Open access Research

# BMJ Open Quantitative examination of the bone health status of older adults with intellectual and developmental disability in Ireland: a cross-sectional nationwide study

Éilish Burke, <sup>1</sup> Rachael Carroll, Máire O'Dwyer, James Bernard Walsh, <sup>3</sup> Philip McCallion, 4 Mary McCarron<sup>5</sup>

To cite: Burke É. Carroll R. O'Dwyer M, et al. Quantitative examination of the bone health status of older adults with intellectual and developmental disability in Ireland: a cross-sectional nationwide study. BMJ Open 2019;9:e026939. doi:10.1136/ bmiopen-2018-026939

Prepublication history and additional material for this paper are available online. To view these files, please visit the journal online (http://dx.doi. org/10.1136/bmjopen-2018-026939).

Received 27 September 2018 Revised 10 January 2019 Accepted 27 February 2019

#### **ABSTRACT**

Objectives (1) To investigate the prevalence of osteopenia and osteoporosis among adults with intellectual disabilities (IDs) and (2) to examine alternative optimal bone screening techniques.

**Design** Observational cross-sectional study. Setting Wave 2 (2013–2106) of the Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing. **Participants** A national representative sample of 604 male and female persons with ID aged 43 years and over. In total, 575 participants completed quantitative ultrasound (QUS) measurements for one or both feet.

Outcome measures Participants underwent health assessments consisting of eight objective health measures including the standardised QUS of the calcaneus bone using a GE Lunar Achilles. A preinterview questionnaire and face-to-face interview were also completed. Results Objectively QUS identified poorer rates of bone

health in people with ID overall with 74% indicating evidence of osteopenia (33.2%) or osteoporosis (41%). Females scored lower than males in the QUS t-scores -2.208 (±1.77) versus -1.78(±1.734). Bone status was stratified by gender (p=0.114), age (p=0.003), level of ID (p<0.0001) and living circumstance (p<0.0001).

Conclusions This study has shown the prevalence of poor bone health in people with ID is substantial implying an increased risk of fracture due to reduced skeletal integrity. QUS screening has been shown to be useful when combined with clinical risk factors.

### Check for updates

@ Author(s) (or their employer(s)) 2019. Re-use permitted under CC BY-NC. No commercial re-use. See rights and permissions. Published by BMJ.

For numbered affiliations see end of article.

### **Correspondence to**

Dr Éilish Burke; eburke7@tcd.ie

#### INTRODUCTION

Osteoporosis is a progressive bone disease characterised by low bone mineral density (BMD) and microarchitectural deterioration, leaving bone susceptible to fragility fractures. 1 Due to the silent nature of the condition, many people are unaware of its presence and yet Ireland has no related national osteoporosis strategy.<sup>2</sup> Studies among people with intellectual disability (ID) investigating osteoporosis have been relatively restricted

#### Strengths and limitations of this study

- ► The participant sample is representative of the national intellectual disability (ID) population in Ireland and sufficiently large to be statistically significant.
- The completion of a detailed questionnaire and interview by participants provided a means to examine potential confounders.
- The use of GE Lunar Achilles quantitative ultrasound (QUS) as an alternative to dual energy X-ray densitometry to evaluate the bone health for people with ID could alleviate challenges of bone assessment in this vulnerable population. While it is acknowledged that QUS is not the 'gold standard' for measurement, this chosen method complied with the standards and position defined by the International Society of Clinical Densitometry.
- The participant's reported their doctor's diagnosis of osteoporosis therefore human error is possible. Fracture data were collected from medical records. Additional unrecognised or unreported fractures could have occurred, especially in those with a severe/profound level of ID who may have been unable to report.
- For the purposes of this paper, exploring risk factors fell outside the remit and were not considered.

to small unrepresentative samples.3 4 Findings in Ireland have ranged from 1% to 8% of those assessed having a preexisting diagnosis of osteoporosis.<sup>5</sup> Zylstra et al<sup>6</sup> did investigate the prevalence of osteoporosis in a larger group of adults with ID living in the community (n=298) and identified rates of osteoporosis of the femur at 17.1% and osteopenia (low bone mass) at 51%. Diagnosis is challenging as many people with ID are unable to express their symptoms, may fail to report symptoms or may find assessment and diagnostic processes, for example, dual energy X-ray densitometry (DXA), difficult or frightening.<sup>78</sup> Health challenges such as complex comorbidity and communication difficulties often contribute to undiagnosed and unmet health needs among people with ID, <sup>910</sup> increasing their risk of undiagnosed osteoporosis. 11-13 Lohiya *et al* 14 reports that almost 40% of their study participants were unable to have BMD measured due to physical and behavioural issues. Given the challenges of assessment and the sometimes unsuitability of DXA, there is no robust assessment protocol for people with ID. Alternate screening methods are rarely utilised and many people with ID progress without proper bone health assessment. 12 15 In recent decades, new technology has developed for the evaluation of skeletal health, namely, QUS. These low-cost, radiation free devices give complete measurements in 10s. As an alternate method of bone health screening, the OUS can give an indication of bone quality, contribute to identifying the risk of fracture and combined with assessment of clinical risk factors, can contribute to the implementation of prevention strategies and/or treatment. 16 17

#### Osteoporosis assessment and quantitative ultrasound (QUS)

Establishing and identifying people at risk of osteoporotic fracture reflects a comprehensive clinical approach to osteoporosis management. DXA, particularly of the femoral neck, is the 'gold standard' and is commonly used in the identification and classification of osteoporosis and osteopenia. Incorporated with widely used clinical risk factor assessment tools such as FRAX, DXA is also utilised for monitoring any skeletal changes overtime. 18-21 The International Society of Clinical Densitometry (ISCD) established an official position on QUS in the management of osteoporosis<sup>22</sup> where the use of OUS is justified in the absence of DXA. However, they acknowledge the technological diversity between DXA and QUS and between the varieties of QUS devices that are available as well as the difficulties in equal comparison. The only validated skeletal site recommended for an ultrasound scan by ISCD is the os calcis.

Using validated devices in conjunction with assessment of clinical risk factors, QUS is affirmed to provide a reliable and convenient method for the evaluation of osteoporosis risk. <sup>17</sup> <sup>23–25</sup>

The life expectancy of people with ID has extended and the potential for conditions associated with ageing, such as osteoporosis, is increasing. Frequently, osteoporosis is diagnosed post an initial clinical fracture, by which time bone quality is already substantially compromised. For people with ID, fracture can contribute to an already established impairment and impact on overall health, quality of life and loss of independence. Considering that investigations among the ID population are few, establishing prevalence will contribute to building a greater understanding of the clinical picture of the bone health status of adults with ID in Ireland. Therefore, the primary objectives of this paper are:

1. To investigate the prevalence of osteopenia and osteoporosis among adults with ID.

2. To examine an alternative bone screening technique.

#### METHODS Study design

The data for this study were drawn from the Intellectual Disability Supplement to The Irish Longitudinal Study on Ageing (IDS-TILDA). IDS-TILDA is a nationally represented longitudinal study examining the health and well-being of adults of all levels of ID aged 40 years and over. All participants were randomly selected from the National Intellectual Disability Database (NIDD) which is a national service-planning databases where individuals with ID register to inform decision-making in relation to planning of specialist services. It is managed by the Health Research Board of Ireland and captures demographic data, level of ID and current service provision. The NIDD provided the sampling frame for the IDS-TILDA study.

#### **Sample**

All participants from IDS-TILDA (n=708) were invited to participate in a suite of health assessments as part of the second wave of data collection. In total, 604 participated in the overall health assessments. Of those 575 participants completed QUS of one or both feet. See online supplementary figure 1.0 for QUS participation flowchart.

#### **Data measurements and data collection**

All participants invited to the health assessments received easy read explanatory information prior to attending. On the day of the assessments, full explanation was given using accessible material, demonstration and alternate communication methods such as hand over hand. Written consent was obtained from those participants capable; however, it was also recognised that some participants were unable to provide written consent. For these individuals, the family member/carer supporting them would consent on their behalf and provide support to the researcher on best communication methods. The assessor also used a system of process consent whereby consent was reaffirmed before each assessment was conducted.

IDS-TILDA collects data every 3 years, data collection tools used include a preinterview questionnaire which captures general demographic information, age, gender, level of ID and living circumstance; doctor's diagnosis of chronic conditions, prescribed medications, personal and family history of fracture and healthcare utilisation. The face-to-face interview captures physical and behavioural health, mental and cognitive health, social participation, functional limitation, physical activity and occupation and finally the newly included objective health assessment. The health assessment consisted of eight objective health measures which included QUS of the calcaneus bone using a GE Lunar Achilles. The fieldwork was carried out by researchers with extensive experience working with people with ID who received comprehensive training on the overall study protocol, interviewing techniques and consent.

A pilot study was carried out on the entire research design of the health assessments. The IDS-TILDA team also worked closely with focused groups involving people with ID in developing layout and easy read material. The Scientific Advisory Committee of the study, who are a group of experts working with people with ID, provided advice on development of protocols, questionnaires, ethics and design of material and instruments used within the study to ensure content and face validity was established. The QUS device utilised in this study met the official position of the ISCD. For quality control and the evaluation of precision, the QUS device was calibrated on a daily basis by using a phantom during the period of screening. 22

#### **Determining bone quality**

A GE Lunar Achilles Ultrasonometer was utilised in this study to measure ultrasound variables of the peripheral skeleton namely the os calcis and a standardised procedure was employed for each participant. All measurements were conducted by the same assessor adding to the validity of the process. Full easy-read information was presented to the participant. Where necessary the participant observed a demonstration of the QUS measurement. The measurement took 10s to complete.

The GE Lunar device has built-in reference values based on age and gender of a healthy female Caucasian adult. The references were supplied by the manufacturer and the assessor calibrated the device based on these reference values. The device provides three QUS parameters namely broadband ultrasonic attenuation (dB/MHz), speed of sound (in m/s) and stiffness index (SI, %) which the machine calculated and expressed as a QUS t-score. For the purposes of this study, the t-score calculated from the SI identified by the device are divided into three outcome categories. These classifications are used for research purposes only and not in a diagnostic capacity.

- 1. Normal: low risk of fracture.
- 2. Osteopenia: moderate risk of fracture.
- 3. Osteoporosis: at high risk of fracture.

Previous studies have noted a difference in QUS results between the left and right foot. The non-dominant foot is recommended for testing; however, people with ID found it difficult to identify conclusively their non-dominant foot. Therefore, at the time of their QUS assessment, both feet were screened where possible. Where obvious contraindications by pathology were noted only one foot measurement was taken.

Great care was taken in standardising the method of measurement and ensuring quality control in accordance to the manufacturer's instructions. As stated at the start of each health assessment day, the QUS was calibrated and quality assessed using the phantom procedure as advised by the manufacturer.

#### **Categorising medicines**

A pharmacist researcher provided medication data capture training to field researchers to enable verification

of medicines data. In the pre interview questionnare (PIQ), participants/proxies were asked 'Can you tell me what medications (including prescribed and over the counter, herbal medicines) you take on a regular basis like every day or every week? <sup>27</sup> These medication data were then confirmed by field researchers at the face-toface interview. In most cases (92.8%), these data were recorded by proxy. Medicines were recorded by brand or generic name, including prescription and non-prescription and over-the-counter medicines, dose of medications and duration. All data were anonymised and medications data received were coded using the WHO Anatomical Therapeutic Chemical (ATC) classification system, and checked by two pharmacists. Medications that specifically decreased or improved bone health were further grouped into three categories:

- Taking at least one medicine that contributed to bone loss.
- Taking at least one treatment medicine for osteoporosis.
- 3. Taking supplementation.

#### Statistical analysis and data quality

All data were entered into the SPSS V.22 for analysis. A quality checking and data integrity procedure was employed which involved the checking of five designated identifiers of the total sample against the hard copy record of each participant by the assessor and an independent investigator. With regards missing values only valid percentages are presented in this paper. The researcher then examined the results of the QUS and the lower of the overall individual scores was chosen as the score of choice. The scores were then categorised as previously stated. Correlation coefficients were applied to evaluate the relationship between QUS score with demographics, history of fracture, attendance for DXA scan, medication use and doctor's diagnosis of osteoporosis. Alpha was set at ≤0.05. The Strengthening the Reporting of Observational Studies in Epidemiology standardised reporting guidelines for cross-sectional studies has been followed.<sup>2</sup>

#### Patient and public involvement

IDS-TILDA advances public involvement through a person-centred approach which empowers people with ID, their carers, service providers and families to be visible partners in cocreated research. This includes active involvement in priority setting, cocreating easy-read and accessible research materials (eg, computer-aided personal interview questionnaires) the conduct of research and sharing and applying research results. The study includes a Public, Patient Involvement Working Group to develop a public patient involvementstrategy.

#### **RESULTS**

A total of 575 (81.2% of total Wave 2) participants engaged in QUS, 57.4% (n=330) were female and the majority of participants were between the age of 50 and 64 years

(50.8%, n=229). Objectively measured QUS identified poorer rates of bone health overall, over 74% indicating objective evidence of osteopenia (33.2%, n=191) or osteoporosis (41%, n=236). In total, 20.9% (n=116) reported a history of fracture. The mean score of the stiffness index was 72.122 ( $\pm 23.186$ ). With regards QUS t-score the mean score was -2.02 ( $\pm 1.76$ ) with females scoring lower than males, -2.208 ( $\pm 1.77$ ) versus -1.78 ( $\pm 1.734$ ). Full participant profile and skeletal health can be viewed in table 1.

As seen in table 2, objectively measured bone status was stratified by age, level of ID and living circumstance. Overall, proportionately more females (44.5%, n=147) were identified as having objective evidence of osteoporosis than men (36.3%, n=89). There was a definite age gradient within the osteoporosis category not in the osteopenia category. Level of ID appeared to influence osteoporosis scores with over 60% of those with severe/profound level of ID presenting within the osteoporosis category (62.7%, n=96).

DXA attendance data (n=142) and history of fracture (n=116) are presented in table 3. Overall, 18% (n=38) of men and 35.4% (n=104) of women reported having a DXA scan. Fewer people who attended reported a severe/profound level of ID (20.1%, n=29) compared with those with mild (31.5%, n=34) or moderate level of ID (31.3%, n=68). Nearly equal proportions of those living independently/family attended as those living in community group home or in residential settings. Almost a quarter presented with objective evidence of osteopenia (23.4%, n=39) and over a third of those with objective evidence of osteoporosis (34.6%, n=72) reported having a DXA scan. Positively, the majority of those who had a doctor's diagnosis of osteoporosis also had a DXA scan (83.1%, n=64/77).

Of those who reported a fracture (116 participants), 35.5% (n=38) reported having a DXA scan. Hence, 67.2% (n=78) of participants did not; however, the question asked included any history of fractures, so it is difficult to ascertain if these were specifically osteoporotic type fractures. Overall, hip fracture was one major osteoporotic type established and was reported as 9.5% (n=11) of the total reported fractures. Examining the history of fracture, there were almost equal reported prevalence among males, 21.7% (n=51) versus females, 20.4% (n=65). Increasing age was statistically associated with experiencing a fracture (p=0.03) and having objective evidence of poor bone health (osteopenia or osteoporosis), p=0.005.

On examination of the medications, 41% (n=239) of participants reported taking antiepileptic medicines (AEDs), 25.4% (n=146) were prescribed proton pump inhibitors (PPI) and a fifth, 20.3% (n=117) reported receiving selective serotonin reuptake inhibitors (SSRI, ATC Code N06AB), that is, medicines considered to contribute to bone loss (table 4). In total, 24.9% (n=143) of participants reported treatment for osteoporosis, most taking a calcium and vitamin D combination 21.4% (n=123), followed by bisphosphates (7.1%, n=41). Of all those who reported their

**Table 1** Profile and skeletal health profile of participants who had quantitative heel ultrasound (n=575)

Demographic profile	n	%	Totals*
Gender			
Male	245	42.6	575
Female	330	57.4	
Age (years)			
43–49	163	28.3	575
50–64	229	50.8	
65+	120	20.9	
Level of ID			
Mild	123	23.3	528
Moderate	252	47.7	
Severe/profound	153	29.0	
Living circumstance			
Independent/family	87	15.3	569
CGH	243	42.7	
Residential	239	42.0	
Skeletal health profile			
Doctor's diagnosis of osteoporosis			
Yes	81	14.4	562
Objective measured Ql	JS		
Normal	148	25.7	575
Osteopenia	191	33.2	
Osteoporosis	236	41.0	
Attended DXA scan			
Yes, within 2 years	102	18.2	559
Yes, over 2 years	40	7.2	
No	363	64.9	
Don't know	54	9.7	
History of fracture			
Yes	116	20.9	554
Accident and emergency	attendance		
Fracture	13	2.4	552
No	438	79.1	
Sprain	8	1.4	
Multiple injuries	2	0.4	

Quantitative ultrasound	Mean±SD
BUA (dB/MHz)	97.092±17.937
SOS (m/s)	1522.64±51.242
Stiffness index	72.122±23.186
QUS t-score	-2.026±1.767
Male QUS t-score	-1.78±1.734
Female QUS t-score	-2.208±1.770

<sup>\*</sup>Not all participants provided information on all questions asked.

BUA, broadband ultrasonic attenuation; CGH, community group home; ID, intellectual disability; QUS, quantitative ultrasound; SOS, speed of sound.

 Table 2 Objectively measured QUS bone status stratified by gender, age, level of ID and living circumstances

 Normal
 Osteopenia
 Osteoporosis

 Demographic (n= 575)
 Total
 n
 %
 n
 %
 n
 %
 P value\*

 Gender
 0.114
 Male
 245
 71
 29.0
 85
 34.7
 89
 36.3

 Female
 330
 77
 23.3
 106
 32.1
 147
 44.5

 Age (years)
 0.003

Male	245	71	29.0	85	34.7	89	36.3	
Female	330	77	23.3	106	32.1	147	44.5	
Age (years)								0.003
43–49	163	54	33.1	56	34.4	53	32.5	
50-64	292	74	25.3	100	34.2	118	40.4	
65+	120	20	16.7	35	29.2	65	54.2	
Level of ID†								<0.0001
Mild	123	46	37.4	43	35.0	34	27.6	
Moderate	252	71	28.2	92	36.5	89	35.3	
Severe/profound	153	18	11.8	39	25.5	96	62.7	
Living circumstances†								<0.0001
Indep/family	87	40	46.0	25	28.7	22	25.3	
CGH	243	68	28.0	91	37.4	84	34.6	
Residential	239	36	15.1	70	31.0	129	54.0	

<sup>\*</sup> $\alpha$ =0.05 with statistically significant values in bold.

medication use, 4% (n=23) were taking supplementation in the form of vitamin D.

The association of medication use with objective evidence of low bone quality as measured by the QUS, history of fracture and doctor's diagnosis of osteoporosis was also examined (table 5). In total, 64% (n=368) of those who completed QUS measures reported taking medicines that contribute to bone loss. Of those who had objective evidence of osteopenia (n=191), almost two-thirds (61.3%, n=117) reported medicines that contribute to bone loss; and of those who had objective evidence of osteoporosis (n=236), over three-quarters, 77.5% (n=183), were prescribed medication such as AEDs, PPIs or SSRIs. Conversely, for those with objective evidence of osteopenia or osteoporosis, 25.1% (n=48) and 30.1% (n=71), respectively, were being treated for osteoporosis, with a further 5.2% (n=10) and 3.0% (n=7), respectively, on vitamin D supplementation.

In summary, when examining the association of medication use with history of fracture, 71.6% (n=83) were prescribed medicines that contribute to bone loss, 29.3% (n=34) were on treatment for osteoporosis and 3.4% (n=4) were taking supplementation. Of those who reported doctor's diagnosis of osteoporosis (n=81), 80.2% (n=65) were taking medicines with potential to induce bone loss. Three-quarters of those with a diagnosis (61 participants, 75.3%) were on treatment for osteoporosis and 9.9% (n=8) were taking vitamin D supplement alone.

## DISCUSSION Principal findings

This study assessed the bone status of participants in IDS-TILDA. In addition to the objective measurement, participants reported if they had a doctor's diagnosis of osteoporosis, had a history of fracture, had attended for DXA and the medications they were prescribed. The findings suggest an increase in the prevalence of osteoporosis in people with ID. While this was independent of gender, dependencies on age, level of ID, residential type accommodation and medication effect were observed, as well as a potential limitation of DXA use for people with ID.

### Strengths, weaknesses and results compared with other studies

#### Objectively measured bone health

Wave 1 of the IDS-TILDA study identified a prevalence of doctor's diagnosis of osteoporosis at 8.1% which had doubled to 16.4% by Wave 2. 30 With regards to objective evidence of osteoporosis, over 4 in 10 presented with an at risk and concerning QUS t-score  $\leq$  -2.5, confirming previous findings of 43.7% by Bastiaanse *et al*. 41 who also utilised an Achilles QUS. This differs greatly from the osteoporosis prevalence of 16% identified by Vice *et al*. 42 based on BMD identified through a chart review of DXA diagnosis. However, this could be underestimated because of difficulties people with ID have accessing DXA. In this study, there was no significant gender effect on prevalence of QUS measured osteoporosis with 36.3% males versus 44.5% females. Conversely, the study of Bastiaanse

<sup>†</sup>Obs missing=notall participants provided level of ID or living circumstance.

CGH, community group home; DXA, X-ray densitometry; ID, intellectual disability; QUS, quantitative ultrasound.



Table 3 Profile of those who had a DXA scan and of those who reported a history of fracture

	Attend	ded for DX	A scan		Has a	a history o	f fracture	
Demographic (n= 575)	n	%	No in sample*	P value	n	%	No in sample*	P value <sup>α</sup>
Gender				0.0001				0.705
Male	38	18.0	211		51	21.7	235	
Female	104	35.4	294		65	20.4	319	
Age (years)				0.0001				0.030
43–49	23	16.7	138		23	14.6	157	
50–64	65	25.3	257		70	24.9	281	
65+	54	49.1	110		23	19.6	116	
Level of ID*				0.044				0.125
Mild	34	31.5	108		33	27.3	121	
Moderate	68	31.3	217		49	20.0	245	
Severe/profound	29	20.1	144		26	17.4	149	
Living circumstances*				0.632				0.204
Indep/family	18	23.7	76		23	26.4	87	
CGH	62	29.4	211		42	17.8	236	
Residential	62	28.4	218		51	22.1	231	
OM bone status				0.025				0.005
Normal	31	23.8	140		21	15.0	140	
Osteopenia	39	23.4	167		32	17.3	185	
Osteoporosis	72	34.6	208		63	27.5	229	
Doctor's diagnosis of osteoporosis				0.0001				0.189
Yes	64	83.1	77		20	26.3	76	
No	77	18.2	422		93	19.7	471	
History of fracture				0.040				
Yes	38	35.5	107		-	-	-	-
No	98	25.5	385		-	-	-	

\*Not all participants provided all data.  $\alpha$ =0.05.

CGH, community group home; DXA, X-ray densitometry; ID, intellectual disability; OM, objectively measured.

et  $al^{31}$  in Rotherdam reported significant prevalence rates of 38.7% males and 49.5% females.

This study also established that increasing age is significantly associated with reduced bone health, although a definitive age gradient was only found for the presence of QUS categorised osteoporosis. However, poor bone health in this study was not a condition solely of old age but seen right across all ages from 43 years onwards. In Ireland, comparative general population prevalence rates based on QUS measurement are available through TILDA: 'The Irish Longitudinal Study on Ageing'. 33 The rates of QUS measured osteoporosis in this study are substantially higher than TILDA who report 3% of males and 13% of females compared with 36.3% and 44.5% in this study. These considerable differences demonstrate the extent of the underlying and unrecognised issue of poor bone health in the Irish population of adults with ID. Of great concern is the unrecognised high prevalence

among men. The International Osteoporosis Foundation reported projections that place men on a trajectory of continuous increased risk of fracture in comparison to women.<sup>34</sup> A similar picture is emerging in this study with a profound message for men with ID and their carers that osteoporosis is not exclusively a female condition.

A significant association was found between poorer bone quality and more severe level of ID with objective evidence of osteoporosis in over 60% of those with severe/profound ID, compared with 35% with moderate ID and 27.6% with mild ID. This is not surprising as those with a severe/profound level of ID are more likely to have mobility issues and greater physical impairment and are more likely to present with polypharmacy and multimorbidity. The grant of the level of ID individuals with ID are at risk of osteoporosis. However, those with severe/profound are at greater risk. Another factor to consider is the issue of immobility, which contributes greatly to bone

Table 4 Medicines promotion a	nd inhibiting	bone loss
Type of medicine	n	%
Contributors to bone loss (CBL)		
Anti-epileptic (AEDs)	239	41.6
Proton pump inhibitors	146	25.4
SSRI	117	20.3
Barbiturates	19	3.3
Lithium	16	2.8
Oral prednisolone	5	0.9
Tamoxifen	2	0.3
Methotrexate immunosuppressant	2	0.3
Anastrazole	2	0.3
Medroxyprogesterone	2	0.3
Taking one or more type of CBL	medicine	
Totals	368	64.0
Treatment for osteoporosis		
Calcium and vitamin D	123	21.4
Bisphosphates	41	7.1
Calcium only	15	2.6
Desosumab	8	1.4
Teripartide	1	0.2
Taking one or more treatment me	edicine	
Totals	143	24.9
Supplementation		
Vitamin D only	23	4.0
Totals	23	4.0

AEDs, antiepileptic medicines; SSRI, selective serotonin reuptake inhibitors.

status.<sup>38</sup> Those with a more severe/profound ID are more likely to be immobile. The implications of lack of exercise in this group as a contributor to poor bone status needs to be highlighted with carers and support workers.

Those living in residential type accommodation were more likely to present with evidence of osteoporosis (54%). This is expected because those living in more supported type of accommodation tend to be of an older generation and can present with more severe/profound level of ID and have increased health needs. <sup>26</sup> <sup>39</sup>

Several studies in the general population have identified an association of AED, PPI and SSRI medications particularly psychotropic medicines with compromised bone integrity. These medicines interfere with calcium and vitamin D absorption contributing to bone loss. The longer these medicines or a combination of such medicines are taken, the higher the risk. The evidence emerging in this study is that there is a significant association between taking medicines that contribute to bone loss, with over three-quarters of those exposed to these medicines presenting within the QUS category

osteoporosis. While two-thirds of the participants were on these medications, less than a third with objective evidence of poor bone health were on treatments such as calcium and vitamin D combinations or bisphosphonates. Disturbingly levels of vitamin D supplementation were poor at 4% (n=23). Sunshine levels are inadequate in Ireland and Irish national health policy supports supplementation. For those with ID who may infrequently go out in the sun, the likelihood of obtaining sufficient sunshine to synthesis sufficient vitamin D is even more limited. However, Tohill and Laverty<sup>43</sup> highlight the difficulties and sometimes impracticality of sun exposure for people with ID, especially those with more severe/profound ID. They suggest that diet is more crucial in these cases and by raising awareness of the importance of vitamin D and incorporating a vitamin D rich diet and exercise into people's daily care plan, improvement to bone health can be achieved.

#### **Consequence of poor bone health**

Fracture occurs as a direct result of bone compromise from trauma. Frequently, osteoporotic fracture is a result of low trauma. Consequently, this will result in a hospital admission which will contribute to distress and pain. Identified fracture rates in this study are high. Over one-fifth of participants had a history of fracture, yet only 27.9% (n=38/116) with a history of fracture attended for diagnostic assessment (DXA). Fracture is the direct result of bone fragility and can lead to reduced quality of life, pain and often surgical intervention. In Ireland, the Health Service Executive projects that for the general population the number of hospital admissions directly as a result of falls and fractures will almost double if current trends continue and the impact on an already overburdened health service could be overwhelming. There are no specific national figures for people with ID being hospitalised in Ireland. In IDS-TILDA, the number of Accident and Emergency (A&E) admissions for people with ID at 18.7%, as reported by McCarron and colleagues,<sup>27</sup> is substantially higher than TILDA rates (14.9%) with fracture as the main reason for A&E attendance. Males had the highest prevalence of fracture yet males in this study were least likely to be identified with bone health concerns.

Globally, between 30% and 40% of all osteoporotic fractures occur among men including a quarter of all hip fractures, the most serious complication of osteoporosis with a higher level of associated mortality when compared with women. 44 Osteoporotic fractures account for more disability adjusted life years lost than most common cancers (except for lung cancer) and among men the life time risk of experiencing an osteoporotic fracture is almost three times higher than presenting with prostate cancer. 45

#### Screening and bone status

Fracture begets fracture. The need for robust skeletal assessment and management especially post initial

Table 5 Associations of medications with QUS results, history of fracture and those who reported a doctor's diagnosis of osteoporosis

											,					
	Medi	Medications														
	Contr	ibuting to	Contributing to bone loss	ss		Treatn	Treatment for osteoporosis	steopor	osis		Supple	Supplementation	u			Totals
																No in
	Yes		No		P value Yes	Yes		No		P value	Yes		No		P value	sample
Variable*	u	%	u	%		u	%	u	%		u	%	u	%		
QUS measures					0.0001					0.009					0.492	
Normal	89	45.9	80	54.1		24	16.2	124	83.8		9	4.1	142	95.9		148
Osteopenia	117	61.3	74	38.7		48	25.1	143	74.9		10	5.2	181	94.8		191
Osteoporosis	183	77.5	53	22.5		71	30.1	165	6.69		7	3.0	229	97.0		236
Total	368	0.49	207	36.0		143	24.9	432	75.1		23	4.0	552	0.96		575
History of fracture					0.072					0.198					0.669	
Yes	83	71.6	33	28.4		34	29.3	82	7.07		4	3.4	112	9.96		116
No	274	62.6	164	37.4		103	23.5	335	76.5		19	4.3	419	95.7		438
Total	357	64.0	197	35.6		137	24.7	417	75.3		23	4.2	531	92.8		554
Doctor's diagnosis of osteoporosis					0.002					<0.0001					0.005	
Yes	92	80.2	16	19.8		61	75.3	20	24.7		œ	6.6	73	90.1		81
No	299	62.2	182	37.8		81	16.8	400	83.2		15	3.1	466	6.96		481
Total	364	64.8	198	35.2		142	25.3	420	74.7		23	4.1	539	62.9		562

 $\alpha$ =0.05, statistically significant values in bold. \*Not all participants answered all questions. QUS, quantitative ultrasound.

fracture can stop the fragility fracture cycle. Clinically diagnosis has become dependent on DXA BMD. However, this study shows that the majority of participants did not attend for DXA scanning (65%, n=363), while DXA may be contraindicated in some cases. The literature recognises that DXA scanning poses challenges for many people with ID, such as the need to travel to a hospital which can contribute to increased anxiety, non-compliance during the assessment and need to use sedation, all of which increase costs creating additional barriers. Utilising alternate methods such as the QUS, an easier approach and one recommended in the absence of DXA proved acceptable to people in this study.<sup>21</sup>

There are no definitive guidelines for screening people with ID although previous studies have made recommendations. Tyler *et al* suggest screening should begin before the age of 50, as people with ID should be considered a high-risk group. This is supported by Dreyfus *et al* who identified low rates of screening especially among men. Although it must be noted that routine screening in the general population is not recommended, evidence from this study and others <sup>5 46</sup> indicate the need to consider targeted purposeful screening for people with ID.

#### **Future research**

As the aim of screening is to identify risk of fracture and instigate treatment where necessary, further investigation must identify the optimal age at which to screen people with ID and inform the development of a risk matrix. Furthermore, given that screening can contribute to improved health, increased awareness of overall health screening, especially bone status screening among service providers, is required. Osteoporosis screening for men globally is quite low. It is more probable that asymptomatic men are less likely to be screened by comparison to asymptomatic women therefore highlighting the issues of screening for men.

More work is needed to confirm the rates of osteoporosis among men with ID, inform guidelines for screening and increase related education. Awareness and education on the management and amelioration of osteoporosis are not reaching its intended audience. Considering the challenges with literacy and education that people with ID experience and the higher levels of poor bone health identified in this study, the many campaigns undertaken by health authorities are less likely to influence people with ID and their carers. This must be addressed through reasonable adjustment of health promotion, increased service provider awareness and increased emphasis on education for individuals with ID and their support workers.

#### **CONCLUSION**

The prevalence of poor bone health has been identified in this study as considerable, implying that people with ID are at risk of reduced skeletal integrity and subsequent fracture. This may be due to a cascade of disparity, poor health behaviours and lack of assessment. The assessment technique utilised in this study was successful in engaging and establishing the bone health status of a large representative sample of older adults with ID. However, in consideration to cautions specified by clinical bodies such as ISCD, QUS measurement alone given the lack of standardised protocols and the proliferation of different devices, is insufficient for diagnosis. However, there is evidence that with such high prevalence of poor bone health and known difficulties with other screening methods, the successful use of QUS reported here confirms its usefulness when used in combination with clinical risk factors. Further investigations are required to establish confirmed risks for this vulnerable population.

#### **Author affiliations**

<sup>1</sup>School of Nursing and Midwifery, Trinity College Dublin, The University of Dublin, Dublin, Ireland

<sup>2</sup>School of Pharmacy and Pharmaceutical Sciences, Trinity College Dublin, The University of Dublin, Dublin, Ireland

<sup>3</sup>Centre for Medical Gerontology, Trinity College Dublin, The University of Dublin, Dublin, Ireland

<sup>4</sup>School of Social Work, Temple University, Philadelphia, Pennsylvania, USA
 <sup>5</sup>Dean of the Faculty of Health Sciences, Trinity College Dublin, The University of Dublin, Dublin, Ireland

Acknowledgements The authors would like to thank the people with ID who participated in this study, their families, carers, the services involved, the IDS-TILDA Scientific Advisory Committee and the Intellectual Disability Consultative Groups for their support. The authors would also like to acknowledge funding for the IDS-TILDA study from the Health Research Board and the Department of Health.

Contributors EB, RC, MO, JBW, PM and MM contributed to the overall conception and design of the study. EB and RC undertook the data extraction. EB and RC undertook the statistical analyses, EB wrote the first draft of the manuscript. EB, JBW, PM and MM revised the manuscript. MO was the researcher pharmacist who conducted the coding and trained the field researchers in medicine recording. All authors had full access to all of the data (including statistical reports and tables) in the study and can take responsibility for the integrity of the data and the accuracy of the data analysis. All authors contributed to the interpretation of results and drafting of the manuscript. All authors read and approved the final manuscript. EB and MM are the guarantors.

**Funding** This research was funded by the Department of Health in Ireland and managed by the Health Research Board. Grant Number HRA-PHS-2012-14. The funding bodies did not play a role in the study design or writing of the manuscript.

**Disclaimer** The views expressed are those of the authors and are not necessarily those of the Department of Health, The Health Research Board or Trinity College Dublin.

Competing interests None declared.

Patient consent for publication Not required.

**Ethics approval** Ethics for the IDS-TILDA study, which included the health assessments, was obtained from the faculty ethics committee and all participating service providers.

Provenance and peer review Not commissioned; externally peer reviewed.

**Data sharing statement** Due to the nature and small population from which this sample was drawn the participants could possibly be identifiable. All data are held within an encrypted secured drive only accessed by assigned personnel. Those wishing to access may do so with the permission of the PI. All interested researchers may apply to the study.

Open access This is an open access article distributed in accordance with the Creative Commons Attribution Non Commercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work is properly cited, appropriate credit is given, any changes made indicated, and the use is non-commercial. See: http://creativecommons.org/licenses/by-nc/4.0/.

#### **REFERENCES**

- Kanis JA, Melton LJ, Christiansen C, et al. The diagnosis of osteoporosis. J Bone Miner Res 1994;9:1137–41.
- Laffoy M. Strategy to prevent falls and fractures in Ireland's ageing population summary, conclusions and recomendations. Health Service Executive, Dublin 2008.
- Arvio M, Kilpinen-Loisa P, Tiitinen A, et al. Bone mineral density and sex hormone status in intellectually disabled women on progestininduced amenorrhea. Acta Obstet Gynecol Scand 2009:88:428–33.
- Center J, Beange H, McElduff A. People with mental retardation have an increased prevalence of osteoporosis: a population study. Am J Ment Retard 1998;103:19–28.
- Burke EA, McCallion P, Carroll R, et al. An exploration of the bone health of older adults with an intellectual disability in Ireland. J Intellect Disabil Res 2017;61:99–114.
- Zylstra RG, Porter LL, Shapiro JL, et al. Prevalence of osteoporosis in community-dwelling individuals with intellectual and/or developmental disabilities. J Am Med Dir Assoc 2008;9:109–13.
- Angelopoulou N, Matziari C, Tsimaras V, et al. Bone mineral density and muscle strength in young men with mental retardation (with and without Down syndrome). Calcif Tissue Int 2000;66:176–80.
- Srikanth R, Cassidy G, Joiner C, et al. Osteoporosis in people with intellectual disabilities: a review and a brief study of risk factors for osteoporosis in a community sample of people with intellectual disabilities. J Intellect Disabil Res 2011;55:53–62.
- Jasien J, Daimon CM, Maudsley S, et al. Aging and bone health in individuals with developmental disabilities. Int J Endocrinol 2012;2012:1–10.
- Krahn GL, Hammond L, Turner A. A cascade of disparities: health and health care access for people with intellectual disabilities. *Ment Retard Dev Disabil Res Rev* 2006;12:70–82.
- ÉA B, Carroll R, O'Dwyer M, et al. Osteoporosis and People with Down Syndrome: A Preliminary Descriptive Examination of the Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing Wave 1 Results. Health 2018;10:1233.
- Lennox NG, Diggens JN, Ugoni AM. The general practice care of people with intellectual disability: barriers and solutions. J Intellect Disabil Res 1997;41:380–90.
- Kirschner KL, Breslin ML, lezzoni LI. Structural impairments that limit access to health care for patients with disabilities. *JAMA* 2007;297:1121–5.
- Lohiya GS, Tan-Figueroa L, Iannucci A. Identification of low bone mass in a developmental center: finger bone mineral density measurement in 562 residents. J Am Med Dir Assoc 2004;5:371–6.
- Michael J, Richardson A. Healthcare for all: the independent inquiry into access to healthcare for people with learning disabilities. *Tizard Learning Disability Review* 2008;13:28–34.
- Njeh CF, Boivin CM, Langton CM. The role of ultrasound in the assessment of osteoporosis: a review. Osteoporos Int 1997;7:7–22.
- Hans D, Hartl F, Krieg MA. Device-specific weighted T-score for two quantitative ultrasounds: operational propositions for the management of osteoporosis for 65 years and older women in Switzerland. Osteoporos Int 2003;14:251–8.
- Kanis JA, Oden A, Johansson H, et al. FRAX and its applications to clinical practice. Bone 2009;44:734–43.
- Kanis JA. Diagnosis of osteoporosis and assessment of fracture risk. The Lancet 2002;359:1929–36.
- Kanis JA, Johansson H, Oden A, et al. Assessment of fracture risk. Eur J Radiol 2009;71:392–7.
- Kanis JA, Burlet N, Cooper C, et al. European guidance for the diagnosis and management of osteoporosis in postmenopausal women. Osteoporos Int 2008;19:399–428.
- Krieg MA, Barkmann R, Gonnelli S, et al. Quantitative ultrasound in the management of osteoporosis: the 2007 ISCD Official Positions. J Clin Densitom 2008;11:163–87.
- Hans D, Dargent-Molina P, Schott AM, et al. Ultrasonographic heel measurements to predict hip fracture in elderly women: the EPIDOS prospective study. Lancet 1996;348:511–4.
- Collinge CA, Lebus G, Gardner MJ, et al. A comparison of quantitative ultrasound of the calcaneus with dual-energy x-ray absorptiometry in hospitalized orthopaedic trauma patients. J Orthop Trauma 2010;24:176–80.
- Glüer CC, Eastell R, Reid DM, et al. Association of five quantitative ultrasound devices and bone densitometry with osteoporotic

- vertebral fractures in a population-based sample: the OPUS Study. *J Bone Miner Res* 2004;19:782–93.
- Kinnear D, Morrison J, Allan L, et al. Prevalence of physical conditions and multimorbidity in a cohort of adults with intellectual disabilities with and without Down syndrome: cross-sectional study. BMJ Open 2018;8:e018292.
- McCarron M, Swinburne J, Burke E, et al. Growing older with an intellectual disability in Ireland 2011: First results from the Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing (IDS-TILDA). School of Nursing and Midwifery, Trinity College Dublin 2011.
- Hourigan S, Fanagan S, Kelly C. HRB Statistics Series 37 Annual Report of the National Intellectual Disability Database Committee, 2017
- von Elm E, Altman DG, Egger M, et al. The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) Statement: guidelines for reporting observational studies. Int J Surg 2014;12:1495–9.
- Burke E, McCarron M, McCallion P. Advancing years, Different challenges: Wave 2 IDS-TILDA: findings on the ageing of people with an intellectual disability: an intellectual disability supplement to the Irish Longitudinal Study on Ageing. Dublin: Trinity College Dublin, 2014.
- Bastiaanse LP, Mergler S, Evenhuis HM, et al. Bone quality in older adults with intellectual disabilities. Res Dev Disabil 2014;35:1927–33.
- Vice MA, Nahar VK, Ford MA, et al. Risk Factors for Low Bone Mineral Density in Institutionalized Individuals with Developmental Disabilities, Health promotion perspectives. 2015;5:147.
- Barrett A, Burke H, Cronin H, et al. Fifty plus in Ireland 2011: first results from the Irish Longitudinal Study on Ageing (TILDA, 2011.
- 34. Ebeling P. Osteoporosis in men: Why change needs to happen. World Osteoporosis Day Thematic Report International Osteoporosis Foundation, Nyon 2014.
- 35 O'Dwyer M, Peklar J, McCarron M, et al. Prevalence, patterns and factors associated with polypharmacy and excessive polypharmacy in an ageing population with intellectual disability in Ireland. Irish Journal of Medical Science 2013;182:S210. 236 GRAYS INN RD, 6TH FLOOR, LONDON WC1X 8HL, ENGLAND, SPRINGER LONDON LTD.
- McCarron M, Swinburne J, Burke E, et al. Patterns of multimorbidity in an older population of persons with an intellectual disability: results from the intellectual disability supplement to the Irish longitudinal study on aging (IDS-TILDA). Res Dev Disabil 2013;34:521–7.
- O'Dwyer M, Peklar J, McCallion P, et al. Factors associated with polypharmacy and excessive polypharmacy in older people with intellectual disability differ from the general population: a cross-sectional observational nationwide study. BMJ Open 2016;6:e010505.
- 38. Wagemans AM, Fiolet JF, van der Linden ES, et al. Osteoporosis and intellectual disability: is there any relation? *J Intellect Disabil Res* 1998;42:370–4.
- Hess M, Campagna EJ, Jensen KM. Low bone mineral density risk factors and testing patterns in institutionalized adults with intellectual and developmental disabilities. *J Appl Res Intellect Disabil* 2018;31 Suppl 1:157–64.
- Sato Y, Kondo I, Ishida S, et al. Decreased bone mass and increased bone turnover with valproate therapy in adults with epilepsy. Neurology 2001;57:445–9.
- Targownik LE, Lix LM, Metge CJ, et al. Use of proton pump inhibitors and risk of osteoporosis-related fractures. CMAJ 2008;179:319–26.
- Bolton JM, Targownik LE, Leung S, et al. Risk of low bone mineral density associated with psychotropic medications and mental disorders in postmenopausal women. J Clin Psychopharmacol 2011;31:56–60.
- 43. Tohill C, Laverty A. Sunshine, diet and mobility for healthy bones an intervention study designed to implement these standards into the daily routine in an at risk population of adults with intellectual disability. J Intellect Dev Disabil 2001;26:217–31.
- Bliuc D, Nguyen ND, Milch VE, et al. Mortality risk associated with low-trauma osteoporotic fracture and subsequent fracture in men and women. JAMA 2009;301:513–21.
- Johnell O, Kanis JA. An estimate of the worldwide prevalence and disability associated with osteoporotic fractures. Osteoporos Int 2006;17:1726–33.
- Dreyfus D, Lauer E, Wilkinson J. Characteristics associated with bone mineral density screening in adults with intellectual disabilities. *J Am Board Fam Med* 2014;27:104–14.