Effectiveness of endoscopic cricopharyngeal myotomy in adults with neurological disease: systematic review

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Abstract

Objective: To determine the effectiveness of endoscopic cricopharyngeal myotomy on upper oesophageal sphincter dysfunction in adults with upper oesophageal sphincter dysfunction and neurological disease.

Data sources: Published and unpublished studies with a quasi-experimental design investigating endoscopic cricopharyngeal myotomy effects on upper oesophageal sphincter dysfunction in humans were considered eligible. Electronic databases, grey literature and reference lists of included studies were systematically searched.

Review methods: Data were extracted by two independent reviewers. Methodological quality was assessed independently using the PEDro scale and MINORS tool.

Results: Of 2938 records identified, 2 studies were eligible. Risk of bias assessment indicated areas of methodological concern in the literature. Statistical analysis was not possible because of the limited number of eligible studies.

Conclusion: No determinations could be made regarding endoscopic cricopharyngeal myotomy effectiveness in the cohort of interest. Reliable and valid evidence on the following is required to support increasing clinical usage of endoscopic cricopharyngeal myotomy: optimal candidacy selection; standardised post-operative management protocol; complications; and endoscopic cricopharyngeal myotomy effects on aspiration of food and laryngeal penetration, mean upper oesophageal sphincter resting pressure and quality of life.

Key words: Deglutition Disorders; Esophageal Sphincter, Upper

Introduction

The upper oesophageal sphincter (UOS) is a 3-4 cm intraluminal high-pressure zone^{1,2} separating subatmospheric oesophageal pressure from atmospheric pharyngeal pressure.³ The prevalence of dysphagia, a characteristic of UOS dysfunction, is high in neurological disease cohorts. If conservative measures fail to improve UOS functioning, surgery may be proposed.

Cricopharyngeal myotomy may be performed via open, transcervical or endoscopic approaches.⁴ Endoscopic cricopharyngeal myotomy was initially performed using potassium-titanyl-phosphate lasers; however, surgical trends have recently favoured use of the carbon dioxide (CO₂) laser given the possible benefits regarding reduced thermal damage and blood vessel coagulation.^{5,6} Endoscopic cricopharyngeal myotomy is performed transorally; it involves cricopharyngeal diverticuloscope visualisation and exposure, with pulsed or non-pulsed laser incision to the buccopharyngeal fascia.^{5,7,8} Buccopharyngeal fascia and retropharyngeal space violation increase the risk of mediastinitis because of air extravasation and the potential for contamination with abscess formation.^{5,7} If the buccopharyngeal fascia is damaged, the surgical bed is closed using fibrin glue or endoscopic stapling.^{5,9}

Lawson *et al.*¹ retrospectively evaluated endoscopic cricopharyngeal myotomy effects in a neurological disease cohort, a post-chemoradiation therapy cohort and a cricopharyngeal spasm cohort. Fibre-optic endoscopic swallowing and videofluoroscopic swallowing were evaluated pre- and post-operatively, along with self-assessments of symptom improvement. The authors reported no complications, yet recurrence of clinical symptoms was noted in 62.5 per cent of neurological disease patients. Linke *et al.*¹⁰ retrospectively reviewed outcomes of

Linke *et al.*¹⁰ retrospectively reviewed outcomes of patients with neuromuscular diseases, reporting 100 per cent enteral feeding tube removal post-operatively. However, cricopharyngeal myotomy was combined

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with glottopexy and glottal closure in 58 per cent of patients, thus confounding the results.

Lawson *et al.*¹¹ reviewed endoscopic cricopharyngeal myotomy outcomes in an unspecified population, although five patients presented with neurological disease diagnoses such as amyotrophic lateral sclerosis, stroke and myopathy. Results were confounded by the fact that 13.8 per cent of participants underwent endoscopic cricopharyngeal myotomy following failed open or transcervical cricopharyngeal myotomy. The authors reported significant objective improvement and no complications. However, only short-term follow up was assessed and there was no information on longterm symptom evolution.

Bergeron and Chhetri⁶ retrospectively reviewed outcomes in a mixed population that included patients with UOS dysfunction following stroke and neuronal damage. All participants demonstrated significant objective improvements post-operatively. Several patients required concurrent dilation and endoscopic cricopharyngeal myotomy; however, details regarding who received this procedure were not available. In addition, assessment was non-blinded, which increased the risk of detection bias.

Hazarika and colleagues¹² retrospectively investigated combined endoscopic cricopharyngeal myotomy and dilation in a mixed population that included neurological disease patients. Objective improvement was reported in 87.5 per cent of patients. However, one participant died as a result of mediastinitis caused by intra-operative perforation, indicating the morbidity and mortality potential of endoscopic cricopharyngeal myotomy.

There is controversy in the literature regarding endoscopic cricopharyngeal myotomy.¹³ This is related to under-reporting of complications,^{9,14} and limited rigorous efficacy, coupled with varying clinical indications and follow-up durations.¹¹ Therefore, large prospective studies with rigorous clinical indicators and validated assessment tools are required^{11,14} to direct further research and clinical practice in the area. This systematic review aimed to examine the evidence for endoscopic cricopharyngeal myotomy as an intervention for UOS dysfunction in patients with dysphagia associated with neurological disease.

Research questions

These were: (1) does endoscopic cricopharyngeal myotomy performed for UOS dysfunction reduce or eliminate aspiration of food and/or fluids and laryngeal penetration in adults with UOS dysfunction and neurological disease?; (2) does endoscopic cricopharyngeal myotomy performed for UOS dysfunction reduce mean UOS resting pressure in adults with UOS dysfunction and neurological disease?; (3) does endoscopic cricopharyngeal myotomy performed for UOS dysfunction change oral intake status in adults with UOS dysfunction and neurological disease?; (4) what are the adverse events associated with endoscopic cricopharyngeal myotomy performed for UOS dysfunction in adults with UOS dysfunction and neurological disease?; and (5) does endoscopic cricopharyngeal myotomy performed for UOS dysfunction result in a change in quality of life (QoL) for adults with UOS dysfunction and neurological disease?

Materials and methods

The process of conducting and reporting this systematic review was guided by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses ('PRISMA') statement.¹⁵

Eligibility criteria

A systematic search of eight electronic databases and grey literature, from inception to June 2015, was conducted (Table I).

A highly sensitive search strategy was employed using filters, Medical Subject Headings and key-text terms, developed with assistance from Trinity College Dublin librarians. No language or date limitations were applied. This search examined all published and unpublished studies with a quasi-experimental design that investigated endoscopic cricopharyngeal myotomy effects on upper oesophageal sphincter (UOS) dysfunction in humans

Resource categoryResource title (dates searched)Bibliographic databases (electronic searches)Ovid Medline (1946 – January 2015) Elsevier Embase (1947 – January 2015) Elsevier Science Direct (1823 – January 2015) Elsevier Scopus (1823 – January 2015) Ebsco AMED (1985 – January 2015) Thompson Reuters Web of Science (1900 – January 2015) Ebsco CINAHL (1981 – January 2015) Conference proceedings abstracts – European Society for Swallowing Disorders (2011–2014)* – Dysphagia Research Society (1992–2014)* Proceed Discoption of the searches (1906 – January 2015)	TABLE I RESOURCES SEARCHED		
Bibliographic databases (electronic searches)Ovid Medline (1946 – January 2015) Elsevier Embase (1947 – January 2015) Elsevier Science Direct (1823 – January 2015) Elsevier Scopus (1823 – January 2015) 	Resource category	Resource title (dates searched)	
Produest Dissertations & Theses database (1/16 – January 2015)	Bibliographic databases (electronic searches) Grey literature (hand searches)	Ovid Medline (1946 – January 2015) Elsevier Embase (1947 – January 2015) Elsevier Science Direct (1823 – January 2015) Elsevier Scopus (1823 – January 2015) Ebsco AMED (1985 – January 2015) Thompson Reuters Web of Science (1900 – January 2015) Ebsco CINAHL (1981 – January 2015) Conference proceedings abstracts – European Society for Swallowing Disorders (2011–2014)* – Dysphagia Research Society (1992–2014)* ProQuest Dissertations & Theses database (1716 – January 2015)	

*Both published in *Dysphagia*. AMED = Allied and Complementary Medicine Database; CINAHL = Cumulative Index to Nursing and Allied Health Literature

and which satisfied the inclusion and exclusion criteria (Table II). $^{16-18}$

Studies that investigated endoscopic cricopharyngeal myotomy combined with another intervention were omitted as this hindered clear data analysis. Studies comprising participants of mixed demographics were considered if extraction of data for participants aged 19 years and over was possible.

Study selection process

Searches were independently conducted by two authors (OG and PK). Following the deletion of duplicate articles, the double screening of titles, key words and abstracts was conducted (by ÓG and PK) to minimise reporting bias and human error. References were managed using the bibliographic management system Zotero (free online software). Hand searches of conference abstracts from the European Society for Swallowing Disorders and the Dysphagia Research Society were also conducted (by OG and PK), as these societies primarily aim to investigate normal and abnormal swallowing physiology, and dysphagia and its related functions. The reference lists of eligible studies were searched (by ÓG) to identify further relevant articles. A third independent reviewer (SB) mediated disputes if they occurred.

Data extraction process

Data were extracted electronically (by ÓG and PK independently). The extraction process was piloted on a representative sample of studies to ensure appropriateness and reviewer competency.¹⁹ A random sample of 20 per cent of data was reviewed by a third author (SB) to mitigate discrepancies. Disputes were documented and resolved via discussion of rationale, with input from an independent reviewer (SB) if required.

Data regarding both objective and patient-reported outcomes were extracted.²⁰ Primary outcome data extracted included objective post-operative improvements in laryngeal penetration and aspiration of food, oral intake status, and objective mean manometric UOS resting pressure reductions. Secondary outcome data extracted included adverse events directly related to surgery, such as increases in dysphagia, compromised medical health, negative psychological or social consequences, hospitalisation, or death.

TABLE II INCLUSION AND EXCLUSION CRITERIA

Inclusion criteria

- Adults aged 19+ years (male & female) seen in any setting - Documented UOS dysfunction (as rated on objective assessment) & neurological dysphagia (acute, progressive or non-progressive) at all severity levels
- Exclusion criteria
- Congenital neurological conditions¹⁶
- History of head & neck cancer¹
 History of radiation therapy^{17,18}

UOS = upper oesophageal sphincter

Post-operative QoL changes were also considered, to assess psychosocial intervention effects.

Time frames for analysis of effects (i.e. immediate, less than 1 month; medium, 1-6 months; and longterm, greater than 24 months) were chosen to capture long-term intervention effects.²¹

Data were stratified into comparative subgroups using Microsoft[™] Excel[®] spreadsheets, with planned statistical analysis performed using IBM[™] SPSS[®] software.

Risk of bias assessment

Risk of bias was independently assessed by two authors (ÓG and PK) using the PEDro (Physiotherapy Evidence Database) scale²² and MINORS (Methodological index for non-randomised studies) tool,²³ with conflicts resolved via input from a third author (SB).

The PEDro scale assesses methodological quality across the study design spectrum, with sufficient reliability for use in systematic reviews.¹⁶ Maher et al.²² reported an inter-rater reliability generalised kappa statistic of between 0.40 and 0.75 for the PEDro scale, indicating moderate to good agreement between reviewers.

The MINORS tool is a methodological index that assesses risk of bias and quality of non-randomised surgical trials, providing requisite sensitivity to facilitate meta-analysis.²³ It has been widely used in systematic reviews of surgical trials because of high levels of intra-reliability and test-retest reliability, internal consistency, and validity.

The issue of missing data was addressed by contacting authors, for studies published within the last five years. In the case of no response following two contact attempts, the study was excluded.

Endpoints and data synthesis

A meta-analysis of primary and secondary endpoints (using calculations of risk ratios and 95 per cent confidence intervals for dichotomous outcomes, and standardised mean differences and 95 per cent confidence intervals for continuous outcomes) was planned if sufficient numbers of studies were identified via searches, and if adequate homogeneity (as indicated by a chisquare test of homogeneity) was present. If heterogeneity was present, causative factors were to be explored via a subgroup analysis of heterogeneity sources. The authors planned to construct a forest plot to display effect estimates and confidence intervals for dichotomous outcomes. A subgroup analysis considering longand short-term intervention effects, with stratification as per acute versus progressive neurological disease and specialty training of the operating surgeon, was also planned.

Results

Study selection

A search of 8 databases and grey literature yielded 2938 articles (Figure 1). No additional articles were





Preferred Reporting Items for Systematic Reviews and Meta-Analyses ('PRISMA') flow diagram. CINAHL = Cumulative Index to Nursing and Allied Health Literature; AMED = Allied and Complementary Medicine Database

identified from searching the reference lists of included studies. After deleting duplications, 1711 records remained. Of these, 1609 records were excluded at the screening stage as they did not satisfy the inclusion criteria. Authors subsequently examined 102 full-text records. One author (ÓG) sought access to and/or clarification of missing data. Eight authors responded, leading to 12 exclusions on the basis of ineligible participants, inappropriate interventions or insufficient available data. Three studies were excluded as the authors were unable to access the full contents of the articles. After full-text reviews and author contact, two studies ultimately satisfied the inclusion criteria and were included in the systematic review.

Study characteristics

Study participants. The two studies that satisfied the inclusion criteria presented data regarding five patients with confirmed neurological disease (Tables III–V).^{11,19}

Only one study provided the overall average age of participants.¹¹ The average age of neurological disease patients was not provided in either study. The pooled age range for both studies was 38–83 years. Northern European, single-centre university hospital clinics were the setting for both studies. No details regarding participants' nationalities were provided.

Study design. Both included studies were published in the English language and both were non-randomised.

ENDOSCOPIC CRICOPHARYNGEAL MYOTOMY IN NEUROLOGICAL DISEASE PATIENTS

TABLE III STRATIFIED CLINICAL DEMOGRAPHICS
Acute - 2 stroke (unspecified type) ¹¹ Progressive - 1 ALS (unspecified type) ¹¹ - 1 myopathy (unspecified type) ¹¹ Unspecified - 1 'central nervous system disease' ¹⁹
ALS = amyotrophic lateral sclerosis

One was a retrospective patient survey,¹⁹ and one was a retrospective case series with a chart review spanning the years 1995 to 2000 (Table IV).¹¹

Neither study recruited a comparison or control group; the inclusion of such a group would have enhanced internal validity.^{20,24} Study duration ranged from three years and eight months¹¹ to approximately five years.¹⁹ Lawson *et al.*¹¹ reported their 'experience with [endoscopic cricopharyngeal myotomy] in 29 cases', and Brøndbo¹⁹ evaluated 'functional [operative] results'. Brøndbo¹⁹ did not specify follow-up periods, while Lawson *et al.*¹¹ considered the immediate postoperative stage, with a median long-term follow up of 21 months (range, 1–44 months).

Interventions. Both studies utilised continuous mode CO₂ laser-assisted endoscopic cricopharyngeal myotomy. Lawson *et al.*¹¹ used fibrin glue to conduct primary mucosal closure. It is unclear what procedure was used by Brøndbo.¹⁹ Only Lawson reported statistically significant effects of endoscopic cricopharyngeal myotomy (Table V).¹¹

Outcomes

As discussed, the outcomes considered were related to the research questions and were as follows: objective post-operative reductions in aspiration of food and laryngeal penetration and in mean upper oesophageal sphincter (UOS) resting pressure, reductions in adverse events, and improvements in oral intake status and QoL.

Primary outcomes. Only Lawson *et al.*¹¹ addressed objective post-operative reductions in aspiration of food and laryngeal penetration, utilising both videofluoroscopic swallowing studies and fibre-optic endoscopic swallowing evaluations to enhance objectivity.²⁵ The median frequency of aspiration events, calculated using a non-validated scale, was significantly reduced from 3 (uncommon) (inter-quartile range, 2–4) to 4 (none) (inter-quartile range, 4–4).

Manometry, which is used to examine changes in mean UOS resting pressure pre- and post-surgery, was not used in either study. No post-operative changes in oral intake status were reported in either study.^{11,19}

			TA	ABLE IV				
			STUDIES THAT FULFI	LLED INCLUSION	CRITERIA			
Study (year)	Country	Study design	Aim	Setting	Study duration	Follow-up timeframes considered	Quality rating	5
							MINORS tool	PEDro scale
Brøndbo ¹⁹	Norway	Retrospective	To evaluate functional results of endoscopic	University	1995 - 2000	Unspecified	3/16	2/11
(2000) Lawson <i>et al.</i> ¹¹ (2003)	Belgium	survey Retrospective case series	cricopharyngean myouonny To report experience with endoscopic cricopharyngeal myotomy in 29 cases	nospital clinic University hospital clinic	January 1998– September 2001	Immediate (2–3 wk) post-op stage, with median-length follow up of 21 mth (range, 1–44 mth)	8/16	4/11
Wk = weeks; pos	t-op = post-	-operatively; mth $= 1$	months					

Secondary outcomes. Both included studies referenced post-operative adverse events; however, no post-operative adverse events were documented and no post-operative changes to QoL were discussed (Table V).

Intervention efficacy

Given the low numbers and heterogeneity of eligible studies, meta-analysis could not be conducted. Similarly, subgroup analysis of long- and short-term operative effects, along with stratification according to acute versus progressive neurological disease and specialty training of operating surgeons, could not be conducted. Only Lawson *et al.*¹¹ specified follow-up timeframes, reporting both short- and long-term outcomes (with follow-up assessments at week 2–3 and at a median of 21 months, respectively).

The two included studies reported on five patients eligible for inclusion (two with acute disorders, two with progressive diseases and one with an unspecified neurological disease); however, details regarding the location, severity and type of the presenting conditions were not provided. Thus, there was insufficient evidence to determine the effectiveness of endoscopic cricopharyngeal myotomy in this population of neurological disease patients.

Risk of bias

Independent reviewers reached consensus regarding risk of bias ratings, without dispute. The study by Brøndbo¹⁹ was given an overall score of 3 out of a possible 16 using the MINORS tool,²³ and the study by Lawson *et al.*¹¹ was given a score of 8 (Table VI).

Neither of the two included studies recorded whether blinding occurred; hence, there was a possibility of detection and performance bias. Lawson *et al.*¹¹ provided information regarding the statistical significance of the results. According to the PEDro scale,²² neither study was rated 'high' in terms of quality (Table VII).

Discussion

No conclusions can be drawn regarding the effectiveness of endoscopic cricopharyngeal myotomy in adult neurological disease populations. There is insufficient evidence to determine whether this procedure is associated with the post-operative reduction or elimination of aspiration of food and laryngeal penetration. Lawson *et al.*¹¹ noted that the median aspiration frequency was significantly reduced, which is in agreement with research by Takes et al.14 in nonneurological disease populations. Takes et al.14 found that endoscopic cricopharyngeal myotomy significantly reduced aspiration in adults with unspecified cricopharyngeal bar disorders. However, Dawe et al.24 reported that the aspiration of food and laryngeal penetration in adults with an upper oesophageal sphincter (UOS) dysfunction following chemoradiation therapy was not significantly improved with endoscopic cricopharyngeal myotomy, as measured using the Modified Barium Swallow Impairment Profile.²⁶ Aspiration

		H	ENDOSCOPIC	C CRICOPHA	RYNGEAL MYOTOMY II	NTERVENTIO	N, OUTCOM	E, RESULTS AND ADVERSE EF	rects	
Study	Overall n	Age range (mean); years	Gender (n)	Nationality	Endoscopic cricopharyngeal myotomy intervention	Comparison group?	Specialty of operating surgeon	Outcomes considered	Results	Adverse events
Brøndbo ¹⁹	17	52-83 (not provided)	11 F; 6 M	Not provided	CO ₂ laser (continuous mode)	No	Unclear	Subjective improvement in swallowing, length of time to oral intake, weight gain, complications	 All patients progressed to soft diet from 3rd post-op day Descriptive, non-significant changes in swallowing abilities 	None reported
Lawson et al. ¹¹	29	38–81 (62)	7 F; 22 M	Not provided	CO, laser (continuous mode), with primary mucosal closure using fibrin glue	No	Unclear	Objective reduction or elimination of food aspiration & laryngeal penetration, time to oral intake, symptom recurrence, complications	& weight change 1. Statistically significant improvements in aspiration & dysphagia frequency, & extent of pooling in pyriform sinus	None reported
F = females;	M = males	; $CO_2 = carbo$	n dioxide; po:	st-op = post-of	serative					

TABLE VI MINORS SCORES FOR INCLUDED STUDIES			
MINORS criteria	Study		
	Brøndbo ¹⁹	Lawson <i>et al.</i> ¹¹	
A clearly stated aim	1	1	
Inclusion of consecutive patients	2	2	
Prospective collection of data	0	0	
Endpoints appropriate to study aim	0	2	
Unbiased assessment of study endpoints	0	0	
Follow-up period appropriate to study aim	0	1	
Loss to follow up $<5\%$	0	1	
Prospective calculation of study size	0	1	
Overall score (out of 16)	3	8	

Note regarding scoring: 0 = not reported; 1 = reported but inadequate; 2 = reported and adequate

pneumonia costs the US healthcare system approximately \$4.4 billion per annum.^{27,28} Therefore, it is essential to investigate endoscopic cricopharyngeal myotomy outcomes to determine if the relative risk of laryngeal penetration and aspiration of food, and the associated morbidity and mortality, is reduced postoperatively.

Endoscopic cricopharyngeal myotomy can result in changes in diet. For example, Bergeron and Chhetri⁶ investigated post-operative dietary changes in a mixed population who had undergone endoscopic cricopharyngeal myotomy and dilation, and found that Functional Outcome Swallowing Scale²⁹ scores significantly improved, with 50 per cent of neurological disease patients who received pre-operative enteral feeding progressing to oral intake post-operatively. Linke *et al.*¹⁰ reported that all neuromuscular disease patients who received enteral feeding pre-operatively progressed to varying levels of oral intake following combined endoscopic cricopharyngeal myotomy, glottopexy and glottal closure.

Endoscopic cricopharyngeal myotomy is reported to greatly improve oral intake in those with nonneurological UOS dysfunction. Takes et al.¹⁴ reported a reduction, from 100 per cent to 40 per cent, in patients with idiopathic dysfunction requiring dietary alterations following surgery. In a study by Silver and Gal,⁸ approximately 67 per cent of patients with UOS dysfunction following chemoradiation therapy who received total enteral feeding progressed to soft diets, and 17 per cent progressed to a regular diet. Similarly, 100 per cent of those on a liquid diet pre-operatively progressed to a regular diet post-operatively. Bachy et al.¹⁷ reported dietary changes in an unspecified population, stating that 75 per cent of patients reported post-operative improvements in swallowing liquids, while 81 per cent experienced improvements in swallowing solids, as measured using the Deglutition Handicap Index.¹⁸ Ho *et al.*³⁰ investigated an unspecified population, noting that both endoscopic cricopharyngeal myotomy and open or transcervical cricopharyngeal myotomy resulted in comparably significant Functional Outcome Swallowing Scale² score changes.

The authors of the studies included in this systematic review did not detail changes in pre- and post-operative oral intake levels, and therefore no determination regarding intervention effects may be made. As UOS dysfunction often necessitates dietary changes,³¹ it is essential to address this in future studies to aid informed decision making and treatment planning.

Both of the included studies considered the adverse events associated with surgery, noting that no complications occurred. There is a dearth of evidence

TABLE VII PEDRO SCORES FOR INCLUDED STUDIES				
PEDro criteria	Study			
	Brøndbo ¹⁹	Lawson <i>et al.</i> ¹¹		
Eligibility criteria specified?	Yes	No (criteria provided but source not provided)		
Subjects randomly allocated to groups? (In a crossover study, subjects randomly allocated in order in which treatments received?)	No	No		
Allocation concealed?	No	No		
Groups similar at baseline regarding most important prognostic indicators?	No	No		
Blinding of all subjects?	No	No		
Blinding of all therapists who administered therapy?	No	No		
Blinding of all assessors who measured at least 1 key outcome?	No	No		
Measures of at least 1 key outcome obtained from >85% of subjects initially allocated to groups?	No	Yes		
All subjects for whom outcome measures were available received treatment or control condition as allocated, or, where this was not the case, data for at least 1 key outcome was analysed by 'intention to treat'?	Yes	Yes		
Results of between-group statistical comparisons reported for at least 1 key outcome?	No	Yes		
Study provides both point measures & measures of variability for at least 1 key outcome?	No	Yes		
Overall score (out of 11)	2	4		

pertaining to endoscopic cricopharyngeal myotomy complications, which may relate to publication bias. With increasing clinical usage, the reporting of complications has proportionally increased, although this issue remains contentious.⁹ In the USA and Australia, up to 16.6 per cent of hospital admissions across clinical specialties (e.g. orthopaedics, neurosurgery, thoracic and cardiac surgery, and general medicine) result in complications, with 7 per cent of complications producing permanent disabilities and 14 per cent causing death.^{21,32–35} Therefore, reliable reporting of complications must be promoted.^{36,37}

Regarding QoL, there is a small body of evidence to indicate positive changes following cricopharyngeal myotomy in non-neurological disease populations.^{12,32} Bachy *et al.*¹⁷ retrospectively investigated post-operative QoL outcomes in an unspecified population. These authors found that endoscopic cricopharyngeal myotomy significantly improved psychosocial wellbeing, as measured using the Deglutition Handicap Index.¹⁸ Gervais and Dorion³⁷ investigated QoL outcomes in patients presenting with oculopharyngeal syndrome who underwent open or transcervical cricopharyngeal myotomy or blepharoplasty. They reported significant QoL improvements following open or transcervical cricopharyngeal myotomy.

Neither of the studies included in this review addressed post-operative QoL changes. Nevertheless, consideration of the relationships between QoL, health needs and patient satisfaction³⁶ are important for future research studies.

Review limitations

This study has some limitations. For example, combined treatments were not considered, resulting in the exclusion of studies that may have enhanced findings. Bergeron and Chhetri⁶ found that the post-operative outcomes of those receiving intra-operative dilation were not significantly different to those receiving endoscopic cricopharyngeal myotomy alone. Therefore, future reviews may be justified in expanding criteria to accommodate treatment combinations.

Another limitation relates to the inability to conduct a meta-analysis to produce a unified statistical estimate of clinical effectiveness for endoscopic cricopharyngeal myotomy in adults with UOS dysfunction and neurological disease, because of the limited numbers of eligible studies identified. This limitation reflects a larger issue at the level of the wider literature relating to this field, and indicates an urgent need for methodologically rigorous and evidence-based research pertaining to endoscopic cricopharyngeal myotomy for UOS dysfunction.

Recommendations

High-quality evidence is required to support clinical decision-making. Directions for future research lie in reporting and conducting research studies. For example, trials should be reported according to either

the Consolidated Standards of Reporting Trials ('CONSORT') statement³⁸ or the Strengthening the Reporting of Observational studies in Epidemiology ('STROBE') guidelines.³⁹ Trials should provide precise patient demographic details. Outcome measures should be psychometrically robust, and assess the physiological, functional and psychological impact of impairment. Furthermore, trials should prioritise blinding where appropriate, to reduce detection and performance bias. In addition, patients should be followed up at immediate, short-term and long-term time points post-operatively.

Conclusion

There is insufficient evidence to determine the effectiveness of endoscopic cricopharyngeal myotomy in adults with upper oesophageal sphincter (UOS) dysfunction and neurological disease. Despite this, one cannot assume that endoscopic cricopharyngeal myotomy is an ineffective treatment, rather that there is a need for high-quality research to support this procedure. It is essential that the methodological quality of future trials is addressed comprehensively in order to: optimally select candidates for surgical intervention, safeguard patients against adverse effects, and ultimately improve QoL and overall dysphagia management in this vulnerable population.

Despite the discussed limitations, this review provides a clear description of the current status of literature pertaining to this topic. It should act as a point of departure for future methodologically rigorous research that can inform evidence-based practice guidelines for the care of adults with UOS dysfunction and neurological disease.

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