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The incidence of healthcare use, ill health and mortality in adults with intellectual disabilities and mealtime support needs

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Abstract

Background Adults with intellectual disabilities (ID) experience a wide range of eating, drinking and/or swallowing (EDS) problems, for which they receive diverse mealtime support interventions. Previous research has estimated that dysphagia (difficulty swallowing) affects 8% of all adults with ID and that 15% require some form of mealtime support. People with ID (whether they require mealtime support or not) also experience a greater burden of ill health and die younger than their peers in the general population with no ID.
Methods Using an exploratory, population-based cohort study design, we set out to examine health-related outcomes in adults with ID who receive mealtime support for any eating, drinking or swallowing problem, by establishing the annual incidence of healthcare use, EDS-related ill health, and all-cause mortality. This study was conducted in two counties in the East of England.

Results In 2009, 142 adults with mild to profound ID and a need for any type of mealtime support were recruited for a baseline survey. At follow-up 1 year later, 127 individuals were alive, eight had died and seven could not be contacted. Almost all participants had one or more consultations with a general practitioner (GP) each year (85–95%) and, in the first year, 20% reportedly had one or more emergency hospitalizations. Although their annual number of GP visits was broadly comparable with that of the general population, one-fifth of this population's primary healthcare use was directly attributable to EDS-related ill health. Respiratory infections were the most common cause of morbidity, and the immediate cause of all eight deaths, while concerns about nutrition and dehydration were surprisingly minor. Our participants had a high annual incidence of death (5%) and, with a standardized mortality ratio of 267, their observed mortality was more than twice that expected in the general population of adults with ID (not selected because of mealtime support for EDS problems).
Conclusions All Annual Health Checks now offered to adults with ID should include questions about respiratory infections and EDS functioning, in order to focus attention on EDS problems in this

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population. This has the potential to reduce life-threatening illness.

Keywords deglutition disorder, incidence, intellectual disability, mealtime support, morbidity, mortality

Introduction

Adults with intellectual disabilities (ID) are known to have difficulty accessing high-quality primary and secondary care, making them more vulnerable to undetected and inadequately managed health conditions than their non-disabled peers (Krahn *et al.* 2006). Studies have shown that epilepsy (McGrother *et al.* 2006), gastro-oesophageal reflux disease (GORD) (Bohmer *et al.* 2000), mental health problems (Costello & Bouras 2006), respiratory issues and oral health problems (McCarthy & O'Hara 2011) are overrepresented among people with ID. This distinct epidemiological profile and greater burden of ill health (Emerson & Baines 2010), coupled with communication and self-care difficulties, can complicate the provision of health care for these individuals, with potentially tragic results (Mencap 2007). It is therefore imperative that health and social care professionals, caregivers and others responsible for supporting adults with ID are well informed and proactively monitor their health needs. Premature mortality is another significant and persistent concern, and although their life expectancy is increasing, individuals with ID still die 25 years earlier than people without ID. The median age at death among people with ID in England was 56 years old in 2010, with variation according to aetiology, compared with 81 for the general population (Glover & Ayub 2010; Emerson *et al.* 2012). Respiratory infections are the most common cause of death in this group, and many of these deaths are thought to be linked to dysphagia (swallowing problems) (Tyrer & McGrother 2009; Heslop *et al.* 2013).

Dysphagia is common in people with ID, and an important factor contributing to the need for mealtime support (Table 1). Approximately 15% of adults with ID require mealtime support (Ball *et al.* 2012) and at least 8% (Chadwick & Jolliffe 2009) have dysphagia. Although such problems are thought to be more common in people with ID, at all ages, than in the general population, nationally representative prevalence estimates are still lacking. However, there is good evidence that the epidemiology of these problems differs between adults with and without ID (Sheppard 1991; Chadwick & Jolliffe 2009). Among individuals with ID, eating, drinking and/or swallowing (EDS) problems commonly begin in childhood, are more likely to persist into adulthood and just as likely to worsen or change with older age and the onset of other conditions, such as dementia and loss of dentition (Sheppard 2006).

EDS problems can make mealtimes lengthy, difficult and potentially life threatening. They may also result undernutrition, dehydration and

Table 1 Definitions of key terms, as used in the present study

Eating, drinking and/or swallowing (EDS) problems: A range of issues including dysphagia, a lack of independent feeding skills and behavioural or psychological problems that result in difficulty eating and drinking and interfere with mealtimes.
EDS-related health issues: Any condition or illness that could result from or affect a person's ability to eat and drink. Specific questions were asked about general practitioner and hospital visits for respiratory infections, malnutrition, dehydration and weight loss, as well as specialist referrals for dysphagia or enteral feeding via percutaneous endoscopic gastrostomy (PEG) or jejunostomy (PEJ) tube. Other EDS-related issues and illnesses included gastro-oesophageal reflux disease (GORD), regurgitation, urinary tract infections (UTIs) and bowel problems explicitly linked to dehydration or food refusal by informants.
Mealtime support: The diverse range of assistance that enables safer mealtimes for individuals with eating, drinking and/or swallowing problems. Categorized into four groups: (1) *minimal support* (able to eat independently, with limited support, such as occasional pacing advice, adapted cutlery/crockery and help cutting up food); (2) *moderate support* (able to get food to the mouth with or without help, but requiring more substantial support, e.g. with pacing, positioning and adapted cutlery, to ensure safety/adequate nutrition); (3) *full oral support* (unable to feed self without caregiver support); and (4) *full enteral support* (feeding wholly or partially via PEG/PEJ tube). Broadly defined, individuals requiring minimal or moderate support could feed themselves, whereas those requiring full oral or enteral support could not.

aspiration pneumonia (Chadwick & Jolliffe 2009). For adults with ID, EDS problems are usually managed by caregivers, sometimes under the direction of multidisciplinary community teams, incorporating specialist input from speech and language therapists (SLTs), dietitians, occupational therapists (OTs) and other healthcare practitioners. Previous research has found relatively high levels of caregiver adherence to dysphagia management guidelines (between 77 and 83% overall) (Chadwick *et al.* 2003; Crawford *et al.* 2007), indicating that once problems are recognized and recommendations made, they are taken seriously; yet, they may be under-recognized (Chadwick & Jolliffe 2009).

To our knowledge, no previous studies have investigated EDS problems in relation to mortality or primary and secondary care use among adults with ID, and few studies have explored the health issues attributable to eating and drinking in this population (Chadwick & Jolliffe 2009). A better understanding of this population's current healthcare use and their burden of morbidity and mortality is essential for planning more effective health and social care. With this in mind, we sought to establish the annual incidence of EDS-related healthcare use, health problems and all-cause mortality in adults with ID and EDS problems, as well as the proportion of overall healthcare use attributable to EDS problems. This descriptive study was also intended to precede further analysis identifying predictors of negative outcomes, which will highlight areas for intervention and the prevention of key health issues.

Methods

Study design, setting and data collection procedure

Data collection, ethical considerations and greater detail regarding the types of eating and drinking difficulties experienced by our participants and the assistance required have all been reported previously (Ball *et al.* 2012). This exploratory, descriptive cohort study consisted of two surveys, administered 1 year apart. Potential participants were adults (18+ years of age) who had an ID and required support for any problem with eating, drinking or swallow-

ing. All individuals who met these criteria were identified from the population known to local specialist services for people with ID, or community learning disability teams (CLDTs), in all of Cambridgeshire and North Essex, over 1 year beginning on 31 July 2008 (Cambridgeshire) and 5 December 2008 (Essex). In summary, the process of identifying potential participants involved contacting managers of all CLDTs and providers of social care (nursing homes, group residential care, supported living, domiciliary care and day centres) for adults with ID in Cambridgeshire and North Essex. All eligible individuals were invited to participate in the study. Consent was sought for interviews with caregivers (family carers, paid support workers or care home managers) and healthcare practitioners involved in the provision of mealtime support, and for access to health notes. A sample size calculation was not appropriate for this study, as it is a descriptive exploration of the incidence and prevalence of multiple outcomes in people with ID and mealtime support.

Baseline

Baseline data were gathered in face-to-face interviews with caregivers between January and September 2009, and cross-checked against written care plans and CLDT records. A structured interview form was used to collect cross-sectional information regarding five groups of variables: socio-demographic characteristics, disability-related characteristics, mealtime support needs, indicators of dysphagia and additional illnesses/disabilities (see Table 2 for variables). Severity of ID was taken from CLDT records, although these categories should be viewed as approximate (World Health Organization 1996).

The form also included retrospective questions addressing the 12 months preceding baseline (year 1) on numbers of GP visits (in general and for EDS problems), numbers of emergency and planned hospital visits (for EDS problems), number of emergency hospital visits *not* related to eating/drinking issues and specialist input for EDS problems. We also asked whether participants had *ever* been referred to a specialist for an eating/drinking issue, and about the reasons for all EDS-related GP visits, hospital visits and specialist input, as well as the

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Table 2 Participant characteristics

Participant characteristics		Frequency (column %) (N = 127, missing data vary by variable)
Socio-demographic characteristics	Age at baseline (years):	
	18–45	59 (46.5)
	46–90	68 (53.5)
	Male	70 (55.1)
	County of residence:	
	Cambridgeshire	62 (48.8)
	Essex	65 (51.2)
	Living arrangements:	
	Group social care (including nursing homes)	86 (67.7)
	Supported living	23 (18.1)
	Private accommodation (own/family home)	18 (14.2)
Disability-related characteristics	Severity of ID (N = 126):	
	Mild	15 (11.9)
	Moderate	27 (21.4)
	Severe	68 (54.0)
	Profound	16 (12.7)
	Aetiology of ID:	
	Cerebral palsy	38 (29.9)
	Down's syndrome	18 (14.2)
	Other/unknown cause	71 (55.9)
	Has a physical disability	102 (80.3)
	Extent of mobility:	
	Mobile	36 (28.3)
	Limited mobility	49 (38.6)
	Not mobile	42 (33.1)
	Has difficulty self-feeding	55 (43.3)
Mealtime support factors	Level of mealtime support required:	
	Minimal	19 (15.0)
	Moderate	65 (51.2)
	Full (oral)	31 (24.4)
	Full (enteral)	12 (9.4)
	Stability of mealtime support (N = 126):	
	Stable	63 (50.0)
	Increasing	63 (50.0)
Indicators of dysphagia	Has diagnosed dysphagia (N = 125)	45 (36.0)
	Has carer-reported swallowing problems	58 (45.7)
	Had any clinical features of dysphagia in year 1	73 (57.5)
Other illness / disability	Has dementia (or suspected dementia)	10 (7.9)
	Has epilepsy	42 (33.1)

ID, intellectual disabilities.

advice received from specialists and the length of each hospital admission.

Follow-up

Follow-up interviews were conducted by phone between January and September 2010, 1 year after baseline. We asked participants/informants for the same information as at baseline, excluding questions addressing emergency hospital visits not for eating/drinking, socio-demographic and disability-related characteristics. Questions were added about new dysphagia diagnoses, changes to enteral feeding and each participant's vital status (including, if they had

died, the date and cause). Mortality information was verified using death certificates issued by the UK's General Register Office (GRO). We gathered the same follow-up information from deceased and surviving participants' caregivers; however, we analysed these data separately.

Appendix S1 provides further details regarding the structured interview form and the computation of all variables.

Statistical analysis

We used the Pearson chi-squared test and the two-tailed Fisher's exact test to describe and examine associations between categorical variables. The independent samples *t*-test and Mann–Whitney *U*-test were used, as appropriate, to explore differences between groups. Participants with missing information for specific variables were excluded from analysis on a case-by-case basis. The statistical significance of changes in healthcare use and morbidity from year 1 to 2 were assessed using McNemar's chi-squared and Wilcoxon signed-rank tests for repeated measures. All hypothesis tests were two sided and P-values ≤ 0.05 were considered statistically significant. Point estimates are presented with 95% confidence intervals (CI), using Wilson CIs for proportions (Agresti & Coull 1998). Statistical analysis was performed using IBM SPSS version 19.0 (IBM Corp., Armonk, New York, USA) and Stata version 9.2 (StataCorp, College Station, Texas, USA).

Results

Participants

Recruitment and retention of participants

All 726 people (327 in Cambridge and 399 in Essex) identified in the prevalence study were invited to participate (Fig. 1). However, agreement for participation was received for only 20% (142), all of whom completed the baseline survey. Eight participants died between baseline and follow-up (year 2). We achieved 12-month follow-up for 127 (95%) of the remaining 134 participants. The seven people lost to follow-up and the eight known to have died were excluded from the baseline and

follow-up calculations presented below. For the 127 followed up survivors, the mean length of time between baseline and follow-up was 53.5 weeks [standard deviation (SD) 5.1] with a median of 52 weeks (range: 48–73). Six (4.5%) of the final 127 provided no information about GP visits, but answered all other questions.

Participant characteristics at baseline (year 1)

Table 2 presents the baseline characteristics of the 127 participants who were alive and took part in follow-up at the end of year 2. The mean age of the sample at baseline ($N = 127$) was 46.6 years old (SD 17.7) and the median was 47.0 (range: 18–90 years old). Older adults, aged 46–90, had significantly less severe ID: almost half (43%, 29/68) had a mild/moderate ID, compared with less than one-quarter (22%, 13/58) of 18- to 45-year-olds [$N = 126$, $\chi^2(1) = 4.9$, $P = 0.027$]. Diagnosed dysphagia was not associated with age: 35% of both age groups had dysphagia [$N = 125$, $\chi^2(1) = 0.00$, $P = 1.00$]; or ID severity: 36% of individuals with mild/moderate ID (15/42) and severe/profound ID [30/83, $N = 125$, $\chi^2(1) = 0.00$, $P = 1.00$] were affected. However, diagnosed dysphagia was strongly associated with ID aetiology [$N = 125$, $\chi^2(2) = 13.9$, $P = 0.001$], affecting only about one-quarter of participants with Down's syndrome (DS, 5/18) or unknown/other aetiologies (18/71), compared with nearly two-thirds (22/36) of individuals with cerebral palsy (CP), supporting the well-established relationship between dysphagia and CP (Rogers 2004; Calis *et al.* 2008).

Follow-up (year 2)

One year later, nearly one-third of participants (30%, 38/127) reportedly required more mealtime support than at baseline, although only four people (3% of 127) had newly diagnosed dysphagia and the same number had newly inserted percutaneous endoscopic gastrostomy (PEG) or percutaneous endoscopic jejunostomy tubes.

Incidence and causes of healthcare use

General practitioner visits (all-cause and eating, drinking and/or swallowing related)

Almost all participants saw their GP at least once in either year 1 (94%) or year 2 (85%), with only four
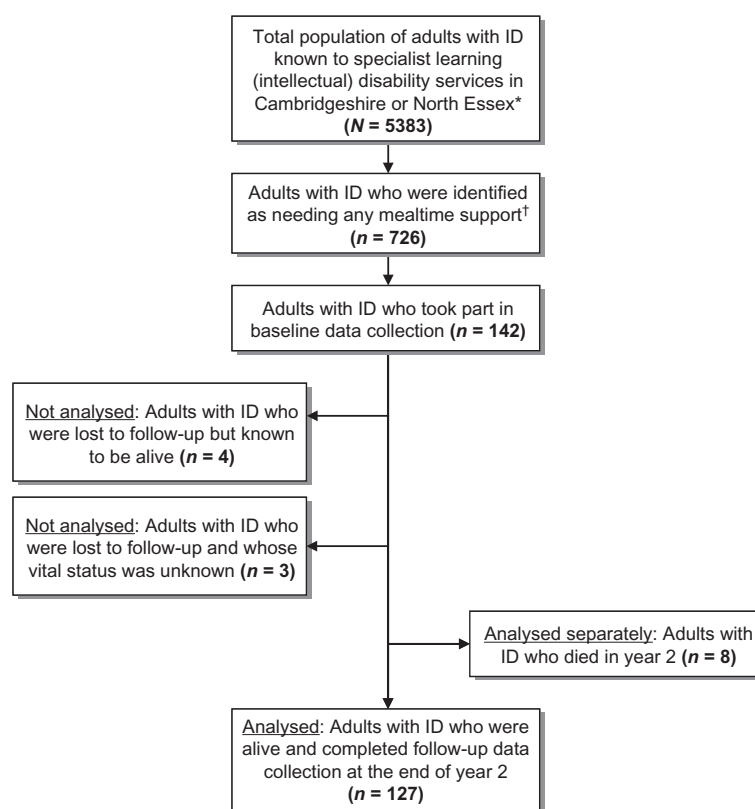
Figure 1 Study population and participant flow.
*Our estimate of the total number of adults with intellectual disabilities (ID) known to specialist services in North Essex (the area currently covered by the West Essex, Mid Essex and North East Essex Clinical Commissioning Groups) was adapted from the 4500 people total known to the Essex County Council (ECC) Learning Disabilities Register on 31 July 2007, taken from: The Essex Partnership (2008) *Essex Joint Strategic Needs Assessment: People Living with Disabilities*. When this total population of 4500 for the entire county of Essex is restricted only to the geographic areas covered by our study (excluding Basildon, Brentwood, Castle Point, Rochford and Thurrock and Southend Unitary Authorities in South/South-east Essex), the population is reduced to 3153. In Cambridgeshire, 2230 adults with ID were known to the Cambridgeshire Learning Disability Partnership on 31 July 2008.
†Identified through clinicians and social care providers over a 1-year period beginning 31 July 2008 in Cambridgeshire and 5 December 2008 in Northeast Essex.

(3% of 120) not consulting their GP for any reason during the 24-month study period. We only gathered data on causes of EDS-related GP visits, where respiratory infections predominated in both years: approximately 80% of all participants who saw the GP for an EDS-related problem (47/58 in year 1 and 30/38 in year 2) reported that respiratory infection was the primary cause.

Although the annual mean number of GP visits (all-cause and EDS related) declined slightly from year 1 to 2 (Table 3), the pattern across age groups remained similar, with stable use from 18 to 74 years, only increasing in the over-75s (Fig. 2). This contrasts with GP consultation data for the general (non-ID population) drawn from the nationally representative 2007 General Household Survey (GHS), wherein the annual rate of consultations in each age group increases up to age 65 (see Fig. 2). Moreover, GHS values for the general population mostly lie between our sample's figures in years 1 and 2. (See Appendix S2 for details.)

Table 3 Health service utilization over a 2-year period: Proportion of the sample with any healthcare contact and the average annual rate of health service use

| Type of health service use	Proportion with any visits/input n (%, 95% Wilson confidence interval)			Mean number of visits per person mean (95% confidence interval)		
	Year 1 (N = 127)	Year 2 (N = 127)	McNemar's χ^2 P-value	Year 1 (N = 127)	Year 2 (N = 127)	Wilcoxon Z P-value
All-cause GP visits	119* (94.4%, 88.7–97.4)	103[†] (85.1%, 77.7–90.4)	0.013	6.08* (5.19–6.97)	4.22[‡] (3.53–4.92)	0.001
GP visits related to EDS	58 (45.7%, 37.3–54.3)	38[†] (31.4%, 23.8–40.1)	0.009	1.24* (0.79–1.68)	0.97[§] (0.54–1.40)	0.148
Specialist input related to EDS	67 (52.8%, 44.1–61.2)	42 (33.1%, 25.5–41.6)	<0.001	No information collected on number of visits to specialists.		
Emergency hospitalization related to EDS	6 (4.7%, 2.2–9.9)	18 (14.2%, 9.2–21.3)	0.012	0.063 (0.010–0.116)	0.157 (0.085–0.229)	0.040
Planned hospitalization related to EDS	9 (7.1%, 3.8–12.9)	10 (7.9%, 4.3–13.9)	1.000	0.087 (0.028–0.145)	0.110 (0.040–0.181)	0.418

* Sample (N) for this figure is 126.
[†] N = 121.
[‡] N = 118.
[§] N = 113.
EDS, eating, drinking and/or swallowing; GP, general practitioner.
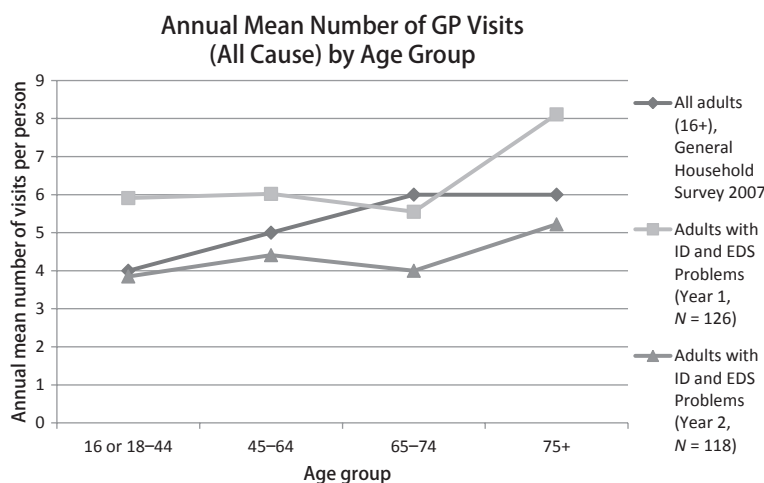
Figure 2 Age-stratified average annual numbers of GP visits.

Specialist referrals related to eating, drinking and/or swallowing

Specialists include SLTs, dietitians, OTs, clinical psychologists and other healthcare practitioners (excluding GPs) who address EDS problems in hospital or community settings. Although 80% (104/127) of our participants had been referred to a specialist for an EDS problem at any time prior to baseline, recent input was less common: only half to one-third of our participants received specialist input in year 1 or 2, respectively (see Table 3). Of the sample as a whole ($N = 127$), 77 (61%) individuals received specialist input at least once during the 2-year study period; 32 (25%) had input in both years.

Of the 42 participants who received specialist input for EDS problems in year 2, 79% (33) had seen an SLT, and 19% (eight) a dietitian. Input from clinical psychologists, psychiatrists and others, particularly for the management of mealtime behaviours, was less common (three cases, 7%). Some participants saw multiple specialists in both years. Reasons for input also overlapped: in year 2, 14 people (33% of 42) were regularly reviewed or monitored, while 29 (69%) reported non-routine input for a range of specific issues and worries, such as difficulty swallowing/suspected dysphagia (11 people, 26%) and behavioural problems (4, 10%). Five participants (12%) received input for quality-of-life issues (e.g. introducing

'oral tasters', or small amounts of food and/or fluids, for three PEG-fed participants, and integrating toast into the breakfast of one person on a liquidised diet, at his request). This holistic consideration of emotional well-being and quality of life alongside physical health is in line with current thinking in SLT practice (Macleman 2005; Chadwick 2014).

Hospitalizations (all-cause and eating, drinking and/or swallowing related)

Emergency hospitalizations encompass emergency department (ED) attendances alone, as well as admissions through emergency, while planned hospitalizations refer to elective appointments and admissions. Numbers of all-cause emergency hospitalizations were only collected in year 1. In the first year, 27 participants (21% of 127) experienced 35 emergency hospitalizations for any reason (median: 1 visit; range: 1–3).

With respect to emergency hospitalizations for EDS problems, in year 1, six participants experienced eight emergency hospitalizations for EDS problems (mean: 1.3 visits per person; median: 1; range: 1–2) while in year 2, 18 had a total of 21 emergency hospitalizations for EDS problems (mean: 1.2 visits per person; median: 1; range: 1–2). Individuals who had EDS-related emergency hospitalizations were older than the sample average

(mean age: 59.0 in year 1 and 51.9 in year 2), although this was not statistically significant [mean difference, year 1: −13.0, 95% CI −27.5 to 1.5, $t(125) = -1.769$, $P = 0.079$; mean difference, year 2: −6.1, 95% CI −15.0 to 2.8, $t(125) = 0.175$, $P = 0.297$]. In contrast with the decline in GP visits and specialist referrals from year 1 to 2, the incidence of emergency hospitalizations for EDS problems increased, while planned appointments and admissions remained steady in this period (see Table 3).

Respiratory infections were the most commonly reported single reason for EDS-related emergency hospitalizations in year 2. Nine (43%) of the 21 EDS-related emergency hospitalizations were for respiratory infections, four of which were specifically identified as aspiration pneumonia. Although the duration of one (11%) of these nine hospitalizations was not reported, the others were generally long, inpatient admissions, lasting a mean of 27.1 days (SD 29.0; 95% CI 2.9–51.3; range: 1–84 days). Assuming that the ninth hospitalization was an ED attendance (not admission), and was therefore zero days long gives us a shorter, but still extended, mean length of stay for respiratory infections of 24.1 days ($N = 9$; SD 28.6; 95% CI 2.2–46.1; range: 0–84 days). The stress of extended hospital admissions may have additional repercussions for participant health; for example, the individual with the longest admission (84 days) reportedly experienced related muscle wastage, further reducing his ability to self-feed. In addition to the clear human costs of these hospitalizations, we have estimated the financial cost of these eight admissions and one attendance, using a conservative costing strategy, at approximately £37 000 (see Appendix S2 for further details).

GP visits for respiratory infections were strongly associated with emergency hospital visits for the same reason. Of the nine participants who experienced an emergency hospitalization primarily due to respiratory infection in the second year, seven had seen the GP at least once for this reason in the first year (78% compared with 34% of the 118 people with no such hospital visits, Fisher's exact test $P = 0.013$), six of whom also saw the GP for a respiratory infection in year 2 (67% compared with 21% of the remaining 112 participants, $N = 121$,

Fisher's exact test $P = 0.007$). In addition, in the first year, these nine individuals consulted their GPs for respiratory infections (median: 2 visits) significantly more often than the remaining 118 participants (median: 0; Mann–Whitney $U = 791.0$, $P = 0.005$).

Proportion of healthcare use attributable to eating, drinking and/or swallowing problems

Approximately one-fifth of this population's healthcare use was attributable to EDS-related issues: eight (23%) of the 35 all-cause emergency hospitalizations and 156 (21%) of the 756 all-cause GP visits in the first year related to eating/ drinking.

Incidence and causes of health issues related to eating, drinking and/or swallowing

All information on EDS-related health issues is derived from questions about GP and hospital visits for specific problems, including respiratory infections, weight loss, dehydration, malnutrition and/or other problems that could be caused by or affect eating and drinking, such as GORD. Around one-third of our surviving participants had a respiratory infection each year (see Table 3), while one-fifth (26/122) experienced respiratory infections in both years. Fewer participants had nutritional concerns (regarding undernutrition or dehydration): 16% (20/ 127) of participants in the first year, declining to 11% (13/124) in the second ($N = 124$, McNemar's exact test $P = 0.286$). Only five individuals (4% of 124) had such concerns in both years.

Incidence and causes of mortality (year 2)

Eight (5.6%) of the 142 people who participated in the baseline study are known to have died between baseline and follow-up. Of the other 134, 131 (92%) are known to have survived, whereas the vital status of the remaining three (2%) is unknown. Using all 142 baseline participants as a denominator, and knowing that between 8 and 11 people died in year 2 provides a crude death rate (CDR) of between 5.6% (95% Wilson CI 2.9–10.7%) and 7.7% (4.4–13.3%). Using the lower figure, the annual

CDR is approximately 56.3 deaths per 1000 adults with ID and EDS problems.

We used age-specific mortality estimates (Emerson & Hatton 2008) to calculate and sum expected numbers of deaths in our population, by age decile, and compared these to the observed number of deaths. This resulted in a standardized mortality ratio (SMR) of 267 (exact 95% CI 115–526), meaning that, even after controlling for the potential effects of our sample's older age, almost three times as many of our participants died as would be expected in the 'general' ID population (see Appendix S2 for details).

Respiratory infections were the immediate cause of death, specified by the GRO death certificate, in all eight cases. Three people died from *pneumonia*, two from *bronchopneumonia*, two from *aspiration pneumonia* and one simply from a *chest infection* with no further specification.

Discussion

Focusing on one area of England, we have described the incidence of healthcare use, health problems, such as respiratory infections, and mortality. Each year, the majority of our participants (85–95%) visited their GP at least once, while around 20% attended hospital for any emergency reason. Around one in five GP and emergency hospital visits were for EDS problems. Respiratory infections were common, affecting approximately one-third of all participants each year, and 20% (26/122) in both years. Nutritional issues were rare, affecting between 11 and 16% of participants each year, and only 5% in both years. Compared with other adults with ID, our subgroup of adults with ID *and* EDS problems had a higher CDR, and an SMR nearly three times what would be expected in the 'general' ID population. Improved management of EDS problems in adults with ID may have the potential to reduce the burden of respiratory infection and prevent hospital admissions and premature death.

Comparison with other studies

Most of the literature on the health service utilization of adults with ID focuses on primary care.

With respect to consultation with primary healthcare providers, one UK-based study found that 74% (58/78, 95% CI 64–83%) of individuals with ID had any annual contact with their GP (Whitfield *et al.* 1996), while the figure from a more recent Dutch study was 82% (79–84%) (Straetmans *et al.* 2007). Comparing these estimates to ours from Table 3, a slightly higher proportion of our participants had seen the GP for any reason: between 85% (year 2, 95% CI 78–90%) and 95% (year 1, 89–97%). With respect to mean annual numbers of GP consultations, early UK-based studies reported relatively low rates of contact: 2.7 (Wilson & Haire 1990) and 2.2 visits per year for people with unspecified ID or 3.5 for people with DS (no CI provided) (Howells 1986). This appears to have increased over time, with more recent studies reporting higher rates: 4.1 (3.3–4.8) (Stein & Ball 1999) and 5.4 (no CI provided) (Felce *et al.* 2008), although the latter may be inflated slightly by the inclusion of contact with any members of the primary care team. These more recent numbers are very similar to our estimates: our participants had a mean of 6.1 (5.2–7.0) all-cause GP visits in the first year and 4.2 (3.5–4.9) in the second. Although there is wide variation in previous estimates, across time periods and according to the population with ID examined, it appears that our participants had significantly more contact with their GPs than other adults with ID in the first year of study; however, the figures for the second year were not statistically different from previous reports at a 5% level of significance.

Although some existing research addresses ED visits and hospital admissions among people with ID in general, most of this work is from North America, which may limit its comparability with UK health system data (Morgan *et al.* 2000; Janicki *et al.* 2002; Balogh *et al.* 2005; Venkat *et al.* 2011). In the United Kingdom, it is thought that about one-quarter of all adults ID are admitted to hospital each year, although whether this is emergency or planned is not specified [National Patient Safety Agency (NPSA) 2004]. Although not perfectly comparable, this is broadly in line with the 20% of our participants who reported one or more all-cause emergency hospitalizations in the first year. Recent studies have also identified that people with ID have a different pattern of hospital admissions than those

without ID, including more emergency admissions and higher rate of hospitalization for ambulatory care-sensitive conditions (ACSCs) (Balogh *et al.* 2005; Glover & Evison 2013). High rates of ACSCs (such as convulsions and epilepsy, dehydration and gastroenteritis, constipation, GORD, and influenza and pneumonia), which are considered amenable to primary care intervention, may expose gaps in the quality or provision of primary healthcare (Glover & Evison 2013). Although in our study we only have information on the causes of EDS-related hospitalizations, almost half of these 21 emergency hospitalizations in the second year were for two ACSCs: pneumonia/respiratory infections (nine) or convulsions/seizure (one), indicating that they are potentially amenable to intervention.

Although respiratory infections, such as aspiration pneumonia, are an important health issue for all adults with ID (Hollins *et al.* 1998), our study highlights their increased importance for individuals with ID and EDS problems. Around 30% (13/44) of our surviving participants with diagnosed dysphagia had recurrent or persistent respiratory infections (two or more GP and/or hospital visits for respiratory infections) in both years 1 and 2, while around half reported having at least one respiratory infection serious enough to seek medical attention (year 1: 53%; year 2: 43%). Our findings support and expand on previous reports that 40% of adults with ID and clinically diagnosed dysphagia experience recurrent respiratory infections (Chadwick & Jolliffe 2009). Consistent with existing research on the general population with and without ID (Glover & Evison 2013), emergency hospitalizations for respiratory infections were also long (mean duration: 24 days). The common occurrence of respiratory infections in the sample as a whole, alongside the observed emergency hospitalizations and extended length of admissions for respiratory infection, suggest that many participants suffer from chronic problems, which regularly develop into acute episodes requiring substantial medical management. Although we cannot determine the quality of the health care received by each participant, lengthy hospital admissions may be exacerbated by gaps in mainstream healthcare provision, such as failing to accommodate the different needs of people with ID through 'reasonable adjustments' to services. There is evidence that

staff in acute general hospitals, in particular, may not have the skills, experience and confidence required to appropriately and effectively care for adults with ID (Sowney & Barr 2006; Gibbs *et al.* 2008; Backer *et al.* 2009), to the extent that this was identified as a serious safety issue [National Patient Safety Agency (NPSA) 2004].

Our participants also had a higher CDR than other adults with ID. Using data from a study of adults with moderate to profound ID in Leicestershire and Rutland, which found that 409 participants died during 23 000 years of follow-up (Tyrer *et al.* 2007), we calculated a CDR of 17.8 deaths/1000 person-years (95% Poisson CI 16.1–19.5). In contrast, the CDR in our sample of adults with mild to profound ID and mealtime support needs was approximately 56.3 deaths/1000 person-years (24.5–111.8). Despite wide CIs, the estimates from the two studies do not overlap; therefore, we have concluded that our participants had a significantly higher CDR than other adults with ID. Using indirect standardization to adjust for differing age structures, we found that if our participants were subject to the same age-specific mortality rates as the 'general' ID population, they would have an SMR of 267. This figure indicates that participants in our study were more than two and a half times more likely to die than they would have been had they not required mealtime support.

With respect to cause of death, respiratory infections were the primary cause of death in all eight deceased individuals, two (25%) of which were specifically identified as aspiration pneumonia. Using death certificates in London, Hollins *et al.* (1998) found that approximately 50% of adults with ID died as a result of respiratory disease, compared with 15% of men and 17% of women without ID. A nationally representative study of death certificates from 2004 to 2008 found that pneumonia or lower respiratory tract infection was the specified cause of death for 35% of people with ID, and that 14% of deaths in people with ID were due to lung problems caused by aspiration of solids or liquids (Glover & Ayub 2010). Although all adults with ID are at an increased risk of death from respiratory infections, our data suggest that individuals with ID and EDS problems are particularly vulnerable, and therefore merit more intensive monitoring and prevention strategies.

Comparative Age Distribution of Adults with ID
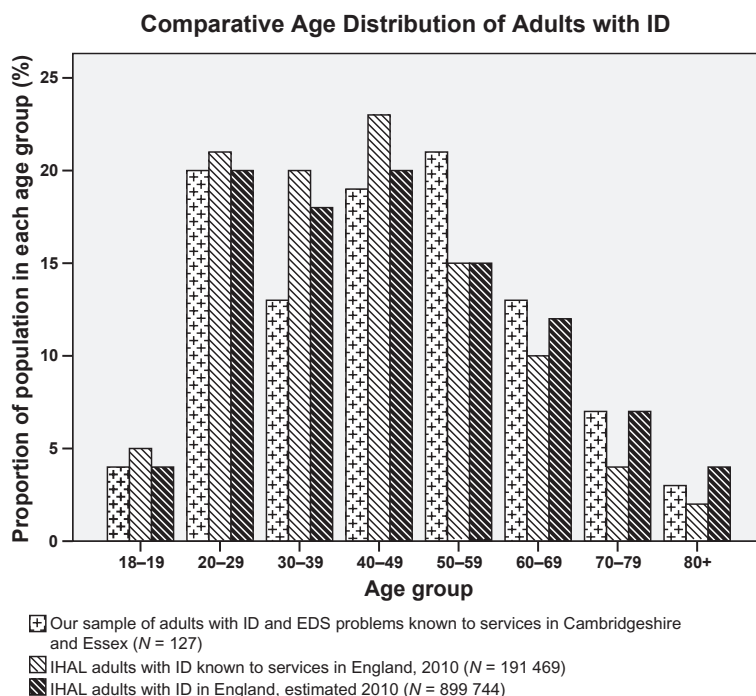
Figure 3 Comparing the age distribution of our participants [adults with intellectual disabilities (ID) and eating, drinking and/or swallowing (EDS) problems] to that of other adults with ID (estimated by Emerson *et al.* 2011 for *Improving Health and Lives: Learning Disabilities Observatory*).

Strengths and limitations

The primary nature of our data and broad inclusion criteria allowed us to explore issues that would not have been possible using large databases of routinely collected GP or hospital data. Our recruitment strategy did not rely on pre-existing specialist referrals for clinically diagnosed dysphagia; therefore, the resulting sample had a broad range of EDS problems and mealtime support. Our data on health issues are potentially underestimates, as they were derived from reported primary and secondary healthcare use for specific problems, and illnesses that did not result in an appointment with a healthcare practitioner will have been excluded. Additionally, our retrospectively reported data may be subject to recall bias, although this was likely mitigated by cross-checking against health and social care records.

As our original aim was to include all people identified as needing mealtime support in this research, sampling was not undertaken and involvement in our study depended on the willingness and ability of individuals to participate. Around 20% of those contacted became participants: selection bias

could therefore have affected the representativeness of our study population, although the direction of bias is unclear. This relatively low response rate reflects the well-documented difficulty involved in identifying and contacting people with ID for participation in primary research (Oliver *et al.* 2002; Lennox *et al.* 2005; Nicholson *et al.* 2013). Because of ethical constraints on access to data regarding non-participants, we were unable to rigorously assess how representative our sample was.

Using estimates published by the Learning Disabilities Observatory, Fig. 3 compares the age distribution of our study population (adults with ID and EDS problems) with both the population of adults with ID known to services in England and the total population of adults with ID (Emerson *et al.* 2011). Although our population approximates the age distribution of the general population of adults with ID, individuals between 30 and 49 years old are slightly underrepresented and older adults (aged 50+) overrepresented, most likely reflecting the onset of age-related dysphagia. We did not find any variation in the prevalence of diagnosed dysphagia according to participant age, likely because of our inclusion criteria. However, EDS problems are

known to increase in complexity and severity with increasing age, largely due to the onset of co-morbid conditions, such as dementia (Lazenby 2008), which is supported by the finding that older participants were more likely to report *increasing* mealtime support needs (Ball *et al.* 2012).

Implications for research and practice

Despite its limitations, this study addressed an under-researched area, which has gained renewed attention in policy and practice. We recommend that future research focuses on the level and nature of all-cause morbidity and health service use in this population (the 80% not attributable to EDS-related issues), in order to clarify broader issues of health needs and access in this subgroup of adults with ID. Future research should include a control group of adults with ID and no confirmed EDS problems, to situate the incidence and prevalence of healthcare use and illnesses in better context. In support of National Patient Safety Agency (NPSA) guidelines, we recommend that carers and healthcare providers should remain vigilant to repeat respiratory infections [National Patient Safety Agency (NPSA) 2007a,b], which are a substantial cause of morbidity for adults with ID and EDS problems and, in the most serious cases, may result in death. Potential links with eating and drinking (e.g. aspiration, choking or coughing around mealtimes) should be investigated thoroughly and followed up on a regular basis, for example, as part of the Annual Health Checks offered to all adults with ID (Kerr *et al.* 2012).

Ethical approval

The project received ethical approval from the NHS Cambridgeshire 3 Research Ethics Committee, UK.

All participants with the capacity to do so gave consent to participate in the study. For individuals invited to take part who lacked the capacity to give or withhold consent, a consultee was appointed, in accordance with the Mental Capacity Act 2005, who was able and willing to make a decision regarding whether the individual would wish to take part, if he or she had the capacity to decide. For all participants who lacked capacity, a consultee agreed to their participation.

Conflict of interest

None of the authors have any conflicts of interest.

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Supporting Information

Additional Supporting Information may be found in the online version of this article at the publisher's web-site:

Appendix S1. Additional methodological details.
Appendix S2. Additional calculations.
Table S1. Details and costs of emergency hospital visits for respiratory infections in year 2.
Table S2. Indirectly age standardizing our observed mortality rates using age-specific mortality estimates for adults with ID in England.
Table S3. Standardized mortality ratio (SMR) and calculation of 95% upper and lower confidence limits (95% CI), using Equation 1 and Equation 2.