 (14.4%) autistic people, compared with 4.2% general population.

Future plans Analyses will determine the extent of premature mortality, causes of death, and avoidable deaths, profile of health conditions and cancers, healthcare quality and screening and determinants of mortality and healthcare.

STRENGTHS AND LIMITATIONS OF THIS STUDY

⇒ Ninety-four per cent Census completion for a whole country population, and >92% successful record linkage.
⇒ Record linkage across multiple data sources.
⇒ Limitations include the use of routinely collected data.
⇒ Reliance on recorded cause of death on death certificates.
⇒ Self/proxy reporting of intellectual disabilities and autism, and other Census data.

INTRODUCTION

People with intellectual disabilities are thought to die about 20 years earlier than other people,[1] or 28 years earlier for people with Down syndrome,[2] but most studies are small. A large English study found a standardised mortality ratio (SMR) of 3.18 (95% CI 2.94 to 3.43).[3] Two large Scottish studies reported an SMR of 2.24 (95% CI 1.98 to 2.49) in adults overall; 5.28 (95% CI 3.98 to 6.57) for Down syndrome adults, and 1.93 (95% CI 1.68 to 2.18) for adults without Down syndrome,[4] and an SMR of 11.6 (95% CI 9.6 to 14.0) in children overall.[5] Autism is also thought to be associated with premature death, though there have been few studies, and co-occurring intellectual disabilities may account for some of the reported findings.[6–9] The most common causes of death of people with intellectual disabilities are likely to differ from other people. For example, aspiration pneumonia which is avoidable, may occur quite commonly,[3 4 10] and SMRs have been reported as 2.4 (95% CI 1.3 to 3.8) for colorectal cancer, and 2.3 (95% CI 1.0 to 4.3) for female genital cancers.[3] Additionally, probably about 40% of early mortality would have been treatable by good quality healthcare, which may be double the amount that occurs in other people.[4 10 11] Causes of death of autistic people may also differ from those

in the general population, with deaths related to the nervous system, and injury and poisoning reported to be the most common causes.[8] These inequalities are a major injustice that need addressing. An important step towards this is to gain a better understanding of health, mortality, healthcare and their determinants in people with intellectual disabilities and autistic people.

The health profiles of people with intellectual disabilities and autistic people differ from other people, which has a bearing on mortality. People with intellectual disabilities have, for example, high rates of epilepsy, mental illness, gastrointestinal disease, antipsychotic use and polypharmacy.[12–14] They may have higher diabetes rates,[15 16] due to sedentary lifestyles, obesity and antipsychotics.[17 18] Cancer profiles appear to differ from other people, with colorectal, stomach, oesophageal, brain and uterine cancer, and leukaemia all having been reported to have higher incidence in people with intellectual disabilities, but with some inconsistencies between studies, and wide CIs due to study sizes.[19–21] Higher incidence of gastrointestinal cancers is plausible, due to poor diets, lifestyle and high rates of *Helicobacter pylori* and gastro-oesophageal reflux disorder, and lower screening uptake. Multimorbidity is typical in people with intellectual disabilities,[13 14] and poor health is reported for people with intellectual disabilities across the full life-course.[22] In autistic people, general health is also reported to be poor across the lifespan.[23 24] Autistic people commonly have comorbid physical conditions, and some are more prevalent than in the general population, including sleep problems, epilepsy, sensory impairments, atopy, autoimmune disorders and obesity, but not asthma.[25] However, there are substantial gaps in the evidence-base on the health of autistic people, and some findings are inconsistent.[25]

Despite this higher burden of health need and early mortality, healthcare has been reported to be poorer for people with intellectual disabilities,[26] and for autistic people,[27] and barriers in access to healthcare have been reported.[26 27] Adults with intellectual disabilities are reported to participate less in cervical screening and breast screening,[28] and bowel screening[29] than other people. There is also some indication that people with intellectual disabilities and autistic people are more likely to be hospitalised for ambulatory care-sensitive conditions than other people; these are conditions that should be fully manageable in primary healthcare settings, and so such hospitalisations are a further marker for poor care.[30 31] These factors may impact on poorer outcomes and survival, due to later stage disease when treatment is initiated, and poorer management of disease.

Internationally, one of the challenges in investigating these issues at a population level is the lack of data on representative samples of people with intellectual disabilities, or autistic people. Important studies have used novel methods to link routinely collected administrative data and identify people with intellectual disabilities via their use of educational services, services designed for adults with intellectual disabilities and/or though identifying diagnostic codes in health records likely to indicate the presence of intellectual disabilities or autism.[32–35] These approaches provide valuable data and also challenges, as, after leaving education/school, not all adults with intellectual disabilities use intellectual disabilities services, and nor do autistic adults, and there are coding issues in health records which are completed by multiple professionals and may be both incomplete and overinclusive for syndromes that are associated with intellectual disabilities in some but not all people. Additionally, caseness may be variably defined, with the term intellectual and developmental disorders being commonly used and which may or may not be a broader definition of neurodevelopmental disorders combining, for example, some or all of intellectual disabilities, autism, attention deficit hyperactivity disorder and specific learning disabilities.[35] A further approach to population studies is using national surveys conducted with samples of the population, but has the limitation that people with intellectual disabilities or autism are represented in only small numbers and so restricts the analyses that can be undertaken, and in longitudinal research they tend to have higher cohort attrition and more biased cohort retention than other people.[35 36] In Scotland, the national Census in 2011 included questions on both intellectual disabilities and on autism, and so presents an ideal data-source for research with these populations. We are not aware of any other national Census data which has included data on intellectual disabilities and autism, other than Irish data which has the limitation of not distinguishing intellectual disabilities from specific learning disabilities. Record-linkage of Scotland's Census, 2011 to Scotland's other rich routinely collected health data sets and national statistics provides an excellent vehicle for detailed investigation of these issues. Scotland's Census, 2011 systematically questioned whether every person in the country had intellectual disabilities, and/or autism, and distinguished intellectual disabilities from dyslexia and other specific learning disabilities. It included data on 94% of the whole Scottish population, and imputed the remaining 6% (5 295 403 people in total). Due to its large size, it has power, when linked to other databases to answer important questions on health, causes of mortality, determinants and healthcare in people with intellectual disabilities, and autistic people. This is an important step leading towards a reduction in morbidity, early mortality and avoidable deaths and improvements in healthcare of people with intellectual disabilities and autistic people.

In Scotland's Census, 2011, of the whole Scottish population, 0.5% were recorded as having intellectual disabilities (0.4% women and 0.6% men), and 0.6% were recorded as being autistic people (0.3% women and 1.0% men). Regarding autism, 1.6% of all Scottish children and young people (<25 years of age) were recorded to have autism (0.7% women and 2.5% men), while 0.2% of adults aged 25 years and over were recorded to have autism (0.1% women and 0.3% men), reflecting the diagnostic broadening of autism spectrum disorders in

recent years, and greater awareness of the condition. This also accounts for why, of the autistic children and young people, 15.0% additionally had intellectual disabilities, while of the autistic adults aged 25 years and over, 29.4% additionally had intellectual disabilities. In total, 1.1/1000 people had both autism and intellectual disabilities, and are represented in both the intellectual disabilities and the autism cohorts. There is some evidence to suggest that autism is less recognised in women than in men,[37] but less so for people with more severe intellectual disabilities.[38]

We created two data sets, with record linkage to several of Scotland's health data sets. Linkage was undertaken for the 94% of the Scottish population who completed the Scottish Census, 2011. Of the people with intellectual disabilities recorded in Scotland's Census, 2011, 22 538 (92.9%) were successfully linked with their health records. Of the people with autism recorded in Scotland's Census, 2011, 27 741 (94.6%) were successfully linked with their health records. Regarding the general population who had neither intellectual disabilities nor autism, of the 15% randomly selected, 700 437 (95.1%) were successfully linked to their health records; while of the 5% randomly selected, 233 378 (95.1%) were successfully linked to their health records. Hence, each data set includes data on 22 538 people with intellectual disabilities, 27 741 autistic people, and a randomly selected comparison general population with neither intellectual disabilities nor autism. For ethical reasons, two data sets were necessary to ensure data minimisation, as our research questions on cancer incidence and mortality required a larger comparison group to reach adequate power, due to lower incidence, than was required to answer the other research questions. The minimal number of variables was requested for each data set: in data set 1, this therefore did not include ethnicity, nor general or mental health variables, as there is not sufficient power to investigate these factors in relation to causes of deaths and cancers.

- Data set 1 includes 22 538 people with intellectual disabilities, 27 741 autistic people and a random sample of 700 437 (15%) of the Scottish population. It will be used to answer research questions on SMRs, causes of deaths, treatable deaths, cancer standardised incidence ratios (SIRs) and cancer SMRs.
- Data set 2 includes 22 538 people with intellectual disabilities, 27 741 autistic people and a random sample of 233 378 (5%) of the Scottish population. It will be used to answer research questions on independent predictors of deaths and treatable deaths, comparison of uptake of cervical screening and bowel screening, descriptions of population health burden and quality of healthcare.

The sample size for the comparison sample in data set 1 was determined using the Scottish crude annual incidence rates and mortality rates for bowel cancer, oesophageal cancer and uterine cancer as examples, and previously published SIRs and SMRs for people with intellectual disabilities.[3 19 21] The figure 1 shows examples

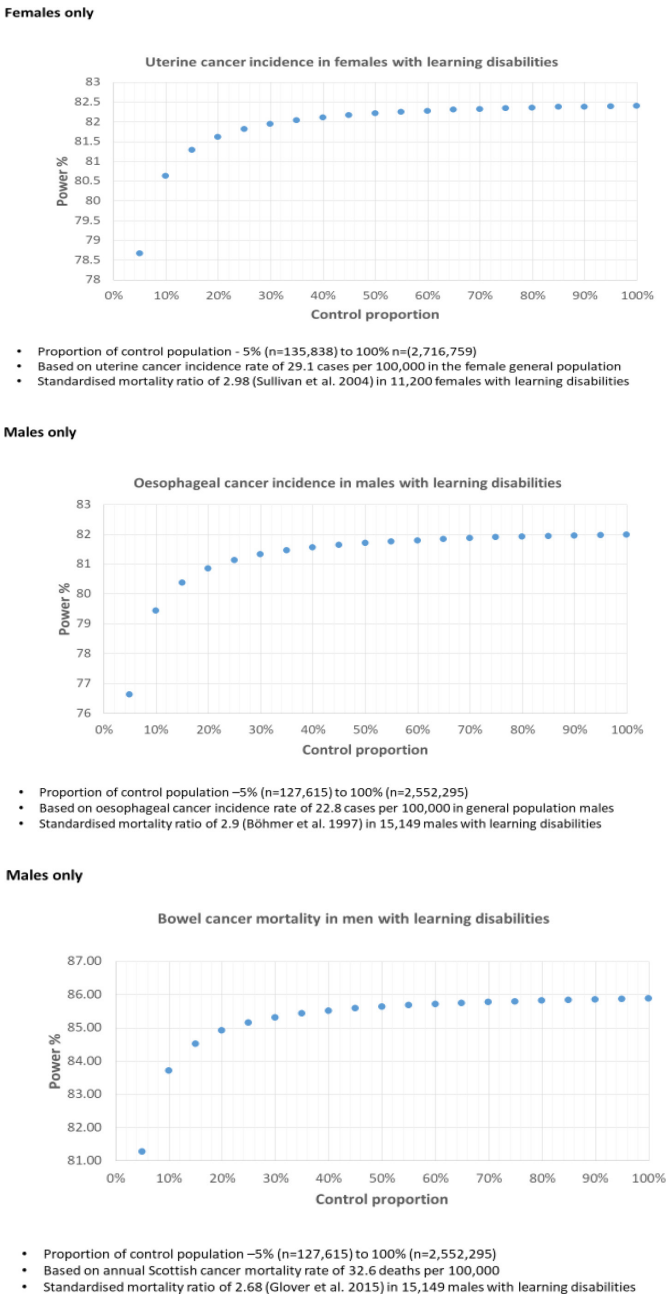

Figure 1 Example power calculations across comparison sample sizes.

of some of the power graphs produced which led to the decision to use 15% of the Scottish population as the comparison group.

The data sets we linked together and variables we selected were based on these specific research questions posed on (a) people with intellectual disabilities, and (b) autistic people, compared with other people:

1. What are the age and sex-SMRs?
2. What are the common causes of death, and what proportion are treatable deaths?
3. Do cancer incidence and mortality differ?
4. What personal (including diseases identified via medication and admissions), household and service-related

factors (including hospital admissions) are markers/predictors of deaths and treatable deaths?
5. Do uptake of the national bowel and cervical cancer screening programmes differ?
6. What is the population health burden (identified via medication and admissions)?
7. Do people with diabetes* or other ambulatory care-sensitive conditions, receive as good healthcare, and are diabetes or other ambulatory care-sensitive conditions more likely to result in-hospital admissions and deaths?

*Diabetes was selected as an exemplar due to Scotland's Register of Diabetes Care.

The purpose of this report is to describe the e-cohorts we have created, the record-linkages that have been made, and the variables that are available within the data sets to investigate the above research questions.

COHORT DESCRIPTION

Data sharing agreements are in place with the data controllers of all the linked data sets.

Table 1 shows the data sources which were linked, the dates of the data collection, the intended research purposes for the data and the data controllers.

Table 1 Linked data sources included in the two data sets

Resource	Time period	Use	Data controller
Data set 1			
Scotland's Census, 2011	27.3.11	To identify the populations with intellectual disabilities, and autism, and the comparison population, and their basic demography.	National Records of Scotland.
Register of Deaths	27.3.11–15.08.20	To identify deaths, causes of deaths and treatable deaths.	National Records of Scotland.
Scottish Morbidity Records 06	27.3.11–15.08.20	To identify cancer incidence, mortality and treatments (quality of healthcare).	NHS National Services Scotland, Information Services Division.
Data set 2			
Scotland's Census, 2011	27.3.11	To identify the populations with intellectual disabilities, and autism, and the comparison population, their basic demography, household, employment and long-term conditions data (potential determinants of mortality).	National Records of Scotland.
Register of Deaths	27.3.11–15.08.20	To identify deaths.	National Records of Scotland.
Prescribing Information System	27.3.11–27.9.11	To identify people with: asthma/chronic obstructive pulmonary disease; angina/congestive heart failure/hypertension; peptic ulcer/reflux; constipation; diabetes; thyroid disorder; depression; bipolar disorders; anxiety/sleep; psychosis; ADHD; epilepsy; glaucoma (health burden).	NHS National Services Scotland, Information Services Division.
Scottish Morbidity Records 01	27.3.11–15.08.20	To identify general/acute hospital admissions and causes, and ambulatory care sensitive admissions.	NHS National Services Scotland, Information Services Division.
Scottish Morbidity Records 04	27.3.11–15.08.20	To identify mental health admissions and their causes.	NHS National Services Scotland, Information Services Division.
SCI-diabetes	27.3.11–15.08.20	To identify quality of diabetic care and diabetic outcomes (quality of healthcare).	Caldicott guardians.
National Bowel Screening Programme	27.3.11–15.08.20	To identify screening uptake.	NHS National Services Scotland, Information Services Division.
Cervical screening	2016–15.08.20	To identify screening uptake.	NHS National Services Scotland, Information Services Division.

ADHD, attention deficit hyperactivity disorder; NHS, National Health Service; SCI-diabetes, Scottish Care Information - Diabetes Collaboration.

The variables we selected to include from each of the data sources were outcomes, exposures or potential confounders. A table of the full list of variables is included in online supplemental file 1. Data linkage of Scotland's Census, 2011, and de-identification, were carried out by the National Records of Scotland (NRS) Indexing Service (a Trusted Third Party Linkage Service) to the NRS Indexing Spine, which includes each person's Community Health Index. The Community Health Index is the unique National Health Service identifier given to everyone in Scotland; hence this indexing then enabled linkage to all the other selected health databases. Online supplemental file 2 shows the process. The linked data sets are held in Scotland's national safe haven, where access is provided to the approved members of our team.

Scotland's Census, 2011 includes information on people staying in Scotland on 27 March 2011. It includes people in both private households and communal establishments (eg, care homes). One householder on behalf of all occupants in private households, or the manager on behalf of all occupants of communal dwellings, was required to complete the Census questionnaire. Non-responders were followed-up, and help was provided to complete the form if needed. It was a legal requirement to complete the Census questionnaire, and a statement on the form said that non-completion, or supplying false information could attract a £1000 fine. It was estimated to have achieved a 94% response rate.[39] During the original data processing, the Census team adjusted for the 6% for whom data were missing, using a Census Coverage Survey (including around 40 000 households) to estimate numbers and characteristics. Self/proxy reporting was used to identify people with intellectual disabilities, autistic people and people with other long-term conditions from the Census 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. Cognitive question testing with retrospective probing was undertaken on question 20 prior to the Census, to test whether the questions were answered accurately and willingly by respondents, and to identify what changes might be required to improve data quality and/or the acceptability of the response options. The pilot testing was conducted with 102 participants with a mix of gender and age, both with and without health conditions/disabilities (including people with more than one of the conditions), 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 the version included in the actual Census, to accurately capture the data on autism, in keeping with feedback from the pilot testing with people with autism. (Henceforth, throughout this paper we refer to 'autism', rather than the exact wording on the Census described above). Importantly, question 20 distinguished autism from learning disability (which in the UK is synonymous to the international term 'intellectual disabilities'), learning difficulty (which in the UK is synonymous to the international term 'specific learning disability' such as dyslexia) and mental health conditions. All people with a positive response to question 20 on intellectual disabilities and/or autism were included in our record-linked data sets.

The data sources linked to the variables from Scotland's Census, 2011 are described below. All provide routinely collected data for the whole of Scotland. The Scottish Morbidity Record (SMR) routinely records information from episodes of healthcare.

1. SMR 01 provides data on general/acute hospital admissions and causes using the international statistical classification of diseases and related health problems, 10th edition (ICD-10) codes.[40]
2. SMR 04 provides data on mental health admissions and causes also using ICD-10 codes.
3. SMR 06 is the Cancer Registry and provides information on the disease, its treatments, and outcomes.
4. Prescribing Information System provides information on all medicines that are prescribed and dispensed in the community in Scotland, or issued in hospitals and dispensed in the community, and prescriptions written in Scotland that were dispensed elsewhere in the UK, using British National Formulary codes.
5. Scottish Care Information–Diabetes Collaboration is a national diabetes register that collects data on the quality of care, and outcomes for all patients with diabetes.
6. The national bowel, and cervical screening programmes collect data on screening uptake.
7. Scotland's national death register collates information from death certificates including cause of death as recorded by the certifying clinician, using ICD-10 codes.

Table 2 reports the characteristics of the cohorts with intellectual disabilities, and autism, and the random samples of the general population with neither of these conditions.

Patient and public involvement

This resource and the programme of research it will support, was designed to respond to the growing concern expressed by people with intellectual disabilities, autistic people, their families, and third sector organisations about premature deaths, and poor healthcare. The Scottish Learning Disabilities Observatory, where this research is being undertaken, has a specific remit for such work. Its steering group includes partners from third sector organisations, including Down syndrome Scotland, and people with intellectual disabilities, who approved the work plan for this project prior to it commencing.

Table 2 Characteristics of the cohorts

Variable	Category	Data set 1 and 2 intellectual disabilities N=22 538 (100%)	Data set 1 and 2 autism N=27 741 (100%)	Data set 1 15% of general population* N=700 437 (100%)	Data set 2 5% of general population* N=233 378 (100%)
Sex	Male	12 837 (57.0)	21 390 (77.1)	335 226 (47.9)	111 497 (47.8)
	Female	9701 (43.0)	6351 (22.9)	365 211 (52.1)	121 881 (52.2)
Age	0–15	4509 (20.0)	15 387 (55.5)	117 980 (16.8)	39 220 (16.8)
	15–24	3546 (15.7)	6790 (24.5)	76 184 (10.9)	25 574 (11.0)
	25–34	2976 (13.2)	2024 (7.3)	83 868 (12.0)	27 721 (11.8)
	35–45	3277 (14.5)	1300 (4.7)	97 886 (14.0)	32 599 (14.0)
	45–54	3664 (16.3)	1053 (3.8)	108 052 (15.4)	36 164 (15.5)
	55–64	2494 (11.1)	617 (2.2)	92 698 (13.2)	31 022 (13.3)
	65–74	1330 (5.9)	300 (1.1)	67 532 (9.6)	22 166 (9.5)
	75+	742 (3.3)	270 (1.0)	56 237 (8.0)	18 912 (8.1)
Ethnicity†	White	22 022 (97.7)	26 976 (97.2)	Not available	225 626 (96.7)
	Asian/Asian Scottish/Asian British	368 (1.6)	425 (1.5)	Not available	5181 (2.2)
	African	35 (0.2)	80 (0.3)	Not available	928 (0.4)
	Caribbean or black	21 (0.1)	24 (0.1)	Not available	213 (0.1)
	Mixed/multiple ethnic groups	66 (0.3)	189 (0.7)	Not available	888 (0.4)
	Other ethnic group	26 (0.1)	47 (0.2)	Not available	542 (0.2)
SIMD	1—most deprived	6200 (27.5)	6748 (24.3)	130 354 (18.6)	42 828 (18.4)
	2	5481 (24.3)	5901 (21.3)	135 311 (19.3)	45 407 (19.5)
	3	4516 (20.0)	5678 (20.5)	142 238 (20.3)	47 886 (20.5)
	4	3693 (16.4)	5128 (18.5)	147 866 (21.1)	49 429 (21.2)
	5—most affluent	2648 (11.7)	4286 (15.5)	144 668 (20.7)	47 828 (20.5)
General health status†	Very good	3389 (15.0)	10 510 (38.0)	Not available	122 511 (52.4)
	Good	7942 (35.2)	9608 (34.6)	Not available	69 575 (30.0)
	Fair	7715 (34.2)	5387 (19.4)	Not available	28 273 (12.1
	Bad	2356 (10.5)	1533 (5.5)	Not available	9885 (4.2)
	Very bad	1136 (5.0)	703 (2.5)	Not available	3134 (1.3)
Mental health condition†	Yes	4755 (21.1)	3998 (14.4)	Not available	9710 (4.2)
	No	17 783 (78.9)	23 743 (85.6)	Not available	223 668 (95.8)

*General population with neither intellectual disabilities nor autism.
†Data not available for data set 1 to ensure data minimisation, as it does not have sufficient power to investigate these factors in relation to causes of deaths and cancers.
SIMD, Scottish Index of Multiple Deprivation.

FINDINGS TO DATE

To date, research has been completed on the cross-sectional Scottish Census, 2011 data only. We have analysed, compared with the general population, the independent contributions of having intellectual disabilities, and being autistic, on general health. We have also reported general health comparisons between the general population, with autistic people who also have intellectual disabilities. Further, we have reported mental health data compared with the general population on the people with intellectual disabilities (whether or not they were also autistic); autistic people (whether or not they also had intellectual disabilities); and autistic people who also have intellectual disabilities. We then reported on visual and hearing impairments, and physical disabilities in autistic people (whether or not they also had intellectual disabilities); and in autistic people who also had intellectual disabilities. These distinctions are important given the high proportion of overlap between intellectual disabilities and autism.

We derived a dichotomised variable of poor general health from the five response general health question in

Scotland's Census, 2011. In comparison with the general population, in children/youth, both intellectual disabilities (OR=18.34 (95% CI 917.17 to 19.58)), and autism (OR=8.40 (95% CI 8.02 to 8.80)) were independently associated with poor general health. In adults, both intellectual disabilities (OR=7.54 (95% CI 7.02 to 8.10)), and autism (OR=4.46 (95% CI 4.06 to 4.89)) were also independently associated with poor general health.[41] People with co-occurring intellectual disabilities and autism combined were particularly vulnerable to poor general health.[42]

In comparison with the general population, mental health conditions (answered as 'yes' or 'no') were also more common in people with intellectual disabilities (OR=7.1 (95% CI 6.8 to 7.3)),[43] autistic adults (OR=8.6 (95% CI 8.2 to 9.0))[44] and people with co-occurring intellectual disabilities and autism (OR=130.8 (95% CI 117.1 to 146.1)).[45] Poor general health was also associated with poor mental health in people with intellectual disabilities.[43]

Visual impairment, hearing impairment and physical disability were also more common than in the general population. For autistic adults, and autistic children/youth, respectively, visual impairment (OR=8.5 (95% CI 7.9 to 9.2) and OR=8.9 (95% CI 8.1 to 9.7)); hearing impairment (OR=3.3 (95% CI 3.1 to 3.6) and OR=5.4 (95% CI 5.1 to 5.6)); and physical disability (OR=6.2 (95% CI 5.8 to 6.6) and OR=15.8 (95% CI 14.1 to 17.8)) were considerably more common.[44 46] For people with both intellectual disabilities and autism, visual impairment (OR=65.9 (95% CI 58.7 to 73.9)); hearing impairment (OR=22.0 (95% CI 19.2 to 25.2)); and physical disability (OR=157.5 (95% CI 144.6 to 171.7)) were even more common.[44]

Raising awareness of these issues is important, as these conditions are disabling, and in some cases life limiting if left untreated. In addition to the communication needs that people with intellectual disabilities and autistic people have due to their neurodevelopmental condition, comorbidity renders differential diagnosis and treatments more complex, particularly if unrecognised, hence it is essential that clinicians are aware of the additional conditions that their patients may have.

STRENGTHS AND LIMITATIONS

Strengths of the e-cohorts include their large scale, national Census coverage of the whole country of Scotland, with 94% uptake, and a high level of successful record linkage (>92%), hence limiting bias, and the systematic enquiry about intellectual disabilities and autism of the whole population. The questions on intellectual disabilities and autism were subject to cognitive question testing prior to use, to ensure that they accurately captured the conditions, and were acceptable to the population. Additionally, specific learning disabilities were distinguished from intellectual disabilities or autism, which is important given lay terms in current use, for example, in the UK and Ireland. We are not aware of any other national Census data which has included data on these conditions, other than Irish data which has the limitation of not distinguishing intellectual disabilities from specific learning disabilities. Scotland has a rich profile of routinely collected health data which has been successfully used many times in previous research on the general population, and linkage of Scotland's Census, 2011 to these data, has provided large scale, whole country cohorts of intellectual disabilities, and of autism.

Limitations include that routinely collected data lack disease-specificity and granularity, and cause of death data are infrequently verified by post-mortem. Ascertainment of intellectual disabilities and autism was based on self or proxy reporting. Scotland is a high-income country with good country-wide educational and diagnostic services, and diagnosis of neurodevelopmental conditions attracts additional support for learning so is advantageous for the child. If a child's learning is delayed, schools have a statutory responsibility to seek assessment, so such children will have received a formal diagnosis. The families of children with learning delay are integral to the assessment and diagnostic care pathway for the child, and the development of the co-ordinated support plan for their additional support needs. Proxy reporting is the basis for much of the healthcare provided for people with intellectual disabilities and autistic people who cannot self-report, and overall, proxy reports are concluded to be a useful addition to determine aspects of well-being when the need arises.[47] Those adults for whom a diagnosis of autism was recorded will reflect the contemporary diagnostic practices in place during their childhood; and the concept of the autism spectrum has broadened in recent years.

COLLABORATION

The breadth of the data sets created means they are suitable to be repurposed to answer many additional questions to the ones we outline in our current programme of research. Collaboration is encouraged, and as we are not the data controllers, this would be subject to approval by Scotland's Public Benefit and Privacy Panel for Health, Scotland's Statistics Public Benefit and Privacy Panel and ethical approval by an appropriate ethical committee.

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Contributors S-AC conceived and designed the programme, and wrote the first draft of the manuscript. AH and DK contributed to the design, and project management. DM, MF and GSS designed and supervised the statistics. LAH-M, ER and KD analysed data. JPP contributed expertise on data linkage and data sets. CM oversees programme management. All contributed to drafting the manuscript, and approved the final version. S-AC is the guarantor.

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Competing interests None declared.

Patient and public involvement Patients and/or the public were involved in the design, or conduct, or reporting, or dissemination plans of this research. Refer to the Cohort Description section for further details.

Patient consent for publication Not applicable.

Ethics approval This programme of research was approved by Scotland's Public Benefit and Privacy Panel for Health (reference: 1819–0051), Scotland's Statistics Public Benefit and Privacy Panel (1819–0051), and the University of Glasgow's College of Medical, Veterinary, and Life Sciences Ethical Committee (reference: 200180081).

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Data availability statement Data may be obtained from a third party and are not publicly available. Data access is available through approval by Scotland's Public Benefit and Privacy Panel for Health, Scotland's Statistics Public Benefit and Privacy Panel, and an appropriate ethical committee.

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ORIGINAL ARTICLE

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Trends and variations in per capita expenditure on adult intellectual disabilities health and social care across Scotland, and by urban/rural class

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Abstract

Background: Following the global financial crisis in 2007/08, the UK implemented an austerity programme which may impact on services. Scotland comprises both densely populated urban conurbations and highly dispersed remote rural and island communities.

Method: Expenditure data were extracted from Scottish Government statistics. Per capita expenditure was calculated using adjusted Scotland's 2011 census data.

Results: There was a 3.41% decrease in real term expenditure on adult intellectual disabilities services between 2012/13 and 2014/15 (>£32 million). In 2014/15, per capita expenditure on adult intellectual disabilities health care ranged from £1,211 to £17,595; social care from £21,147 to £83,831; and combined health and social care expenditures from £37,703 to £85,929. Per capita expenditure on combined health and social care was greater in rural areas, with more on intellectual disabilities social care, though less on health care.

Conclusions: Scottish expenditure on adult intellectual disabilities services has not kept abreast of rising living costs. It varies considerably across the country: a post-code lottery.

KEYWORDS
geographic variation, health services, intellectual disabilities, per capita expenditure, social services, urban/rural variations

1 | INTRODUCTION

The 2007/08 financial crisis pushed major economies in the world into recession. In response, after initial austerity measures in late 2008, the UK introduced an austerity programme in 2010 aimed at addressing national debt levels (Reeves, Basu, McKee, Marmot, & Stuckler, 2013). Adults with intellectual disabilities experience significant health and social inequalities, including higher rates of multimorbidity and earlier death than the general population (Hughes-McCormack et al., 2017a,b; Kinnear et al., 2018; O'Leary et al., 2017). They require state funding for dedicated provision of support for health and social care to achieve aspirations for quality of life. It would be of societal concern if year-on-year austerity measures reduced care provision, potentially widening existing inequalities.

Few studies have assessed the specific impact of cuts in public finance on the allocation of health and social care funding levels for this population. Despite noting the poor quality of data on funding of intellectual disabilities services in England, one study concluded that the unit costs of intellectual disabilities social care (after adjusting for inflation) between 2004/05 and 2008/09 have actually risen by 38% for residential care compared to 32% for home care, 21% for day care and an increase by about 24% for health service costs (Royal Mencap Society/Learning Disability Coalition, 2010). Past reports have shown substantial variation in expenditure on intellectual disabilities services across England, with rural areas spending less

on intellectual disabilities health services (Forsyth & Winterbottom, 2002), and Local Authority services (social services; Moscone, 2011). There are some overlaps in the management and delivery of care and support for people with intellectual disabilities across NHS and Local Authority boundaries, and geographic variations in NHS or Local Authorities may be accounted for by local arrangements with the other.

In Scotland, the Government allocates funding to its Health Boards and Local Authorities taking account of local population size, demographics and additional service-delivery demands. The Health Boards and Local Authorities then allocate resources within their geographic areas to their range of populations/services, based on the underpinning principle of the assessed needs of individuals (not, for example, on a fixed daily rate per person with intellectual disabilities). There has not been an explicit linear cut in public spending on disability during the period studied, but increased demand on services might have led to the application of higher eligibility thresholds to prioritize access to services whilst still meeting statutory requirements.

The global financial crisis and subsequent austerity drives have led Western liberal democracies, including the devolved Scottish Government, to consider how best to address persistent health and social inequalities in the context of reduced public spending (Smith, Bambra, & Hill, 2016). In 2016, Scotland brought about the integration of health and social care services, requiring NHS Health Boards and Local Authorities to merge their budgets for adult community and preventative health and social care services. It is therefore important to examine expenditure prior to the implementation of integration and any geographic variations in this, to establish the baseline against which any future changes can be compared, whilst austerity continues.

Scotland is a geographically large country, which includes high-density large urban conurbations, as well as dispersed remote and rural areas including small island communities. The challenges to service delivery in such contrasting areas are likely to differ and could have implications on both expenditure and service delivery models.

The primary aims of this study are to investigate whether there are any variations in Health Board and Local Authority expenditure on adult intellectual disabilities services across Scotland, and whether there is a year-on-year impact of austerity over a 3 year period, 2012/13–2014/15, prior to budgetary integration. Secondary aims are to investigate whether local expenditure on intellectual disabilities services is related to the number of adults with intellectual disabilities, and/or the extent of rurality of the area.

2 | METHOD

Expenditure data for 2012/13, 2013/14 and 2014/15 on adult intellectual disabilities services were extracted from the Scottish Local Government Financial Statistics and the Scottish Health Service Costs (The Scottish Government, 2016; NHS Scotland, 2016), from which data are published annually. The Scottish Local Government Financial Statistics is a publication of the social care expenditure for all Local Authorities in Scotland, whilst the Scottish Health Service Costs is a publication on healthcare expenditure in Scottish Health Boards. All Local Authorities and Health Boards are required to make these annual returns to the Scottish Government on their expenditure in order to enable monitoring and benchmarking comparisons between localities. Expenditure is reported in pounds sterling (£).

Data from all 32 Local Authorities were extracted and from 13/14 regional NHS Health Boards. One regional Health Board (NHS Western Isles) was excluded from this study as the datasource did not contain information on its expenditure (on request, the Board responded that it employs one nurse and a small proportion of a psychologist for people with intellectual disabilities, via mental health services).

Information on the number of adults with intellectual disabilities (aged 16 years or over) in each Scottish Health Board and Local Authority in 2011 was taken from Scotland's Census, 2011 (Scottish Learning Disabilities Observatory, 2016). The estimations for subsequent years (2012–2015) were derived by applying the percentage change in the total adult population (National Records of Scotland, 2017a,b). The Scottish population is relatively static in size, growing by only 0.6% over this period, with similar rates of emigration (0.5%) and immigration (0.7%), and a slight drop in birth rate and rise in lifespan accounting for the overall marginal increase (National Records of Scotland, 2017c). There is currently no national data on survival at birth and lifespan specifically for people with intellectual disabilities, but assume these will have slightly increased over the same time period, as for the general population (O'Leary et al., 2017). Therefore, it is considered to be the best estimate of the size of the population with intellectual disabilities. The total adult population estimates for Health Boards were based on the 2014 NHS Board configuration, accounting for the Health Board boundary changes that occurred within the study period. All annual population estimates were then averaged to obtain the 2012/13, 2013/14 and 2014/15 estimated population size for adults with intellectual disabilities. For example, the population size for 2012 and 2013 was averaged to provide a figure for the 2012/13 population of adults with intellectual disabilities for each Health Board and Local Authority. Per capita spending was then calculated for each Health Board and Local Authority, for each of the 3 years, by dividing the amount of adult intellectual disabilities services expenditure by the estimated number of adults with intellectual disabilities for that year.

2.1 | Measures and variables

The Health Board expenditure included adult intellectual disabilities in-patient, outpatient, day-patient, and community intellectual disabilities team expenditures and resource transfer. Resource transfer refers to monies transferred from the NHS to finance the provision

of health care by Local Authorities. The Local Authority expenditure included employee and operating costs, transfer payments, support service costs and revenue contribution to capital with adjustments for interaccount and interauthority transfers.

Rurality was classed using the sixfold Scottish Government urban/rural classification (Scottish Government, 2014b). This is based on the population size and accessibility of an area. Our classification shows where the major proportion of the total population in the Health Boards/Local Authorities resides, thus describing the extent of urbanization, ruralization and remoteness. The six classification groups are as follows:

1. Class 1, large urban areas: majority of settlements of 125,000 people or more.
2. Class 2, other urban areas: majority of settlements of 10,000–124,999 people.
3. Class 3, accessible small towns: majority of settlements of 3,000–9,999 people, within 30 min drive of a settlement of 10,000 or more.
4. Class 4, remote small towns: majority of settlements of 3,000–9,999 people, with a drive time of more than 30 min to a settlement of 10,000 or more.
5. Class 5, accessible rural area: majority of settlements of less than 3,000 people, within 30 min drive of a settlement of 10,000 or more.
6. Class 6, remote rural areas: majority of settlements of less than 3,000 people, with a drive time of more than 30 min to a settlement of 10,000 or more.

2.2 | Data analysis

Statistical analyses were conducted using Stata software version 14.1 (Stata). All data on expenditure was adjusted for inflation, so as to express them in 2014/15 terms, using the HM Treasury deflator updated on 30.6.17 (HM Treasury, 2017). Real term expenditure (expenditures adjusted for inflation) is therefore expressed in 2014/15 terms, by multiplying the cost of health and/or social care intellectual disabilities services by the HM Treasury adjustment factor, obtained from the deflators: 96.953 for 2012/13, and 98.552 for 2013/14, with respect to 100 for 2014/15.

Variations and trends in the per capita expenditure on adult intellectual disabilities services across Health Boards and Local Authorities were examined using bar charts. The distributions of expenditure on intellectual disabilities services for the years 2012/13 to 2014/15 were not normal. The relationship between per capita expenditure on adult intellectual disabilities services by Health Boards and Local Authorities and the number of adults with intellectual disabilities in localities was examined using Spearman's correlation. Prior to this, the variables were explored using scatterplots. Trends in per capita expenditure were then further analysed by stratifying the Health Boards and Local Authorities based on their major urban/rural class. One-way analysis of variance and Tukey post hoc tests were conducted to determine whether per capita expenditure differed based on major urban/rural class.

Variation and trends in per capita expenditure on adult intellectual disabilities services were then examined for the combined Health Board and Local Authority for each geographic area, using bar charts. The analyses above were repeated for the combined Health Board and Local Authority data.

3 | RESULTS

In Scottish Health Boards (excluding NHS Western Isles), the number of adults with intellectual disabilities was estimated to be 21,128 in 2012/13; 21,214 in 2013/14; and 21,319 in 2014/15. Across the Health Boards, it ranged from 71 to 5,173 adults in 2014/15. In Local Authorities, the number of adults with intellectual disabilities was estimated to be 21,229 in 2012/13; 21,319 in 2013/14; and 21,426 in 2014/15. Across the Local Authorities, it ranged from 72 to 2,946 adults in 2014/15. These ranges are largely accounted for by the differences in total population size across the different areas of Scotland, with the smallest Health Board comprising only 0.40% of Scotland's population, and the largest 21.49%; and the smallest Local Authority comprising only 0.40% and the largest 11.38%.

Scotland's total real term expenditure (expressed in 2014/15 terms) on adult intellectual disabilities services by all Health Boards was £260,929,976 in 2012/13; £255,332,930 in 2013/14; and £250,188,001 in 2014/15; a decrease of 4.29% in real terms across the 3 years. Scotland's total real term expenditure on adult intellectual disabilities services by all Local Authorities was £718,388,291 in 2012/13; £698,990,381 in 2013/14; and £697,310,000 in 2014/15; a decrease of 3.02% in real terms over the 3 years. There is extensive variation in both the total and the per capita expenditure on adult intellectual disabilities services, across Health Boards and Local Authorities (Tables 1 and 2; Figures 1 and 2).

The total real term expenditure on adult intellectual disabilities services by Health Boards ranged from £77,888 to £77,633,539 in 2012/13; £168,177 to £75,111,798 in 2013/14; and £85,848 to £68,101,740 in 2014/15. There is a large variation in per capita expenditure on adult intellectual disabilities services across Scottish Health Boards, ranging from £ 1,105 to £ 19,072 in 2012/13; £2,379 to £17,768 in 2013/14; and £1,211 to £17,595 in 2014/15.

The total real term expenditure on adult intellectual disabilities services by Local Authorities ranged from £3,109,754 to £80,975,318 in 2012/13; £3,563,601 to £71,639,338 in 2013/14; and £3,317,000 to £68,617,000 in 2014/15. There is a large variation in per capita expenditure on adult intellectual disabilities services across Local Authorities, ranging from £24,692 to £84,090 in 2012/13; £24,471 to £89,058 in 2013/14; and £21,147 to £83,831 in 2014/15.

There was a statistically significant positive correlation between per capita expenditure on adult intellectual disabilities services and the number of adults with intellectual disabilities across Health Boards in 2012/13: Spearman's rank correlation = 0.588, $p = 0.035$;

TABLE 1 Total and per capita expenditure on adult intellectual disabilities services by Scottish Health Boards in 2012/13, 2013/14, 2014/15

	Total healthcare expenditure (real terms expenditure in 2014–15 terms, £)			Per capita healthcare expenditure (real terms expenditure in 2014–15 terms, £)		
Health Board	2012/13	2013/14	2014/15	2012/13	2013/14	2014/15
NHS Ayrshire & Arran	17,441,892	17,203,234	17,021,884	10,167	10,035	9,941
NHS Borders	4,635,763	4,937,606	4,858,455	9,539	10,140	9,966
NHS Dumfries & Galloway	8,153,682	8,430,424	8,898,810	13,685	14,160	14,948
NHS Fife	24,416,915	22,797,092	22,622,878	19,072	17,768	17,595
NHS Forth Valley	11,687,895	11,861,670	12,257,535	10,551	10,667	10,952
NHS Grampian	24,931,907	24,983,212	25,046,742	12,225	12,133	12,075
NHS Greater Glasgow & Clyde	77,633,539	75,111,798	68,101,740	15,147	14,610	13,166
NHS Highland	6,869,270	6,170,812	6,100,538	5,439	4,866	4,800
NHS Lanarkshire	30,611,680	30,104,291	29,726,524	12,153	11,926	11,750
NHS Lothian	33,509,781	32,496,546	33,332,905	10,726	10,314	10,467
NHS Orkney	514,881	500,251	496,995	5,966	5,778	5,718
NHS Shetland	77,888	168,177	85,848	1,105	2,379	1,211
NHS Tayside	20,444,883	20,567,817	21,637,147	11,924	11,946	12,514
All of Scotland[a]	260,929,976	255,332,930	250,188,001	12,350	12,036	11,735

[a]Excluding NHS Western isles.

but there was no significant correlation in 2013/14: Spearman's rank correlation = 0.528, $p = 0.064$; nor in 2014/15: Spearman's rank correlation = 0.473, $p = 0.103$. Per capita expenditure on adult intellectual disabilities services showed no significant correlation with the number of adults with intellectual disabilities across Local Authorities: Spearman's rank correlation = −0.070, $p = 0.702$ in 2012/13; Spearman's rank correlation = −0.238, $p = 0.190$ in 2013/14; Spearman's rank correlation = −0.180, $p = 0.324$ in 2014/15.

Regarding urban/rural class, five Health Boards were classed as "large urban" areas, four as "other urban" areas, one as an "accessible rural" area and three as "remote rural" areas. None were classed in the "accessible small towns" or "remote small towns" groups. The mean per capita Health Board expenditure on adult intellectual disabilities services across the urban/rural classes for each of the 3 year periods is shown in Table 3. A statistically significant difference between the urban/rural classes in Health Board expenditure was found by one-way analysis of variance: $F = 6.84$, $p = 0.011$ in 2012/13; $F = 8.70$, $p = 0.005$ in 2013/14; and $F = 9.80$, $p = 0.003$ in 2014/15, with per capita expenditure lower in rural areas. The Tukey test showed that per capita expenditure by Health Boards was statistically significantly lower in "remote rural" areas than in "large urban" areas, and in "remote rural" than in "other urban" areas across all 3 years, with no significant differences between other classes (Table 4).

Regarding urban/rural class, nine Local Authorities were classed as "large urban" areas, fourteen as "other urban" areas, four as "accessible rural" areas and five as "remote rural" areas. None were classed in the "accessible small towns" or "remote small towns" groups. The mean per capita Local Authority expenditure on intellectual disabilities services across the urban/rural classes for each of the 3 year periods is shown in Table 3. A statistically significant difference between the urban/rural classes in Local Authority expenditure was found by one-way analysis of variance: $F = 4.15$, $p = 0.015$ in 2012/13; $F = 4.76$, $p = 0.008$ in 2013/14; and $F = 4.49$, $p = 0.011$ in 2014/15), with per capita expenditure higher in rural areas. The Tukey test showed that per capita expenditure by Local Authorities was statistically significantly higher in "remote rural" areas compared to "other urban" areas across all 3 years, with no significant differences between other classes (Table 4).

3.1 | Combined Health Board and Local Authority expenditure

Scotland's combined health and social care real term expenditure on adult intellectual disabilities services was £ 975,904,242 in 2012/13; £950,759,710 in 2013/14; £943,692,001 in 2014/15; a decrease of 3.41% in real terms across the 3 years. There is extensive variation in both the total and the per capita combined health and social care expenditure on adult intellectual disabilities services across Scotland, shown by geographic area in Table 5 and Figure 3.

The total health and social care real term expenditure on adult intellectual disabilities services ranged across areas from £3,624,636 to £232,879,896 in 2012/13; £4,344,921 to £219,021,612 in 2013/14; and £3,813,995 to £209,199,740 in 2014/15. There is a large variation in the per capita combined health and social care expenditure on adult intellectual disabilities services across areas. Per

TABLE 2 Total and per capita expenditure on adult intellectual disabilities services by Scottish Local Authorities in 2012/13, 2013/14, 2014/15

	Total social care expenditure (real terms expenditure in 2014–15 terms, £)			Per capita social care expenditure (real terms expenditure in 2014–15 terms, £)		
Local Authority	2012/13	2013/14	2014/15	2012/13	2013/14	2014/15
Aberdeen City	31,436,882	30,776,646	31,506,000	37,247	36,152	36,773
Aberdeenshire	40,393,799	42,078,294	41,331,000	48,891	50,446	49,156
Angus	12,454,488	11,894,228	11,604,000	25,732	24,471	23,787
Argyll & Bute	14,525,595	14,672,457	14,266,000	48,121	48,196	47,065
Clackmannanshire	4,579,539	4,164,299	4,431,000	27,985	25,450	27,006
Dumfries & Galloway	23,989,975	22,786,955	23,357,000	40,320	38,327	39,289
Dundee City	20,528,504	20,717,997	22,396,000	29,971	30,205	32,643
East Ayrshire	16,349,159	18,183,294	15,682,000	28,740	31,998	27,621
East Dunbartonshire	16,459,522	16,476,581	15,224,000	45,538	45,348	41,690
East Lothian	12,945,448	17,296,453	16,463,000	32,407	42,991	40,567
East Renfrewshire	12,459,645	12,467,530	12,943,000	35,580	35,356	36,449
Edinburgh City	62,852,104	63,172,741	63,940,000	36,463	36,298	36,325
Eilean Siar	3,414,025	3,563,601	3,806,000	35,303	36,988	39,695
Falkirk	15,736,491	16,123,468	17,874,000	25,771	26,301	29,013
Fife	59,942,446	55,670,103	56,451,000	46,823	43,390	43,906
Glasgow City	80,975,318	71,639,338	68,617,000	27,838	24,528	23,292
Highland	34,194,919	25,893,944	28,440,000	35,806	27,047	29,573
Inverclyde	11,354,986	9,397,070	8,929,000	26,670	22,161	21,147
Midlothian	10,411,230	9,672,051	10,332,000	26,634	24,471	25,755
Moray	11,570,555	11,828,273	10,242,000	31,513	31,818	27,334
North Ayrshire	15,080,503	15,283,302	14,899,000	24,757	25,122	24,527
North Lanarkshire	48,230,586	44,058,974	44,813,000	37,326	34,035	34,537
Orkney Islands	3,109,754	3,844,671	3,317,000	36,234	44,657	38,374
Perth & Kinross	15,362,083	15,309,684	16,218,000	28,326	28,039	29,447
Renfrewshire	22,569,699	21,557,147	22,859,000	29,289	27,941	29,530
Scottish Borders	20,846,183	20,275,591	18,595,000	42,906	41,647	38,154
Shetland Islands	5,989,500	6,361,109	6,004,000	84,090	89,058	83,831
South Ayrshire	17,126,855	17,294,423	16,956,000	31,892	32,197	31,600
South Lanarkshire	36,545,543	36,333,103	36,444,000	30,377	30,113	30,122
Stirling	11,293,101	11,188,002	10,614,000	33,995	33,480	31,412
West Dunbartonshire	11,427,186	12,372,149	12,526,000	30,232	32,823	33,262
West Lothian	14,232,669	16,636,902	16,231,000	24,692	28,728	27,804
All of Scotland	718,388,291	698,990,381	697,310,000	33,851	32,805	32,568

capita expenditure ranged from £38,471 to £86,084 in 2012/13; £36,852 to £92,379 in 2013/14; and £37,703 to £85,929 in 2014/15. Per capita combined health and social care expenditure was considerably greater in NHS Shetland, a "remote rural" area, for each of the 3 years, and in general, higher for other rural areas, but not consistently so across Scotland, for example, NHS Orkney is also a "remote rural" area, whilst NHS Fife is an "other urban area."

The mean per capita combined health and social care expenditure on adult intellectual disabilities services across the urban/rural classes for each of the 3 year periods is shown in Table 3. There was no statistically significant difference between the urban/rural classes in per capita combined health and social care expenditure using one-way analysis of variance: $F = 0.44$, $p = 0.729$ in 2012/13; $F = 0.63$, $p = 0.615$ in 2013/14; and $F = 0.42$, $p = 0.742$ in 2014/15. The Tukey test also showed no significant differences between the urban/rural classes.

The change in per capita combined health and social care expenditure between 2012/13 and 2014/15 ranged from −£5,607 to

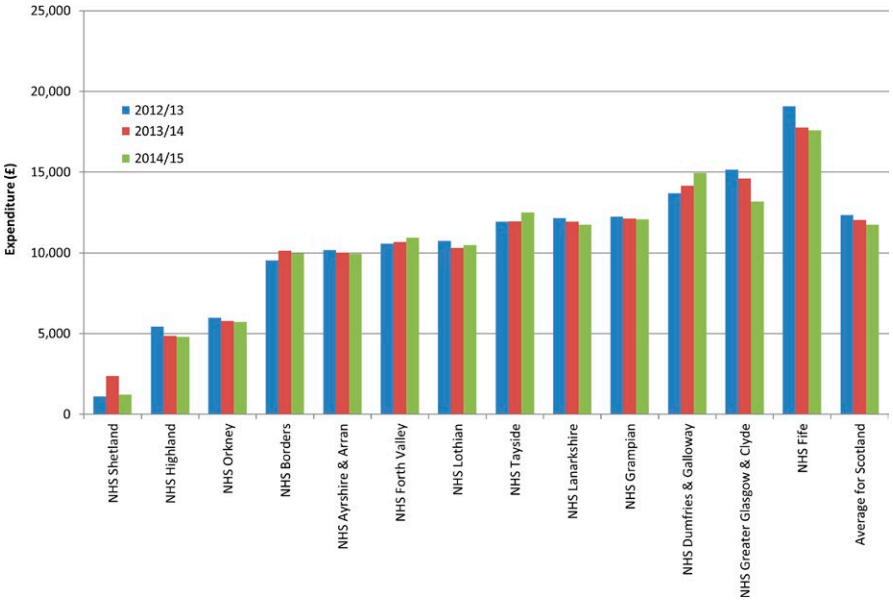

FIGURE 1 Per capita real terms expenditure on adult intellectual disabilities services by Scottish Health Boards, 2012/13, 2013/14, 2014/15 [Colour figure can be viewed at wileyonlinelibrary.com]

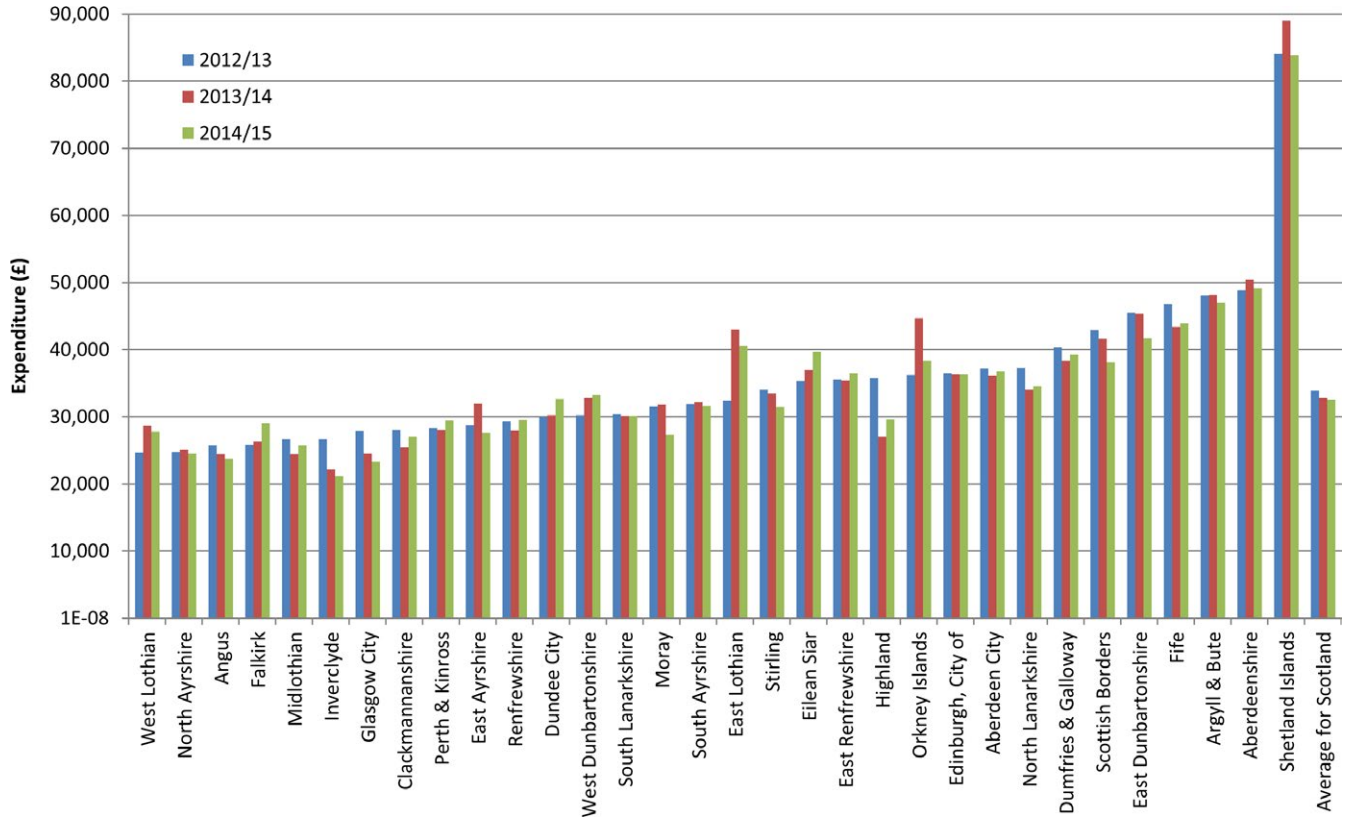

FIGURE 2 Per capita real terms expenditure on adult intellectual disabilities services by Scottish Local Authorities, 2012/13, 2013/14, 2014/15 [Colour figure can be viewed at wileyonlinelibrary.com]

£1,881. For five of the areas, it had increased over this 3 year period, but for eight areas, it had decreased. Year-on-year reduction in expenditure was seen in two areas, NHS Greater Glasgow and Clyde, which is the largest area in Scotland, accounting for 21.49% of the total Scottish population, and NHS Borders, which is one of the smaller areas.

Based on the size of the whole adult population in Scotland, the cost of the combined adult intellectual disabilities health and social care services to each adult in the general population was £223 in 2012/13, £216 in 2013/14 and £213 in 2014/15. It varies across areas, ranging from £176 to £322 in 2012/13; £175 to £346 in 2013/14; and £182 to £322 in 2014/15 (Table 6).

TABLE 3 Mean per capita expenditure on intellectual disabilities services across the urban/rural classes for 2012/13, 2013/14, 2014/15

	Mean per capita health care expenditure (real terms expenditure in 2014–15 terms, £)			Mean per capita social care expenditure (real terms expenditure in 2014–15 terms, £)			Mean per capita combined health and social care expenditure (real terms expenditure in 2014–15 terms, £)		
Urban/rural class	2012/13	2013/14	2014/15	2012/13	2013/14	2014/15	2012/13	2013/14	2014/15
Large urban areas (Class 1)	12,435	12,186	11,994	34,387	33,632	33,833	45,472	44,724	44,411
Other urban areas (Class 2)	13,369	13,158	13,359	30,194	29,589	29,460	49,350	48,052	48,438
Accessible rural areas (Class 5)	9,539	10,140	9,966	38,929	41,725	38,803	52,436	51,777	48,112
Remote rural areas (Class 6)	4,170	4,341	3,910	47,911	49,189	47,707	57,364	59,806	56,071
All of Scotland[a]	10,592	10,517	10,393	35,233	35,306	34,709	49,945	49,771	48,625

[a]Excluding NHS Western Isles for healthcare expenditure.

TABLE 4 Tukey post hoc test showing only statistically significant differences in mean per capita expenditure across the urban/rural classes for 2012/13, 2013/14, 2014/15

Major urban/rural class	Year	Contrast	Std. Error	*p* value	Lower 95% CI	Upper 95% CI
Health boards						
6 versus 1	2012/13	−8,265.080	2,118.036	0.016	−14,877.160	−1,652.998
6 versus 1	2013/14	−7,844.741	1,787.247	0.008	−13,424.170	−2,265.316
6 versus 1	2014/15	−8,084.713	1,777.680	0.006	−13634.270	−2,535.155
6 versus 2	2012/13	−9,199.120	2,215.095	0.011	−16,114.200	−2,284.039
6 versus 2	2013/14	−8,816.501	1,869.148	0.005	−14,651.600	−2,981.398
6 versus 2	2014/15	−9,449.208	1,859.142	0.003	−15,253.080	−3,645.341
Local authorities						
6 versus 2	2012/13	17,716.940	5,160.587	0.010	3,626.926	31,806.950
6 versus 2	2013/14	19,600.280	5,535.511	0.007	4,486.607	34,713.940
6 versus 2	2014/15	18,247.880	5,124.383	0.007	4,256.722	32,239.050

4 | DISCUSSION

4.1 | Principal findings and interpretation

Scotland's combined health and social care expenditure on adult intellectual disabilities services was £975,904,242 in 2012/13; £950,759,710 in 2013/14; and £943,692,001 in 2014/15. This is a real term decrease of 3.41% across the 3 years, and >£32 million. Per capita expenditure varies considerably across the country, revealing a postcode lottery. Whilst per capita expenditure on adult intellectual disabilities services may appear a lot (on average £44,265 in 2014/15), the cost to each adult in the general population is not (on average £213 in 2014/15).

Within context, the 4.29% real term decrease in intellectual disabilities health care expenditure and 3.02% real term decrease in intellectual disabilities social care expenditure between 2012/13 and 2014/15 sits within a real term decrease of 2.60% in the Scottish health budget between 2012/13 and 2014/15, and real term decrease of 10.69% in Scottish local government budget between 2012/13 and 2014/15. These Scottish figures are derived from the GDP deflators by HM Treasury (2014) on the cash terms budgets for 2012/13, 2013/14 and 2014/15 (Scottish Government, 2012, 2013, 2014a). This suggests that compared to expenditure on other services, intellectual disabilities health expenditure is being disproportionally reduced, whereas intellectual disabilities social care expenditure (whilst reduced) is not disproportionately so.

Per capita expenditure on health care was strongly positively correlated with the number of adults with intellectual disabilities in 2012/13. However, this relationship was not seen for healthcare expenditure in 2013/14 and 2014/15, nor for social care expenditure across all the three years, suggesting no consistent economy of scale for the larger Health Boards or Local Authorities.

The extent to which these real term decreases in expenditure on intellectual disabilities services have impacted on health and social

TABLE 5 Total and per capita expenditure on combined adult intellectual disabilities health and social care services by geographic (Health Board) area, 2012/13, 2013/14, 2014/15

	Total health and social care expenditure (real terms expenditure in 2014–15 terms, £)			Per capita health and social care expenditure (real terms expenditure in 2014–15 terms, £)		
Geographic area	2012/13	2013/14	2014/15	2012/13	2013/14	2014/15
NHS Ayrshire & Arran	65,998,410	67,964,253	64,558,884	38,471	39,646	37,703
NHS Borders	25,481,946	25,213,196	23,453,455	52,436	51,777	48,112
NHS Dumfries & Galloway	32,143,656	31,217,380	32,255,810	53,950	52,435	54,183
NHS Fife	84,359,362	78,467,195	79,073,878	65,893	61,156	61,499
NHS Forth Valley	43,297,025	43,337,440	45,176,535	39,087	38,973	40,365
NHS Grampian	108,333,143	109,666,425	108,125,742	53,119	53,259	52,127
NHS Greater Glasgow & Clyde	232,879,896	219,021,612	209,199,740	45,436	42,601	40,443
NHS Highland	55,589,784	46,737,214	48,806,538	44,011	36,852	38,404
NHS Lanarkshire	115,387,809	110,496,368	110,983,524	45,808	43,775	43,870
NHS Lothian	133,951,232	139,274,694	140,298,905	42,877	44,205	44,058
NHS Orkney	3,624,636	4,344,921	3,813,995	41,997	50,186	43,878
NHS Shetland	6,067,388	6,529,286	6,089,848	86,084	92,379	85,929
NHS Tayside	68,789,957	68,489,726	71,855,147	40,119	39,779	41,558
All of Scotland[a]	975,904,242	950,759,710	943,692,001	46,191	44,817	44,265

[a]Excluding NHS Western Isles.

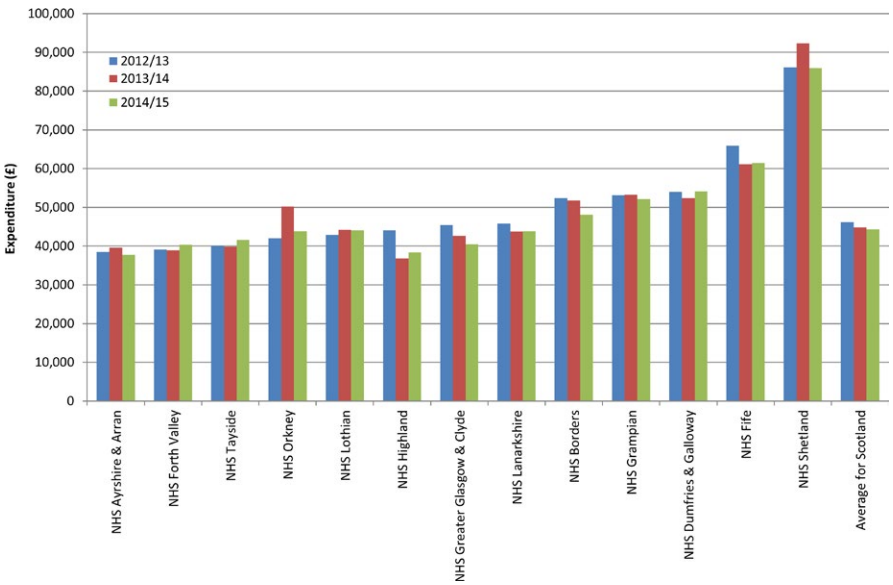

FIGURE 3 Per capita real terms expenditure on combined adult intellectual disabilities health and social care services by geographic (Health board) area, 2012/13, 2013/14, 2014/15 [Colour figure can be viewed at wileyonlinelibrary.com]

inequalities experienced by adults with intellectual disabilities is not known. Having identified the decrease in expenditure, it will be important to further investigate this. In addition, the impact, if any, that budgetary integration will have on expenditure nor on health and social inequalities is not known. However, this study now provides baseline data against which changes may be measured.

Health Board per capita expenditure is less in "remote rural" and "accessible rural" area than urban areas, significantly so for "remote rural" areas compared with "large urban" and "other urban" areas. Conversely, Local Authority per capita expenditure is greater in "remote rural" and "accessible rural" areas than urban areas, significantly so for "remote rural" areas compared with "other urban" areas. This may be explained as social care is needed every day, so it needs to be delivered locally with greater cost in remote rural areas due to distances covered, whereas it may be possible to travel for health care even though it is less accessible. Indeed, there is comprehensive coverage of multidisciplinary intellectual disabilities community teams across the "large urban" and "other urban" areas in Scotland, but limited services in "remote rural" areas, with many relying on intellectual disabilities nursing only. Though not significant, combined health and social care costs were marginally greater in "other urban" compared to "large urban" areas. This possibly reflects lower service

TABLE 6 Total expenditure on combined adult intellectual disabilities health and social care services by total adult population in each geographic (Health Board) area, 2012/13, 2013/14, 2014/15

	Total expenditure per adult in the general population (real terms expenditure in 2014–15 terms, £)		
Geographic area	2012/13	2013/14	2014/15
NHS Ayrshire & Arran	213	220	209
NHS Borders	269	266	247
NHS Dumfries & Galloway	255	248	256
NHS Fife	279	259	260
NHS Forth Valley	176	175	182
NHS Grampian	226	227	222
NHS Greater Glasgow & Clyde	246	231	219
NHS Highland	209	175	183
NHS Lanarkshire	216	207	207
NHS Lothian	191	196	196
NHS Orkney	201	241	210
NHS Shetland	322	346	322
NHS Tayside	200	199	207
All of Scotland[a]	223	216	213

[a]Excluding NHS Western Isles.

delivery costs in "large urban" areas compared to "other urban" areas. It should be noted though, that the social care finding was disproportionately influenced by the Shetland islands who are an outlier for per capita expenditure on social care; it is not mirrored to such an extent in the other "remote rural" areas, the Orkney islands, Eilean Siar (Western isles), Highland, or Argyll and Bute, although three of these four are all above the mean per capita expenditure for Scotland, and the other (Highlands) is approximately at the mean. Hence, per capita social care expenditure is more in "remote rural" areas, reflecting the particular challenges these areas face in delivering care to very dispersed communities.

These urban/rural findings for health mirror those previously reported for England (Forsyth & Winterbottom, 2002), but differ with regards to social care (Moscone, 2011). A previous report from Scotland found that adults with intellectual disabilities living in rural areas had more contact with primary and secondary healthcare services, and dentists and opticians, than adults with intellectual disabilities in a large urban area (Nicholson & Cooper, 2011). Our study has examined specifically intellectual disabilities health services, rather than the generic health services studied by Nicholson and Cooper (2011), accounting for this difference. It seems that in rural areas, there is a greater reliance on generic health services.

In Scotland, out of area placements are a very small percentage of the total population of adults with intellectual disabilities and so have negligible effect on population estimates. Learning Disability Statistics Scotland (Scottish Consortium for Learning Disability, 2017) reports that only 3.6% of adults with intellectual disabilities known to Scottish Local Authorities are subject to out of area placements. Therefore, these do not have any significant impact on the per capita spending figures. Scotland differs from, for example, England in this respect, as none of the Local Authorities are exclusively areas of prohibitive housing costs; and remote and rural island and highland communities would be very unlikely recipients of out of area placements.

Whilst the 2007/2008 financial crisis spread across much of the world, government responses to it have differed. Our results are set within the UK context of an austerity programme, so may not reflect experiences in other countries. However, our study findings are important and have implications for policy developments across high-income countries.

4.2 | Strengths and limitations

The study does have limitations. The individual proportions of health and social care expenditure out of combined health and social care varied considerably across Scotland, no doubt influenced by local factors which studies such as ours cannot capture. There may be some differences in recording expenditure in different Health Boards or Local Authorities, which are not identified in the secondary analysis of Scottish Government routinely collected administrative expenditure data submitted by each Health Board and Local Authority. Our study was restricted to adults only, as it is not possible to disaggregate expenditure on child/young persons intellectual disabilities services across Scotland; as this is integrated within expenditure on children's services more widely. Regarding the estimates of Scotland's population with intellectual disabilities, it is possible, but unlikely, that the proportion of people with intellectual disabilities within the general population has marginally fallen over the 3 year study period; indeed, it may have risen if the extent of the inequality gap in lifespan reduced. Hence, it is unlikely that this would make any meaningful difference to measures of per capita expenditure; this approach is unlikely to either over- or under-estimating the intellectual disabilities population size, and this methodological approach is considered to be the best possible. In an important manner, in support of this approach, it should be noted that Scotland has not been subject to immigration at the same levels as some other European countries. The study's strengths are the inclusion of the whole of the country, contemporaneous records of actual expenditure which was adjusted for inflation to allow yearly comparisons, and the uniqueness of Scotland's Census, 2011, in identifying all adults with intellectual disabilities in the country, hence allowing per capita expenditure to be calculated.

5 | CONCLUSION

We conclude that austerity has impacted on Scotland's expenditure on adult intellectual disabilities services, by not keeping abreast of rises in living costs, and with much variation across areas. With the

exception of social care expenditure in "remote rural" areas, the variation in expenditure across Scotland—a postcode lottery—is of particular concern, suggesting that health and social care is not equitable across the country for adults with intellectual disabilities. In Scotland, health and social care integration was effected in 2016, with Integration Authorities responsible for funding local services, previously separately managed by NHS Health Boards and Local Authorities. The full benefits and disadvantages of the new arrangements are yet to be seen; this study now provides baseline expenditure data for the whole country with which trends/changes post-integration can be measured, to support future research on health and social care outcomes.

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RESEARCH

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A study on prescriptions contributing to the risk of high anticholinergic burden in adults with intellectual disabilities: retrospective record linkage study

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Abstract

Background: People with intellectual disabilities may face a disproportionate risk of experiencing high anticholinergic burden, and its negative sequalae, from a range of medications, and at younger ages than the general population, but there has been little previous study. Our aim was to determine the source of anticholinergic burden from prescribed medication.

Methods: Retrospective matched observational study using record linkage. Adults with ($n=4,305$), and without ($n=12,915$), intellectual disabilities matched by age-, sex- and neighbourhood deprivation. The main outcome measure was the prescription of long-term (approximately 12 months use) anticholinergic medications overall (classified according to the Anticholinergic Risk Scale [ARS]), by drug class, individual drugs, and polypharmacy.

Results: Adults with $n=1,654$ (38.4%), and without $n=3,047$ (23.6%), intellectual disabilities were prescribed medications long-term with anticholinergic effects. Of those on such drugs, adults with intellectual disabilities were most likely to be on central nervous system (62.6%), gastrointestinal (46.7%), and cardiovascular (28.4%) medications. They were prescribed more central nervous system, gynaecological/urinary tract, musculoskeletal, and respiratory medications, and less cardiovascular, infection, and endocrine medications than their matched comparators. Regardless of age, sex, or neighbourhood deprivation, adults with intellectual disabilities had greater odds of being prescribed antipsychotics (OR = 5.37 [4.40–6.57], $p<0.001$), antiepileptics (OR = 2.57 [2.22–2.99], $p<0.001$), and anxiolytics/hypnotics (OR = 1.28 [1.06–1.56], $p=0.012$). Compared to the general population, adults with intellectual disabilities were more likely to be exposed to overall anticholinergic polypharmacy (OR = 1.48 [1.33–1.66], $p<0.001$), and to psychotropic polypharmacy (OR = 2.79 [2.41–3.23], $p<0.001$).

Conclusions: Adults with intellectual disabilities are exposed to a greater risk of having very high anticholinergic burden through polypharmacy from several classes of medications, which may be prescribed by several different prescribers. There is a need for evidence-based recommendations specifically about people with intellectual disabilities with multiple physical and mental ill-health conditions to optimise medication use, reduce inappropriate prescribing and adverse anticholinergic effects.

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Keywords: Intellectual disabilities, Anticholinergic burden, Polypharmacy, Psychotropics

Background

Adults with intellectual disabilities experience an increased risk of adverse effects associated with high anticholinergic burden [1–4]. Previous literature has focused predominantly on the older adult general population and found high anticholinergic burden to be associated with a range of poor health outcomes including cognitive impairment and dementia [5]; emergency health service use [6], and all-cause mortality [7–9]. Common adverse effects of anticholinergic burden include constipation, dry mouth, blurred vision, cognitive impairment, urinary retention, and increased fall risk. Some recent studies have also included younger adults. A UK Biobank study investigating 502,538 healthy adults found high anticholinergic burden in 27–73 year olds was modestly associated with cardiovascular events, hospital admissions (due to fall/fractures, and dementia/delirium) and mortality [9]. The cumulative impact of anticholinergic burden is, therefore, a pertinent issue in the pharmacotherapy of all patients across the lifespan.

People with intellectual disabilities have poorer health than the general population, and often have long-term health problems. Higher multimorbidity and more complex health needs occur at younger ages and continue throughout the lifespan, with poorer health outcomes and premature mortality [10]. Subsequently, people with intellectual disabilities are more likely to experience polypharmacy (taking multiple medication concomitantly, e.g., 5+) [11] with complex pharmacotherapy, that can result in a significantly increased risk of adverse effects. The mean number of physical health conditions experienced by adults with intellectual disabilities is 11.04 [12], as well as 40% experiencing mental ill-health [10]. Unsurprisingly, one study reported that for adults with intellectual disabilities living in rural areas, in addition to a median of 8 (IQ = 4–11) contacts with primary health care over a year, 43.6% also had contacts with secondary or tertiary health care, 10.3% had hospital admissions, 10.3% had emergency GP contacts, and 20.5% presented to accident and emergency departments [13]. To a lesser extent, the figures were also high for participants in urban areas [13]. Hence, many adults with intellectual disabilities are treated by multiple prescribers. Moreover, prescriptions initiated in hospital settings are managed in the community, and many general practitioners in primary care, report being concerned changing anticholinergic medications initiated by specialists [14].

Whilst some increased medication use is appropriate, given the higher prevalence of physical and mental ill-health, there are an increasing number of guidelines and initiatives aimed at reducing inappropriate prescribing, considering medication review and de-prescribing where appropriate [15–17]. Previous evidence has indicated psychotropics to be the main drug class responsible for high anticholinergic burden in older adults with intellectual disabilities (aged 40+) [3]. Within this class of medications, a large focus of attention has been on the off-label use of antipsychotics to manage 'challenging behaviour' in this population [18]. This is despite high anticholinergic risk, lack of substantial evidence base of effectiveness in this context, and no clear indication for antipsychotic use [18]. If there is no diagnosis of mental ill-health, best practice guidelines recommend short-term use only with review of efficacy after 3–4 weeks [16], and as part of a multimodal treatment approach, i.e., non-pharmacological psychosocial or behavioural interventions [19].

This paper reports on the contribution of different classes of drugs to the risk of high anticholinergic burden in adults with intellectual disabilities compared to those in the general population, and also investigates psychotropic prescriptions in further detail.

Methods

Study cohort

The research was approved by Scotland's Public Benefit and Privacy Panel for Health and Social Care (reference 1516–0281). The National Health Service (NHS) Greater Glasgow and Clyde Primary Care Intellectual Disabilities Register of 2014 was created from multiple sources (e.g., general practitioners who were financially incentivised to identify all their patients with intellectual disabilities (100% did so) and community intellectual disabilities teams) and updated annually thereafter. It was used to identify all adults (aged 17+) with intellectual disabilities within the defined geographical boundary of NHS Greater Glasgow and Clyde, which forms almost a quarter of the Scottish population. Adults with intellectual disabilities were each matched to three general population adults based on age (year of birth), sex, and neighbourhood deprivation (Scottish Index of Multiple Deprivation: SIMD2016 using the postcode area). Whilst health in the general population is closely linked to neighbourhood deprivation [20], previous research suggests this gradient is not evident in the intellectual disabilities population [21]. However, people with intellectual disabilities are more likely to live in the more deprived areas. This distinction is important, as public health

initiatives designed for the general population that are focussed on more deprived areas build in disadvantage to people with intellectual disabilities living in more affluent areas, but who have health inequalities. Therefore, we chose to match the groups to reflect the difference structure of the intellectual disabilities population compared to the general population. In Scotland, patient health records are identified using their unique patient identifier; the Community Health Index (CHI), which is held centrally by National Services Scotland (NSS). The CHI database was used for this matching and linkage process. NSS also holds the Prescribing Information System database, which was used to identify 1 year of anticholinergic medication prescriptions between September 2016 and August 2017 for the study groups. This data source records all community prescribed and dispensed medications in Scotland and classifies them via British National Formulary (BNF) categories. NHS Scotland's Information Services Division completed data linkage and extraction in August 2017. The study group consisted of 17,228 but during data cleaning the research team identified two general population adults as having intellectual disabilities. Since each adult with intellectual disabilities was matched with general population adults 1:3 to create one 'cluster', all participants from these matched clusters were excluded ($n=8$), leaving a sample size of $n=17{,}220$. Further details on sampling and linkage have previously been reported [4].

Anticholinergic medications were identified using the modified Anticholinergic Risk Scale (ARS) which was updated by the authors [4]. The ARS list classifies anticholinergic medicines as moderate (risk category 1), strong (risk category 2), and very strong (risk category 3), and is one of the more conservative anticholinergic burden scales [9]. There are numerous anticholinergic burden scales, although there is consensus of a cumulative total score of 3 or greater to be clinically at risk of adverse effects. To determine the main contributors to anticholinergic burden, prescriptions were only included if there were 3 or more repeat prescriptions constituting approximately 9–12 months usage during the year being studied. Anticholinergic drugs were those listed in the updated ARS and categorised according to the BNF chapter, and psychotropic sub-chapters.

Statistical analysis

Data were summarised descriptively by group (those with, and without intellectual disabilities), for each medication outcome. Outcomes investigated were individual medication classes, psychotropic prescribing (antipsychotics, antiepileptics, antidepressants, anxiolytics/hypnotics) and polypharmacy. Group comparisons were made using mixed-effects binary logistic regression models with a fixed effect for group, and a random effect for the matched cluster. Group effects are reported from models adjusted for known confounders (sex, age category, SIMD quintile), with the general population as reference and intellectual disabilities cohort as effect level.

Interaction terms between group and each main effect (sex, age, SIMD quintile) were added to adjusted models and when significant, explored using both odds ratios (OR) and marginal effects at representative values (MERs). OR [95% Confidence Intervals] and p values are reported for the adjusted group effect tested within each level of the main effect, e.g., significant interaction between group (adults with and without intellectual disabilities) and age examined by modelling the group effects within each age category. Using the full model (main effects and interaction term), the MER analyses shows the magnitude of effects allowing us to obtain predicted probabilities of the group effect on each outcome. Adjusted MERs describe the strength of an association between a risk factor (e.g., group) and the outcome (e.g., antipsychotic use), allowing us to see how the marginal effect differs across that range (e.g., change across age categories), at representative values of covariates (held constant) [22], e.g., the predicted probability of being on an antipsychotic if an adult is in the intellectual disabilities group and aged 55–64 (within group analysis). Values were chosen on the basis of no statistically significant effects of that covariate in the group interaction term (e.g., average of sex and SIMD for group differences in antipsychotic prescribing). Statistical analyses were conducted in Stata (StataCorp 2017).

Results

Demographics

The full data set included $n=4{,}305$ adults with intellectual disabilities and $n=12{,}915$ general population adults. General demographics for this full sample can be seen in Table 1. Of the full sample, there were 1,654 (38.4%) adults with intellectual disabilities prescribed long-term anticholinergic medication, compared to 3,047 (23.6%) adults without intellectual disabilities. These groups comprise our population for all analyses (Table 1).

Anticholinergic drugs across medication classes

For the adults with intellectual disabilities prescribed anticholinergic medication, these were most likely to be central nervous system (62.6%), gastrointestinal (46.7%), cardiovascular (28.4%), and respiratory system medications (13.4%) (Table 2). The pattern differed for the general population (though may not be representative of the whole general population who are older, have equal sex ratio, and a different distribution of SIMD, due to the matching with the intellectual disabilities group).

Table 1 Descriptive statistics for total sample and subgroup of those prescribed long-term anticholinergic medication (≥ 3 repeat prescriptions over 12 months)

Characteristic	Total		AC medications	
	ID	GPop	ID	GPop
Population	4,305	12,915	1,654	3,047
Sex				
Male	2,504 (58.2%)	7,512 (58.2%)	925 (55.9%)	1,628 (53.4%)
Female	1,801 (41.8%)	5,403 (41.8%)	729 (44.1%)	1,419 (46.6%)
Age categories				
17–24	403 (9.4%)	1,209 (9.4%)	96 (5.8%)	38 (1.3%)
25–34	811 (18.8%)	2,433 (18.8%)	224 (13.5%)	172 (5.6%)
35–44	673 (15.6%)	2,019 (15.6%)	210 (12.7%)	282 (9.3%)
45–54	965 (22.4%)	2,895 (22.4%)	418 (25.3%)	726 (23.8%)
55–64	794 (18.4%)	2,382 (18.4%)	366 (22.1%)	859 (28.2%)
65–74	464 (10.8%)	1,392 (10.8%)	235 (14.2%)	645 (21.2%)
75 +	195 (4.5%)	585 (4.5%)	105 (6.4%)	325 (10.7%)
SIMD				
1-most deprived	2,293 (53.3%)	6,879 (53.3%)	867 (52.4%)	1,821 (59.8%)
2	848 (19.7%)	2,544 (19.7%)	331 (20.0%)	605 (19.9%)
3	545 (12.7%)	1,635 (12.7%)	233 (14.1%)	335 (11.0%)
4	349 (8.1%)	1,047 (8.1%)	135 (8.2%)	173 (5.7%)
5-least deprived	270 (6.3%)	810 (6.3%)	88 (5.3%)	113 (3.7%)

AC anticholinergic, *GPop* general population, *ID* intellectual disabilities

Compared to the matched general population, the adults with intellectual disabilities were more likely to be prescribed central nervous system (2.23 [1.96–2.53]), obstetrics, gynaecology, urinary tract (2.82 [1.83–4.34]), and musculoskeletal (2.49 [1.69–3.67]) medications and were less likely to be prescribed cardiovascular (0.61 [0.53–0.71]), infections (0.43 [0.21–0.68]), and endocrine (0.45 [0.29–0.68]) medications.

Table 3 shows the most commonly prescribed anticholinergic drugs (ranked 1–10) for the intellectual disabilities group; 5 are psychotropics, 3 are cardiovascular, and the remaining 2 are gastrointestinal.

Psychotropic prescribing

Of the anticholinergic central nervous system drugs that were prescribed, the majority (> 99%) were psychotropics (antipsychotics, antiepileptics, antidepressants, anxiolytics/hypnotics). Compared to the adults without intellectual disabilities, regardless of sex, age or neighbourhood deprivation, adults with intellectual disabilities had greater odds of being prescribed antipsychotics (5.37 [4.40–6.57]); antiepileptics (2.57 [2.22–2.99]); and anxiolytics/hypnotics (1.28 [1.06–1.56]), all statistically significant at $p < 0.01$. The likelihood of anticholinergic antidepressant prescribing was similar between the groups ($p = 0.174$). Table 4 shows the number and percentage of adults with, and without, intellectual disabilities prescribed psychotropic drugs for each of these subcategories.

Extending these findings to include the interaction terms between group and the main effects (sex, age, neighbourhood deprivation), revealed some statistically significant results. The interaction between group and age was significant for antipsychotics ($p < 0.001$), antidepressants ($p < 0.001$) and anxiolytics/hypnotics ($p = 0.01$),

Table 2 Number and percentage of adults with intellectual disabilities prescribed anticholinergic medication by medication class (BNF chapter), in comparison with general population adults

BNF chapter	Adults prescribed anticholinergic medications ($n = 4,701$)		OR [95% CI], p value*
	Intellectual disabilities ($n = 1,654$)	General population ($n = 3,047$)	
Gastro-intestinal system	773 (46.7%)	1,505 (49.4%)	1.00 [0.88–1.13], $p = 0.985$
Cardiovascular system	477 (28.4%)	1,419 (46.6%)	0.61 [0.53–0.71], $p < 0.001$
Respiratory system	222 (13.4%)	373 (12.3%)	1.06 [0.88–1.27], $p = 0.547$
Central nervous system	1,037 (62.6%)	1,231(40.4%)	2.23 [1.96–2.53], $p < 0001$
Infections	9 (0.54%)	53(1.7%)	0.43 [0.21–0.86], $p = 0.017$
Endocrine system	29 (1.8%)	120 (3.9%)	0.45 [0.29–0.68], $p < 0.001$
Obstetrics, gynaecology and urinary-tract disorders	51 (3.1%)	40 (1.3%)	2.82 [1.83–4.34], $p < 0.001$
Musculoskeletal and joint diseases	65 (3.9%)	40 (1.3%)	2.49 [1.69–3.67], $p < 0.001$

All regression models are adjusted for sex, age, and SIMD (neighbourhood deprivation)

* *OR* Odds Ratio, 95% *CI* Confidence Interval and corresponding *p* value for the Intellectual Disabilities group effect with the General Population as the reference group

Table 3 Top 10 anticholinergic medicines prescribed in adults with intellectual disabilities compared to general population controls

Medication (ARS risk category)	ID ($n=1,654$)	Rank	GPop ($n=3,047$)	Rank
Omeprazole (1)	26.5%	1	29.0%	1
Carbamazepine (1)	16.4%	2	1.5%	29
Lansoprazole (1)	12.9%	3	12.6%	4
Ramipril (1)	11.9%	4	15.3%	3
Lamotrigine (1)	10.6%	5	1.1%	34
Risperidone (1)	10.2%	6	0.9%	38
Amlodipine (1)	9.6%	7	17.1%	2
Sertraline (2)	9.0%	8	9.3%	6
Diazepam (2)	7.9%	9	5.7%	11
Bendroflumethiazide (1)	6.2%	10	9.9%	5

Percentages are presented from the subgroup sample taking long-term anticholinergic medicines

ARS Anticholinergic Risk Scale, *ID* Intellectual Disabilities, *GPop* General Population

Table 4 Number and percentage of adults with intellectual disabilities prescribed anticholinergic psychotropic medication by subcategory, in comparison with general population adults

Psychotropic subcategory	Intellectual disabilities ($n=1,654$)	General population ($n=3,047$)	OR [95% CI], p value*
Antipsychotics	411 (24.9%)	173 (5.7%)	5.37 [4.40–6.57], p < 0.001
Antiepileptics	486 (29.4%)	393 (12.9%)	2.57 [2.22–2.99], p < 0.001
Antidepressants	206 (12.5%)	383 (12.6%)	0.87 [0.71–1.06], p = 0.174
Anxiolytics/hypnotics	222 (13.4%)	289 (9.5%)	1.28 [1.06–1.56], p = 0.012

Numbers and percentages are presented from the subgroup sample taking long-term anticholinergic medicines. All regression models are adjusted for sex, age, and SIMD

* *OR* Odds Ratio, 95% *CI* Confidence Interval and corresponding *p* value for the Intellectual Disabilities group effect with the General Population as the reference group

whilst the interaction between group and neighbourhood deprivation was significant for antiepileptics ($p<0.001$). ORs for the subgroup analyses by age (Additional file 1: Table S1) and deprivation (Additional file 1: Table S2) are reported. Figure 1 plots the MERs by group and age, averaging across sex and SIMD, demonstrating the change in risk factors that affect the probability that the individual is on a specific type of psychotropic (1 representing a 100% likelihood).

Prescribing of antipsychotics in the general population decreased from younger to older adults, but for adults with intellectual disabilities it peaked at 55–64 years, hence the extent of the difference between the groups increased with age (Fig. 1a). Antipsychotic statistically significant group differences start from age group 25–34 (2.48 [1.34–4.60]), rising over each age group, to age 75 + (61.07 [13.11–284.44]) (Additional File 1: Table S1). Age-related prescribing patterns of antidepressant and anxiolytic/hypnotics showed fewer statistically significant results. Whereas the general population show a decrease in antidepressant prescriptions from younger to older adults, the intellectual disabilities group fluctuated; ORs were significantly lower for the intellectual disabilities group at ages 17–24 (0.06 [0.01–0.27]), and higher for ages 65–74 (2.21 [1.35–3.64]) (Fig. 1b, Additional File 1: Table S1). Differences in anxiolytic prescribing between the groups were statistically significant, with increased prescribing for the intellectual disabilities group for 17–24 years (2.76 [1.00–7.63]) and 55–64 years (1.97 [1.28–3.03]) but did not differ at other ages (Additional File 1: Table S1). With regard to antiepileptics, the extent of differences between groups showed a positive gradient across SIMD, with the smallest difference in the most deprived neighbourhood (1.94 [1.59–2.37]), and the greatest difference being in more affluent areas (8.77 [3.64–21.15]) (Additional File 1: Table S2 and Figure S1). This was due to higher prescriptions of antiepileptics in more deprived neighbourhoods in the general population, but not in the adults with intellectual disabilities.

Polypharmacy

The range of different anticholinergic medicines a person was prescribed was between 1 and 11 for both groups, with a median of 2. Anticholinergic polypharmacy (concurrent use of 2 + anticholinergic medications) was significantly higher in the adults with intellectual disabilities group with 61.3% ($n=1,013$) prevalence compared to 54.7% ($n=1,668$) (OR = 1.61 [1.41–1.83], $p<0.001$).

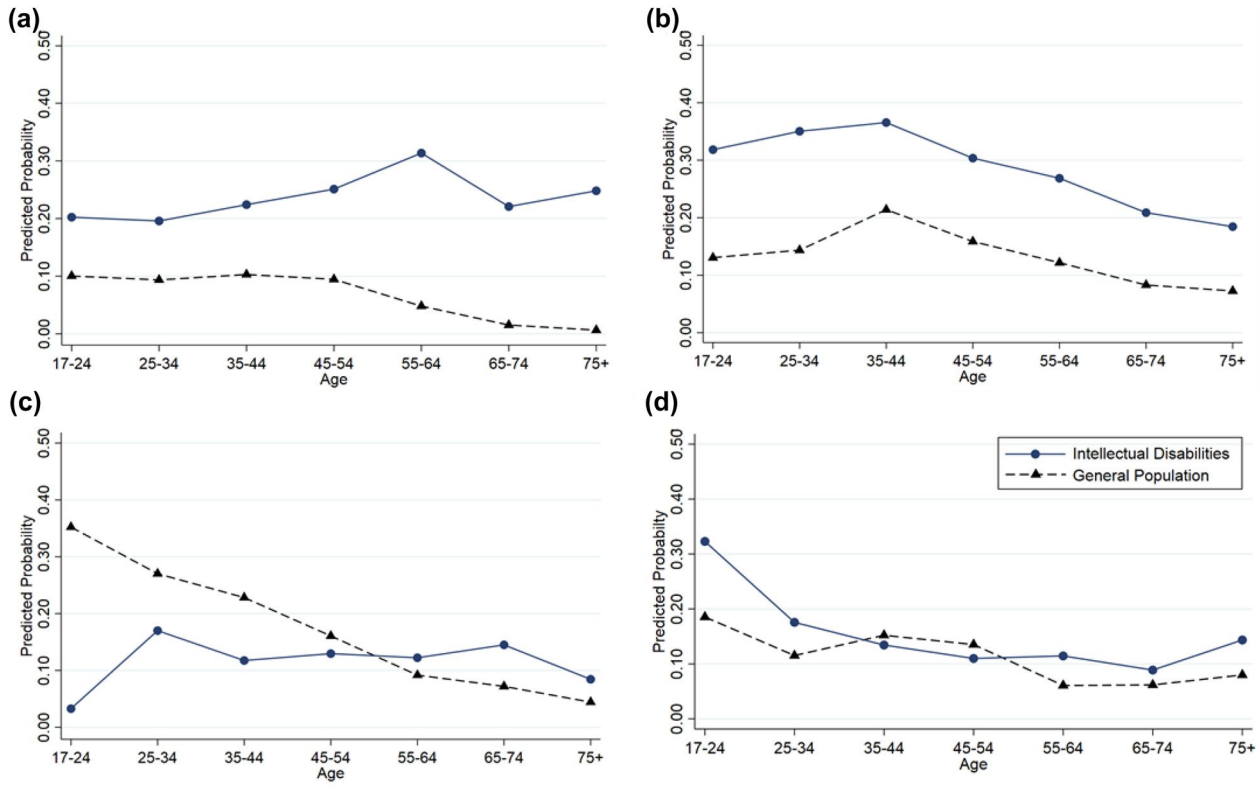

Fig. 1 Influence of age and groups' MERs for predicted probability of being on **a** antipsychotics, **b** antiepileptics, **c** antidepressants, and **d** anxiolytics/hypnotics

Psychotropic polypharmacy (concurrent use of 2 + psychotropic medications) was also significantly higher in the adults with intellectual disabilities group with 33.7% ($n = 558$) prevalence compared to 14.3% ($n = 435$) (OR = 2.79 [2.41–3.23], $p < 0.001$). These results are adjusted for age, sex, and neighbourhood deprivation. Adults with intellectual disabilities are almost three times more likely to be exposed to psychotropic polypharmacy compared to the general population.

Discussion

Principal findings and interpretation

Our findings are important, as they demonstrate that the high rates of anticholinergic burden experienced by adults with intellectual disabilities at all ages (not just older ages) are due to several classes of medications, not only psychotropic, and which may be prescribed by several different clinicians. Whilst the general practitioner has an overview, it is important for all clinicians to be cognisant of this issue. Of the 38.4% of adults with intellectual disabilities on long-term anticholinergic prescriptions, the medications most frequently prescribed were central nervous system medications (62.6%), gastro-intestinal medications (46.7%), and cardiovascular medications (28.4%), and polypharmacy (both anticholinergic and psychotropic) was more common than monotherapy.

Examining psychotropic contributions to anticholinergic burden showed that adults with intellectual disabilities were more likely to be prescribed antipsychotics, antiepileptics and anxiolytics/hypnotics compared to adults without intellectual disabilities, and had similar levels of antidepressant prescriptions. Results on antipsychotic medication use support previous evidence of an age-related increase for those with intellectual disabilities [18, 23–25], in contrast to the general population who showed a decreased use across age (Fig. 1). Whilst we find statistically significant differences between the groups from ages 25 onwards, the smaller numbers in older age groups incur large confidence intervals (Additional File 1: Table S1). The current results and previous evidence [18, 26], indicate long-term antipsychotic use, despite associated health risks, including anticholinergic effects, and best practice guidelines advocating non-pharmacological approaches in the long-term [16]. These findings suggest antipsychotics comprise a significant contribution to high anticholinergic burden.

Regarding anticholinergic antiepileptics, the greatest group difference in the most affluent neighbourhoods (Additional File 1: Table S2, Figure S1). This reflects the higher rates of prescribing in more deprived areas in the general population, which is a typical finding [10]. Prescribing is high across all neighbourhoods for the intellectual disabilities population, with no obvious deprivation gradient. This highlights the need for medication reviews and service provision in all neighbourhoods regardless of extent of deprivation.

Polypharmacy

Anticholinergic polypharmacy across all drug classes was present, and at 1.6 greater odds for the adults with intellectual disabilities. The combination of multiple lower risk drugs (ARS score 1) and high polypharmacy in adults with intellectual disabilities contributes to the overall clinical risk of high anticholinergic burden. Non-anticholinergic licensed alternatives could be explored in place of the more commonly used anticholinergic medicines. A realistic medicine patient centred approach is key and the prescription of psychotropic medication should be "informed by a comprehensive biopsychosocial assessment" [19]. Rates of multimorbidity (physical and mental ill-health) are high in people with intellectual disabilities [10], so polypharmacy (including psychotropic) can be indicated and appropriate. For example, around 25% of adults with intellectual disabilities have epilepsy requiring antiepileptic medications [12]. Although, antiepileptic drugs are commonly prescribed as mood-stabilisers and not only seizure-control [27], which can increase the risk of high anticholinergic burden.

Clinical services are typically organised around single conditions, which can pose additional risks for polypharmacy. Comprehensive, regular, and targeted medication reviews would help to identify excess prescribing. To see where medications can be rationalised one drug at a time, to establish support for treatment adherence, to enable good communication between the person's different health care professionals/teams if they are accessing multiple services, and not focus simply on single-disease care pathways. This is well-recognised in the general population, e.g., recent NICE clinical guideline on multimorbidity [28], but has received little attention for people with intellectual disabilities. For example, a review on the effectiveness of medication reviews for those with intellectual disabilities included only 8 studies, with just 3 of good quality; all of which evidenced the reduction in medication-related problems after multidisciplinary medication reviews [29].

Comparison with previous literature

Inappropriate polypharmacy is a problem among adults with intellectual disabilities and has a high risk of adverse effects. Previous evidence from representative population samples of adults with intellectual disabilities report a high prevalence of medicine use (average of 4–7 prescriptions) and polypharmacy (overall between 21% and 38%, psychotropic polypharmacy between 23% and 41%) [30–33]. Higher rates of polypharmacy (54%) and psychotropic polypharmacy (66%) have been reported in older adults with intellectual disabilities (aged 40+) [2, 34]. Our data are comparable to these results; similar to Axmon et al. [1], we find that most adults take at least 2 anticholinergic medications irrespective of age or sex. Our data include only anticholinergic prescriptions, therefore, are likely an underestimation of overall prescriptions and polypharmacy, e.g., many adults with intellectual disabilities take the antiepileptic sodium valproate, which is psychotropic but not anticholinergic. Results show adults with intellectual disabilities were 2–3 times more likely to experience long-term polypharmacy compared to their peers. Our findings support previous work reporting high psychotropic polypharmacy in this patient population to be predominantly due to antipsychotic and antiepileptic medication use. [3, 24]

Polypharmacy and anticholinergic burden are interrelated, but both show an independent dose–response relationship with all-cause hospital admissions and mortality in general population adults [9, 35]. Polypharmacy prevalence is increasing; a recent UK Biobank study reported anticholinergic burden to be 3–9 times higher between 1990 and 2015 [36]. Inappropriate prescriptions pose a health risk, adults with intellectual disabilities have a 2.7 times greater odds of hospitalisation due to psychotropic adverse medication events [37]. More generally, there is evidence that every prescribed drug leads to an increase in having a potential drug–drug interaction of clinical significance for this population (OR = 0.87 [0.72–1.00]) [31]. People with intellectual disabilities often experience multimorbidity which may necessitate polypharmacy, but this can also mean increased exposure to potential drug–drug interactions. Moreover, both polypharmacy and multimorbidity were independent significant predictors for mortality in older (aged 50+) adults with intellectual disabilities [38]. The precise relationship between polypharmacy, anticholinergic burden, multimorbidity, and mortality is yet to be clarified.

Strengths and limitations

The strength of the current study lies in the sample; ours was a large representative group of adults with intellectual disabilities age-, sex-, and neighbourhood

deprivation-matched to general population adults without intellectual disabilities within Scotland's largest NHS health board. The use of this record linkage allows for robust information on prescribed medication with anticholinergic effects, although only a year of data was analysed. The study's specific focus is on anticholinergic medication, and not the total rates of encashed prescriptions; so conclusions on overall prescribing cannot be drawn. In addition, as this is prescription data, there is no clarity of patient usage, dosage, or clinical indication. Therefore, an important limitation is the lack of clinical data on the conditions, diseases, or indeed, actual anticholinergic burden as measured by serum activity assays. There is also a lack of information on the severity of the intellectual disabilities. Finally, we were unable to distinguish between regular or pro re nata (PRN) medication in our database. Whilst our conservative definition of only including medications with 3 + repeat prescriptions over the 12 months is highly likely to have included only long-term anticholinergic medication use, we cannot say with 100% certainty that it did not include some people with very high use of PRN (as required) medication. In addition, this means that we may have underestimated the extent of exposure to anticholinergic medication in view of the use of PRN antipsychotics in this population which did not meet our threshold definition.

Conclusions

In line with evidence from the general population, our data clearly show that drugs with a moderate anticholinergic risk from different drug classes contribute to the risk of clinically high anticholinergic burden [39]. In several cases, prescriptions of these drugs are unavoidable, for example, to manage complex epilepsies, however, given the multiple prescribers, an overview review by a clinician identified as responsible is essential to reduce or discontinue drugs when no longer needed, and avoid unnecessary repeat prescriptions. People with intellectual disabilities are particularly vulnerable, with commonplace psychotropic polypharmacy as well as overall anticholinergic polypharmacy. Many people with intellectual disabilities are on antipsychotic medication over years and sometimes decades [18, 26], despite associated health risks, including anticholinergic effects. However, following NHS England's "Stopping over-medication of people with a learning disability, autism or both (STOMP)" initiative [15], there is some evidence of successful antipsychotic deprescribing [40], but these prescriptions may be being replaced with other psychotropics, namely, antidepressants [41], which could contribute to anticholinergic burden similarly. There is a need for robust evidence-based recommendations for all prescribers specifically about people with intellectual disabilities with multiple physical and mental ill-health conditions to optimise medication use. Although pharmacotherapy for those with intellectual disabilities is particularly complex, prescribers may not prioritise reducing the risk of anticholinergic burden. We report on the anticholinergic side effects; an important next research step will be to study the extent of anticholinergic burden in people with intellectual disabilities as measured by serum anticholinergic activity. Our results show that we should be cautious about the concurrent use of multiple low risk anticholinergic medications. It is imperative that anticholinergic prescribing, and particularly psychotropic prescribing, should be considered alongside the known health risks, and subsequently reviewed for efficacy, tolerability, and safety.

Supplementary Information

The online version contains supplementary material available at https://doi.org/10.1186/s12991-022-00418-x.

Additional file 1: Table S1. Odds ratios for comparison of adults with intellectual disabilities and the general population adults for psychotropics with a significant interaction between group and age. **Table S2.** Odds ratios for comparison of adults with intellectual disabilities and the general population adults for antiepileptics by neighbourhood deprivation (SIMD quintile). **Figure S1.** Influence of neighbourhood deprivation (SIMD quintile) on groups' MERs for predicted probability of being an antiepileptics.

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Author contributions

LW, SA-C, AH and DC contributed to the concept and design of the work. LW led the analysis with the support of BS and NG. All authors contributed to the interpretation of data. LW led the original draft writing with the help of S-AC, CP, AH and DC. All authors revised, edited, and approved the manuscript for publication for important intellectual content. All authors gave the final approval of the version to be published. All authors read and approved the final manuscript.

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Availability of data and materials

All data generated or analysed during the study are included in this published article (and its additional files).

Declarations

Ethics approval and consent to participate

The research was approved by Scotland's Public Benefit and Privacy Panel for Health and Social Care (reference 1516-0281). As this was a data linkage project using electronic health records data, there was no consent to participate required. Data were linked and shared with the research team within Scotland's local Safe Haven following the necessary legal and governance approvals.

Consent for publication

Not applicable.

Competing interests
The authors declare that they have no competing interests.

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Rates and causes of mortality among children and young people with and without intellectual disabilities in Scotland: a record linkage cohort study of 796 190 school children

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ABSTRACT

Objectives To investigate mortality rates and causes in children and young people with intellectual disabilities.

Design Retrospective cohort; individual record linkage between Scotland's annual pupil census and National Records of Scotland death register.

Setting General community.

Participants Pupils receiving local authority-funded schooling in Scotland, 2008 to 2013, with an Additional Support Need due to intellectual disabilities, compared with other pupils.

Main outcome measures Deaths up to 2015: age of death, age-standardised mortality ratios (age-SMRs); causes of death including cause-specific age-SMRs; avoidable deaths as defined by the UK Office of National Statistics.

Results 18 278/947 922 (1.9%) pupils had intellectual disabilities. 106 died over 67 342 person-years (crude mortality rate=157/100 000 person-years), compared with 458 controls over 3 672 224 person-years (crude mortality rate=12/100 000 person-years). Age-SMR was 11.6 (95% CI 9.6 to 14.0); 16.6 (95% CI 12.2 to 22.6) for female pupils and 9.8 (95% CI 7.7 to 12.5) for male pupils. Most common main underlying causes were diseases of the nervous system, followed by congenital anomalies; most common all-contributing causes were diseases of the nervous system, followed by respiratory system; most common specific contributing causes were cerebral palsy, pneumonia, respiratory failure and epilepsy. For all contributing causes, SMR was 98.8 (95% CI 69.9 to 139.7) for congenital anomalies, 76.5 (95% CI 58.9 to 99.4) for nervous system, 63.7 (95% CI 37.0 to 109.7) for digestive system, 55.3 (95% CI 42.5 to 72.1) for respiratory system, 32.1 (95% CI 17.8 to 57.9) for endocrine and 14.8 (95% CI 8.9 to 24.5) for circulatory system. External causes accounted for 46% of control deaths, but the SMR for external-related deaths was still higher (3.6 (95% CI 2.2 to 5.8)) for pupils with intellectual disabilities. Deaths amenable to good care were common.

Conclusion Pupils with intellectual disabilities were much more likely to die than their peers, and had a different pattern of causes, including amenable deaths across a wide range of disease categories. Improvements are needed to reduce amenable deaths, for example, epilepsy-related and dysphagia, and to support families of children with life-limiting conditions.

Strengths and limitations of this study

- Novel use of education records and record linkage to death records to study mortality in an unselected cohort of children and young people with intellectual disabilities.
- Due to the use of a whole country population, these results are well-powered and generalisable.
- Despite comprising a whole country population, our study was not large enough to delineate cause-specific mortality ratios by sex.
- This study was limited by lack of demographic and clinical diagnostic information, including the severity or cause of intellectual disabilities.
- Reliance on death certificate data is limited by inconsistencies in reporting of cause of death

INTRODUCTION

Children and young people with intellectual disabilities have a much higher prevalence of physical and mental ill-health compared with the general population.[1–3] The life expectancy of people with intellectual disabilities has been reported to be about 20 years shorter than in the general population, or 28 years shorter specifically for people with Down syndrome.[4–7] While the actual number of deaths in childhood is smaller than in adults, mortality studies comparing people with intellectual disabilities with the general population have tended to show increased risk ratios in younger age groups compared with adults. However, the reported excess risk varies considerably between studies, and not all studies are comparable due to, for example, reporting deaths within different

age ranges, and additionally, some have small sample sizes and wide CIs. Reported standardised mortality ratios (SMRs) comparing people with and without intellectual disabilities, have ranged from 3.3 (95% CI 2.1 to 5.0) in young people aged 10 to 19 years[8] to 17.3 (95% CI 9.4 to 29.0) in young people aged 10 to 17 years[9]; from 2.6 in males aged 2 to 19 years and 1.7 in females aged 2 to 19 years,[10] to 21.6 (95% CI 10.8 to 38.7) in males aged 0 to 19 years and 18.1 (95% CI 3.7 to 53.0) in females aged 0 to 19 years;[11] and have been reported to be 30.4 (95% CI 18.4 to 47.5) in children aged 0 to 9 years.[9] We have summarised all previous studies to our knowledge which report mortality ratios for children and young people under age 25 years, with and without intellectual disabilities, where they are reported separately from older age groups (online supplementary appendix 1).

Most of these studies do not report causes of death among children and young people with intellectual disabilities. Bourke *et al*[12] reported the most common causes of death in children, young people and adults with intellectual disabilities aged 1 to 25 years to be respiratory infection (34%), with an additional 10% having an aspiration-related cause, congenital heart defects (15%) and accidents (11%). Compared with children and young people who did not have intellectual disabilities, their causes of death by International Classification of Diseases, tenth revision, (ICD-10) chapter were more likely to be attributed to the nervous system, endocrine, nutritional and metabolic diseases, or congenital malformations, and less likely to be attributed to conditions originating in the perinatal period, external causes, or injury or poisoning.[12] Patja *et al*[10] reported respiratory diseases to be the most common underlying/immediate cause of death in children and young people with intellectual disabilities aged 2 to 19 years, with a relative risk of 5.8 (95% CI 4.4 to 15.6) in males and 4.3 (95% CI 0.3 to 4.7) in females, and did not find any other causes (infectious diseases, tumours, vascular diseases, diseases of digestive system, accidents and poisonings, or other causes) to differ from those expected in the general population. However, the study was limited by small sample size. Durvasula *et al*[13] reported 7 of 14 deaths among young people with intellectual disabilities aged 10 to 24 years were attributed to the respiratory system (pneumonia and aspiration).

Adults with intellectual disabilities are over-represented in deaths which would have been amenable to treatment by timely and effective healthcare.[4 5 9] However, there is limited evidence on whether children and young people with intellectual disabilities also experience such amenable deaths more commonly than other children and young people, as most authors who have reported cause-specific mortality did so by grouping across all ages due to sample sizes.

Overall, as shown in online supplementary appendix 1, studies on mortality in children with intellectual disabilities are mostly small in size, and results are variable. Studies of causes of death exclusively in children and young people with intellectual disabilities are also limited. Hence, the aim of this cohort study is to compare all-cause and cause-specific mortality in Scotland's school attending population with and without intellectual disabilities.

METHODS

We used education data from Scotland's annual pupil census between 2008 and 2013 to establish a cohort of children and young people with and without intellectual disabilities. We used individual record linkage based on probabilistic record matching (on date of birth, sex and postcode) to the Community Health Index, Scotland's list of all unique patient identifiers, including the National Records of Scotland (NRS) deaths register, to ascertain all deaths up to February 2015 in Scotland.

The Scottish annual pupil census is completed in September each year and provides information on all children attending local authority-funded primary, secondary and special schools in Scotland, or funded placements in alternative schools, which includes 95% of the entire population of children and young people in Scotland. This information includes whether the child has a record of Additional Support Needs, and the type of Additional Support Need. It is held by the Scottish Exchange of Educational Data (ScotXed).

The record linkage methodology required date of birth, sex and postcode; however, since names were not used to link pupil records to the health data, we excluded non-singleton births (available for Scottish-born pupils only, identified from linkage to maternity records). Unlikely matches were excluded and the most likely match was selected as the correctly linked pupil record. We also excluded any records with duplicate pupil records or where the linkage was tied with another patient. We included in the study all pupils with records of Additional Support Need due to intellectual disabilities between 2008 and 2013, between the ages 4 and 19 years old, on entry. Pupils were also censored on reaching age 25, if they reached this age during the observation period; so the maximum follow-up age was 24 years. Only pupils with intellectual disabilities recorded in at least two different school years were included in the intellectual disabilities group, to ascertain that they were correctly identified. Pupils who were included in at least two pupil census over the study period and had no record of intellectual disabilities or autism were used as the comparison group. Pupils with only autism were also excluded from controls, to eliminate potential mislabelling of support need for either autism or learning disability in the absence of clinical diagnoses.

The pupil census also includes data on age, sex, ethnicity and Scottish Index of Multiple Deprivation 2012 (SIMD).[14] Derived from individual pupil postcode of residence, SIMD is a composite of seven indices to indicate the extent of neighbourhood deprivation. SIMD was divided into quintiles according to the general population. Data on disability requirements including physical

(eg, visual, hearing or physical impairments), communication or curriculum needs are also listed.

Non-modifiable descriptive data on sex, ethnicity and SIMD, were taken from each pupils' first year in the census. For disability requirements, all records across multiple pupil census years were used to define whether having ever received adaptation requirements. Explorative statistical analyses using t-tests and χ^2 tests were employed to investigate characteristics of pupils with intellectual disabilities compared with their peers in the comparison group. Differences in age of death were explored using t-tests. Crude mortality rates were calculated using the censor date 13 February 2015 or date of death. Since only those pupils who attended school in at least 2 years over our observed study period were eligible, the period between the first and second record introduced an immortal time bias, where no deaths could have occurred, and therefore the entry to the study was defined as the date of their second pupil census record. For indirect standardisation, observed deaths were assumed to be independent and vary with the Poisson distribution. The mortality rates were indirectly standardised for both males and females using the expected age-specific mortality rates per 1-year age group, using Stata's 'strate' command, to calculate age-SMRs for pupils with versus without intellectual disabilities. The 95% CIs were calculated based on the quadratic approximation of the log likelihood. Expected rates were calculated using fixed age and sex-specific rates from the large control population. The SMRs were subsequently calculated stratified by age into childhood (aged 5 to 14 years) and young people (aged ≥15 years), and by sex. The SMRs were also calculated for all deaths, excluding for external causes. This was to investigate whether the over-representation of female deaths in people with intellectual disabilities compared with the general population[12 15 16] is related to the large proportion of male deaths from external causes in the general population.[17]

For all-cause mortality, Kaplan-Meier survival curves were plotted for the overall time period for both groups. Cox proportional hazards models are also presented, adjusted for age and sex.

For cause of death analyses, the underlying cause of death is defined internationally[18] as the disease or injury which initiated the chain of morbid events leading directly to death, or the accident/act which produced the fatal injury. We also used a broader definition to analyse all-contributing causes, that included all deaths, with any mention on the death certificate related to the cause; combining both the underlying cause with secondary or contributing factors. While the same ICD-10 codes are used, it is important to note that one death may have several other additional causes as contributing factors, all of which are counted in figures reporting 'all-contributing causes'.

For the underlying causes of death, the total number of deaths in each ICD-10 chapter were collated, and this was then repeated for specific causes listed within chapters. Any errors or ambiguous deaths were listed as an unknown cause. All deaths where the underlying cause was ill-defined or defined by ICD-10 WHO guidelines[18] as codes in Chapter 18 excluding R95, were also re-classified as 'unknown'. Next, the breakdown of all-contributing causes were analysed by collating number of deaths in each ICD-10 chapter. For cause-specific SMRs, indirect age-standardisation was also performed, but using 5-year age bands to age-standardise rates and robust standard errors were used. For categories which had fewer than 10 deaths, no calculation was attempted due to lack of reliability in the small number of deaths. Furthermore, in keeping with the Office of National Statistics (ONS) mortality methodology,[19] all mortality rates between 10 and 20 deaths were labelled as unreliable. The ONS revised definition of avoidable mortality for children and young people[20] defined avoidable mortality as either amenable mortality (avoidable through good quality healthcare even after a condition has developed) or preventable mortality (avoidable through incidence reduction via public health interventions) or both. This list of ICD-10 causes was used to determine the occurrence of avoidable deaths. The rates and age-SMRs (age-standardised using 5-year age bands) for avoidable, amenable and preventable mortality were calculated using robust errors, except where there were fewer than 10 deaths per chapter. In keeping with the ONS avoidable mortality methodology,[19] all mortality rates based on fewer than 20 deaths were labelled as unreliable.

Sensitivity analysis

A sensitivity analysis was carried out using wider inclusion criteria from the education data for both groups; the intellectual disabilities group included all pupils with at least one record of support at school due to intellectual disabilities. The control group included all pupils with at least one census record, and without support records for intellectual disabilities or autism. There were no other methodological changes made to the age standardising process or censor dates, but entry date was changed to the date of the first record of support need for pupils with intellectual disabilities or the first census date for pupils without intellectual disabilities.

All statistical analyses were undertaken using Stata, V.15.0 (StataCorp).

Personal and patient involvement

This study was undertaken in the Scottish Learning Disabilities Observatory due to the growing concern among people with intellectual disabilities and their families around mortality. Its steering group includes people with intellectual disabilities and partners from third sector organisations. Results from this study will be disseminated to people with intellectual disabilities and their families in an easy-read version via the Scottish Learning Disabilities Observatory website and newsletters.

Table 1 Demographic information for pupils with and without intellectual disabilities

Demographic information*	Intellectual disabilities		Controls		P value†
Total, n (person-years)	18278	(67 342)	777912	(3 672 224)	
Male sex, n (%)	11891	(65%)	389160	(50%)	p<0.001
Age, person-years (%)					
<10	12518	(19%)	995297	(27%)	
10 to 14	28297	(42%)	1332123	(36%)	
15 to 19	23672	(35%)	1178608	(32%)	
19 to 24	2855	(4%)	166196	(5%)	
Disability adaptations, n (%)					
Physical adaptation, ever received	1971	(11%)	1837	(0.2%)	p<0.001
Curriculum adaptation, ever received	6623	(36%)	6341	(0.8%)	p<0.001
Communication adaptation, ever received	3553	(19%)	1760	(0.2%)	p<0.001
SIMD quintile, n (%) at first census					
1 (most deprived)	5822	(32%)	169038	(22%)	
2	3888	(21%)	149290	(19%)	
3	3397	(19%)	152415	(20%)	
4	2896	(16%)	158228	(20%)	
5 (least deprived)	2275	(12%)	148941	(19%)	p<0.001
Ethnicity, n (%)					
White‡	16553	(91%)	708941	(91%)	p<0.001
Asian‡	514	(3%)	23791	(3%)	
Mixed or multiple ethnicities	144	(1%)	8035	(1%)	
African, Caribbean or black	87	(<1%)	4710	(<1%)	
Other ethnic groups	92	(<1%)	4665	(<1%)	
Not disclosed / or unknown	888	(5%)	27770	(4%)	

*Data taken from first census record, except for disability adaptation, which includes any record across census years.
†χ^2 test for intellectual disabilities compared with control group (For SIMD, χ^2 test was performed across all categories, overall p value).
‡(White: Scottish, British and other) (Asian: Indian/British/Scottish, Pakistani/British/Scottish, Bangladeshi/British/Scottish, and Chinese/British/Scottish).
SIMD, Scottish Index of Multiple Deprivation.

RESULTS

Out of 947922 pupils in the census between 2008 and 2013 who were successfully linked to health records, there were 27140 pupils who had ever registered as having an Additional Support Needs due to intellectual disabilities, and of these, 18278 (1.9% of pupils) met the criteria of having at least two records of support. The remaining 8862 pupils with a single support record were excluded, except for the sensitivity analysis. There were 909688 pupils without any records of intellectual disabilities or autism. Of these, 131776 were excluded due to appearing in only 1 year of the census, except for the sensitivity analysis. The remaining 777912 pupils attended school for at least 2 years over the study period and were designated as controls.

Using data from the pupils' first year in the census, pupils with intellectual disabilities were more likely to be male, more likely to reside in areas of greater neighbourhood deprivation and to have registered for free school meals, compared with their peers (table 1). Pupils with intellectual disabilities were also more likely to require adaptations in school, including physical adaptations, communication and curriculum adaptations. The majority of the study population were identified as having white (Scottish, British or other) ethnicity.

Missing education support records

There were 11329 pupils (62%) of the intellectual disabilities group who appeared in certain census years without having a record of support. The majority, 70%, (n=7970) were before the accrual of the first record; these pupils had a median two pupil census records prior to receiving their support (IQR 1, 3). There were 3359 pupils or 18% of the entire study group who went on to have census records without support records, after having received intellectual disabilities support provision. These pupils had a median 1 subsequent year (IQR 1, 2) without support out of a median 4 remaining years (IQR 3, 6) in the census.

Figure 1 Forest plot of age-standardised and sex-specific mortality ratios for pupils with intellectual disabilities.

Mortality analysis

Linking the pupil census population to the NRS register of deaths up to February 2015 resulted in the equivalent of 3 739 568 person-years of follow-up. There were 564 deaths identified in the study population during this period. There were 106 deaths (0.6%) among children and young people with intellectual disabilities over 67 342 person-years, which translated to a crude mortality rate of 157 deaths per 100 000 person-years (95% CI 130 to 190). In the control group, there were 458 deaths (<0.1%) over 3 672 224 person-years, which translated to a crude mortality rate of 12 deaths per 100 000 person-years (95% CI 11 to 14). The mean age of death among children and young people with intellectual disabilities was 14.3 years (95% CI 13.4 to 15.1), which was significantly lower (p<0.001) than controls where the mean age of death was 16.1 years (95% CI 15.8 to 16.5). Sixty-two per cent of deaths among children with intellectual disabilities occurred in males, which was equivalent to the sex distribution in the whole intellectual disabilities cohort (p=0.545). Among controls, 61% of deaths occurred in males in spite of them accounting for only 50% of this group (p<0.001). Over 50% of deaths among pupils with intellectual disabilities occurred during childhood (<15 years old), compared with 29% of deaths among controls.

The all-cause age-SMR was 11.6 (95% CI 9.6 to 14.0), as shown in figure 1. The SMR was higher for female pupils than male pupils with intellectual disabilities; female SMR of 16.6 (95% CI 12.2 to 22.6) versus male SMR of 9.8 (95% CI 7.7 to 12.5). Exclusion of external causes of death resulted in a considerable increase in the all-cause SMR for both females and males with intellectual disabilities; overall SMR was 21.6 (95% CI 17.8 to 26.3), female SMR of 25.6 (95% CI 18.8 to 34.9) versus male SMR of 19.6 (95% CI 15.3 to 25.2). This produced a relative increase of 10 more deaths overall for pupils with versus without intellectual disabilities, which was similar in females (+9.0 increase) and males (+9.8 increase). The childhood (aged 5 to 14 years) SMR was 21.6 (95% CI 16.6 to 28.2) and was higher for females than males with intellectual disabilities; female SMR of 30.3 (95% CI 19.8 to 46.5) versus male SMR of 18.4 (95% CI 13.1 to 25.7). For young people (≥15 years old), SMR was 7.7 (95% CI 5.9 to 10.2) and was also higher for females than males with intellectual disabilities; female SMR of 11.1 (95% CI 7.1 to 17.4) versus male SMR of 6.5 (95% CI 4.6 to 9.3). Hence, the difference from the control pupils was greater in children rather than young people for both females and males.

The Cox proportional HR for all-cause mortality, adjusted for age and sex, was found to be very similar; HR: 11.97 (95% CI 9.64 to 14.86). Proportional hazards assumption was met (p=0.422). Kaplan-Meier survival curves for the overall time period are found in online supplementary data (online supplementary appendix 2).

Cause of death

Cause of death data was available for over 95% of deaths among pupils with intellectual disabilities and over 91% deaths among controls. Table 2 shows the underlying causes of death and all-contributing causes of death by ICD-10 chapter. There were major differences between pupils with intellectual disabilities and controls with regard to the most common underlying causes. Among pupils with intellectual disabilities, these were diseases of the nervous system (33%), congenital malformations, deformations and chromosomal abnormalities (22%), followed by nutritional, metabolic and endocrine diseases (8%), of which, most were conditions which were the cause of the pupils' intellectual disabilities, for example, neuronal ceroid lipofuscinosis or ornithine metabolism disorders. These were followed by respiratory diseases (8%) and neoplasms (7%). The most common underlying cause of death among control pupils was death due to external causes (46%), which made up a higher proportion of all deaths than in the pupils with intellectual disabilities (5%). Among controls, 71% of deaths due to external causes occurred in boys compared with 100% in the intellectual disabilities group.

There were also differences in the most common all-contributing causes of death (table 2). These chapters were not mutually exclusive, since one death could be included in several categories. Of the 106 deaths among pupils with intellectual disabilities, diseases of the nervous system contributed to 56 and diseases of the respiratory system contributed to 55. The 56 deaths which included diseases of the nervous system included 34 due to cerebral palsy and 16 due to epilepsy. The 55 deaths which included diseases of the respiratory system included 27 due to pneumonia, 9 due to pneumonitis associated with food and gastric contents, 17 due to respiratory failure, and 15 other respiratory disorders. In comparison, the control pupils had diseases of the nervous system contributing to 39 out of the total 458 deaths, and diseases of the respiratory system contributing to 51 of 458 deaths which

Table 2 By ICD-10 chapter, the underlying causes of death as well as all-contributing factors in death, and cause-specific crude mortality rates per 100 000 person-years for pupils with and without intellectual disabilities

	Underlying cause of death						All-contributing factors in death					
	Intellectual disabilities			Controls			Intellectual disabilities			Controls		
ICD-10 chapter*	n (%)	CMR	95% CI	n (%)	CMR	95% CI	n	CMR	95% CI	n	CMR	95% CI
Ch. 6. Diseases of the nervous system	**35** (33%)	51.9	37.3 to 72.4	**19** (4%)	0.5[U]	0.3 to 0.8	**56**	83.2	64.0 to 108.1	**39**	1.1	0.8 to 1.5
Ch. 17. Congenital malformations, deformations and chromosomal abnormalities	**23** (22%)	34.2	22.7 to 51.4	**13** (3%)	0.4[U]	0.2 to 0.6	**32**	47.5	33.6 to 67.2	**18**	0.5[U]	0.3 to 0.8
Ch. 4. Endocrine, nutritional and metabolic diseases	**9** (8%)			**15** (3%)	0.4[U]	0.2 to 0.7	**11**	16.3[U]	9.0 to 29.5	**18**	0.5[U]	0.3 to 0.8
Ch. 10. Diseases of the respiratory system	**8** (8%)			**17** (4%)	0.5[U]	0.3 to 0.7	**55**	81.7	62.7 to 106.4	**51**	1.4	1.1 to 1.8
Ch. 2. Neoplasms	**7** (7%)			**92** (20%)	2.5	2.0 to 3.1	**8**	NA		**94**	2.6	2.1 to 3.1
Ch. 20. External causes of morbidity and mortality	**5** (5%)			**210** (46%)	5.7	5.0 to 6.6	**16**	23.8[U]	14.6 to 38.8	**231**	6.3	5.5 to 7.2
Ch. 9. Diseases of the circulatory system	**<5** (5%)			**24** (5%)	0.7	0.4 to 1.0	**15**	22.3[U]	13.4 to 37.0	**54**	1.5	1.1 to 1.9
Ch. 11. Diseases of the digestive system	**<5** (5%)			**6** (1%)			**13**	19.3[U]	11.2 to 33.3	**11**	0.3[U]	0.2 to 0.5
Ch. 1. Certain infectious and parasite diseases	**<5** (5%)			**12** (3%)	0.3	0.2 to 0.6	**8**			**29**	0.8	0.5 to 1.1
Ch. 5. Mental and behavioural disorders	**<5** (5%)			**<5** (1%)			**6**			**17**	0.5[U]	0.3 to 0.7
Ch. 3. Diseases of the blood, blood-forming organs and immune mechanism	**<5** (5%)			**<5** (1%)			**<5**			**8**		
Ch.14. Diseases of the genitourinary system	**<5** (5%)			**<5** (1%)			**<5**			**<5**		
Ch. 13. Diseases of the musculoskeletal system and connective tissue	**0**			**<5** (1%)			**<5**			**7**		
Ch. 8. Diseases of the ear and mastoid process	**<5** (5%)			**0**			**<5**			**0**		
Ch. 15. Pregnancy, childbirth and puerperium	**0**			**<5** (1%)			**0**			**<5**		
Ch. 18. Symptoms, signs and abnormal clinical and laboratory findings	**NA**			**NA**			**28**			**76**		
Ch.19. Injury, poisoning and certain other consequences of external causes	**NA**			**NA**			**10**			**219**		
Unknown cause or error in underlying code	**5** (5%)			**39** (9%)			**NA**			**NA**		
TOTAL	**106**			**458**			**NA**			**NA**		

U rates based on 10 to 20 deaths labelled 'U' for unreliable.
*n <5 repressed due to statistical disclosure.
CMR, crude mortality rate - reported for ≥10 deaths; ICD-10, International Classification of Diseases, tenth revision.

Table 3 The top 10 specific underlying causes of death and all-contributing causes of death for pupils with and without intellectual disabilities

Intellectual disabilities				Controls			
Underlying cause of death	n	All-contributing factors	n	Underlying cause of death	n	All-contributing factors	n
Cerebral palsy	19	Cerebral palsy	34	All neoplasms	92	Signs and symptoms: injury	114
Brain deformity	9	Pneumonia	27	Traffic accident	76	All neoplasms	94
All neoplasms	7	Respiratory failure	17	Self-harm	54	Traffic accidents	76
Muscular dystrophy	6	Epilepsy	16	Accidents, other	41	Self-harm	54
Epilepsy	5	Respiratory disorders	15	External, undetermined intent	25	Signs and symptoms: asphyxiation	51
Chromosomal abnormalities	5	Brain deformity	12	Asthma	14	Accident, other	43
Neuronal ceroid lipofuscinosis	<5	Chromosomal abnormalities	10	Assault	13	Signs and symptoms: poisoning	29
Pneumonia, including influenza	<5	Pneumonitis due to food and gastric contents	9	Infections	12	All infections	29
Congenital heart disease	<5	All neoplasms	8	Epilepsy	8	External, undetermined intent	26
Accidents, other	<5	All infections	8	Cystic fibrosis	8	Pneumonia	21
Unknown causes	5	Ill-defined or ambiguous death	8	Unknown causes	39	Ill-defined or ambiguous death	58

included 21 due to pneumonia. The most common all-contributing causes of death for the control pupils were, as found for the underlying cause, external causes at 50% compared with 15% among pupils with intellectual disabilities.

Table 2 reports these data by presenting the cause-specific crude mortality rates by ICD-10 chapter for all pupils. As recommended by the ONS,[19] avoidable mortality rates based on low numbers are labelled as unreliable and marked 'U'.

The top 10 individual leading causes of death are shown in table 3. Among pupils with intellectual disabilities, the highest number of individual underlying cause of deaths were cerebral palsy (18%), followed by congenital brain deformities (8%) and neoplasms (7%). Where there were fewer than five individual deaths per cause, these causes were not reported due to statistical disclosure control. For the majority of deaths in pupils with intellectual disabilities, this was the case; 85% of specific causes could not be disclosed. Among control pupils, the highest number of individual underlying cause of deaths were neoplasms (20%), and road traffic accidents (17%). In relation to their peers, only three of the top 10 underlying causes of death among children with intellectual disabilities featured in the top 10 list for the controls—neoplasms (7% vs 20% of controls), epilepsy (5% vs 2% controls) and accidents (non-road traffic related,<5% vs 9% controls).

Cause-specific SMRs, indirectly standardised using 5-year age bands and robust errors, are shown in figure 2. For underlying causes, this was only possible for the two largest categories (by ICD-10 chapters); congenital malformations, deformations and chromosomal abnormalities, and diseases of the nervous system. For the all-contributing causes, the age-SMR for seven chapters were calculated. For congenital malformations, deformations and chromosomal abnormalities, the SMR was 98.8^{U} (95% CI 69.9 to 139.7), and for diseases of the nervous system was 76.5 (95% CI 58.9 to 99.4). The ratios were also high for diseases of the digestive system at 63.7^{U} (95% CI 37.0 to 109.7); and for diseases of the respiratory system at 55.3 (95% CI 42.5 to 72.1). Despite external causes contributing to a larger proportion of deaths among the control group, the mortality rate was still higher in the intellectual disabilities group than in the controls; the crude rate was 23.8^{U} per 100 000 person-years, compared with 6.3^{U} per 100 000 for the controls for external cause of death (either as the underlying cause or as a contributing factor). This produced an SMR of 3.6^{U} (95% CI 2.2 to 5.8), demonstrating there is considerable over-representation in the intellectual disabilities group versus the controls.

Avoidable mortality

According to the UK ONS definition of avoidable mortality, (deaths which are amenable, preventable or both), 19% of deaths in the intellectual disabilities cohort were classed as avoidable; 15% of deaths were amenable to treatment and 6% were preventable. The majority of avoidable deaths (80%) were considered amenable to

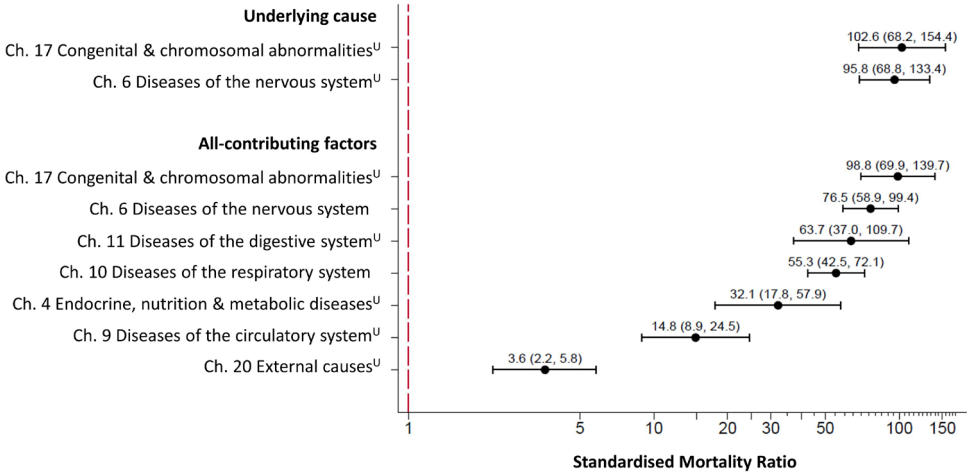

Figure 2 Forest plot of cause-specific age-SMRs for pupils with intellectual disabilities by ICD-10 chapter for underlying cause of death and for all-contributing factors of death. Footnote: Age-SMRs and 95% CI were calculated using 5-year age bands for all ICD-10 chapters with ≥10 deaths. SMRs which were calculated using low numbers (between 10 and 20 deaths) are labelled 'U' for unreliable. ICD-10, International Classification of Diseases, tenth revision; SMR, standardised mortality ratio.

treatment for their age group, including epilepsy, pneumonia and neoplasms. Among the control pupils, 63% of deaths were classed as avoidable, 16% were amenable to treatment and 48% were preventable. The crude avoidable mortality rate for pupils with intellectual disabilities was higher at 29.7[U] (95% CI 19.2 to 46.0) per 100 000 in pupils with intellectual disabilities, compared with 7.8 (95% CI 7.0 to 8.8) per 100 000 in the control pupils. The SMR was 3.6[U] (95% CI 2.3 to 5.5). Further breakdown of avoidable rates was possible for deaths that were amenable to healthcare; in the intellectual disabilities group, the amenable mortality rate was 23.8[U] (95% CI 14.6 to 38.8) per 100 000 versus 2.0[U] (95% CI 1.6 to 2.5) per 100 000 in controls; and the SMR was found to be 11.5[U] (95% CI 7.0 to 18.8).

Among pupils with intellectual disabilities, there were additional causes of death that the authors of this paper consider would have been amenable to healthcare: aspiration pneumonia; otitis media; megacolon; gastrointestinal haemorrhage; gastroenteritis; and contributing causes of death including gastro-oesophageal reflux and urinary tract infections. These are not currently included within the ONS list of underlying causes.

Sensitivity analysis

Of 27 140 pupils with at least one record of support due to intellectual disabilities, 65% were male, and compared with the main analysis group, there were significant reductions in frequency of school adaptations (physical disability reduced from 11% vs 9%, (p<0.001), curriculum adaptations from 36% to 31% (p<0.001) and communication adaptations from 19% to 16% (p<0.001)). There were higher numbers of pupils in this group with years without intellectual disabilities support. There were 156 deaths in the intellectual disabilities group (134 per 100 000 person-years (95% CI 114.2 to 156.3)) compared with 684 deaths (13.8 per 100,000 (95% CI 12.8 to 14.8)) among the control group. The SMR for this sensitivity analysis was 9.5 (95% CI 8.1 to 11.1), a change of −2 excessive deaths compared with the main analysis SMR. Mean age of death was similar in the sensitivity group, being 14.4 years (95% CI 13.7 to 15.1) in the intellectual disabilities group and 16.2 years (95% CI 15.9 to 16.5) in the control group. The ratio of deaths by sex were also very similar, with no difference for the intellectual disabilities group; 61% deaths were in males, similar to the proportion of males in the group (p=0.306) and an increase in male deaths among controls; 63% deaths were in males, whereas only 50% in the control group were male (p<0.001).

DISCUSSION

Principle findings and interpretation

Our study is one of very few that has reported mortality rates among children and young people with intellectual disabilities, and is highly novel in reporting underlying, all-contributing and the most common individual causes of death at this age, including cause-specific SMRs. We have demonstrated that children and young people in Scotland with intellectual disabilities have a 12-fold risk of death compared with their peers, rising to 22-fold on excluding external causes. Pupils with intellectual disabilities were also over-represented in deaths that were amenable to healthcare, and were approximately 3.6 times more likely to experience an avoidable death (although calculated using unreliably low rates). Children aged 5 to 14 years with intellectual disabilities had a higher risk relative to peers (SMR 21.6) than the young people aged ≥15 years with intellectual disabilities (SMR 7.7). This difference reflects that, in the general population, there were considerably more deaths in young people than in children, especially for males, as opposed to more deaths of children than young people with intellectual disabilities. The SMR was higher for female pupils in both age groups,

reflecting the higher death rate of males in the controls. Nervous system and respiratory causes of death were most common among children and young people with intellectual disabilities, including deaths that would have been amenable to quality healthcare, such as epilepsy, pneumonia and pneumonitis due to food and gastric contents. It is highly important to identify amenable deaths so that actions can be devised and taken. Causes of death among children and young people with intellectual disabilities were higher across several disease categories than for other children and young people, including diseases of the nervous system, digestive system, respiratory system, endocrine, nutrition and metabolic diseases, diseases of the circulatory system and external causes.

Previous studies have demonstrated that there is an increased risk of sudden unexpected death in epilepsy among people with intellectual disabilities; however, in our study, the majority of deaths which listed epilepsy as a contributing factor, also listed pneumonia, so this does not appear to account for our findings.

While external causes of deaths accounted for the greatest proportion of deaths among control children and young people (46%), especially in males, we found that external causes of death were still over-represented among children and young people with intellectual disabilities compared with their controls (partly due to inhalation of gastric contents and inhalation of objects obstructing breathing). Trollor *et al*[17] hypothesised that higher SMRs in adult women than men with intellectual disabilities may be driven by the larger proportion of male deaths in the general population due to external causes and the lack of equivalent deaths in males with intellectual disabilities. However, in our population of children and young people, when we re-calculated SMRs to exclude external causes, the observed increase in risk for females remained. Hence, at this age range, this is only a partial explanation for the sex differences in SMRs, and there are other risk factors and vulnerabilities which require further exploration. It should be noted, however, that in children and young people with intellectual disabilities, not all studies report a higher SMR in females compared with males.[10 11]

Comparison with previous studies

Two previous studies[9 12] have reported a higher SMR for children than for young people. Glover *et al*[9] reported results separately for children aged 0 to 9 years (SMR 30.4) and young people aged 10 to 17 years with intellectual disabilities (SMR 17.3). The Australian study by Bourke *et al*[12] reported a higher adjusted HR (aHR) for children aged 6 to 10 years (aHR 12.6) than young people aged 11 to 25 years (aHR 4.9). The SMRs we report are lower than those reported by Glover *et al*,[9] but the extent of difference between the children and young people is similar, although for differently defined age groups. The CIs reported in our study are narrower due to the larger sample size. The SMRs we report are higher than those previously reported from small scale studies in Finland and USA,[8 10 15] and a larger one in Ireland,[16] yet lower than a study reported from England[9] and a small study from Canada.[11] These differences may be due to actual international differences or due to methodological differences between studies including: the method and source of identification of the population with intellectual disabilities; age ranges included; and study size with several of the previous studies having produced results with wide CIs. All of these studies report a higher SMR in females than in males, except the studies conducted in Canada and Finland.

The only previous study that has reported cause of death for children and young people aged 1 to 25 years reported the most common causes of death to be infections in 50% (particularly respiratory infections in 34%), birth defects in 19% (particularly cardiac defect in 15%) and accidents in 11%, although by ICD-10 chapter, deaths due to diseases of the respiratory tract were reported for 4.6%, infections and parasitic diseases for 3.1% and external causes for 7.7%; and the most common were congenital malformations, deformations and chromosomal abnormalities in 29.1%, and diseases of the nervous system in 27.6%.[12] They did not report cause-specific SMRs by ICD-10 chapters, but crude numbers were proportionally higher for children with intellectual disabilities for diseases of the nervous system, endocrine, nutritional and metabolic diseases, and congenital malformations, and lower for conditions originating in the perinatal period, external causes, or injury or poisoning.[12] We demonstrated diseases of the nervous system and respiratory system to be the most common causes of death, and that cause-specific SMRs were higher across all congenital malformations, deformations and chromosomal abnormalities, diseases of the nervous system, digestive system, respiratory system, endocrine, nutrition and metabolic diseases, circulatory system and external causes.

Glover *et al* graphed avoidable deaths in his study of children and adults.[9] We are unaware of any previous studies numerically quantifying amenable deaths among children and young people with intellectual disabilities.

Strengths and limitations

Our study drew on data from an entire country, collected annually, and linked to national death records. It was large in scale, including over 18 000 children and young people with intellectual disabilities and a large control population. A record of intellectual disabilities at school brings an entitlement to additional support and so is likely to drive good recording in high-income countries like Scotland. However, it only uses a binary definition for intellectual disabilities; therefore, the study could not investigate mortality among people with different causes and severities of intellectual disabilities. Our study was not large enough to delineate cause-specific mortality ratios by sex nor to study whether there are any ethnic variations. Use of death certificate data is known to have limitations,[19] including inconsistent reporting and no reporting of severity of conditions. There may be some

diagnostic overshadowing in death certificate data for people with intellectual disabilities, obscuring the events leading to death.[21–23] The ONS list of avoidable deaths does not include some that appear important among children and young people with intellectual disabilities, such as aspiration pneumonia, otitis media, megacolon, gastrointestinal haemorrhage and gastroenteritis, which featured as an underlying cause of death in our data. Additionally, death certificate data does not include wider determinants of health and death that may be implicated, such as being the target of discrimination or neglect.

Additionally, while we believe this population to be highly representative of children with intellectual disabilities across Scotland, we acknowledge that we were unable to access data on children not in school; there may be some under-ascertainment of children with intellectual disabilities with exceptional and complex health needs unable to attend school.

Conclusion and future directions

It is extremely important to study deaths among children and young people with intellectual disabilities, especially as so few studies have previously done so. Among the studies that have, there exists wide variation in the extent of reported inequality compared with other children and young people, and wide CIs, but all show a higher SMR. Our large study provides robust data that quantifies the extent of the difference; children and young people have a 12 times higher risk of death. A larger body of research exists for adults (rather than children and young people) with intellectual disabilities, and demonstrates substantial inequalities and a high proportion of amenable deaths that could be addressed via reasonable adjustments in care provision. In our study, we have now reported that children and young people with intellectual disabilities also experience inequalities and experience amenable deaths. This is important, and we need a better understanding of it so that targeted improvements in care can be developed and delivered to reduce this inequality. Heslop *et al*[24] conducted a confidential inquiry into deaths of people with intellectual disabilities and made recommendations for improvements to practice regarding respiratory deaths, including aggressive monitoring and treatment of gastro-oesophageal reflux as well as postural and physical therapies. We have found that this is also important for children and young people with intellectual disabilities, if we are serious about improving life expectancy. Additionally, Scotland now offers influenza vaccines to all primary school-age children to reduce pneumonia; we therefore need to understand uptake by children with intellectual disabilities, and its determinants, to gauge whether this will change mortality findings.

The results of this study should be used to inform and direct multidisciplinary healthcare teams, as well as educators and carers to the associated risks of mortality in childhood and generate greater awareness around potential areas of improvement. Our countrywide study had a mean follow-up of around 5 years, and given that the pupil census is recorded annually, it presents the framework for further work to investigate both mortality trends in children and young people with intellectual disabilities, and a more detailed understanding of these. Future studies could consider looking at predictors of death in children and young people to inform translation of findings into clinical benefit for people with intellectual disabilities.

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Contributors GSS analysed the data, interpreted findings and wrote the first draft of the manuscript. MF developed record linkage, analysed the data, interpreted findings and contributed to the manuscript. JPP developed record linkage, interpreted findings and contributed to the manuscript. DK, AH and CM interpreted data and contributed to the manuscript. S-AC conceived the study, analysed and interpreted the data and contributed to the manuscript. All authors approved the final version of the manuscript.

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Competing interests None declared.

Patient and public involvement Patients and/or the public were involved in the design, or conduct, or reporting, or dissemination plans of this research. Refer to the Methods section for further details.

Patient consent for publication Not required.

Ethics approval This study received approval from the NHS National Services Scotland Privacy Advisory Committee and Public Benefit and Privacy Panel -(PBPP) approval no. 1617-0259.

Provenance and peer review Not commissioned; externally peer reviewed.

Data availability statement No data are available. This study linked patient information held across several administrative health datasets within Information Services Division (ISD) of NHS National Services Scotland (NSS), with externally held data held by the Scottish Government (ScotXed education) and National Records of Scotland. Linkage and de-identification of data was performed by ISD. A data processing agreement between NHS NSS and University of Glasgow and a data-sharing agreement between ScotXed and University of Glasgow were 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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Rates, causes, place and predictors of mortality in adults with intellectual disabilities with and without Down syndrome: cohort study with record linkage

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ABSTRACT

Objectives To investigate mortality in adults with intellectual disabilities: rates, causes, place, demographic and clinical predictors.

Design Cohort study with record linkage to death data.

Setting General community.

Participants 961/1023 (94%) adults (16–83 years; mean=44.1 years; 54.6% male) with intellectual disabilities, clinically examined in 2001–2004; subsequently record-linked to their National Health Service number, allowing linkage to death certificate data, 2018.

Outcome measures Standardised mortality ratios (SMRs), underlying and all contributing causes of death, avoidable deaths, place, and demographic and clinical predictors of death.

Results 294/961 (30.6%) had died; 64/179 (35.8%) with Down syndrome, 230/783 (29.4%) without Down syndrome. SMR overall=2.24 (1.98, 2.49); Down syndrome adults=5.28 (3.98, 6.57), adults without Down syndrome=1.93 (1.68, 2.18); male=1.69 (1.42, 1.95), female=3.48 (2.90, 4.06). SMRs decreased as age increased. More severe intellectual disabilities increased SMR, but ability was not retained in the multivariable model. SMRs were higher for most International Statistical Classification of Diseases and Related Health Problems, 10th Revision chapters. For adults without Down syndrome, aspiration/reflux/choking and respiratory infection were the the most common underlying causes of mortality; for Down syndrome adults 'Down syndrome', and dementia were most common. Amenable deaths (29.8%) were double that in the general population (14%); 60.3% died in hospital. Mortality risk related to percutaneous endoscopic gastrostomy/tube fed, Down syndrome, diabetes, lower respiratory tract infection at cohort-entry, smoking, epilepsy, hearing impairment, increasing number of prescribed drugs, increasing age. Bowel incontinence reduced mortality risk.

Conclusions Adults with intellectual disabilities with and without Down syndrome have different SMRs and causes of death which should be separately reported. Both die younger, from different causes than other people. Some mortality risks are similar to other people, with earlier mortality reflecting more multimorbidity; amenable deaths are also common. This should inform actions to reduce early mortality, for example, training to avoid aspiration/choking, pain identification to address problems before they are advanced, and reasonable adjustments to improve healthcare quality.

Strengths and limitations of this study

- ► Thorough methods of case ascertainment for intellectual disabilities at baseline.
- ► Individual verification of intellectual disabilities and its severity, and detailed health assessments at baseline.
- ► Longitudinal design.
- ► Large cohort size and study duration, and successful record linkage for 94% of participants.
- ► Limitations include that the study was conducted in only one part of Scotland, and the reliance on recorded cause of death from death certificates.

INTRODUCTION

People with intellectual disabilities die at a younger age than other people; on average, 20 years younger,[1] or 28 years younger specifically for people with Down syndrome.[2] It has been demonstrated that people with intellectual disabilities receive poorer management of their long-term conditions within primary healthcare services compared with the general population,[3] and it is conceivable that this is one contributor to earlier mortality. It has been suggested that as many as 40% of deaths of people with intellectual disabilities may have been amenable to good quality healthcare.[4–6] There has been a recent increase in research on mortality in people with intellectual disabilities, but very little research has distinguished people with

intellectual disabilities with and without Down syndrome, or investigated the factors associated with risk of mortality, and causes of mortality.

Previous studies on death in people with intellectual disabilities had limitations such as small sample sizes, or non-representative populations. More recently, there have been large-scale studies which are more representative, having been drawn from intellectual disabilities registers, or social security or primary care data with record linkage to death certification. These have been undertaken in parts of Sweden, Australia, England, Finland, Canada, Ireland and USA(online supplementary table 1).[5–19] These studies fairly consistently report standardised mortality ratios (SMRs) to be high for people with intellectual disabilities, more so at younger ages and higher for women than men. Adult studies have tended to report SMRs in the region of 2–4, although in some, SMR is only slightly above 1.[10 16 19] However, direct comparison between studies is not always possible, due to the different age ranges studied and methods of reporting.

In view of the methods that studies have used for population identification (typically, routine administrative data linked to death certifications), they provide little information on the socioclinical factors that influence SMR, or the risk factors associated with death, beyond that of age and sex. Three studies reported SMR by level of intellectual disabilities, with, broadly speaking, higher SMR with more severe intellectual disabilities.[7 10 17] Only three studies (different studies to those that reported on level of intellectual disabilities) were able to report data separately for adults with intellectual disabilities with and without Down syndrome; two found higher mortality rates for adults with Down syndrome (SMR=7.6[9] and HR=9.21[5]) than for adults without Down syndrome, or an OR showing Down syndrome as a risk of death.[12]A further study reported SMR=5.5 for children and adults (combined) with Down syndrome, but did not report SMR for those with intellectual disabilities without Down syndrome.[20] Two studies reported adults with intellectual disabilities to have higher SMRs if they have the comorbidities of epilepsy,[5 7] and cerebral palsy,[7] as opposed to not having these comorbidities. One study reported adults with intellectual disabilities with comorbid autism to have lower risk of mortality than those without comorbid autism.[5] One study reported the risk factors for mortality in a population with intellectual disabilities to be age, Down syndrome, cerebral palsy, blindness/low vision, technological dependence/medical fragility, wheelchair dependence, mobility impairment without wheelchair dependence, and epilepsy.[12] Factors not found to be risks, if any, were not reported, and a further limitation was that factors were reported by agency staff, rather than the individuals undergoing health assessments.[12] We have not identified any other studies that investigated risk factors for time to mortality in adults with intellectual disabilities.

There is less consistency regarding the most common certified underlying causes of death in adults with intellectual disabilities, partly as some studies do not report these separately for children and adults, or by age ranges. Additionally, studies group causes of death in different ways (eg, pneumonia vs respiratory system), which can affect prevalence rankings between studies. Pneumonia, other respiratory diseases and diseases of the nervous system were reported to be the most common in one study,[11] diseases of the circulatory system and respiratory systems in another,[5] heart disease, neoplasm and Alzheimer disease in a third,[18] and diseases of the circulatory system, neoplasm and the nervous system in a fourth.[19] In adults with intellectual disabilities, cause-specific SMRs have been reported to be high across most groups of disorders.[5 11] These studies did not report cause of death separately for adults with and without Down syndrome. Given the different health profile of people with Down syndrome compared with people with intellectual disabilities of other causes, this is an important limitation.[21] In people with Down syndrome, most studies on mortality have been conducted with child populations, and report the most common causes of death to be congenital heart disease, and pneumonia/diseases of the respiratory system.[2]

Overall, the existing body of literature on mortality in adults with intellectual disabilities does not include more detailed information on level of intellectual disabilities, nor separate out the population with, from those without, Down syndrome (for whom causes of death may differ), nor investigate health and demographic predictors of death other than age and sex, and is inconsistent with regard to causes of death. A better understanding of these factors may provide a pathway to action to reduce the observed earlier mortality in adults with intellectual disabilities.

This study aims to investigate the rates, causes, place and demographic and clinical associations with mortality in adults with intellectual disabilities, with and without Down syndrome.

METHODS

Participants

The adult (aged 16+ years) intellectual disabilities population living within the NHS Greater Glasgow area was identified through multiple sources between 2000 and 2001. General practitioners were financially incentivised to identify their registered patients with intellectual disabilities, and all 631 (100%) did so. Adults were also identified via the intellectual disabilities health and social work services including day services, the Health Board register and records of financial payments for any service by social work. This process led initially to an overidentification, such as people with IQ scores in the 70–80 range with additional complex health needs. All were systematically reviewed by nurses in the intellectual disabilities health service, and this group were removed. Thus, a register was compiled, and subsequently updated annually via general practices, with central support from the intellectual disabilities health service, until 2017 when

services were redesigned. The identified adult prevalence of intellectual disabilities within the area was 3.33 per 1000 in 2000–2001.

Process and data collection

With initial piloting in 2001, each participant had a detailed assessment of their general and mental health, and demographic factors, completed 2002–2004. One of the six specially trained, registered nurses reviewed each person's primary health care records, then used a semi-structured tool, the C21st Health Check, to assess clinical factors and the level and cause of intellectual disabilities. In addition to a review of existing health problems and all bodily health systems, a physical examination was undertaken, including assessment of vision and hearing, measurement of height and weight and a phlebotomy protocol followed. All information was then reviewed by the nurse with one of three general practitioners with a special interest in intellectual disabilities, and any further investigations that were indicated were completed. Previously known, and newly identified, conditions were then classified using the *International Statistical Classification of Diseases and Related Health Problems, 10th Revision (ICD-10)*.[22] Anyone identified to have possible, probable or definite mental ill-health, autism or problem behaviours was then fully assessed by the project's intellectual disabilities psychiatrists. Each person's assessment findings were then case conferenced by the two Consultant psychiatrists, and diagnoses were derived and agreed according to clinical diagnoses, *ICD-10 (Diagnostic Criteria for Research)*,[23] *Diagnostic and Statistical Manuel of Mental Disorders-IV-TR*[24] and *Diagnostic Criteria for Psychiatric Disorders for use with Adults with Learning Disabilities*.[25] Information was also collected on demographics, and community, hospital, and social service use. Further details are provided elsewhere.[26 27] The data were entered into a database by two dedicated data-entry staff.

Each person in Scotland is given a number unique to them at birth or first registration with a general practitioner, which is used in almost all subsequent health service encounters, and on certification of death. The numbers are held on the Community Health Index (CHI) database at National Services Scotland. These CHI numbers provided a means to record link each participant with National Records for Scotland death certification data. This linkage was performed in 2018, and the linked data were held in the NHS Greater Glasgow & Clyde (NHS GG&C) Safe Haven. Data on immediate, underlying and contributory causes of deaths by ICD-10 codes, age at death and place of death were extracted.

In order to provide finer granularity of cause of death, two clinical academics then grouped specific causes of death into narrower groupings than those provided by ICD-10 chapter headings (online supplementary table 2). This approach was also in view of the recognised issue of variation between health staff in distinguishing and recording immediate causes of death, and because some causes occurred in low numbers so could not be individually reported due to the risk of statistical disclosure. Additionally, some conditions likely to be the same are spilt between different ICD-10 chapters, for example, dementia in Alzheimer disease (F00) and unspecified dementia (F03) in the ICD-10 mental and behavioural disorders chapter, and Alzheimer's disease (G30) and Alzheimer's disease, unspecified (G30.9) in the ICD-10 diseases of the nervous system chapter. A list of related conditions was generated by one of the clinical academics and then checked by the second.

Analyses

All statistical analyses were conducted using R for Windows V.3.3.0 or SAS V.9.3 and were performed within the NHS GG&C Safe Haven environment. Due to disclosure principles of the Safe Haven, results with counts of less than five cannot be released; these have been referred to as <5 throughout. Similarly, if it is deemed possible that participants may be identified from the results, these may be omitted. Details are provided if this occurred.

Data were summarised for the population of adults aged 16+ years with intellectual disabilities. Categorical variables were summarised with the number and percentages of people falling into each category and the number of missing data. Continuous variables were summarised with the number of observations and those missing, the mean and SD, and the minimum and maximum values, unless otherwise stated.

Participant characteristics were summarised overall and for those alive and those deceased. For those who are deceased, their data including age at death, underlying/contributing causes of death, and location of death were summarised for those with and without Down syndrome. Location codes for place of death are provided where available. We assumed those with the code for non-institutional location to have died at home. Due to small numbers, location codes have been grouped together for NHS hospitals, home, and other hospitals/care facilities including hospices.

Mortality incidence rates have been calculated using the number of deaths in the cohort divided by the number of person years alive within the study period multiplied by 100 000, overall and for those with and without Down syndrome. SMRs were calculated using population data for those aged 15 and over within NHS GG&C in 2010.[28 29] Death rates for males and females by 5-year band ages groups spanning from 15 to 20 years old to 90 years and over were summed to form the expected death rates for the general population. The observed death rate for adults with intellectual disabilities was taken from our study results. The observed/expected death rates were calculated for the intellectual disabilities cohort overall then separately by age group, sex, ability level, and for the adults with and without Down syndrome, and ICD-10 chapter for cause of death, and compared with the general population.

Deaths were also analysed for those that could be considered as deaths that would have been avoidable.

The Office for National Statistics (ONS) published a definition of avoidable mortality,[30] which lists the causes of amenable deaths (deaths that should not occur in the presence of good healthcare, eg, respiratory disease) and causes of preventable deaths (eg, from diseases that could have been avoided by prior immunisation), by ICD-10 codes. Causes of death for the adults with intellectual disabilities have been summarised by ONS definition of avoidable deaths.

To determine the demographic and clinical factors associated with death in adults with intellectual disabilities, time to event analyses were explored using univariate Cox Proportional Hazards models. Variables were selected as potentially relevant on the basis of what is known on causes of death in people with intellectual disabilities, the 20 most common physical health conditions reported in the adult population with intellectual disabilities,[21] and other factors hypothesised as potentially clinically relevant (online supplementary table 3):

- Demographics—nine variables.
- Clinical conditions—33 variables.
- Service use—three variables.
- Prescriptions—five variables.

All 50 variables were then permitted entry in to a single multivariable analysis using stepwise regression methods, in order to identify a model containing the statistically significant factors associated with death. Age at date of the health assessment was entered into the model as a continuous measure. Results from the univariate Cox Proportional Hazards models (online supplementary table 3) and the statistically significant multivariable model from the stepwise results have been presented with HRs with corresponding 95% CIs (HR, 95% CI) and p-values were obtained.

Patient and public involvement

This study was designed to respond to the growing concern expressed by people with intellectual disabilities, their families and third sector organisations about the early deaths of people with intellectual disabilities. The Scottish Learning Disabilities Observatory, where this research was undertaken, has a specific remit for people with intellectual disabilities. Its steering group includes partners from third sector organisations, including Down syndrome Scotland, and people with intellectual disabilities, who approved the work plan for this project prior to it commencing. Results from this study will be disseminated for people with intellectual disabilities in an easy-read version via the Scottish Learning Disabilities Observatory.

RESULTS

Population characteristics

Of note, 962 of the original 1023 (94.0%) adults with intellectual disabilities who were assessed were linked to a CHI number enabling the extraction of relevant death data. Reasons for the unlinked 61 people could be due to moving out of the area, or a recording mistake. One further participant was removed from the analysis due to inaccurate recording of dates, leaving 961 adults in the cohort (93.9%). Of these 961 adults, 294 (30.6%) had a record of death. Table 1 shows the baseline characteristics of the full cohort of 961, the adults who died and those still alive at the time of linkage.

Age at death and mortality incidence

The mean age at death was 61.0 years (SD=7.0 years). Of the 961 adults, 64 (35.8%) of the 179 adults with Down syndrome and 230 (29.4%) of the 782 adults without Down syndrome had a record of death. Their mean age of death was 56.9 years (SD=4.3 years) for the adults with Down syndrome, and 62.2 years (SD=7.5 years) for the adults without Down syndrome. Mortality incidence for the cohort during the study period was 3049.0 per 100 000 person years follow-up, with 3832.1 per 100 000 for those with Down Syndrome and 2885.0 for those without Down syndrome.

Standardised mortality ratios

Compared with the general population, the SMR was 2.24 (1.98, 2.49) overall; 5.28 (3.98, 6.57) for adults with Down syndrome, 1.93 (1.68, 2.18) for adults without Down syndrome; 1.69 (1.42, 1.95) for men and 3.48 (2.90, 4.06) for women. SMRs were higher the more severe the level of intellectual disabilities, with people with profound intellectual disabilities having an SMR of 4.14 (3.11, 5.17). SMR was high for all age groups (though for the 15–25 year age group, the wide CI includes one, perhaps due to the smaller number of deaths in this group); this decreased as age increased. SMRs were high for most ICD-10 chapter groups of conditions, particularly so for congenital malformations at 17.26 (10.75, 23.78), diseases of the digestive system at 16.13 (8.23, 24.04), mental and behavioural disorders at 12.64 (3.27, 22.00) and external causes at 11.08 (3.40, 18.76). Details are shown in table 2.

Causes of death

Cause of death data was available from death certificates for 262 (89.1%) of 294 participants who had died, which include 57 (89.1%) participants with Down syndrome, and 205 (88.7%) participants without Down syndrome. Table 3 shows the underlying causes of death by ICD-10 chapters separately for the adults with and without Down syndrome. For the whole cohort, diseases of the respiratory system were the most common (21.8%), then diseases of the circulatory system (19.1%), then diseases of the nervous system (13.0%) and neoplasms, followed by congenital anomalies (10.3%). For the adults with Down syndrome, congenital anomalies were the most common (in all cases this was a record of 'Down syndrome'), then jointly diseases of the respiratory system and diseases of the circulatory system, then diseases of the nervous system, followed by infections, and mental and behavioural disorders. For the adults without Down syndrome, diseases of the respiratory system were the most common, then diseases of the circulatory system,

Table 1 Cohort characteristics at time of the health assessment, summarised overall and by death status during the follow-up period

Variable	Statistics/groups	All participants (n=961)	Deceased participants (n=294)	Alive participants (n=667)
Age (years)	Mean (SD)	44.1 (14.6)	52.4 (13.6)	40.5 (13.6)
	Min, max	16–83	18–83	16–77
Age group	16–25 years	127 (13.2%)	10 (3.4%)	117 (17.5%)
	26–35 years	153 (15.9%)	26 (8.8%)	127 (19.0%)
	36–45 years	246 (25.6%)	49 (16.7%)	197 (29.5)
	46–55 years	205 (21.3%)	85 (28.8%)	120 (18.0%)
	>55 years	230 (23.9%)	124 (42.0%)	106 (15.9%)
Sex	Male	525 (54.6%)	154 (52.4%)	371 (55.6%)
	Female	436 (45.3%)	140 (47.5%)	296 (44.4%)
Ability level	Mild ID	382 (39.7%)	92 (31.2%)	290 (43.5%)
	Moderate ID	236 (24.5%)	73 (24.7%)	163 (24.4%)
	Severe ID	180 (18.7%)	67 (22.7%)	113 (16.9%)
	Profound ID	163 (17.0%)	62 (21.1%)	101 (15.1%)
Accommodation type	Family carer	374 (38.9%)	70 (23.8%)	304 (45.6%)
	Independent	93 (9.7%)	36 (12.2%)	57 (8.5%)
	Paid support	435 (45.2%)	161 (54.6%)	274 (41.1%)
	Congregate care	59 (6.1%)	27 (9.2%)	32 (4.8%)
Down syndrome	No	782 (81.4%)	230 (78.2%)	552 (82.8%)
	Yes	179 (18.6%)	64 (21.7%)	115 (17.2%)

ID, intellectual disabilities.

then neoplasms, then diseases of the nervous system, followed by diseases of the digestive system. Table 4 presents the most common underlying causes of death by individual causes, or related groups of causes, with finer granularity than ICD-10 chapter headings (groups are shown in online supplementary table 2). Causes are listed in the order of how common they were in the whole cohort. Data are presented separately for the adults with and without Down syndrome. For the whole cohort, the most common cause was aspiration/reflux/choking, then respiratory infection, then other malignancy (non-gastrointestinal), then other condition (mostly unrelated conditions that could not be reported individually or as groups, due to individually occurring at a frequency of <5). For the adults with Down syndrome, Down syndrome was the most common cause, then dementia, then other infection. For the adults without Down syndrome, aspiration/reflux/choking was the most common cause, then respiratory infection, then other malignancy (non-gastrointestinal). For the 21 people whose death certificate listed Down syndrome as their underlying cause of death, the death certificates were reviewed and underlying cause of death reclassified, as a sensitivity check. Following this, the most common underlying causes of death for the adults with Down syndrome were dementia (n=20; 35.1%), then other infection (n=7; 12.3%).

Table 5 shows the all contributing causes of death data, again presenting the most common causes by individual causes, or related groups of causes with finer granularity than ICD-10 chapter headings. Data are presented separately for the adults with and without Down syndrome. For the whole cohort, respiratory infection was the most common cause (27.1%), followed by aspiration/reflux/choking (19.8%), other conditions (15.6%), other cardiovascular conditions (non-acute myocardial nor other ischaemic heart disease: 14.5%), then other respiratory conditions. For the adults with Down syndrome, Down syndrome was the most common, then dementia, then respiratory infection, then aspiration/reflux/choking. For the adults without Down syndrome, respiratory infection was the most common cause, then aspiration/reflux/choking, then other condition, then other respiratory conditions and intellectual disabilities.

Avoidable deaths

According to the ONS list of avoidable deaths, 102 (38.9%) of the 262 deaths were avoidable; most notably, respiratory infection and epilepsies (table 4); 78 (29.8%) were deaths that are amenable to good healthcare, while 51 (19.5%) were preventable deaths, and 27 (10.3%) deaths were classed as both amenable and preventable deaths. This compares to published Scottish death data showing in 2018 that 28% of deaths were avoidable; 14%

Table 2 Standardised mortality ratios

Variable	Groups	SMR (95% CI)
All participants	–	2.24 (1.99 to 2.50)
Age group*	15–25 years	18.73 (0.37 to 37.09)
	26–35 years	4.21 (1.29 to 7.13)
	36–45 years	3.86 (2.28 to 5.44)
	46–55 years	3.77 (2.90 to 4.74)
	>55 years	1.86 (1.60 to 2.12)
Sex	Male	1.69 (1.42 to 1.95)
	Female	3.48 (2.90 to 4.06)
Ability level	Mild ID	1.60 (1.27 to 1.92)
	Moderate ID	2.10 (1.62 to 2.58)
	Severe ID	2.78 (2.11 to 3.44)
	Profound ID	4.14 (3.11 to 5.17)
Down syndrome	No	1.93 (1.68 to 2.18)
	Yes	5.28 (3.98 to 6.57)
Underlying causes of death grouped by ICD-10 chapter†	Congenital malformations, deformations and chromosomal abnormalities	17.26 (10.75 to 23.78)
	Diseases of the blood and blood-forming organs and certain disorders involving the immune mechanism	7.50 (-7.20 to 22.20)
	Diseases of the circulatory system	5.55 (4.01 to 7.09)
	Diseases of the digestive system	16.13 (8.23 to 24.04)
	Diseases of the genitourinary system	3.65 (0.73 to 6.57)
	Diseases of the musculoskeletal system and connective tissue	5.40 (-0.71 to 11.52)
	Diseases of the nervous system	7.73 (5.13 to 10.32)
	Diseases of the respiratory system	6.78 (5.02 to 8.54)
	Diseases of the skin and subcutaneous tissue	2.75 (–2.64 to 8.15)
	Endocrine, nutritional and metabolic diseases	3.43 (1.05 to 5.81)
	External causes of morbidity and mobility	11.08 (3.40 to 18.76)
	Infectious and parasitic diseases	8.93 (1.78 to 16.07)
	Mental and behavioural disorders	12.64 (3.27 to 22.00)
	Neoplasms	6.31 (4.19 to 8.43)
	Symptoms, signs and abnormal clinical and laboratory findings, not elsewhere classified	19.51 (0.39 to 38.63)

*Data used for comparison with General Population (GG&C Health Board) provide data in 5-year age bands therefore 15+. Data on adults with ID are 16+.
†Negative Lower CI and wide CIs indicate low number of observed deaths in study population.
ICD-10, International Statistical Classification of Diseases and Related Health Problems, 10th Revision; ID, intellectual disabilities; SMR, standardised mortality ratios.

amenable, and 24% preventable, similar to the figures in the previous 4 years (data not available prior to 2014).[31] For the 57 deaths of adults with Down syndrome, 17 (29.8%) deaths were avoidable, 15 (26.3%) deaths were amenable to good healthcare, while seven (12.3%) were preventable, and five (8.8%) were both amenable and preventable. For the 205 deaths of adults without Down syndrome, 85 (41.5%) were avoidable, 63 (30.7%) deaths were amenable to good healthcare, while 44 (21.5%) were preventable, and 22 (10.7%) were both amenable and preventable.

Place of death

Of the 262 participants for whom place of death was known, 158 (60.3%) died in an NHS Hospital, 70 (26.7%) died at home, and 34 (13.0%) died within other hospitals/care facilities. This was similar for both the adults with Down syndrome: 31 (54.4%) in an NHS hospital, 17 (29.8%) at home, and nine (15.8%) within other hospitals/care facilities; and the adults without Down syndrome: 127 (62.0%) in an NHS hospital, 53 (25.9%) at home, and 25 (12.2%) within other hospitals/care facilities.

Table 3 Underlying causes of death grouped by ICD-10 chapter, where cause of death is known

ICD-10 chapter	Participants with Down syndrome (n=57)	Participants without Down syndrome (n=205)
Certain infectious and parasitic diseases	5 (8.8%)	<5
Neoplasms	<5	33 (16.1%)
Diseases of the blood and blood-forming organs and certain disorders involving the immune mechanism	<5	<5
Endocrine, nutritional and metabolic diseases	<5	8 (3.9%)
Mental and behavioural disorders	5 (8.8%)	<5
Diseases of the nervous system	7 (12.3%)	27 (13.2%)
Diseases of the eye and adnexa	<5	<5
Diseases of the ear and mastoid process	<5	<5
Diseases of the circulatory system	8 (14.0%)	42 (20.5%)
Diseases of the respiratory system	8 (14.0%)	49 (23.9%)
Diseases of the digestive system	<5	16 (7.8%)
Diseases of the skin and subcutaneous tissue	<5	<5
Diseases of the musculoskeletal system and connective tissue	<5	<5
Diseases of the genitourinary system	<5	5 (2.4%)
Pregnancy, childbirth and the puerperium	<5	<5
Certain conditions originating in the perinatal period	<5	<5
Congenital malformations, deformations and chromosomal abnormalities	21 (36.8%)	6 (2.9%)
Symptoms, signs and abnormal clinical and laboratory findings, not elsewhere classified	<5	<5
External causes of morbidity and mortality	<5	7 (3.4%)
All deaths	57 (100%)	205 (100%)

ICD-10, International Statistical Classification of Diseases and Related Health Problems, 10th Revision.

Table 4 Underlying causes of death grouped by specific individual causes or related groups of causes, where cause of death is known

Causes	Participants with Down syndrome (n=57)	Participants without Down syndrome (n=205)
Aspiration/reflux/choking	<5	22 (10.8%)
Respiratory infection	<5	21 (10.3%)
Down syndrome	21 (36.8%)	<5
Other malignancy	<5	19 (9.3%)
Other condition	<5	17 (8.3%)
Epilepsies	<5	13 (6.4%)
Acute myocardial infarction	<5	13 (6.4%)
Gastrointestinal malignancy	<5	12 (5.9%)
Stroke	<5	11 (5.4%)
Other cardiovascular disease	<5	11 (5.4%)
Other respiratory condition	<5	9 (4.4%)
Other infection	5 (8.8%)	6 (2.9%)
Cerebral palsy	<5	11 (5.4%)
Dementia	9 (15.8%)	<5
Other gastrointestinal disorders	<5	8 (3.9%)
Ulcer/gastrointestinal perforation	<5	7 (3.4%)
Diabetes	<5	7 (3.4%)
Other congenital condition	<5	6 (2.9%)
Other ischaemic heart condition	<5	6 (2.9)
Mental health	<5	<5
Other neurological conditions	<5	<5
Renal failure	<5	<5
All deaths	57 (100%)	205 (100%)

Factors associated with risk of death

The results from the univariate cox proportional hazards models indicated that of the original 50 potential variables, factors associated with risk of death were (online supplementary table 3) as follows:

- Demographics–age at the time of the health assessment, more severe learning disabilities, accommodation type (not living with family carer), not having day-time occupation, and being a smoker (but not sex, the extent of neighbourhood deprivation, civil status, nor Down syndrome, in view of the CIs).
- Clinical conditions–having spastic quadriplegia, hearing impairment, visual impairment, diabetes, percutaneous endoscopic gastrostomy/tube fed, constipation, ataxia/gait disorder, osteoporosis, hypertension, dysphagia, dyspnoea, gastro-oesophageal reflux disorder, lower respiratory tract infection, total number of physical health disorders, not having

Table 5 All contributing causes of death grouped by specific individual causes or related groups of causes, where cause of death is known

Causes	Participants with Down syndrome (n=57)	Participants without Down syndrome (n=205)
Respiratory infection	22 (38.6%)	49 (23.9%)
Aspiration/reflux/choking	11 (19.3%)	41 (20.0%)
Down syndrome	43 (75.4%)	<5
Other condition	8 (14.0%)	33 (16.1%)
Other cardiovascular disease	8 (14.0%)	30 (14.6%)
Other respiratory conditions	<5	31 (15.1%)
Other infection	9 (15.8%)	24 (11.7%)
Intellectual disabilities	<5	31 (15.1%)
Epilepsies	8 (14.0%)	24 (11.7%)
Dementia	24 (42.1%)	<5
Other neoplasms	<5	23 (11.2%)
Cerebral palsy	<5	24 (11.7%)
Acute myocardial infarction	5 (8.8%)	19 (9.3%)
Other gastrointestinal disorders	<5	18 (8.8%)
Diabetes	<5	19 (9.3%)
Other ischaemic heart disease	<5	19 (9.3%)
Renal failure	<5	16 (7.8%)
Stroke	<5	17 (8.3%)
Other congenital condition	<5	15 (7.3%)
Gastrointestinal malignant neoplasm	<5	12 (5.9%)
Ulcer/gastrointestinal perforation	<5	10 (4.9%)
Mental health	<5	10 (4.9%)
Other neurological condition	<5	8 (3.9%)
Heart failure	<5	7 (3.4%)
Injuries and accidents	<5	8 (3.9%)
Medical/surgical complications	<5	<5
Secondary malignancies	<5	<5
Thyroid disorders	<5	<5
Metabolic disorder	<5	<5
All deaths	57 (100%)	205 (100%)

Table 6 Multivariable model results for the outcome time to death

Variable	HR	95% CI	P
Age at time of health assessment	1.056	1.046 to 1.066	<0.0001
Smoker			
No	1	–	
Yes	1.531	1.1011 to 2.128	0.0112
Down syndrome			
No	1	–	
Yes	2.44	1.787 to 3.332	<0.0001
Epilepsy			
No	1	–	
Yes	1.511	1.173 to 1.946	0.0014
Hearing impairment			
No	1	–	
Yes	1.32	1.030 to 1.692	0.0284
Bowel incontinence			
No	1	–	
Yes	0.49	0.376 to 0.640	<0.0001
Diabetes			
No	1	–	
Yes	2.346	1.553 to 3.542	<0.0001
PEG/tube fed			
No	1	–	
Yes	2.346	1.135 to 5.989	0.0024
Lower respiratory track infection			
No	1	–	
Yes	1.782	1.315 to 2.415	0.0002
Total number of prescribed drugs	1.066	1.016 to 1.118	0.0085

PEG, percutaneous endoscopic gastrostomy.

impaired mobility, not having urinary incontinence, not having bowel incontinence, and not having autism (but not epilepsy, body mass index, nail disorder, epidermal thickening, cerebral palsy, fungal infection, musculoskeletal pain, bone deformity, dental/oral problem, eczema/dermatitis, psychosis, affective disorder including bipolar affective disorder, problem behaviour, eating disorder including pica, nor any mental illness).

- Service use—number of general practitioner consultations in the previous 12 months, total number of different types of health professionals providing care at the time of the clinical assessment (but not number of accident and emergency attendances in the previous 12 months).
- Prescriptions—antiepileptic drugs, total number of different types of drugs (but not antipsychotic drugs, antidepressant drugs, nor anxiolytic drugs).

Table 6 shows the final model of the variables retained in the multivariable analysis for time to death. The significant factors indicating an increased risk of death were increased age at the time of the health assessment, smoking, Down syndrome, diabetes, being percutaneous endoscopic gastrostomy/tube fed, lower respiratory tract infection at cohort inception, epilepsy, hearing

impairment, and total number of different types of drugs prescribed, while bowel incontinence showed a reduced risk of death. Of note, level of intellectual disabilities, while significant in the univariate analysis, was not retained in the multivariable model.

DISCUSSION

Principle findings and interpretation

As far as we are aware, this is the first population-based study of adults with intellectual disabilities to report in detail the factors associated with time to death, and to describe their causes of death, and quantify the SMR separately for adults with and without Down syndrome. This is important, since adults with Down syndrome form a notable proportion of all adults with intellectual disabilities (19% in this cohort), and because they have a different pattern of clinical conditions compared with other adults with intellectual disabilities.[21] We found that aspiration/reflux/choking is the most common underlying cause of death in adults with intellectual disabilities, followed by respiratory infection. They are also the most common all contributing causes of death. The profile differed in the adults with Down syndrome for whom 'Down syndrome', followed by dementia, were recorded as the most common underlying cause of death, and all contributing causes of death (or alternatively, dementia, then other infection were the most common underlying causes when 'Down syndrome' deaths were reclassified); with the next most common all contributing cause of death being respiratory infection, then aspiration/reflux/choking. The proportion of deaths that would have been amenable to good care for adults with intellectual disabilities was more than double that seen in the general population. Although aspiration/reflux/choking is not included in the ONS list of avoidable deaths, and therefore not included in the figures we report on amenable deaths, we consider that good care also could have prevented many of these aspiration/reflux/choking deaths. This appears to be very important for adults with intellectual disabilities irrespective of whether they have Down syndrome. Similarly, some other causes of deaths within this cohort (online supplementary table 2), such as constipation/mega-colon, and urinary tract infections do not appear on the ONS list of avoidable deaths.

Clearly, this pattern of causes of death differs from that seen in the general population, in whom the most common underlying causes of death are heart disease, then dementia, then lung cancer in men, and dementia, then heart disease, then stroke in women.[32] When all cancers are grouped together, in the general population, cancer is the leading underlying cause of death in 30% of men and 26% of women, compared with this study reporting 0% for adults with Down syndrome, and 15.2% for adults with intellectual disabilities without Down syndrome—presumably as the adults with intellectual disabilities are dying younger from other causes, and cancers increase with age.

We found an overall SMR of 2.24; 5.28 in the adults with Down syndrome and 1.93 for the adults without Down syndrome. SMRs were higher for most ICD-10 chapter groupings of conditions. It was higher in the women than the men, as has been previously reported in most (online supplementary table 1), but not all[10 19] previous reports. The reason for this is unknown; in the general population, mortality rates have fallen in recent decades, and more so in middle and older aged men than women (ie, the sex gap is narrowing at these ages), but we do not know what trends over time there have been for people with intellectual disabilities. Having intellectual disabilities removes differences in lifespan by sex compared with the general population; but sex was not a predictor of mortality in our study, so the SMR difference may only be because of the difference found in the general population by sex. SMRs were lowest with older age groups, likely to be due to increased illness in the older general population and conversely a healthier group with intellectual disabilities living to older ages compared with those who die younger (as has previously been reported).[33] Although SMR was higher with increasing severity of intellectual disabilities, ability level was not retained within the multivariable model on time to death. The factors that were independently associated with increased risk of death, in order, were being percutaneous endoscopic gastrostomy/tube fed, Down syndrome, diabetes, having a lower respiratory tract infection at entry to the cohort, smoking, epilepsy, hearing impairment, total number of prescribed drugs and age, while bowel incontinence had a reduced risk of death. Some of these predictors are similar to those reported in the general population, suggesting that earlier mortality of adults with intellectual disabilities is largely accounted for by the higher rates of multimorbidities that they experience compared with other people, and amenable deaths.[34]

While accommodation type (not living with a family carer), ability level, not having day-time occupation, having spastic quadriplegia, visual impairment, constipation, ataxia/gait disorder, osteoporosis, hypertension, dysphagia, dyspnoea, gastro-oesophageal reflux disorder, total number of physical health disorders, not having impaired mobility, not having urinary incontinence and not having autism, number of general practitioner consultations in the previous 12 months, total number of different types of health professionals providing care at the time of the health assessment and antiepileptic drugs were related to time of death on univariate analyses, they were not retained in the multivariable model.

The majority of the adults with intellectual disabilities, with and without Down syndrome, died in an NHS hospital.

Comparison with previous literature

The overall SMR we report, higher SMR in women than men and higher SMR at younger age groups is similar to the majority of previous reports. Most mortality studies with people with Down syndrome have been

conducted with children. Previous reports of children and adults (combined) gave an SMR=5.5,[20] and for adults SMR=7.6,[9] compared with our finding for adults with Down syndrome of SMR=5.28. Recent systematic reviews reported people with intellectual disabilities on average died 20 years younger than other people, and people with Down syndrome died 28 years younger, although the majority of the Down syndrome studies were not recent.[1 2] In our study, we found the gap between the age at death of people with intellectual disabilities with and without Down syndrome to be only 5.3 years, possibly reflecting the increasing lifespan of people with Down syndrome exceeding increases in lifespan for people with intellectual disabilities without Down syndrome. Notably, after 'Down syndrome', dementia was the most commonly reported underlying, and all contributing cause of death for the adults with Down syndrome, whereas studies in the past commented on congenital heart disease and respiratory causes.

For the cohort overall, respiratory infection and aspiration/reflux/choking were the most common all contributing causes of death. These conditions feature in previous studies on causes of death,[5 6 8 10 11] although there are inconsistencies between studies. By ICD-10 chapter, our study found the most common underlying causes of death were diseases of the respiratory system, then of the circulatory system, followed by neoplasms. Others reported the most common to be vascular,[10] circulatory,[5] heart disease[17] and jointly circulatory and neoplasm.[19]

Previous research from other countries has highlighted that listing Down syndrome or intellectual disabilities as the underlying cause of death obscures actual causes of death for this population.[35] We therefore presented data on revised cause of death for the 21 people for whom it was listed as Down syndrome (as a sensitivity check), and highlight with interest that in this Scottish cohort, no one had intellectual disabilities listed as underlying cause of death. This may reflect different medical death certificate recording practices in Scotland compared with for example, the USA.

Studies that investigated avoidable deaths in adults with intellectual disabilities found them to be more common than in the general population, due to deaths that would have been amenable to good care. Avoidable deaths have been reported in 44.7% of deaths of people with intellectual disabilities in England (mostly amenable deaths—figure not reported),[6] and in 31% in Australia,[19] compared with our figure of 38.9%. Avoidable deaths that would have been amenable to good care have been reported to occur in 37% of deaths of people with intellectual disabilities in England.[5] Our figure is slightly lower at 29.8% but still more than double that found in the Scottish general population.[31] It should be noted that the ONS list of avoidable deaths was not designed specifically for people with intellectual disabilities, and it may emphasise some causes less relevant, and omit others that might be highly relevant in this population.[5]

Strengths and limitations

The strengths of the study include the thorough methods of case ascertainment for intellectual disabilities at baseline with verification of intellectual disabilities and its severity, suggesting results are generalisable in other high-income countries. While our identification of the population will not have identified everyone with intellectual impairment (an IQ<70), in view of the multiple sources used we believe it will have identified the adults with intellectual disabilities (IQ<70, plus need for support in daily activities, and onset in the developmental period). Additionally, there were detailed clinical assessments at baseline, and a longitudinal design. The size of the cohort and the duration of follow-up is also a strength, as is the successful record linkage for 94% of participants. Our study does have limitations, specifically that the study was only conducted in one region of Scotland, and the reliance on death certificate data to obtain cause of death. Additionally, the characteristics and health of the participants was collected in 2002–2004. The health conditions we investigated tend to be long-standing or remitting/relapsing conditions, and psychotropic prescribing also once initiated tends to be long-standing in people with intellectual disabilities. However, it is possible that extent of neighbourhood deprivation, type of accommodation, employment and civil status (though few marry) might have changed for some people between 2002 and 2004 and 2018; we have no further information to check this. There were no concerns regarding the proportional hazards assumption in the multivariable model. The linkage was also reliant on the accuracy of the CHI number as a sole source of linkage.

Implications

It is important to know the factors that are associated with risk of death, and the common causes of death in this population, as these then inform the actions needed to reduce the unacceptably high SMRs experienced by people with intellectual disabilities. Awareness of these factors may provide a pathway to action to reduce the observed earlier mortality in adults with intellectual disabilities. It is not adequate to solely rely on the public health interventions available to everyone, even when they are accessible. Aspiration, reflux and choking could, and should, be avoided by raising awareness of its consequences (death), and putting in place training on simple measures related to feeding, positioning, food consistency, and when to seek health advice from speech and language therapy, physiotherapy, nursing, and medical advice. Carers need to be aware of how the adults they care for express pain, so that conditions such as gastrointestinal ulcers are attended to, prior to the extreme point of perforation and so treatable conditions such as constipation and urinary tract infections are managed before they lead to respiratory distress and sepsis. Quality of care is important; adults with intellectual disabilities need just as good care for their diabetes and epilepsy (and other conditions) as the rest of the population, with

reasonable adjustments to address accessibility, and accessible smoking cessation programmes.

Future research

Further research on larger samples is needed, particularly with regard to replicating and extending our findings on the factors that are associated with risk of death, and any sex differences in them, so that practitioners can focus on actions to improve the life expectancy of adults with intellectual disabilities, with and without Down syndrome.

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Contributors S-AC is principle investigator, she conceived and managed the project, interpreted data and wrote the first draft of the manuscript. LA contributed to the conception of the project, and project management. NG designed and supervised the statistical analysis, and contributed to data interpretation and drafting of the manuscript. PMcS implemented and refined the statistical analysis, and contributed to data interpretation and drafting of the manuscript. AJ implemented and refined the statistical analysis, and contributed to data interpretation. AH contributed to data linkage and interpretation, and drafting of the manuscript. CMcC provided expertise on data linkage and methods, and drafting of the manuscript. DK contributed to data interpretation and drafting of the manuscript. CM contributed to data interpretation, and drafting of the manuscript. All approved the final version of the manuscript. S-AC is the study guarantor.

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Competing interests None declared.

Patient consent for publication Not required.

Ethics approval Ethical approval was gained from NHS Greater Glasgow Primary Care Trust- Community & Mental Health Research Ethics Committee, and NHS Greater Glasgow and Clyde Safe Haven. Individual consent to participate was taken in line with Scottish law, between 2001–2004.

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BMJ Open

Prevalence of physical conditions and multimorbidity in a cohort of adults with intellectual disabilities with and without Down syndrome: cross-sectional study

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ABSTRACT

Objectives To investigate the prevalence of multimorbidity in adults with intellectual disabilities with and without Down syndrome.

Design Large, population-based cross-sectional study.

Setting The geographical area of one Health Board, Scotland.

Participants All adults (aged 16+ years) known to general practitioners to have intellectual disabilities and adults receiving services provided or paid by intellectual disabilities health or social work services. 1023/1562 potential participants took part (65.5%); 562 (54.9%) men and 461 (45.1%) women, aged 43.9 years (16–83 years). 186 had Down syndrome and 837 did not.

Main outcome measures The prevalence of International Statistical Classification of Diseases, 10th revision, physical health conditions and multimorbidity detected at a comprehensive health assessment.

Results The mean number of physical health conditions/participant was 11.04, and 98.7% had multimorbidity. The most prevalent conditions are painful and/or disabling and, in some cases, life threatening. The five most prevalent were visual impairment, obesity, epilepsy, constipation and ataxic/gait disorders. The pattern of multimorbidity differs from that seen in the general population and is spread across the entire adult life course. The extent of multimorbidity in the adults with Down syndrome was similar to that of the adults without Down syndrome, while the prevalence of individual conditions differed.

Conclusions This robustly designed study with a large population found an extremely high prevalence of multimorbidity in adults with intellectual disabilities across the entire adult life course. This increases complexity of medical management that secondary healthcare services and medical education are not yet geared towards, as these tend to focus on single conditions. This is in addition to complexity due to limitations in communication and understanding. As the physical conditions within their multimorbidity also differ from that seen in the older general population, urgent attention is needed to develop the care pathways and guidelines that are required to inform and so improve their healthcare.

Strengths and limitations of this study

- This is the first study to have reported on multimorbidity in people with intellectual disabilities across the adult life course, where each individual had their health assessed by trained professionals.
- The health assessments were systematic and detailed.
- The study is large and population based, and the participation rate was high.
- A limitation is that the study was only conducted in one area of Scotland.

INTRODUCTION

People with intellectual disabilities have different health needs, shorter life expectancy and other health inequalities compared with the general population.[1–4] Despite this, there is surprisingly little reported on their prevalence of physical ill health and multimorbidity (two or more conditions in addition to intellectual disabilities), and few studies were population based and conducted on a large scale. Multimorbidity is important as its management is more complex than that of single conditions, with risks of drug–drug interactions, drug–disease interactions and disease–disease interactions. However, healthcare systems, and care pathways, are focused on management of single conditions. In the general population, awareness has recently been raised on the importance of multimorbidity, which becomes increasingly prevalent over the age of 50 years.[5]

Only five studies were identified that investigated multimorbidity among adults with intellectual disabilities. Three studies reported high rates of multimorbidity: 71% in 695 older persons with intellectual disabilities,[6] 80% in 1047 older persons receiving

paid support[7] and 40.6% in 8014 adults with intellectual disabilities.[8] However, these studies are limited as two included only older adults,[6 7] one of which relied on self/proxy-reporting of known health conditions out of a list of 12,[6] while the other included 20 conditions,[7] and the third, which was across the adult life course, reported data extracted electronically from primary care case records on 38 conditions, therefore only included conditions that had previously been presented to the general practitioner (GP).[8] Two further studies reported lower rates of multimorbidity (though still higher than in the general population): 22.9% in 14 751 adults with intellectual disabilities aged 18–84 years (vs 13.3% of other people)[9] and 10% in 299 adults with proxy measures of mild intellectual disabilities, aged 16–49 years (vs 5% of other people).[4] The former of these included just 19 long-term conditions (selected on the basis of the UK GP contract, ie, evidenced to be of importance for the general population) and relied on extraction of information on the 19 conditions that had previously been presented to the GP. The latter reported whether people were known to have any of only 15 health conditions and focused only on adults with mild intellectual disabilities who are therefore less dissimilar from the general population than are people with more severe intellectual disabilities.[4] These sampling and methodological differences account for the lower reported rates of multimorbidity in these two studies than in the other three. Only one of these five studies conducted individual health assessments (and only for some of the conditions included in the study),[7] and all five studies reported on only a limited number of preselected conditions.

There is a lack of consistency in reports on the prevalence of single physical health conditions in people with intellectual disabilities, due to the differences in methods used and populations studied. Reported prevalence rates for vision problems, for example, range from 18% to 99%,[10–13] gastro-oesophageal reflux disease ranges from 33% to 50%,[2 14–16] untreated dental caries range from 18% to 84%[17–19] and obesity ranges from 21% to 35%.[20–23] Thus, findings are conflicting. Conceivably, prevalence of physical health conditions may vary by country due to differences in lifestyle, availability, affordability and organisation of healthcare. There is a lack of studies carried out in the UK on the physical health of people with intellectual disabilities.[24] No UK-based data were found on the prevalence of musculoskeletal impairments, constipation or gastro-oesophageal reflux disease among people with intellectual disabilities. A recent systematic review of systematic reviews of the health or healthcare of people with intellectual disabilities also found significant gaps in research on physical health conditions.[25]

In summary, little is known about the extent of multimorbidity and prevalence of physical health problems in adults with intellectual disabilities. This paper reports findings from a large-scale population-based study that was conducted to address this. The aims of this study were to identify in adults with intellectual disabilities with, and without, Down syndrome:

1. the extent of multimorbidity
2. the prevalence of physical ill health
3. the top 20 most prevalent physical health conditions, and their associations with age, gender, level of intellectual disabilities and Down syndrome.

METHODS

Individual consent to participate was taken from each person with intellectual disabilities, as far as that person had decision-making capacity to consent, with consent given by the nearest relative/welfare guardian when the participant lacked such capacity, in keeping with Scottish law. Additionally, for individuals who did not have decision-making capacity to consent, the study was explained to them in keeping with their communicative abilities, and their views were sought and respected.

Participants

The adult population (aged 16 years and over) of people with intellectual disabilities living within the geographical area of Greater Glasgow Health Board, Scotland, were identified and recruited to a cohort study between 2002 and 2004. All persons known to GPs to have intellectual disabilities, persons receiving health, social care, residential, occupational and support services provided by intellectual disabilities health or social work services or any other support hours or services funded through social work or disability allowances were approached to take part in the study.[26] The GPs were financially incentivised to identify their population, and 100% in the area did so. The ascertainment rate was similar to the adult rate reported in a recent meta-analysis on prevalence of intellectual disabilities.[27] Only participants within the strict study boundary were included. Of the 1562 potential participants identified, consent was gained for 1023 adults to take part (65.5%).

Measures and procedure

Six nurses reviewed primary care case records, using a structured format and data collection form. They then completed a comprehensive semi-structured health interview and targeted physical examination and followed a phlebotomy protocol, with the person with intellectual disabilities and their carer, using the *C21st Health Check* (http://www.gla.ac.uk/researchinstitutes/healthwellbeing/research/mentalhealth/research/projects/ucedd/). Physical examination included measurement of height and weight, waist circumference, three recordings of blood pressure, pulse rate, pulse rhythm, communication assessment, oral examination, vision, hearing, peak flow, inhaler technique (if used) and feet and nail assessments, followed by urinalysis, a phlebotomy protocol and referral protocol. Most of the physical examination was protocolled; for example, vision was assessed by first asking a series of nine questions to help detect any possible problems (eg, for persons unable to self-report, carers were asked whether the person screws up his/her

eyes when in bright sunlight), then measuring vision using Kay's pictures at 33 cm and 3 m, and referring persons with possible visual impairment to the University Visual Sciences Department for more detailed, specialist assessment; hearing, likewise, was assessed through a series of questions, then otoscopy, and if the tympanic membrane could be visualised, examination using Warblers at 1/2 m at the level of 30 db/500 Hz, 30 db/1000 Hz, 30 db/2000 Hz and 30 db/4000 Hz, with referral for specialist assessment if there was any suggestion of possible hearing impairment. If the tympanic membrane could not be visualised because of impacted cerumen, drops were first used to clear it. Blindness or low vision was only recorded if it was not corrected by spectacles/best possible correction; and hearing loss was only recorded if it was not corrected by hearing aids. Findings were discussed with one of three GPs who specialised in intellectual disabilities and who classified all the physical health conditions using the International Statistical Classification of Diseases and Related Health Problems, 10th revision (ICD-10).[28] The complete assessment process took about 4 hours per participant, and conditions were recorded if present at the time of assessment (as opposed to historical conditions).

The level of intellectual disabilities of each participant, in keeping with the ICD-10 Classification of Mental and Behavioural Disorders – clinical descriptions and diagnostic guidelines,[29] was derived from recorded assessments or on the basis of the score gained on the health check. A record was made of whether each person had Down syndrome.

Definition of multimorbidity

There is no standard definition for multimorbidity. A recent National Institute for Health and Care Excellence (NICE) guideline on multimorbidity reflected that while multimorbidity is most commonly defined simply as having two or more long-term conditions, this type of definition is not necessarily helpful when providing clinical care.[30] Hence in the NICE guideline, the term multimorbidity refers to the presence of 2 or more long-term health conditions that can include: defined physical and mental health conditions such as diabetes or schizophrenia, ongoing conditions such as learning disability, symptom complexes such as frailty or chronic pain, sensory impairment such as sight or hearing loss and alcohol and substance misuse. The guideline coverage is for adults with two or more long-term physical health conditions and/or adults with one or more mental health conditions and at least one physical health condition. Given that the focus of this study is exclusively on adults with intellectual disabilities, we have used a tighter criteria for multimorbidity of intellectual disabilities plus at least two physical health conditions.

Analysis

Relevant data from the health check were entered into the Statistical Package for Social Services V.22.[31] The number of individuals, age, gender, level of intellectual disabilities and accommodation type were analysed using descriptive statistics. Social deprivation category was based on quintiles of the Carstairs deprivation score. This ranges from 1 (most affluent) to 5 (least affluent).[32] Frequency data were derived to identify the prevalence of multimorbidity and physical health conditions across all ICD-10 chapters. Twenty binary logistic regressions were conducted to determine if there were any associations between each of the 20 dependent variables (each of the 20 most prevalent physical health conditions) and the independent variables of age group, gender, level of ability and Down syndrome.

RESULTS

Demographics

The sample comprised 562 men (54.9%) and 461 women (45.1%) with a mean age of 43.9 years (range 16–83). One hundred and eighty-six (18.2%) had a diagnosis of Down syndrome: 91 men (48.9%) and 95 women (51.1%), with a mean age of 41.1 years. Table 1 describes the demographics and characteristics of the study sample.

The extent of multimorbidity experienced by adults with intellectual disabilities

The highest number of current physical health conditions experienced by an individual was 28. There was a mean number of 11.04 coexisting conditions per participant (*SD*=4.7) (figure 1). A percentage of 99.2 of participants (n=1015) had at least one condition, and 98.7% (n=1010) had two or more conditions. Only eight participants (four males, four females) had no physical health conditions. Multimorbidity was highly prevalent across the whole of the adult life course (figure 2). Figure 2 displays the mean number of physical health conditions by gender, age and level of intellectual disabilities, showing high rates across all groups.

The extent of multimorbidity was similar for the adults with, and without, Down syndrome (figure 3). A gradient across the extent of neighbourhood deprivation was not seen for multimorbidity (figure 4).

The prevalence of physical ill health by ICD-10 chapter

Participants were only counted once if they had more than one condition within each ICD-10 chapter (see figure 5). The most prevalent conditions reported were from the ICD-10 chapters on symptoms and signs: n=772 (75.5%); diseases of the skin and subcutaneous tissue: n=625 (61.09%); diseases of the digestive system: n=573 (56%); endocrine, nutritional and metabolic diseases: n=526 (51.4%); diseases of the nervous system: n=494 (48.3%); diseases of the musculoskeletal system and connective tissue: n=493 (48.2%); and diseases of the eye and adnexa: n=481 (47%). ICD-10 codes within the symptoms and signs chapter include physical health conditions such as ataxic gait and dysphagia.

Table 1 Demographics and characteristics of participants

Participants	N (1023) (n (%))	Without Down syndrome N (837) (n (%))	With Down syndrome N (186) (n (%))
Gender			
Male	562 (54.9)	471 (56.3)	91 (48.9)
Female	461 (45.1)	366 (43.7)	95 (51.1)
Age (years)			
16–24	121 (11.8)	101 (12.1)	20 (10.8)
25–34	156 (15.2)	128 (15.3)	28 (15.1)
35–44	253 (24.7)	192 (22.9)	61 (32.8)
45–54	238 (23.3)	184 (22)	54 (29)
55–64	169 (16.5)	148 (17.7)	21 (11.3)
65 and above	86 (8.4)	84 (10)	2 (1.1)
Level of intellectual disabilities			
Mild	398 (38.9)	321 (38.4)	77 (41.4)
Moderate	248 (24.2)	198 (23.7)	50 (26.9)
Severe	193 (18.9)	159 (19)	34 (18.3)
Profound	184 (18)	159 (19)	25 (13.4)
Accommodation type			
Lives with family carer	390 (38.1)	289 (34.5)	101 (54.3)
Lives independently	102 (10)	94 (11.2)	8 (4.3)
Lives with paid support	467 (45.7)	404 (48.3)	63 (33.9)
Lives in congregate setting	64 (6.3)	50 (6)	14 (7.5)
Deprivation category			
Most affluent	228 (22.3)	179 (21.4)	49 (26.3)
2	92 (9)	71 (8.5)	21 (11.3)
3	66 (6.5)	49 (5.9)	17 (9.1)
4	99 (9.7)	84 (10)	15 (8.1)
Most deprived	538 (52.6)	454 (54.2)	84 (45.2)
White	986 (96.4)	803 (95.9)	183 (98.4)
Non-white	37 (3.6)	34 (4.1)	3 (1.6)
Mean number of physical health conditions	11.04 (100)	10.89 (100)	11.68 (100)

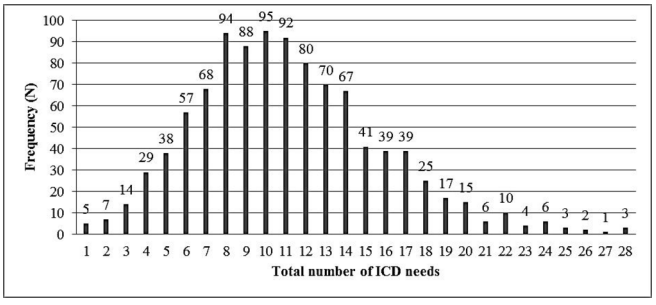

Figure 1 Total number of ICD-10 physical health conditions. ICD-10, International Statistical Classification of Diseases, 10th revision.

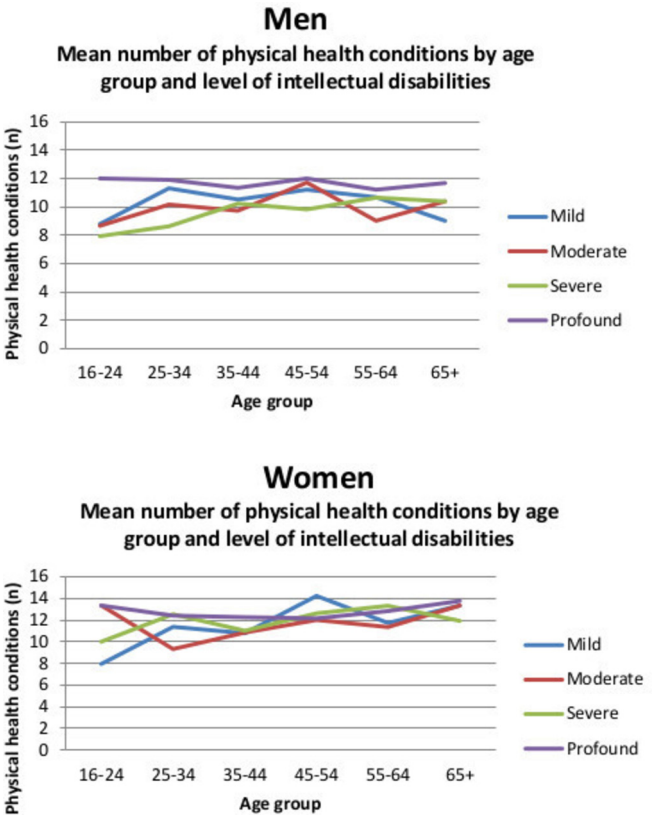

Figure 2 Mean number of physical health conditions by gender, age group and level of intellectual disabilities.

Top 20 most prevalent physical health conditions

Physical health conditions in order of prevalence were: visual impairment, obesity, epilepsy, constipation, ataxic/gait disorders, hearing impairment, nail disorder, epidermal thickening/xerosis, cerebral palsy and other paralytic syndromes, osteoporosis, fungal infection, hypertension, bone deformity, musculoskeletal pain/dorsalgia, eczema/dermatitis, gastro-oesophageal reflux disorder, dysphagia, lower respiratory tract infection, dyspnoea/wheezing and dental/oral (table 2). For adults with Down syndrome, these conditions were also common, but the most prevalent conditions were obesity, visual impairments, hearing impairments, xerosis, nail disorder and constipation, with the first five of these conditions

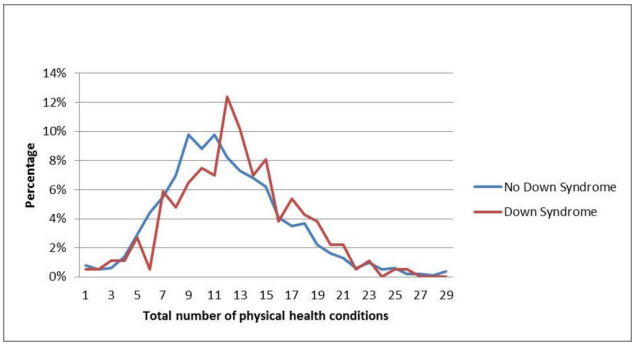

Figure 3 Extent of multimorbidity in individuals with intellectual disabilities with and without Down syndrome.

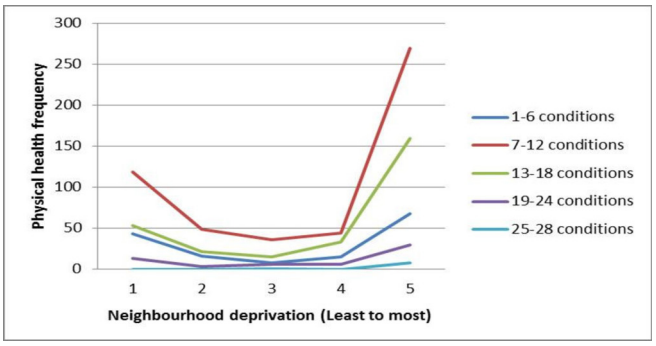

Figure 4 Number of physical health conditions by neighbourhood deprivation.

being more prevalent than in the adults without Down syndrome. Some conditions were much less common than in the adults without Down syndrome: epilepsy, hypertension, ataxia, cerebral palsy and osteoporosis (table 2). While constipation was prevalent in the adults with Down syndrome, it was less so than for the adults without Down syndrome. For both the adults with intellectual disabilities and adults with Down syndrome, these patterns differ from the general population in whom the most prevalent physical health conditions have been reported to be, in order, hypertension, painful condition, asthma, coronary heart disease, irritable bowel, dyspepsia and diabetes.[8]

Table 3 shows the results of the 20 regressions with the top 20 most prevalent physical health conditions as the dependant variables. It presents the ORs for gender, age, level of intellectual disabilities and presence of Down syndrome in independently predicting each of the 20 conditions. Women experienced some conditions more frequently than men, notably: obesity, constipation, epidermal thickening/xerosis, osteoporosis, dyspnoea/wheezing and musculoskeletal pain/dorsalgia. For most conditions, there is not an association with age; however, epilepsy and hearing impairment appear to be less prevalent in older age groups, and osteoporosis and

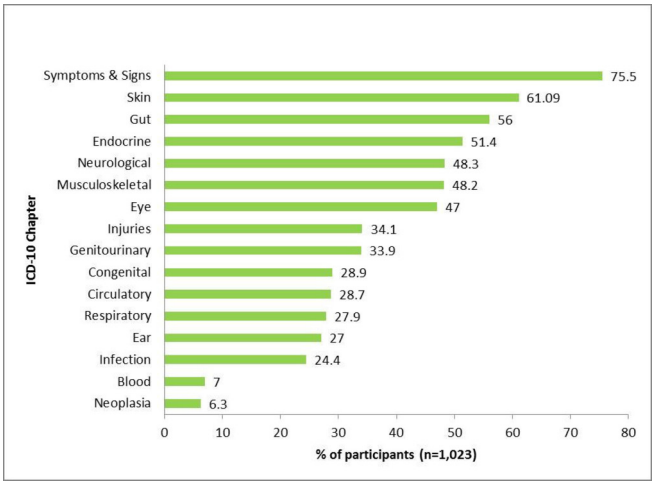

Figure 5 Prevalence (%) of physical ill health by ICD-10 chapter. ICD-10, International Statistical Classification of Diseases, 10th revision.

hypertension are more prevalent in older age groups. Several of the conditions showed a gradient across level of ability, being more prevalent the more severe the intellectual disabilities, including visual impairment, epilepsy, constipation, ataxia, cerebral palsy, osteoporosis, bone deformity, gastro-oesophageal reflux disorder and dysphagia, whilst for obesity, hypertension and dorsalgia, the relationship with ability level was reversed.

DISCUSSION

Principal findings and interpretation

It is believed that this is the first study to have reported on multimorbidity in people with intellectual disabilities across the adult life course in a large population-based sample where each individual had their health comprehensively assessed. A full range of physical health conditions were comprehensively assessed, rather than a shorter list of preselected conditions or only conditions that had already been presented to primary care or proxy measures for conditions. An extremely high prevalence of multimorbidity was reported at 98.7%. As expected, the percentage was much higher than in previous studies due to this methodology. The extent of multimorbidity was similar for both the adults with, and without, Down syndrome, though, as expected, there were some differences in the pattern of conditions. The pattern of multimorbidity also differed from the general population, hence findings from the general population are not transferrable; multimorbidity among people with intellectual disabilities requires specific study.[30] Multimorbidity was prevalent across the entire adult life course, unlike the general population in whom it increases over the age of 50 years,[5] hence healthcare availability is equally essential at all ages. Unlike the general population, a gradient across the extent of neighbourhood deprivation was not seen for multimorbidity as found in previous studies with adults with intellectual disabilities,[8 33] hence focused services are needed in all neighbourhoods.

With regards to single conditions, visual impairment was the most prevalent condition. Previous research has highlighted that carers or health professionals are often not aware of sensory impairments[34]; these are often misattributed to the individual's intellectual disabilities (diagnostic overshadowing)[34] and that people with intellectual disabilities are often unable to communicate that they have a problem.[35] A high index of suspicion is, therefore, needed with regards to visual impairments, particularly as these can be detected by optometrists even in people with profound intellectual disabilities. Epilepsy was also prevalent. Epilepsy among people with intellectual disabilities has previously been reported as much higher than for the general population, with seizures commonly multiple and resistant to drug treatment.[25 36] Uncontrolled epilepsy can be disabling and have serious negative consequences on both quality of life and mortality.[2] It is therefore essential for all healthcare practitioners to be aware of the prevalence and management of a complex and potentially

Table 2 Prevalence of physical health conditions for adults with and without Down syndrome across all ICD-10 chapters

	Physical health condition	Whole cohort (n=1023) n	Whole cohort %	Down syndrome (n=186) (n (%))	Without down syndrome (n=837) (n (%))
1	Visual impairment	481	47	90 (48.4)	391 (46.7)
2	Obesity	415	40.6	105 (56.5)	310 (37)
3	Epilepsy	349	34.1	24 (13)	325 (38.8)
4	Constipation	346	33.8	45 (24.1)	301 (36)
5	Ataxic/gait disorders	306	29.9	30 (16.1)	276 (33)
6	Hearing impairment	276	26.9	73 (39.2)	203 (24.2)
7	Nail disorder (eg, ingrowing nail)	238	23.3	50 (26.9)	188 (22.5)
8	Epidermal thickening/xerosis	217	21.2	69 (37.1)	148 (17.7)
9	Cerebral palsy and other paralytic syndromes	191	18.7	8 (4.3)	183 (21.9)
10	Osteoporosis	189	18.5	11 (5.9)	178 (21.3)
11	Fungal infection	167	16.3	42 (22.5)	125 (14.9)
12	Hypertension	158	15.4	8 (4.3)	150 (17.9)
13	Bone deformity	155	15.1	27 (14.5)	128 (15.3)
14	Musculoskeletal pain/dorsalgia	152	14.9	32 (17.2)	120 (14.3)
15	Eczema/dermatitis	149	14.6	38 (20.4)	111 (13.3)
16	Gastro-oesophageal reflux disorder	148	14.5	26 (14)	122 (14.6)
17	Dysphagia	147	14.4	24 (12.9)	123 (14.7)
18	Lower respiratory tract infection	134	13	34 (18.3)	100 (11.9)
19	Dyspnoea/wheezing	131	12.8	27 (14.5)	104 (12.4)
20	Dental/oral	130	12.7	28 (15)	102 (12.2)

ICD-10, International Statistical Classification of Diseases, 10th revision.

life-threatening condition in the intellectual disabilities population. Constipation was the fourth most prevalent physical health condition. This has been reported as common in adults with intellectual disabilities in institutional settings[37] but has received little research attention in population-based cohorts. Evenhuis[38] reported on the occurrence of constipation in 70 individuals over a 10-year period (mean age 70 years, range 60–92) in a Dutch residential care centre and found that 57% suffered from chronic constipation and 56% were permanently taking laxative treatment. Eight people with chronic constipation had serious side effects (rectal prolapse, diverticula of colon, intestinal obstruction, megacolon and haemorrhoids) and four eventually died of intestinal obstruction. Thus, as well as being painful, constipation may remain undetected for a long time and can cause death due to missed clinical symptoms.[38 39] Many factors can contribute to constipation including immobility, cerebral palsy, neurological disease, certain drugs, poor diet and lack of exercise.[35 40] The high rate reported highlights the importance of this condition. Our study also adds to UK-based data by providing prevalence rates on musculoskeletal impairments, constipation and gastro-oesophageal reflux disease among people with intellectual disabilities, conditions previously unreported in the UK research literature.[24]

Constipation, osteoporosis and dorsalgia were more prevalent in women as seen in the female general population.[40 41] However, the age-related increase in conditions typically seen in the general population is not apparent in our study in adults with intellectual disabilities. On average, the more severe the person's intellectual disabilities, the younger they die,[42] and the more severe a person's intellectual disabilities, the higher the prevalence of many of the conditions, so older age groups have milder intellectual disabilities. A gradient was found across levels of ability for dorsalgia, with lower levels at more severe intellectual disabilities. This seems extremely unlikely given the higher rates of cerebral palsy and bone deformities at more severe levels of intellectual disabilities and suggests that dorsalgia is at risk of underdetection in people with communication problems. High vigilance is therefore needed for this painful condition.

The conditions in table 2 are listed as per the top 20 in the population with intellectual disabilities. It is important to note that this list would be different if it was ordered by the top 20 for the adults with Down syndrome. For example, 24.2% of the participants with Down syndrome

Table 3 Twenty regression analyses showing the independent associations of gender, level of intellectual disabilities, Down syndrome and age, with the top 20 physical health conditions (ORs (95% CIs))

Physical health condition	Gender	Level of disability				Down syndrome	Age					
	Ref=Male	Mild	Moderate	Severe	Profound	Ref: without	16–24	25–34	35–44	45–54	55–64	65+
Vision	0.79 (0.61 to 1.02)	**Ref**	1.12 (0.81 to 1.55)	**1.80 (1.27 to 2.57)**	**2.57 (1.78 to 3.70)**	1.10 (0.79 to 1.53)	Ref	0.83 (0.51 to 1.35)	1.11 (0.71 to 1.73)	1.05 (0.67 to 1.65)	0.88 (0.55 to 1.42)	0.77 (0.43 to 1.36)
Obesity	**1.34 (1.03 to 1.73)**	**Ref**	0.78 (0.56 to 1.08)	**0.65 (0.46 to 0.94)**	**0.30 (0.19 to 0.45)**	**2.06 (1.47 to 2.88)**	**Ref**	1.35 (0.81 to 2.24)	1.06 (0.66 to 1.70)	**1.78 (1.11 to 2.84)**	0.96 (0.58 to 1.60)	1.21 (0.67 to 2.18)
Epilepsy	1.04 (0.79 to 1.38)	**Ref**	**1.57 (1.09 to 2.3)**	**1.78 (1.21 to 2.62)**	**4.49 (3.06 to 6.65)**	**0.21 (0.13 to 0.34)**	**Ref**	**1.74 (1.02 to 2.97)**	**1.68 (1.02 to 2.75)**	1.23 (0.74 to 2.02)	1.02 (0.60 to 1.74)	0.68 (0.35 to 1.30)
Constipation	**1.50 (1.14 to 1.97)**	**Ref**	1.26 (0.88 to 1.82)	**1.85 (1.27 to 2.70)**	**4.30 (2.95 to 6.28)**	**0.56 (0.38 to 0.82)**	Ref	1.38 (0.81 to 2.34)	1.18 (0.72 to 1.94)	1.59 (0.97 to 2.59)	1.13 (0.67 to 1.92)	1.15 (0.62 to 2.15)
Ataxic/ gait disorder	1.23 (0.92 to 1.64)	**Ref**	**2.40 (1.62 to 3.56)**	**3.79 (2.51 to 5.67)**	**6.66 (4.42 to 10.03)**	**0.40 (0.26 to 0.62)**	Ref	1.43 (0.81 to 2.54)	1.37 (0.80 to 2.34)	**1.78 (1.05 to 3.03)**	1.59 (0.90 to 2.78)	**2.59 (1.36 to 4.91)**
Hearing	0.97 (0.73 to 1.30)	Ref	0.94 (0.65 to 1.36)	1.08 (0.73 to 1.61)	0.86 (0.56 to 1.32)	**2.46 (1.74 to 3.49)**	**Ref**	0.71 (0.39 to 1.29)	0.91 (0.54 to 1.54)	1.22 (0.73 to 2.05)	1.37 (0.80 to 2.38)	**4.59 (2.49 to 8.46)**
Nail disorder	1.24 (0.92 to 1.66)	Ref	1.01 (0.69 to 1.49)	1.05 (0.70 to 1.59)	0.9 (0.59 to 1.39)	1.24 (0.85 to 1.80)	**Ref**	**1.95 (1.02 to 3.73)**	1.70 (0.92 to 3.12)	**2.92 (1.61 to 5.29)**	1.79 (0.94 to 3.41)	**2.41 (1.20 to 4.94)**
Epidermal thickening	**1.83 (1.34 to 2.50)**	Ref	**1.49 (1 to 2.21)**	1.23 (0.79 to 1.92)	1.32 (0.85 to 2.07)	**2.74 (1.91 to 3.93)**	**Ref**	**2.87 (1.40 to 5.86)**	**2.29 (1.16 to 4.53)**	**2.94 (1.49 to 5.79)**	**3.25 (1.60 to 6.59)**	**2.34 (1.01 to 5.40)**
Cerebral palsy	0.86 (0.61 to 1.22)	**Ref**	**2.38 (1.41 to 4.04)**	**4.10 (2.43 to 6.86)**	**9.86 (6.02 to 16.15)**	**0.15 (0.07 to 0.32)**	Ref	1.62 (0.86 to 3.06)	1.17 (0.63 to 2.14)	1.25 (0.68 to 2.29)	0.84 (0.43 to 1.63)	0.63 (0.27 to 1.47)
Osteoporosis	**2.34 (1.64 to 3.32)**	**Ref**	**1.68 (1.01 to 2.82)**	**2.67 (1.61 to 4.44)**	**9.66 (6.01 to 15.54)**	**0.22 (0.11 to 0.43)**	Ref	1.59 (0.77 to 3.26)	**2.11 (1.08 to 4.13)**	1.55 (0.78 to 3.08)	**2.40 (1.20 to 4.84)**	**2.85 (1.30 to 6.27)**
Fungal infection	0.85 (0.60 to 1.20)	**Ref**	0.66 (0.43 to 1.03)	0.76 (0.48 to 1.20)	**0.39 (0.22 to 0.69)**	**1.68 (1.11 to 2.53)**	**Ref**	**9.00 (3.09 to 26.20)**	**3.77 (1.29 to 10.99)**	**8.22 (2.89 to 23.39)**	**6.40 (2.18 to 18.79)**	**5.33 (1.66 to 17.11)**
Hypertension	0.93 (0.65 to 1.33)	**Ref**	**0.65 (0.42 to 1.00)**	**0.43 (0.25 to 0.72)**	**0.31 (0.17 to 0.57)**	**0.22 (0.10 to 0.46)**	**Ref**	2.11 (0.78 to 5.66)	**2.52 (1.01 to 6.30)**	**4.49 (1.83 to 10.98)**	**5.31 (2.15 to 13.16)**	**7.74 (2.99 to 19.99)**
Bone deformity	1.31 (0.92 to 1.85)	**Ref**	1.37 (0.85 to 2.21)	1.27 (0.76 to 2.13)	**2.96 (1.87 to 4.70)**	1.03 (0.65 to 1.64)	**Ref**	1.22 (0.6 to 2.47)	1.01 (0.52 to 1.98)	1.58 (0.83 to 3.02)	1.53 (0.77 to 3.04)	2.06 (0.95 to 4.47)
Musculoskeletal	**1.89 (1.32 to 2.70)**	**Ref**	**0.54 (0.35 to 0.85)**	**0.45 (0.27 to 0.74)**	**0.16 (0.07 to 0.34)**	1.14 (0.73 to 1.79)	Ref	2.35 (0.99 to 5.57)	2.08 (0.92 to 4.70)	**3.10 (1.39 to 6.95)**	**3.22 (1.40 to 7.41)**	2.31 (0.90 to 5.96)
Eczema	0.95 (0.66 to 1.35)	Ref	0.62 (0.38 to 1.0)	0.89 (0.55 to 1.45)	0.92 (0.57 to 1.50)	**1.70 (1.12 to 2.59)**	Ref	1.04 (0.55 to 1.97)	0.74 (0.40 to 1.36)	0.890 (0.49 to 1.62)	0.71 (0.36 to 1.38)	0.81 (0.36 to 1.81)
Gastro-oesophageal reflux disorder	1.31 (0.91 to 1.87)	**Ref**	0.85 (0.5 to 1.45)	1.40 (0.84 to 2.35)	**3.36 (2.13 to 5.29)**	0.95 (0.59 to 1.53)	Ref	1.05 (0.49 to 2.22)	1.63 (0.84 to 3.18)	1.80 (0.92 to 3.49)	1.22 (0.58 to 2.55)	1.21 (0.50 to 2.93)
Dysphagia	**1.46 (1.01 to 2.12)**	**Ref**	**2.35 (1.30 to 4.25)**	**3.58 (1.99 to 6.44)**	**10.50 (6.13 to 17.98)**	0.96 (0.58 to 1.59)	Ref	1.24 (0.61 to 2.50)	1.17 (0.60 to 2.27)	1.04 (0.53 to 2.03)	1.39 (0.69 to 2.8)	0.94 (0.38 to 2.32)
Lower respiratory tract infection	0.9 (0.62 to 1.30)	**Ref**	0.78 (0.46 to 1.32)	0.76 (0.43 to 1.34)	**2.51 (1.58 to 3.99)**	**1.87 (1.20 to 2.92)**	Ref	0.68 (0.35 to 1.32)	0.64 (0.35 to 1.17)	0.63 (0.34 to 1.17)	0.63 (0.32 to 1.23)	0.78 (0.34 to 1.78)
Dyspnoea	**2.07 (1.42 to 3.03)**	**Ref**	0.95 (0.59 to 1.52)	1.03 (0.63 to 1.69)	**0.38 (0.19 to 0.75)**	1.12 (0.70 to 1.8)	Ref	1.25 (0.54 to 2.88)	1.29 (0.60 to 2.77)	**2.43 (1.16 to 5.06)**	1.30 (0.57 to 2.94)	2.30 (0.98 to 5.44)
Dental health	0.90 (0.62 to 1.31)	Ref	0.88 (0.54 to 1.42)	1.15 (0.70 to 1.88)	0.65 (0.3 to 1.16)	1.28 (0.80 to 2.04)	Ref	1.90 (0.92 to 3.94)	0.97 (0.47 to 2.01)	1.51 (0.75 to 3.04)	1.21 (0.56 to 2.58)	1.31 (0.54 to 3.15)

Numbers in bold are significant results.

had a thyroid disorder, which is more common than several of the other conditions listed in table 2.

We are unclear why the figures appear to show slightly higher rates of multimorbidity in the 45–54 year group for men with moderate intellectual disabilities and women with mild intellectual disabilities and the apparent high rate for young women with moderate intellectual disabilities.

Strengths and limitations

Strengths of the study are the systematic and detailed health assessments by trained health professionals, the comprehensive ascertainment of the population with intellectual disabilities, large sample size and high participation rate. Of the five adult studies out of 52 studies included in a recent meta-analysis on the prevalence of intellectual disabilities,[27] moderate-to-profound intellectual disabilities were reported to account for 65%–66% of the adults with intellectual disabilities in these studies, compared with 61% in ours; that is, our rates are similar. Although the study was only conducted in one area of Scotland, it is likely that the findings are generalisable to other high-income countries. One drawback of detailed health assessments is that looking for more conditions will result in more conditions being identified. This is both a strength—as conditions are frequently overlooked in this population—but also contributes to the high prevalence of multimorbidity that was identified. We did not include mental health conditions in this study as this information has been previously published elsewhere.[26] Previously published intellectual disabilities papers on multimorbidity varied in terms of whether/the extent to which they included mental health.

Implications of the study for clinicians

In the UK, secondary healthcare is organised around single conditions. This can result in lack of coordination between secondary healthcare providers, impeding patient safety. Medical education is also focused on assessment and management of single conditions, yet management of multimorbidity is far more complex. The most prevalent health conditions in adults with intellectual disabilities differ from those seen in the general population, so the recent work to better understand and address multimorbidity[5] does not transfer readily to the population with intellectual disabilities. This study, therefore, starts to address an urgent need to better understand the pattern of multimorbidity in adults with intellectual disabilities that is important because it impacts on healthcare. For example, osteoporosis, which can lead to multiple fractures and non-healing of bones, is treated by bisphosphonates, but people with gastro-oesophageal reflux disorder are unlikely to tolerate them; both these conditions are in the top 20 list of conditions. People with dysphagia may be unable to take medication in tablet form for a wide range of conditions. Psychotropic drugs are commonly prescribed as mental ill health has a point prevalence of 40.9%[26] in people with intellectual disabilities, but their side effects include visual disturbance, weight gain, lowered seizure threshold, constipation and ataxia—the top five conditions. It is important to note that the top 20 physical health conditions reported are known to be painful, disabling and/or life threatening and can significantly impact on quality of life; the majority of these conditions are amenable to treatment, if high quality care is provided. It is vital that healthcare professionals and carers have increased awareness of the presentation and demographics of commonly occurring conditions in adults with intellectual disabilities so that they can identify and report physical health conditions in a timely manner and thus prevent unnecessary suffering.

NICE guideline 56 on multimorbidity[30] highlights that groups of conditions where treatment is discordant pose more problems of coordination and that people who are usually cared for by specialist services that tend to focus on particular types of morbidity (such as mental health in intellectual disabilities services) pose particular difficulties in management of care. Improved evidence on the multimorbidity experienced by adults with intellectual disabilities, throughout all stages of their adulthood, is therefore crucial. The findings have the potential to support policy and practice change to ensure comprehensive continuity of care in the lives of people with intellectual disabilities especially as more and more begin to live to old age. Improving healthcare provision can only contribute to making the lives of people with intellectual disabilities better.

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