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ORIGINAL COMMUNICATION

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Frequency, timing and outcome of gastrostomy tubes for amyotrophic lateral sclerosis/motor neurone disease

A record linkage study from the Scottish Motor Neurone Disease Register

Abstract *Aims* To describe the frequency, timing and outcome from gastrostomy in amyotrophic lateral sclerosis/motor neurone disease (ALS/MND). *Methods* The Scottish MND Register, a population based disease register (1989–1998), with record linkage to the Scottish Morbidity 1 dataset of hospital discharges coded for gastrostomy procedure was used. Descriptive statistics of patients undergoing gastrostomy were extracted. Survival analysis used Kaplan Meier and Cox proportional hazards methods. *Results* For patients diagnosed between 1989–98, 142 percutaneous endoscopic gastrostomy (PEG) insertion episodes were identified in 1226 patients, 130 of which occurred before the censoring date of 31 December 1999. Annually, on average, 5 % of all revalent patients underwent gastrostomy, and this rate appeared to double between 1989–98. The cumulative incidence of gastrostomy was 11 %. Mean age at PEG tube insertion was 66.8 years, with a mean disease duration of 24 months. Median survival from PEG tube insertion was 146 days. The 1 month mortality after gastrostomy was 25 %. Gastrostomy did not confer a survival advantage compared with no gastrostomy. *Conclusions* We found that gastrostomy feeding tubes are being inserted more frequently in people with ALS/MND. An unexpectedly high early mortality was detected which probably reflects a lack of selection bias compared with previously published data. It is possible that changes in the practice of gastrostomy placement since 1998 result in better outcomes for patients with ALS/MND. Prospective studies are required to assess the risks and benefits of enteral nutrition in ALS/MND.

Key words amyotrophic lateral sclerosis · gastrostomy · survival

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Introduction

In Amyotrophic Lateral Sclerosis/Motor Neurone Disease (ALS/MND) 50–70 % of patients ultimately experience bulbar symptoms such as dysphagia [4, 11] so alternative methods of nutritional support are sought. In recent years, percutaneous endoscopic gastrostomy (PEG) feeding tubes have been proposed as an alternative to nasogastric tubes as a means of circumventing neurogenic dysphagia [15]. In ALS/MND, data to support the use of PEG tubes are conflicting. Following PEG insertion, some authors have reported improved survival compared with controls [5, 14], while others report no such effect [7, 13, 17]. There have been no randomised trials or population-based studies of PEG tube use for ALS/MND [2]. The evidence that exists is based upon case series from referral centres, whose patients survive longer than those identified in population-based studies [1]. Therefore, population-based observational data might provide a more accurate description of the effect of PEG tube use in ALS/MND than a referral centre study.

Using a record-linkage study, we describe the timing, frequency of use, and effect on survival of gastrostomy tubes in all Scottish adults with ALS/MND diagnosed between 1989 and 1998.

Methods

The methods of the Scottish Motor Neurone Disease Register are described elsewhere [1], and will only be briefly summarised here. We identified cases from multiple intersecting sources (a) Referrals from consultant neurologists and neurophysiologists practising in Scotland and Family Care Officers (latterly Nurse Specialists) of the Scottish Motor Neurone Disease Association, (b) Scottish Morbidity Records (SMR1) of discharges from Scottish Hospitals, and (c) Mortality Coding from the Office of the Registrar General for Scotland). We used the modified World Federation of Neurology criteria for people diagnosed between 1989–1993 [1] and the El Escorial criteria for people diagnosed from 1994 onwards [3]. Diagnoses were confirmed pathologically in 69 of 70 autopsies performed in the course of routine management. We recorded symptom onset (bulbar – dysarthria or dysphagia, spinal – limb onset). By convention those with both spinal and bulbar symptoms from onset were classified as bulbar. We took note of any familial cases. Using capture recapture methods [9], we have tested ascertainment and shown it to be of the order of 98% complete.

The original Scottish Motor Neurone Disease Register proforma did not include an entry for gastrostomy tube usage. We performed a record linkage study with the Scottish Morbidity Records (SMR1) of the Information and Statistics Division of the Scottish Office Health Department. The SMR1 dataset covers all Scottish hospitals, and codes all hospital admissions using the ISD-9, ISD-10 coding system for diseases and the OPCS4 system for procedures. We interrogated the SMR1 dataset with identities of patients registered on the Scottish Motor Neurone Disease Register to identify all those who had a hospital admission episode in which a code of gastrostomy or PEG tube procedure occurred. If the date of gastrostomy procedure was ambiguous, we took the mid-point of the hospital admission as the date of procedure. The characteristics of those with an episode coding for at least one gastrostomy tube procedure were compared with those who did not undergo a procedure. We examined survival using Kaplan Meier and Cox proportional hazards methods to compare outcomes of those with and without a gastrostomy tube. For survival analyses, we censored outcomes on 31 December 1999, the last date for which we have death certificate returns from the Office of the Registrar General for Scotland. The rate of gastrostomy was expressed as a rate per prevalent case per year.

The Scottish Motor Neurone Disease Register has full multi-centre medical research ethics approval, and the record linkage study was approved by the Scottish Office.

Results

There were 11,869 hospitalisation episodes, from the Scottish population, between 1989 and 2001 coded with a gastrostomy or PEG tube procedure. Of these, 178 linked to the Scottish MND Register, of which 142 were prior to 31 December 1999. There was a steady increase in the rate of gastrostomy tube episodes (Fig. 1). The overall rate was 5% of prevalent cases per year, and in total 11% of our 1226 registered cases had at least one episode.

The mean age at tube insertion was 66.8 years (95% CI 65–68.7). The mean time from onset to gastrostomy was 730 days (95% CI 606 to 850), and the mean time from diagnosis to gastrostomy tube was 350 days (95% CI 274–247). Two patients appear to have had procedures before the diagnosis of ALS/MND.

Female patients with bulbar onset and a history of treatment with riluzole were significantly more likely to have a gastrostomy. Males with pure lower motor neurone syndromes at presentation were less likely to have a code for this procedure (Table 1). There did not appear to be any difference in the survival of those who had a gastrostomy tube placed compared with those who did not (Log Rank p = 0.52, Fig. 2). Even when the excess of female patients, and bulbar presentations were controlled for in a Cox regression model, there was no discernible effect of gastrostomy tubes on overall survival.

Within 30 days of the gastrostomy procedure, 30% of spinal onset and 22% of bulbar onset patients had died (overall 25% 30 day mortality, Fig. 3). For bulbar patients, the median survival following a first procedure was 186 days (95% CI 40–391), while median survival in spinal onset patients was 99 days (95% CI 26–321). At 6 months and 1 year after the procedure, survival rates were 46% (39% for spinal onset, 51% bulbar onset) and 23% (17% for spinal onset, 27% bulbar onset) respectively. The survival curves suggest that spinal patients may have poorer survival than bulbar patients, but overall there was no difference between the groups (Log Rank, p = 0.82).

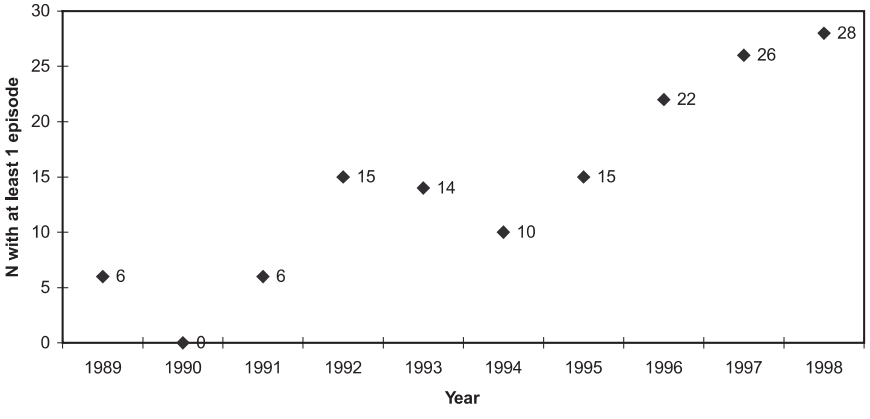

Fig. 1 Numbers of PEG episodes for Scottish MND Register cases 1989–1998

Table 1 Comparison of characteristics of ALS/MND patients with PEG versus no PEG episodes. Scottish MND Register 1989–1998

	PEG Episodes	No PEG Episodes	Statistical test Odds Ratio (95% CI)
Cases	142 (11.5%)	1084	
Males	54	613	0.47 (0.33–0.68)
Females	88	471	
Bulbar	87	343	3.20 (2.22–4.60)
Spinal	54	681	
Lower Motor only	12	170	0.31 (0.27–0.92)
Upper and Lower Motor	121	851	
Riluzole	17	52	2.70 (1.51–4.81)
None	125	1032	
Neurology Assessment	125	860	2.15 (1.10–4.19)
None	10	148	
Mean age at disease onset	64.2	64.8	p = 0.59, T-test
Median survival from onset	2.08 (IQR 1.44 to 2.99)	2.06 (IQR 1.29–3.49)	p = 0.52, log rank
Time from onset to diagnosis	0.75 (IQR 0.42–1.09)	0.84 (IQR 0.50–1.47)	p = 0.04, Kruskall-Wallis

IQR Interquartile range

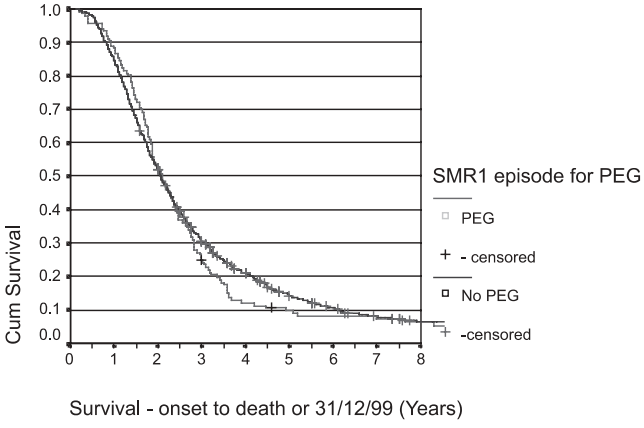

Fig. 2 Survival curves comparing PEG tube versus no PEG tube. Scottish MND Register 1989–1998

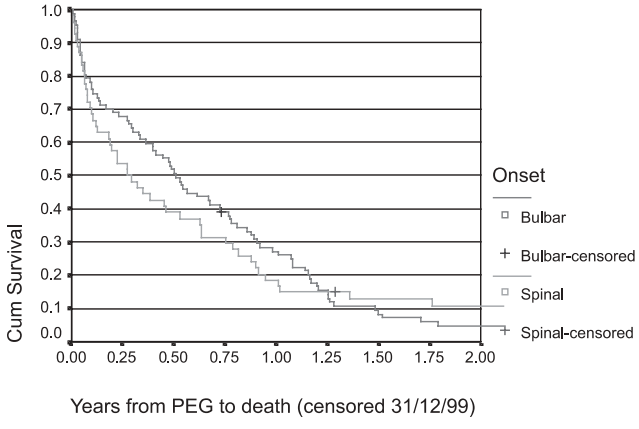

Fig. 3 Survival following PEG tube episode, stratified by bulbar or spinal onset. Scottish MND Register 1989–1998 (Log Rank p = 0.86)

Discussion

This is the first description of gastrostomy tube usage in an unselected population of people with ALS/MND. We found a doubling of the rate of procedures in Scottish adults with ALS/MND in the period 1990–98 (Fig. 1). We were able to validate the SMR1 dataset, as on review of a random selection of hospital records we found that coding for gastrostomy (PEG) tube insertion was accurate.

All studies of PEG in ALS/MND to date have been observational [5–8, 10, 13, 14, 17, 18] and it is difficult to determine whether observed benefits or hazards are the result of bias, random error or a true effect. There was no clear evidence of enhanced survival following PEG tube insertion in our population. This conclusion agrees with other observational studies [7, 13], although it conflicts with Mazzini et al. and Chio et al. who found better survival in those who had undergone a PEG [5, 14] (Table 2). The participants in these positive studies had an unusually long survival from ALS/MND, so were biased towards a favourable outcome through selection bias. One study found evidence of decreased survival following percutaneous gastro-jejunostomy in spinal onset males [17]. We observed a trend towards poorer early survival following gastrostomy in spinal onset cases (Fig. 3). While this did not reach significance, it raises the possibility that there is a sub-group of patients in whom invasive interventions to alleviate dysphagia could be particularly hazardous.

Table 2 Studies evaluating survival following nutritional intervention for ALS/MND

Author (Ref) (Year)	Study participants	Survival 30 day mortality	Median survival from PEG (months)	Overall survival from onset
Mathius-Vliegen et al. [13] (1994)	PEG = 55 No PEG = 13 Mean age 58.8 years Median disease duration 26 months at PEG referral	11.5%	4 (PEG) 3 (Control)	967 days (PEG) 900 days (Control)
Mazzinni et al. [14] (1995)	N = 69 PEG = 31 No PEG = 38 Mean age 60.9 years Mean duration 26 months	9.7%	13 (PEG) 9 (Control)	1155 days (PEG) 912 days (Control)
Kasarskis et al. [10] 1999 (BDNF patients)	PEG = 136 Mean age 59.2 years	9.6%	9 (Mean)	
Kasarskis et al. [10] 1999 (CNTF patients)	PEG = 36 Mean age 56.3 years	6.3%	5 (Mean)	
Strong et al. [17] (1999)	N = 73 Mean age: Bulbar 64.7 years, Limb onset 60.0 years Disease duration: Bulbar 17.7 months, Limb onset 29.9 months	9.6%	No significant survival advantage	669 days (Bulbar) 730 days (Limb onset)
Chio et al. [5] (1999)	N = 68 PEG = 50 No PEG = 18 Mean age 61.7 years Disease duration 25 months	2%	6 (PEG) Increased hazard ratio for patients without PEG tube HR = 1.83 (95 % CI 1.39–2.40)	915 days (PEG) 760 days (Controls)
Desport et al. (2000)	N = 60 PEG = 30 Control = 30 Mean age 65.7 years Disease duration 26 months at PEG	13%	No effect of PEG on survival	
Gregory et al. [8] 2002	N = 33 PEG = 33 FVC < 50 % predicted Mean age 62.8 years	3%	Not stated	204 days
Scottish MND Register	PEG = 142 No PEG = 1084 Mean age PEG = 66.8 years Disease duration = 24 months	25%	5 (PEG)	759 days (PEG) 752 days (No PEG)

This is the only population-based study of the outcome of PEG in ALS/MND performed to date. Our high one-month mortality (25 %) is greater than in other reports (range 2 % – 13 %, Table 2). There are several possible explanations. First, because our study was population-based we probably included patients with rapidly progressive disease who might not have been referred to a tertiary referral centre for specialist neurological assessment. Second, because the study was performed at a time when PEG was being introduced it is possible that the higher mortality rate is explained by technical factors. The management of patients with respiratory failure is particularly relevant. Reports of possible increased mortality associated with PEG procedures in those with an FVC% of less than 50 % [10] did not emerge until after the end of our registration period (1989–1998). These lead to the American Academy recommendation that patients should not have a PEG unless FVC is greater than 50 % [2]. Because this cohort was studied before the introduction of guidelines concerning respiratory assessment before PEG, it is possible that Scottish patients had procedures when their respiratory capacity was sub-optimal. More recently investigators have demonstrated that radiologically guided gastrostomy is associated with a lower rate of technical complications [18]. Ideally, different feeding regimes should be tested in a randomised controlled trial, with either survival or health-related quality of life as an outcome measure. However, in the absence of such trials, prospective population-based studies may demonstrate improvements in survival and quality of life.

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