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Research Article

Prospective Validation of Edinburgh Dysphagia Score as a Triaging Tool beyond the COVID-19 Era

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The Edinburgh Dysphagia Score (EDS) was previously developed to identify patients referred to secondary care with dysphagia, who were most likely to have oesophageal cancer. The aim of this study was to use the EDS prospectively during the COVID pandemic to risk stratify patients to either urgent or routine investigation of dysphagia. Between 1st April and 1st July 2020, 283 patients were referred to NHS Lothian with dysphagia. An EDS score was calculated utilizing information in the GP referral letter or information gained in a "HOT clinic." Patients with a score \geq 3.5 were prioritized for investigation under the "urgent suspicion of cancer" pathway. 243 patients underwent investigations. 18 patients were diagnosed with oesophageal cancer, all of whom had an EDS \geq 3.5 (range 4-10). Approximately one third of patients with dysphagia had a score of <3.5. Using this cut-off, sensitivity was 100% and negative predictive value 100%. This study shows that the EDS can be used prospectively when triaging patients referred to secondary care with dysphagia. The high negative predictive value using the EDS means that patients who have an EDS < 3.5 can be downgraded to a routine waiting list without leading to delays in diagnosing oesophageal malignancy. This will enable faster investigations for patients who remain on the "urgent suspicion of cancer" waiting list. In the age of COVID-19, with increasingly long waiting lists, the EDS is a useful scoring system to identify patients with the greatest need for urgent endoscopy.

1. Introduction

Oesophageal cancer is estimated to affect 9000 people in the UK each year, and the incidence has been rising over the last 20 years [1]. Oesophageal cancer often has an insidious onset with patients being unaware of symptoms until the cancer has spread locally through the layers of the oesophageal wall or metastasized distantly. Dysphagia is a common symptom once the cancer has become large enough to cause significant narrowing of the oesophageal lumen, with resultant difficulty in swallowing certain foods. In the UK, adeno-

carcinoma is the most common subtype of oesophageal cancer, and the UK is estimated to have the highest rate of oesophageal adenocarcinoma in the world, with an incidence of 7.2 per 100,000 men and 2.5 per 100,000 women, respectively [2].

At the end of 2019, SARS-CoV-2 (COVID-19) emerged in Wuhan, China, rapidly becoming a global pandemic and resulting in significant morbidity and mortality. With rising COVID-19 incidence in the UK, the British Society of Gastroenterology issued guidelines to temporarily discontinue aerosol generating procedures including gastroscopies that

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Age (years)		Location of dysphagia		Duration of symptoms		Sex		Reflux		Weight loss	
<40	0										
40-49	4	Neck	-2	\geq 6 months	-1.5	Female	-1	Present	-1	≥3 kg	2
50-59	5										
60-69	6										
70-79	7	Cl t	0		0	M.1.	0	A l	0	42 lan an alaans	0
80-89	8	Chest	0	< 6 months	0	Male	0	Absent	0	<3 kg or absent	0
90-99	9										

TABLE 1: Edinburgh Dysphagia Score calculation.

Note: the EDS is the sum of the score in each category.

were deemed nonemergency and those which were "urgent suspicion of cancer" (USOC) or "two week wait" [3]. At one point in the pandemic, endoscopic activity was reduced to 5% of the prepandemic level [4], and a modeling study estimated that delays in diagnosis could lead to 330-342 additional deaths from oesophageal cancer in England in the five years following diagnosis, an increase of 5.8-6% [5].

Reopening of endoscopy services has been slow with significant reduction in our capacity to meet demands, primarily due to the need for negative pressure environments, for fallow time between procedures and for social distancing within endoscopy units. At 31st December 2020, over 13,000 patients in Scotland were on a waiting list for an OGD, with only 26.8% of procedures carried out within six weeks of referral compared to 67.2% completed within the same timeframe one year earlier [6]. Similar delays in procedures have been found in England where 4.7 million patients were waiting for a procedure or operation in February 2021 [7], an increase of 270,000 patients in 12 months [8].

In order to ensure that patients with the highest risk of significant pathology are investigated quickly, new methods for triaging patients must be identified. The Edinburgh Dysphagia Score (EDS) is a retrospectively validated scoring system, designed to identify patients presenting with dysphagia who are more likely to have oesophageal malignancy [9]. Factors associated with an increased or decreased risk of malignancy included the patient's age, sex, duration of symptoms, location of dysphagia, presence or absence of reflux, and significant weight loss (>3 kg). A scoring system (Table 1) was derived and validated retrospectively against a second cohort of patients. A score of 3.5 or greater is used to identify patients at increased risk of oesophageal malignancy.

The British Society of Gastroenterology has now recommended using the EDS to triage patients [10]. Patients with an EDS \geq 3.5 should be investigated through the "USOC" or "two week wait" pathway, subject to the patient being fit for investigation. In patients with an EDS < 3.5, investigation should be delayed until the lifting of COVID-19 restrictions [10]. We used the EDS prospectively to risk stratify patients to either USOC or routine investigation of dysphagia and investigated the eventual outcomes of these patients at follow-up.

2. Materials and Methods

All referrals to secondary care in NHS Lothian marked for "GI - Upper" between 1st April and 1st July 2020 were reviewed (n = 919). All patients whose referrals included "dysphagia," "difficulty swallowing," or "food or fluids sticking" were included in this study (n = 283). 112 patients attended an appointment in the "HOT clinic." HOT clinics have been used by a number of specialties to reduce admission rates and the duration of inpatient admissions [11, 12]. They rely on assessment of patients with specific symptoms by a senior specialist to triage patients to the most appropriate investigation. During the first COVID-19 lockdown, a HOT clinic for patients with dysphagia was set up in NHS Lothian. An algorithm (Figure 1) was devised with the aim of urgently and accurately triaging patients referred by their general practitioner with dysphagia to either USOC investigation, urgent investigation, or routine investigation. For those requiring USOC investigation, either a barium swallow or an OGD was performed based on the availability of these investigations at the time. Figure 2 illustrates the diagnostic pathway of investigations and results. A senior gastroenterologist took a comprehensive history and determined the patient's EDS, as well as discussing the risks and benefits of investigations. For the patients who did not attend a HOT clinic appointment, the referral letter was reviewed and the EDS calculated from the information provided by the GP before triage. 20 patients did not require investigation as they were determined not to have true oesophageal dysphagia. 20 patients were not investigated either because they declined investigation, did not attend, or died of an unrelated cause prior to investigation. Patients with an EDS ≥ 3.5 were prioritized for urgent investigation.

All OGDs were performed by trained endoscopists or trainees under supervision. All radiological investigations were reviewed by a consultant radiologist. OGD is the preferred method of investigation for dysphagia; however, early in the first wave of the pandemic endoscopic examinations was only carried out in the most urgent of cases; alternative modalities were used in order to minimize delays in diagnosis.

Statistical analysis was performed using RStudio and R (version 4.0.5). IBM SPSS Statistics was used to produce

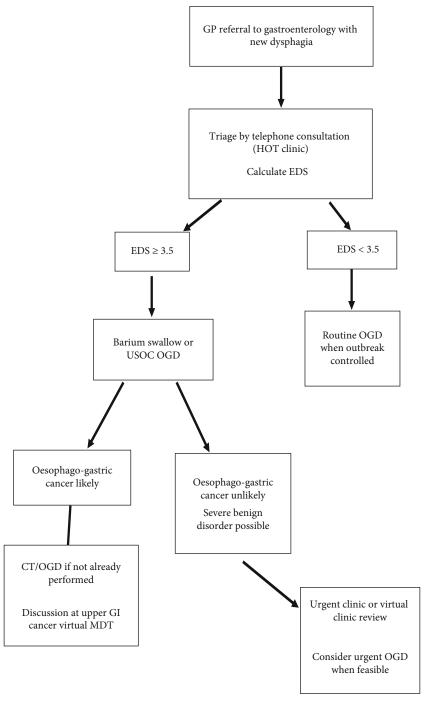


FIGURE 1: Algorithm for patients referred with dysphagia.

the ROC curve (version 27.0.1.0). This study was conducted in accordance with UK research ethics guidelines. After discussion with the research and development director, specific ethical review and approval were not required, as the study was considered a clinical audit for service evaluation and safety using data already obtained for patient care during the COVID-19 pandemic. The data was anonymized and analyzed by researchers who were not involved in clinical care provision at the time of the study.

3. Results

243 patients underwent investigation for dysphagia. 117 patients had an OGD as their first investigation. 119 patients had a barium swallow, with seven undergoing an alternative radiological investigation. 39 patients who had a radiological investigation subsequently underwent an OGD, giving a total of 158 patients undergoing an OGD. Overall, 48.1% of the patients were male. The mean age was 63 (range 28-

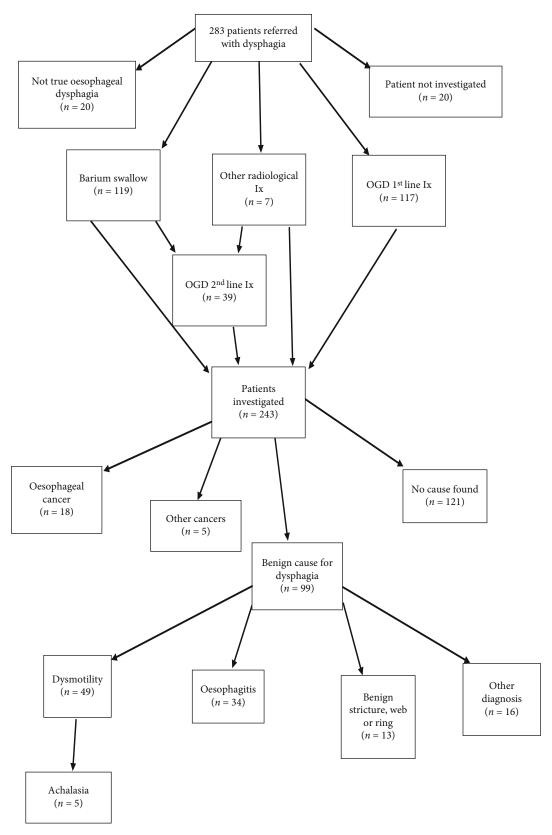


FIGURE 2: Investigation pathway and diagnoses. Some patients had more than one benign cause for dysphagia.

Table 2: Demographic data of all patients.

	EDS ≥ 3.5	EDS < 3.5	p value
Mean age	69 (±1.9)	52 (±2.8)	< 0.001
Sex (male)	56.9%	30.1%	< 0.001
PPI usage prior to symptom onset	51.9%	54.2%	0.83
History of atopy	21.9%	24.1%	0.82

96, 95% CI ± 1.9). Table 2 illustrates the patient demographics.

83 patients had an EDS < 3.5 (34.2%), and 160 patients had an EDS of ≥ 3.5 (65.8%). The scores ranged from -4.5 to 10 (Figure 3). 18 patients were diagnosed with oesophageal malignancy or malignancy of the GOJ (hereafter combined as "oesophageal malignancy"), all of whom had an EDS \geq 3.5 (range 4-10). Using a score of \geq 3.5 to indicate a higher risk of oesophageal cancer, the sensitivity of the EDS was 100% with a 100% negative predictive value (Table 3). The specificity of a patient with a high EDS having an underlying oesophageal malignancy was 37%, and the positive predictive value of the score was 11%. The cut-off of 3.5 or greater indicating a higher risk of oesophageal cancer was based on data from Rhatigan et al. [9]. Analysis was performed to create a ROC curve (Figure 4) with an area under the curve of 0.826% which is similar to both the development cohort (0.834%) and the validation cohort (0.709%) when the scoring system was first designed [9].

3.1. Outcomes of Patients with Oesophageal Cancer. Of the 18 patients diagnosed with oesophageal malignancy, 13 had adenocarcinoma, and five had squamous cell carcinoma. 11 patients had features suggestive of malignancy identified on imaging and subsequently confirmed on histological samples taken at the time of an OGD. Seven patients were only investigated with an OGD. There were no cancers identified on OGD which were not seen on radiological investigation. 16 of the 18 patients with oesophageal malignancy were diagnosed with late-stage disease (American Joint Committee on Cancer stage 3 or greater). Only seven patients were treated with curative intent. Treatments offered included operative management, palliative chemotherapy, placement of an oesophageal stent, and best supportive care. 11 patients died within one year of diagnosis. The mean length of survival for these patients was five months (range 0-11 months, median seven months). Given the small number of oesophageal malignancies identified in this cohort of patients, and all patients had an EDS ≥ 3.5 and thus expedited investigations, it is not possible to determine whether using the EDS identifies cancers at an earlier stage.

3.2. Other Diagnoses. As shown in Figure 2, 49 patients were diagnosed with dysmotility, five of whom were diagnosed with achalasia. Other benign diagnoses which could account for dysphagia included oesophagitis, eosinophilic oesophagitis (EOE), oesophageal candidiasis, and structural abnormalities such as benign strictures. EOE and oesophageal candidiasis were confirmed on samples taken at the time of

OGD. There were five nonoesophageal malignancies diagnosed in this group of patients.

3.3. Follow-Up. The patients who declined investigation, as well as those who only had a radiological investigation, were followed up for 9-12 months following referral (median 11 months). None of these patients subsequently presented with oesophageal malignancy. In addition, five of the 89 patients who only had a radiological investigation represented in the follow-up period with ongoing dysphagia but none were found to have oesophageal cancer. None of the patients with a benign or normal barium swallow had malignancy identified on OGD.

4. Discussion

These results confirm that the EDS is a highly sensitive scoring system which can be used prospectively to identify those patients with dysphagia who are at higher risk of oesophageal cancer, differentiating them from patients at a low risk who do not require urgent investigation. The EDS can be easily calculated from the information on the referral letter to secondary care. A calculator has also been added to the BSG website [13]. Our results using this score as a prospective tool for triage are consistent with the results from the original retrospective study which had a sensitivity value of 97.5% [9].

In our study, 34% of patients had an EDS < 3.5, none of whom were diagnosed with oesophageal cancer. The use of the EDS to identify these low-risk patients could lead to a significant reduction in the number of patients undergoing investigation of dysphagia on an "urgent suspicion of cancer" pathway or "two week wait" pathway. Although downgrading patient referrals may delay investigation, and the possible diagnosis of malignancy, our results indicate that this risk is extremely low. By reducing unnecessary urgent investigations, patients with a high EDS who are more likely to have an oesophageal malignancy can be prioritized for endoscopy. As oesophageal cancer has a 16.2% overall fiveyear survival rate in the UK [14], with 84.5% survival at one year in those diagnosed with stage 1 disease, and only 20.8% survival at one year in those diagnosed with stage 4 disease [15], it is imperative that patients are diagnosed at the earliest possible opportunity. In this study, 61% of patients with oesophageal cancer died within the follow-up period (median survival seven months, IQR 1.5-7.6 months), and only 18% of patients diagnosed with stage 4 oesophageal cancer were still alive at the end of the follow-up period.

In our study, 18 patients were diagnosed with oesophageal cancer, representing 7.4% of patients undergoing investigations. This is consistent with the findings of Rhatigan et al. [9] where 11.5% of patients investigated had oesophageal cancer. Given the timing of our study in relation to the pandemic, it is reassuring that a similar rate of oesophageal cancer has been diagnosed in a similar population. This suggests that patients may not have delayed seeking medical attention for their dysphagia.

A previous study by Prasad et al. [16] found the EDS to have a sensitivity of 89% and a negative predictive value of

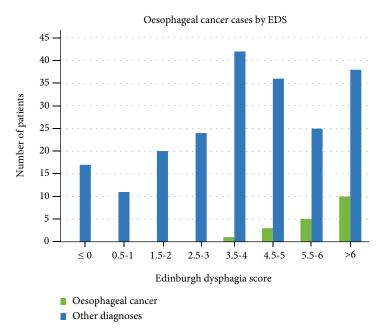


FIGURE 3: Range of Edinburgh Dysphagia Scores and oesophageal cancer diagnoses.

Table 3: Results of EDS and cancer diagnosed on subsequent investigation.

	Oesophageal cancer	No oesophageal cancer	
EDS ≥ 3.5	18	142	PPV = 11%
EDS < 3.5	0	83	NPV = 100%
	Sensitivity = 100%	Specificity = 37%	

PPV: positive predictive value; NPV: negative predictive value.

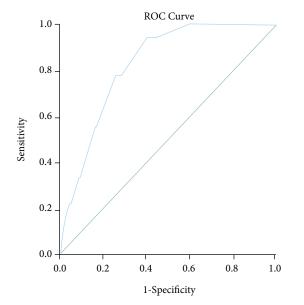


FIGURE 4: Receiver operating characteristic (ROC) curve of the Edinburgh Dysphagia Score.

86%. Their study, however, included other cancers found during OGD investigation, including gastric malignancies and ENT malignancies. We have deliberately excluded the patients with other cancers from our positive finding group as the EDS is only designed to identify patients at higher risk of oesophageal malignancy. It should also be noted that of the 341 patients investigated with dysphagia in the Prasad study, 51.6% were diagnosed with a malignancy which is significantly higher than the 7.4% of patients diagnosed with oesophageal cancer in this study. This difference could explain the disparity in the sensitivities and negative predictive values between these studies. The EDS would therefore be recommended as a triage tool when the incidence of oesophageal cancer in the referral population is low.

4.1. Radiological Investigation. The results also indicate that for many patients, radiological investigation is sufficient in the investigation of dysphagia as less than 6% of these patients who only had a radiological investigation were rereferred within one year with ongoing dysphagia. Initial radiological investigation also identified all patients who had an oesophageal malignancy. The majority of patients in this study who had a radiological investigation had a barium swallow. However, OGD is the gold standard investigation. If there are further waves of the COVID-19 pandemic which require significant reduction in endoscopic investigations, our study provides reassurance that patients who do not have malignancy identified on a barium swallow are unlikely to have an underlying oesophageal cancer. This is particularly pertinent in light of the emergence of the Omicron variant. Utilizing the EDS could also be of benefit in medically underresourced settings. Given the significantly reduced availability of endoscopic procedures at the start of the pandemic, patients were not selected for barium swallow or OGD based on their EDS, rather the investigation available

Age (years)		Location of dysphagia		Duration of symptoms		Sex		Reflux		Weight loss	
< 40 and male	2										
< 40 and female	2.5	Throat	-2	≥6 months	-1.5	Female	-1	Present	-1	≥3 kg	2
40-49	4										2
50-59	5										
60-69	6										
70-79	7	Chest	0	< 6 months	0	Male	0	Absent	0	<3 kg or absent	0
80-89	8										0
> 00	0										

TABLE 4: Updated Edinburgh Dysphagia Score calculation.

Note: the EDS is the sum of the score in each category.

at the time of referral. For this reason, it is not possible to determine if specific EDS would be more suitable for investigation with barium swallow or with OGD.

4.2. Suggested Improvements to the EDS. Consideration was given to any additional factors which could be used to improve the EDS. A history of atopy can be associated with eosinophilic oesophagitis, a benign cause of dysphagia. As the presence or absence of atopy was not significantly different between the two groups of patients, as shown in Table 2, adding this to the EDS calculation would not improve its sensitivity or specificity. This was confirmed in modeling calculations, which showed that giving a factor of -1 for a history of atopy was associated with a sensitivity of 94% and negative predictive value of 98.9%. These results would, therefore, increase the risk of a delay in significant pathology if the EDS was being used to downgrade patients with a low score to routine investigation. We do not, therefore, advise alteration of the score to account for a history of atopy. The patients with an EDS \geq 3.5 were significantly more likely to be male and to be older (Table 2). These differences between the two patient groups were expected as both age and sex are included in the calculation of the EDS—increasing age adds points, and female sex removes a point. Accurate information regarding patient's smoking history and alcohol consumption was not available.

If a patient is under 40 years of age when they are referred to secondary care with dysphagia, it is not possible for the patient to score 3.5 or greater using the EDS. Oesophageal cancer is rare in this age group; however, one study in the US, where the population and their associated risk factors are similar to the UK, found that cases of oesophageal cancer in those under 50 have been increasing by 2.9% per year [17]. A study in Turkey, where squamous cell carcinoma is the predominant oesophageal malignancy, found that 9% of cases of oesophageal cancer occurred in patients under 40 years old [18]. To reduce the risk of missing an oesophageal cancer in patients under 40 years old, we propose changing the number of points from zero points if a patient is under 40, to 2 points for a male patient under 40 and 2.5 points for a female patient under 40. In this study, there were 29 patients referred who were under 40. If the updated EDS (Table 4) were used then only two additional patients would have had an EDS of 3.5 or greater, therefore, using this updated scoring system would not lead to a significant increase in the USOC workload.

The EDS only gives a score for age up to 99 years, and therefore, we propose altering the final age range from 90-99 years to ≥90 years (Table 4). Consideration was given to an additional age range of ≥100 years being added with a value of 10 points; however, even if the patient scored the lowest points for all other factor, the minimum score for a patient \geq 90 years would be 3.5. In this situation, the patient would remain on the USOC pathway; therefore, there is no additional benefit of adding an age range of ≥100 years. The risks of investigating with an OGD in a patient of 100 years or older are increasingly significant at this age, and therefore, we would advise discussion with the patient prior to requesting investigations. The other change which we would suggest to the EDS is the location of dysphagia. Currently, the two options are "neck" or "chest;" however, following feedback, it has been noted that the term "neck" leads to ambiguity and therefore, we would suggest changing this to "throat".

4.3. Implementing the EDS during Triage. The aim of the original study to create and validate the EDS by Rhatigan et al. [9] was to assign priority for investigation of the symptom of dysphagia. During the "first wave" of the COVID-19 pandemic in the United Kingdom, elective NHS services were stopped or significantly reduced, and emergency admissions to hospitals were reduced by 28% compared to the same time period in 2019 [19-24]. At this time, it suddenly became critical to use tools such as the EDS to accurately identify patients at higher risk of underlying malignancy and therefore with the greatest need for urgent investigation. This study proves that the EDS is still a useful tool which can be used beyond the peak of the pandemic and potentially in subsequent waves if endoscopy activity is reduced, to reliably and safely downgrade the priority of endoscopic investigations for approximately one third of patients referred with dysphagia.

During the early stage of the pandemic, setting up a HOT clinic to improve triage of patients referred with dysphagia was possible. However, admission rates have since increased to levels similar to those before the pandemic

[25, 26], and therefore, a consultant-led HOT clinic model is not sustainable. One proposed change to improve triage would be to alter the referral paperwork from primary care to specifically include the information in the EDS on the initial referral, and therefore, the EDS could be quickly calculated by the triaging clinician in all cases. We found that the majority of referrals contained all the required information in the free text; however, location of dysphagia was often poorly described. By using drop-down boxes of set questions, this information could be gathered more efficiently without a decrease in the sensitivity of the EDS.

Another suggestion is that patients with an EDS < 3.5 could undergo a structured telephone assessment by a GI nurse specialist. This could clarify whether red flag signs and symptoms are present, clarify the patient's smoking history and family history, and calculate the GERD-Q or Eckardt scores. Those without concerning features could remain on the downgraded non-USOC pathway or be offered an alternative test which may be more suitable for their symptoms such as barium swallow or Cytosponge. Patients with concerning features could remain on the USOC pathway for urgent investigation of their dysphagia. Further study to determine the optimum method for improving the triage process, without affecting the sensitivity and negative predictive value of the EDS, is required.

5. Conclusions

This study has prospectively validated the Edinburgh Dysphagia Score and has shown that a score < 3.5 has a high negative predictive value for oesophageal cancer in patients who are referred to secondary care with dysphagia. It can, therefore, be used as a tool for risk stratification of patients for endoscopy waiting lists, allowing patients with an EDS < 3.5 to be downgraded to routine waiting lists with a minimal risk of delaying a diagnosis of oesophageal malignancy. Despite endoscopy department activity increasing from the nadir of the pandemic, there is still a significant backlog of patients awaiting endoscopic investigation across the UK, and more patients are being referred each day. Using the EDS will reduce the number of patients with dysphagia requiring investigations on the "urgent suspicion of cancer" or "two week wait" waiting lists by approximately one third. However, oesophageal cancer has few symptoms until the tumor is advanced and therefore, this triage tool does not aid in the diagnosis of early malignancy. The EDS can only be calculated if the required information—age, sex, location of dysphagia, duration of dysphagia, presence of reflux, and significant weight loss—is known. It would be advisable to add this information as a structured set of questions on the referral system for primary care to enable the use of the EDS at the time of triage. The limitations of the EDS when triaging patients under 40 years old have been identified. The score has been updated to reflect that whilst oesophageal cancer is rare in patients under 40, these patients cannot be missed. In future, the updated EDS should be used when triaging patients referred to secondary care with dysphagia.

Data Availability

The data used to support the findings of this study are available from the corresponding author upon request.

Ethical Approval

The authors confirm that the ethical policies of the journal, as noted on the journal's author guidelines page, have been adhered to. No ethical approval was required as the study was considered a clinical audit for service evaluation and safety using data already obtained for patient care.

Conflicts of Interest

There are no conflicts of interest to declare.

Authors' Contributions

Professor John Plevris is the guarantor for the submission. Gail Masterton and John Plevris designed the study. Rahul Kalla, Norma McAvoy, Nicholas Church, Ian Penman, Andrew Williams, Kenneth Trimble, Gail Masterton, and John Plevris reviewed patients in the HOT clinic. Hannah Walton and Dean McAvoy collected and analyzed the data. Hannah Walton wrote the paper. Rahul Kalla, Gail Masterton, and John Plevris edited the paper. All authors approved the final version of the submission.

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