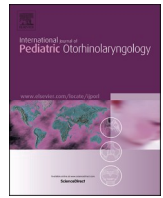




Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinology

journal homepage: www.elsevier.com/locate/ijporl

Endoscopic repair of tracheoesophageal fistulas: A contemporary multi-institutional case series and literature review

Catherine F. Roy^a, Alix Maltezeanu^b, Jean-Martin Laberge^c, Kimberley Kaspy^d, Ana Sant'Anna^e, H el ene Broucqsault^b, Pierre Fayoux^b, Sam J. Daniel^{a,*}

^a Department of Pediatric Otolaryngology, Montreal Children's Hospital, McGill University Health Centre, 1001 Decarie, H4A 3J1, Montreal, Quebec, Canada

^b Department of Pediatric Otolaryngology – Head and Neck Surgery, CHU Lille, F-59000, Lille, France

^c Department of Pediatric Surgery, Montreal Children's Hospital, McGill University Health Centre, 1001 Decarie, H4A 3J1, Montreal, Quebec, Canada

^d Department of Pediatrics, Division of Respiratory Medicine, Montreal Children's Hospital, McGill University Health Centre, 1001 Decarie, H4A 3J1, Montreal, Quebec, Canada

^e Department of Pediatrics, Division of Gastroenterology and Nutrition, Montreal Children's Hospital, McGill University Health Centre, 1001 Decarie, H4A 3J1, Montreal, Quebec, Canada

ARTICLE INFO

Keywords:

Tracheoesophageal fistula
Bronchoscopy
Minimally-invasive surgery

ABSTRACT

Objectives: Recurrent and primary tracheoesophageal fistulas (TEFs) are a challenging surgical pathology to treat, as standard open surgical approaches are associated with high morbidity and mortality. As such, endoscopic modalities have gained interest as an alluring alternative, yet variable success rates have been reported in the literature. The aim of this study was to provide a contemporary update of the literature and describe our institutional experience with the bronchoscopic obliteration of recurrent and primary TEFs.

Methods: Retrospective chart review of all pediatric patients having undergone endoscopic TEF repair at two pediatric academic centers in Montreal, Canada and Lille, France between January 1, 2008 to December 31, 2020.

Results: 28 patients with TEFs (20 recurrent, 8 primary) underwent a total of 48 endoscopic procedures. TEF repair was performed under endoscopic guidance using various combinations of techniques, including fistula de-epithelialization (endoscopic brush, thulium laser, trichloroacetic acid-soaked pledgets or electrocautery), tissue adhesives, submucosal augmentation, esophageal clip and stenting. Successful closure was achieved in 16 patients (57 %), while 12 (43 %) required eventual open or thoracoscopic repair. The mean number of endoscopic procedures was 1.7. There were no major treatment-related complications such as pneumothorax, mediastinitis or death (mean follow-up 50.8 months).

Conclusions: Endoscopic repair of recurrent or primary TEFs is a valuable component of our therapeutic armamentarium and may contribute to decreased surgical morbidity in this complex patient population. Families should be counselled that endoscopic results may be more modest than with open or thoracoscopic approaches, and multiple procedures may be required.

1. Introduction

A tracheoesophageal fistula (TEF), or an aberrant communication between the trachea and esophagus, is a rare pediatric pathology associated with both acquired and congenital etiologies [1]. Congenital TEFs may occur in isolation or in conjunction with esophageal atresia [2]. Although open or thoracoscopic approaches are highly successful in

abating the fistula tract, the reported morbidity of these procedures may be significant, including respiratory complications and mediastinal leaks [3]. As such, endoscopic techniques have gained interest amongst airway surgeons as an alluring, minimally-invasive alternative in the following clinical scenarios [1]: a recurrent TEF (RTEF) after previous open/thoracoscopic repair [2], a missed TEF at the time of initial open/thoracoscopic repair, and [3] an isolated congenital (Gross

* Corresponding author.

E-mail addresses: Catherine.roy6@mail.mcgill.ca (C.F. Roy), alix.maltezeanu@mail.mcgill.ca (A. Maltezeanu), jeanmartinlaberge@hotmail.com (J.-M. Laberge), kimberley.kaspy.med@sss.gouv.qc.ca (K. Kaspy), ana.santanna@mcgill.ca (A. Sant'Anna), Helene.broucqsault@chu-lille.fr (H. Broucqsault), Pierre.fayoux@chu-lille.fr (P. Fayoux), Sam.daniel@mcgill.ca (S.J. Daniel).

<https://doi.org/10.1016/j.ijporl.2024.111960>

Received 5 November 2023; Received in revised form 16 April 2024; Accepted 21 April 2024

Available online 27 April 2024

0165-5876/  2024 The Authors. Published by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

H-type) or acquired fistula [3–5].

A plethora of endoscopic techniques have been explored, with approaches via either the tracheal or esophageal conduits [6,7]. De-epithelialization of the fistulous tract is considered by most authors a quintessential step of the procedure, creating a raw surface to stimulate subsequent fibrosis and healing of the mucosal edges leading to obliteration [5]. This may be achieved with mechanical disruption (brush), cauterization, chemical abrasion or laser [8]. The off-label use of chemical sealants, such as fibrin glue and histoacryl, and submucosal injection of filler materials in the neck of the fistula have also been described with variable success rates [8,9].

To date, the endoscopic TEF literature is replete with case reports or small case series in highly selected patients, providing an overall skewed portrait of expected outcomes. The most recent systematic review included 170 patients from 46 studies, with an overall success rate of 70 % [8]. However, 37 of the included papers represented case reports or small case series of fewer than six patients, entailing an inherent reporting bias and thus highlighting the need for ancillary large-sample studies [8]. We herein describe the surgical outcomes of endoscopic TEF repair at two high-volume tertiary care centers with advanced airway expertise, providing a contemporary account of these techniques in the therapeutic armamentarium of recurrent, missed, isolated, or acquired fistulas.

2. Methods

2.1. Patients, design, and setting

A retrospective chart review of consecutive patients undergoing endoscopic closure of a TEF from January 1, 2008 to December 31, 2020 was performed at the Montreal Children's Hospital in Montreal, Canada and the Lille Hospital University Center, Lille, France.

Patient data was retrieved from both institutions' databases, including demographics, medical history, type of fistula, endoscopic approach, complications and surgical outcomes. Operative reports were thoroughly reviewed, documenting the bronchoscopic description of the fistulous tract, associated airway anomalies, and endoscopic techniques used. All patients underwent a routine post-repair bronchoscopy to document fistula closure. Ethics approval was granted by the McGill University Health Centre Research Ethics Board and Lille institutional review board.

2.2. Literature search

A literature search was performed using the PubMed electronic database from inception to March 1, 2023. Key terms used in the search included *tracheoesophageal fistula*, *broncho-*, *trachea-*, *gastro-*, and *endoscopy*. References of relevant articles found in PubMed were reviewed for additional relevant studies. All studies including five patients or more reporting on outcomes of endoscopic TEF repair were included to mitigate the reporting bias with single cases or small case series. The type of fistula, endoscopic technique, average number of procedures, complications, success rates and length of follow-up were extracted from included studies.

2.3. Statistical analysis

Descriptive statistics (means, frequencies) were calculated for patient demographics, disease characteristics and surgical outcomes. Success rates for primary and recurrent TEFs were compared using a Fisher's exact test. The threshold for statistical significance of two-sided *P* values was set a less than 0.05. All analyses were performed using IBM SPSS (version 29.0.0, Armonk, NY).

3. Results

During the study period, 28 patients (17 from Lille, France and 11 from Montreal, Canada) underwent a total of 48 endoscopic procedures for repair of a TEF (mean 1.7 procedures per patient). The average age at the time of the first endoscopic procedure was 13.7 months (range 1–84). Patient demographics and comorbidities are illustrated in Table 1. Included patients were classified per the Gross classification [10] as follows: 18 type "C", 6 type "D", 2 type "B", 1 double-H fistula. One was classified as type "A" vs. "B" (missed proximal fistula during primary esophageal atresia repair versus acquired fistula during initial open esophageal atresia anastomosis). There were eight primary TEFs, while the remaining 20 TEFs were recurrences in the context of a previous open or thoracoscopic repair. Of mention, four of the patients with primary TEFs were missed fistulas in the context of a previous open repair. Closure of the fistula was ascertained by a post-operative bronchoscopy in all cases. Additionally, if there was any suspicious pouch, methylene blue was instilled via a soft suction catheter in the pouch, with visualization through a concomitant esophagoscopy to ensure there was no residual TEF (Fig. 1).

Endoscopic techniques were successful in obliterating the tracheoesophageal fistula in 16/28 patients (57 %), 10 of which were closed after a single procedure (mean 1.7 procedures, range 1–4). This represented six of the eight primary TEFs (75 %) and ten of the 20 recurrent TEFs (50 %), but the observed difference between primary and recurrent TEFs success rates failed to reach statistical significance ($P = 0.432$). The remaining 12 patients (43 %) underwent a successful open or thoracoscopic repair for a residual or recurrent fistula after a mean of 1.8 endoscopic procedures (range 1–4).

De-epithelialization of the TEF tract was performed in all cases, using a variety of techniques: thulium laser (19 procedures), flexible monopolar cautery (16 procedures), chemical cautery with 20–33 % trichloroacetic acid (5 procedures), mechanical abrasion with an endobronchial brush (7 procedures), and both brushing and flexible monopolar cautery (2 procedures). In 19 procedures, tissue adhesive was applied through the tract (either a fibrin sealant, 2-octyl cyanoacrylate or n-butyl-2-cyanoacrylate), while a submucosal injection of hyaluronic acid into the neck of the fistula to reapproximate the mucosal edges was performed in 11 procedures. Concomitant esophagoscopy during de-epithelialization and application of a tissue adhesive allowed to judge the depth of instrumentation, to ensure the entire length of the fistula was treated while minimizing risk of injury to the distal

Table 1
Patient demographics and characteristics of tracheoesophageal fistulas.

	No. (%)
Sex	
Male	14/28 (50)
Female	14/28 (50)
Associated syndromes	
VACTERL	9/28 (32.1)
Comorbidities	
Cardiovascular	16/28 (57.1)
Gastrointestinal	13/28 (46.4)
Genitourinary	12/28 (42.9)
Neurological	6/28 (21.4)
Musculoskeletal	6/28 (21.4)
Otologic	3/28 (10.7)
Kidney	7/28 [25]
Vertebral	8/28 (28.6)
Respiratory	4/28 (14.3)
Associated airway condition	
Tracheomalacia	23/28 (82.1)
Subglottic stenosis	9/28 (32.1)
Laryngeal cleft	9/28 (32.1)
Laryngomalacia	4/28 (14.3)
Vocal cord paralysis	3/28 (10.7)

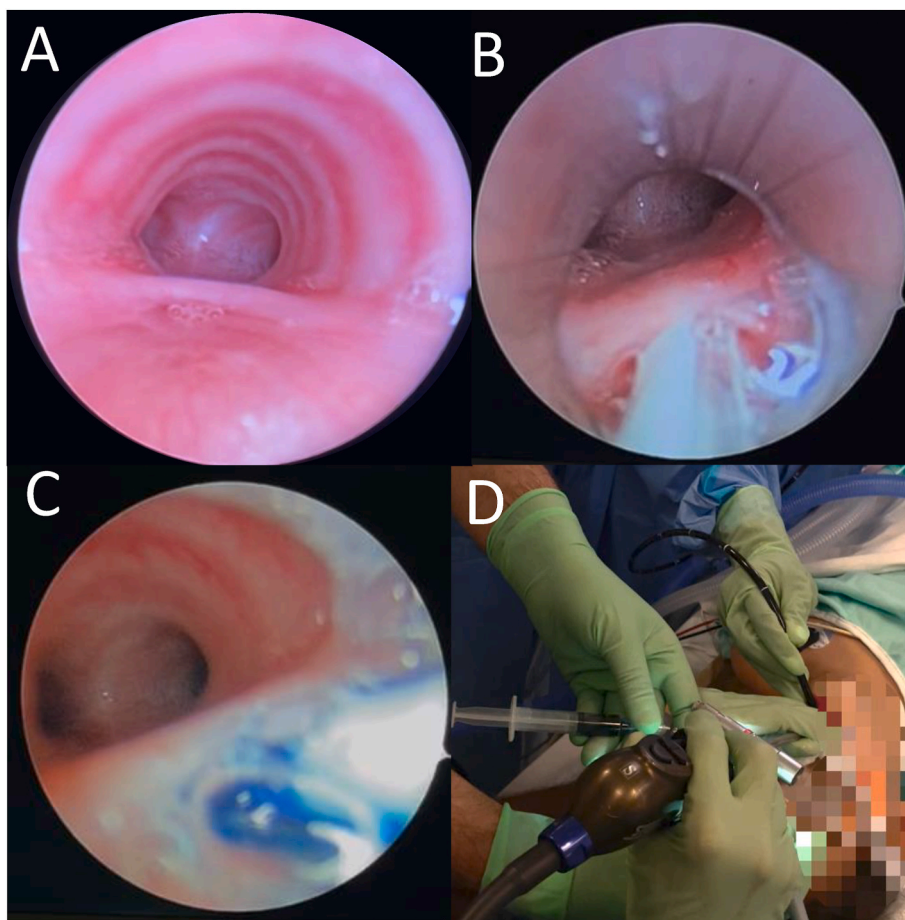


Fig. 1. Methylene blue is instilled through a soft suction catheter gently cannulated in the tracheoesophageal pouch or fistula via the working channel of a rigid bronchoscopy (A–C) with concomitant esophagoscopy (D) to visualize any dye leakage in the esophagus. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

esophageal mucosa. Esophageal clipping as described by Propst et al. was performed in two patients [11], and one patient had a concomitant esophageal endoprosthesis for associated esophageal stenosis at the site of the TEF. While endoscopic techniques varied, the reader may refer to a previously published video by the authors describing combined de-epithelialization with monopolar cautery and a hyaluronic acid submucosal injection [12].

While the procedure itself was performed via a rigid bronchoscope through the trachea, all cases were scheduled on designated triple-endoscopy operative lists with flexible bronchoscopy and esophagoscopy available. This proved useful to ascertain fistula patency pre-operatively (by intubating the fistula with the flexible bronchoscope or confirming methylene blue in the esophagus with the esophagoscope), for rapid removal in the event of an iatrogenic foreign body such as a sealant plug, and esophageal clipping when performed.

There were no major intra-operative or post-operative complications. However, five patients experienced minor respiratory complications attributed to their underlying comorbidities and airway instrumentation as detailed herein. Three of these patients developed post-operative dyspnea (two attributed to their underlying tracheomalacia, one due to minor subglottic edema), but evolved favorably with inhaled and systemic steroids with no need for supplemental oxygen. While almost all patients were transferred to the recovery room extubated, two patients treated at the beginning of our series remained intubated for 24-h as per our initial protocol. One patient was accidentally extubated, but subsequently remained on high-flow cannula and was rapidly weaned to room air after a short course of intravenous steroids. The other patient developed a ventilator-associated pneumonia but was

treated with antibiotics and promptly extubated to room air. There was no death, pneumothorax or mediastinitis. The mean follow-up was of 50.8 months (range 2–168).

4. Literature review and discussion

Tracheoesophageal fistulas remain a challenging pathology to treat, and the rarity of this condition obligates the reporting of surgical series to provide insight into the expected outcomes. First introduced in the German literature in the 1970s, endoscopic approaches have emerged as an elegant and promising therapeutic avenue due the minimally-invasive nature of the procedure [3]. We herein report the largest case series to date, including data from two high-volume tertiary care institutions.

Reported success rates with endoscopic approaches are highly variable, owing to the heterogeneity in patient-specific factors (etiology, size and location of fistula), surgical techniques, and methods used to ascertain fistula closure (i.e. resolution of clinical symptoms, radiological studies or routine post-intervention bronchoscopy). A 2023 systematic review by Ling et al. reported an overall success rate of 70 % in 170 patients treated endoscopically [8]. This rate is perhaps artificially inflated as most included studies represented single case reports or small case series, thus likely introducing a reporting bias [8]. Our review of the literature yielded 13 studies) [4,5,7,9,13–21] reporting on greater than five patients with TEFs managed endoscopically, with an overall success rate of 61.1 % (156 patients, 157 fistulas) (Table 3), commensurate with the current case series.

Recurrent TEFs are typically more challenging to treat due to post-

Table 2
Fistula characteristics, endoscopic techniques and surgical outcomes.

Patient	Fistula type	Primary vs. recurrent	Age at first endoscopic treatment (months)	Endoscopic technique	Successful endoscopic closure	Complications	Follow-up (months)
1	C	Recurrent	9	#1, 2, 3 Laser #4 33 % TCA	Y	Dyspnea in recovery treated with inhaled steroids, no supplemental O2	70
2	B	Primary	1	#1 TCA	Y	None	10
3	D	Primary (missed)	23	#1 Laser	N	None	2
4	C	Recurrent	29	#1 Laser	Y	Accidental extubation, weaned to high-flow nasal cannula then room air	67
5	C	Primary	4.5	#1 Laser + Esophageal clip	Y	None	8
6	D	Recurrent	1.5	#1 Laser + Dermabond	Y	Subglottic edema treated with inhaled and intravenous steroids, no supplemental oxygen	51
7	C	Recurrent	7	#1 Laser	N	None	46
8	C	Recurrent	24	#1 Laser + Esophageal clip	Y	Ventilator-associated pneumonia treated with antibiotics	40
9	C	Recurrent	84	#1 Brush + Dermabond #2 Laser + Dermabond	N	None	12
10	Double-H	Recurrent	4	#1 Laser	N	None	6
11	C	Recurrent	8	#1 Laser #2 Laser	N	None	7
12	C	Recurrent	2.5	#1 Laser	Y	Dyspnea and stridor in recovery treated with inhaled steroids	2
13	D	Primary (missed)	3.5	#1 Laser + Dermabond	Y	None	87
14	C	Recurrent	17.5	#1 Laser + Dermabond	Y	None	56
15	C	Recurrent	1	#1 Laser + Esophageal endoprosthesis	N	None	6
16	C	Recurrent	48	#1 Brush + Dermabond	N	None	39
17	B	Primary (missed)	16.5	#1 33 % TCA	Y	None	23
18	C	Recurrent	3	#1 Brush + Tisseel glue #2 Bugbee + Dermabond #3 Brush + Tisseel glue #4 Bugbee + Tisseel	N	None	90
19	C	Recurrent	5	#1 Bugbee + Tisseel + Deflux #2 20 % TCA + Deflux #3 Bugbee + Dermabond #4 Bugbee + Deflux	N	None	86
20	A vs. B	Primary (missed) vs. acquired	5	#1 Bugbee + Tisseel + Deflux #2 Bugbee	Y	None	60
21	C	Recurrent	68	#1 Brush + Bugbee + EpiDermGlu + Tisseel	Y	None	137
22	C	Recurrent	3	#1 Brush + Tisseel #2 Bugbee + Tisseel	N	None	121
23	D	Primary (missed)	1	#1 Bugbee + Deflux	N	None	39
24	D	Primary (missed)	1	#1 Bugbee	Y	None	37
25	C	Recurrent	3	#1 Brush + Bugbee + Deflux #2 Bugbee + Deflux	N	None	35
26	C	Recurrent	4	#1 Bugbee + Deflux #2 Bugbee + Deflux	Y	None	35
27	D	Recurrent	1	#1 Brush + Histoacryl	Y	None	168
28	C	Recurrent	5	#1 Bugbee + Tisseel #2 Bugbee + Tisseel + Deflux #3 20 % TCA + Deflux	Y	None	83
Overall		8 primary, 20 recurrent	13.7 ± 20.8	Mean # of procedures 1.7 (range 1–4)	16/28 (57.1 %)		50.8 ± 43.5

Deflux®, hyaluronic acid and dextranomer (Palette Life Sciences, Santa Barbara, CA, USA); Dermabond®, fibrin glue and 2-octylcyanoacrylate (Ethicon, Raritan, NJ, USA); Histoacryl®, monomeric n-butyl-2-cyanoacrylate (B. Braun, Melsungen, Germany); Tisseel®, fibrin sealant (Baxter, Deerfield, IL, USA); TCA, Trichloroacetic Acid.

operative adhesions and chronic inflammation [8]. Our series showed a trend towards higher success rates in primary fistulas when compared to recurrent disease, though this did not reach significance. While surgeons should be aware of the potential pitfalls of endoscopic repair in the

recurrent setting, this remains the primary indication for this novel approach. Primary TEFs are often repaired concomitantly with the initial esophageal atresia repair, however congenital H-type or missed TEFs at the time of the initial repair may be amenable to endoscopic

Table 3
Literature review of reported series including 5 patients or more.

Author, year	City, Country	N	Type of fistula	Endoscopic technique	Routine post-procedure bronchoscopy	Avg number of procedures	Complications	Success Rate	Follow-up
Willets, 1998	11 institutions	22	RTEF	FG [11], histoacryl [9], FG and histoacryl [1], combination of histoacryl + sclerosant [2]	No	2.1 [1–4]	–	12/22 (54.5)	Median 107 (3–264)
Bhatnagar, 1999	New Delhi, India	5	3 RTEF, 2 congenital H-type	Diathermy coagulation [3] or Nd:YAG laser [2]	Yes	2 [1–4]	Respiratory distress requiring a 48H intubation [2] and a temporary tracheostomy [1]	3/5 (60 %)	3mo
Montedonico, 1999	Madrid, Spain	9	8 RTEF, 1 congenital H-type	8 histoacryl, 1 fibrin glue	No	2	–	0/9 (0)	18mo
Khurana, 2004	–	6	RTEF	Diathermy coagulation	No	2.2 [1–3]	–	5/6 (83.3 %)	4.42yrs (16mo–8yrs)
Tzifa, 2006	Toronto, Canada	9 patients, 10 TEFs	2 H-type, 1 traumatic, 7 RTEFs (including 1 patient with 2 TEFs)	Electrocautery or mechanical abrasion, histoacryl tissue adhesive	No	1.5 [1,2]	–	9/10 (90 %)	3mo–9yrs
Briganti, 2011	Roma, Italy	5	RTEF	Mechanical abrasion, submucosal Deflux injection	No	2 for two patients in which repair was successful	–	2/5 (40 %)	–
Lelonge, 2016	Santiago, Chile + Saint Etienne, France	14	12 RTEF, 2 primary TEFs missed at time of EA repair	TCA	Yes	2 [1–3]	Postoperative pneumonia treated with IV antibiotics (N = 1)	14/14 (100 %)	41mo (8–71)
Nazir, 2016	Karachi, Pakistan	5	3 congenital TEFs, 2 acquired	Electrocautery, FG	No	1–3	–	5/5 (100 %)	4.2yrs (7mo–10yrs)
Valiyev, 2019	Istanbul, Turkey	9	RTEF	Laser [4], TCA [4] or both [1] with FG	Yes	–	–	1/9 (11.1 %)	28 mo (3mo–5yrs)
Luscan, 2020	Paris, France	11	6 RTEF, 5 H-type	Laser Adhesive surgical glue in one case	Yes	1.2 [1,2]	Necrosis of posterior tracheal wall (N = 1)	4/11 (36.4 %)	Median 24mo (14–72)
Miro, 2020	Valencia, Spain	14	RTEF	Diathermy in all but 3 cases + FG	No	2.1 [1–5]	–	10/14 (71.7 %)	12.1 yrs [10–20]
Sautin, 2021	Minsk, Belarus	12	Isolated or RTEF	Laser	No	2.08 [1–4]	–	8/12 (66.7 %)	Median 3.7yrs
Valero Mamani, 2022	Mexico City, Mexico	7	6 RTEF, 1 primary TEF	Bronchial brush, TCA	Yes	2.2 [1–4]	–	7/7 (100 %)	33mo (9–72mo)
Roy, 2023	Montreal, Canada + Lille, France	28	8 primary, 20 RTEF	Various (see Table 1)	Yes	1.7 [1–4]	–	16/28 (57.1 %)	50.8 months (2–168 mo)
Overall	–	128 patients, 129 fistulas	–	–	–	–	–	96/157 (61.1 %)	–

RTEF = Recurrent tracheoesophageal fistula, FG = Fibrin Glue, TCA = Trichloroacetic acid.

techniques.

Various endoscopic techniques have been described in the literature, most commonly one or more of the following: de-epithelialization, use of a sealant and/or submucosal augmentation material. The aforementioned systematic review by Ling et al. reported high rates of closure with combination therapy (de-epithelialization and sealant) when compared to either in isolation [8]. Of note, a further subgroup analysis showed no statistically significant difference between various de-epithelialization techniques (chemocauterization, laser or electrocautery), nor between the two most commonly used sealants (fibrin glue and histoacryl) [8]. A systematic review of 127 recurrent- and H-type TEFs found de-epithelialization (either alone or in combination) was associated with the highest success rates when compared to sealants

alone, suggesting adhesives may be absorbed prior to adequate fibrosis and healing of the tract [22]. Endoscopic techniques used in our cohorts were highly heterogeneous, and adjuncts such as injection of augmentation material, clips or stents were used selectively in challenging cases thus preventing subgroup analyses and a direct comparison of various methods.

An important consideration in discussing the endoscopic management of TEFs is the frequent need for multiple interventions to achieve a successful closure [22]. While most series report a mean number of procedures approaching two (Table 2), there is no commonly agreed upon “threshold” to abandon endoscopic techniques and consider a conventional open or thoracoscopic repair. Miro et al. [16] suggested up to five attempts may be considered, while Tzifa et al. [5] advise

endoscopic repair be reserved to small, non-patulous fistula for no more than two procedures. Until further clarity is obtained with larger prospective trials, airway surgeons should guide this decision based on their experience, parental preference, and careful consideration of the severity of ongoing TEF symptomatology and the morbidity of available surgical approaches. Additionally, and perhaps most importantly, the value of multi-disciplinary input in this complex and individualized decision-making should be emphasized. At both institutions included in the current paper, patients with TEFs are closely followed by a team including otolaryngologists, pediatric surgeons, gastroenterologists, respirologists, pediatricians, and allied health professionals.

While the success rate of open surgery is undoubtedly superior to that of endoscopic approaches, the associated morbidity of the former must be considered. A systematic review of 165 patients with RTEFs reported a major complication rate of 16 % (mostly anastomotic leaks) and 3.7 % mortality for open surgery, compared to a 5 % rate of respiratory distress and 1.7 % death in patients treated endoscopically [3]. Congenital H-type and missed proximal TEFs (in type B or D) can be approached through a cervical incision if located above the level of the second thoracic vertebrae, with less risk of severe morbidity than a thoracotomy [23]. However, damage to the recurrent laryngeal nerve is a well-recognized risk with open cervical approaches and must be born in mind in the decision-making process [23].

While serious adverse events have been reported with endoscopic treatment, these are overall rare and may be mitigated by refinements in surgical technique and careful use of laser or electrocautery for de-epithelialization. For instance, Luscan et al. described an extensive posterior tracheal wall necrosis following an attempt at endoscopic repair which required open tracheal reconstruction but note this occurred when 212J of laser energy were used (median 34J in this series) [15]. Additionally, when a sealant or augmentation material is used, care should be taken to avoid over injection and optical endobronchial grasping forceps should be available, as endobronchial spillage of a plug necessitating removal has been previously reported [24].

Laryngotracheal anomalies in children with EA and TEFs are highly prevalent, as demonstrated in a recent retrospective review where 89 % of children had tracheomalacia, and 27 % had other airway pathologies including laryngeal clefts, laryngomalacia, vocal cord paralysis and subglottic stenosis [25]. At both centers included in the current study, systematic and routine airway assessment via laryngotracheobronchoscopy is performed in all children with EA-TEFs. These children typically undergo multiple evaluations and other procedures under general anesthesia (including esophageal dilations, gastrostomy tubes, corrective airway surgery for other laryngotracheal anomalies). Thus, it is felt at least one endoscopic attempt at closure in cases of recurrent or missed TEFs is of little added risk when performed under the same anesthesia, with the potential of avoiding more invasive surgery. Nonetheless, in light of the current study and review of the literature, families must be aware of the frequent need for multiple interventions, and possibility of requiring a subsequent open or thoracoscopic repair in cases of failure.

This study is not without limitations, including the inherent challenges of retrospective data including missing data and inconsistent documentation. Additionally, the heterogeneity of the included patients and endoscopic modalities employed limit our ability to provide insight into the optimal surgical technique. Certainly, ancillary prospective and large sample multicentric studies will be required to establish superiority of various endoscopic modalities.

5. Conclusion

We herein present the largest series to date using endoscopic techniques for TEFs from two tertiary care institutions and provide insight on the expected surgical outcomes. The overall success rate of endoscopic approaches cumulating data from this series and all published studies with greater than 5 patients is of 61.1 %. While surgical outcomes are

more modest than standard open and thoracoscopic approaches, the minimal associated morbidity makes endoscopic repair an enticing alternative.

Financial Support

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

CRediT authorship contribution statement

Catherine F. Roy: Writing – review & editing, Writing – original draft, Methodology, Formal analysis, Data curation. **Alix Maltezeanu:** Writing – review & editing, Writing – original draft, Validation, Methodology, Formal analysis, Data curation. **Jean-Martin Labege:** Validation, Supervision, Formal analysis, Data curation. **Kimberley Kaspy:** Writing – review & editing, Supervision, Data curation. **Ana Sant’Anna:** Writing – review & editing, Validation, Supervision, Formal analysis, Data curation. **Hélène Broucqsault:** Writing – review & editing, Validation, Supervision, Data curation. **Pierre Fayoux:** Writing – review & editing, Writing – original draft, Visualization, Validation, Supervision, Methodology, Investigation, Data curation, Conceptualization. **Sam J. Daniel:** Writing – review & editing, Writing – original draft, Visualization, Validation, Supervision, Resources, Methodology, Investigation, Funding acquisition, Data curation, Conceptualization.

Declaration of competing interest

The authors have no competing interests to declare.

Acknowledgements

We would like to acknowledge the contributions of Dr. Rony Sfeir, Dr. Frédéric Gottrand, Dr. Dominique Lévesque, Dr. Adam J. Shapiro, Dr. Véronique Morinville, Dr. Mylène Dandavino, Dr. David Zielinski and Helene Bacha.

References

- [1] R.S. Gutierrez, M. Guelfand, P.V. Balbontin, Congenital and acquired tracheoesophageal fistulas in children, *Semin. Pediatr. Surg.* 30 (3) (2021) 151060, <https://doi.org/10.1016/j.sempepsurg.2021.151060>.
- [2] N.A. McGowan, J. Grosel, An overview of esophageal atresia and tracheoesophageal fistula, *JAAPA* 35 (6) (2022) 34–37, <https://doi.org/10.1097/01.JAA.0000830180.79745.b9>.
- [3] O. Aworanti, S. Awadalla, Management of recurrent tracheoesophageal fistulas: a systematic review, *Eur. J. Pediatr. Surg.* 24 (5) (2014) 365–375, <https://doi.org/10.1055/s-0034-1370780>.
- [4] Y. Lelonge, F. Varlet, P. Varela, et al., Chemocauterization with trichloroacetic acid in congenital and recurrent tracheoesophageal fistula: a minimally invasive treatment, *Surg. Endosc.* 30 (4) (2016) 1662–1666, <https://doi.org/10.1007/s00464-015-4352-1>.
- [5] K.T. Tzifa, E.L. Maxwell, P. Chait, et al., Endoscopic treatment of congenital H-Type and recurrent tracheoesophageal fistula with electrocautery and histoacryl glue, *Int. J. Pediatr. Otorhinolaryngol.* 70 (5) (2006) 925–930, <https://doi.org/10.1016/j.ijporl.2005.10.017>.
- [6] T. Al Lawati, O.I. Saadah, M. Al Sajwani, Outcomes of endoscopic closure of respiratory esophageal fistula in children using the esophageal approach: a case series, *Cureus* 14 (10) (2022) e29985, <https://doi.org/10.7759/cureus.29985>.
- [7] R.J. Valero Mamani, J. Penchyna Grub, G. Blanco Rodríguez, G. Teysier Morales, M. Peña García, Endoscopic management of recurrent tracheoesophageal fistula with trichloroacetic acid in pediatric patients, *Cir. Pediatr.* 35 (3) (2022) 113–117, <https://doi.org/10.54847/cp.2022.03.13>.
- [8] Y. Ling, B. Sun, J. Li, et al., Endoscopic interventional therapies for tracheoesophageal fistulas in children: a systematic review, *Front Pediatr* 11 (2023) 1121803, <https://doi.org/10.3389/fped.2023.1121803>.
- [9] V. Briganti, R. Coletta, G. Giannino, A. Calisti, Usefulness of dextranomer/hyaluronic acid copolymer in bronchoscopic treatment of recurrent tracheoesophageal fistula in children, *Int. J. Pediatr. Otorhinolaryngol.* 75 (9) (2011) 1191–1194, <https://doi.org/10.1016/j.ijporl.2011.06.018>.
- [10] R.E. Grossm the Surgery of Infancy and Childhood, Saunders, United States, 1953.
- [11] E.J. Propst, S.C. Ling, A. Daneman, J.C. Langer, Endoscopic clip for closure of persistent tracheoesophageal fistula in an infant, *Laryngoscope* 124 (9) (2014) 2182–2185, <https://doi.org/10.1002/lary.24650>.

- [12] T. Chen, S.J. Daniel, Endoscopic repair of tracheoesophageal fistula: an interdisciplinary approach (with video), *European annals of otorhinolaryngology, head and neck diseases* 138 (2021) 8–9, <https://doi.org/10.1016/j.anorl.2021.05.011>, 2021/08/01.
- [13] A. Sautin, K. Marakhouski, A. Pataleta, A. Svirsky, V. Averyn, Treatment of isolated and recurrent tracheoesophageal fistula in children: a case series and literature review, *World J Pediatr Surg* 4 (4) (2021) e000316, <https://doi.org/10.1136/wjps-2021-000316>.
- [14] N. Valiyev, M.Y. Erdas, A.P. Ergenekon, G. Kiyani, Bronchoscopic treatment of recurrent tracheoesophageal fistula: is it an effective option? *Dis. Esophagus* 32 (1) (2019) <https://doi.org/10.1093/dote/doz047.91>.
- [15] R. Luscan, F. Simon, N. Khen Dunlop, et al., Thulium LASER for endoscopic closure of tracheoesophageal fistula in esophageal atresia's spectrum: an appropriate tool? *J. Pediatr. Surg.* 56 (10) (2021) 1752–1756, <https://doi.org/10.1016/j.jpedsurg.2020.10.005>.
- [16] I. Miró, C. Gutiérrez, E. Carazo, et al., Fibrin glue treatment associated or not with diathermy for recurrent tracheoesophageal fistula: our results after more than 20 years' experience, *Cir. Pediatr.* 33 (3) (2020) 115–118.
- [17] Z. Nazir, M.A.M. Khan, J. Qamar, Recurrent and acquired tracheoesophageal fistulae (TEF)-Minimally invasive management, *J. Pediatr. Surg.* 52 (10) (2017) 1688–1690, <https://doi.org/10.1016/j.jpedsurg.2017.03.048>.
- [18] S. Khurana, A. Ford, Long-term results of endoscopic diathermy coagulation of recurrent tracheoesophageal fistula, *Pediatr Endosurg Innov Tech* 8 (1) (2004) 31–34, <https://doi.org/10.1089/109264104773513115>.
- [19] V. Bhatnagar, R. Lal, M. Srinivas, S. Agarwala, D.K. Mitra, Endoscopic treatment of tracheoesophageal fistula using electrocautery and the Nd:YAG laser, *J Pediatr Surgery* 34 (3) (1999) 464–467, [https://doi.org/10.1016/s0022-3468\(99\)90500-6](https://doi.org/10.1016/s0022-3468(99)90500-6).
- [20] S. Montedonico, J.A. Díez-Pardo, L. Lassaletta, J.A. Tovar, Adhesivos tisulares en la refistulización de la atresia de esófago [Tissue adhesives in closing of fistulas after surgery of esophageal atresia], *Cir. Pediatr.* 12 (3) (1999) 110–112.
- [21] I.E. Willets, N.E. Dudley, P.K. Tam, Endoscopic treatment of recurrent tracheoesophageal fistulae: long-term results, *Pediatr. Surg. Int.* 13 (1998) 256–258.
- [22] A. Tobia, C. Grau Luque, K. Leitmeye, M. Dorling, N.K. Chadha, Endoscopic treatment in pediatric patients with recurrent and H-type tracheoesophageal fistulas – a systematic review and meta-analysis, *Int. J. Pediatr. Otorhinolaryngol.* 168 (2023), <https://doi.org/10.1016/j.ijporl.2023.111541>.
- [23] A.H. Al-Salem, M.A. Mohaidly, H.M. Al-Buainain, S. Al-Jadaan, E. Raboei, Congenital H-type tracheoesophageal fistula: a national multicenter study, *Pediatr. Surg. Int.* 32 (5) (2016) 487–491, <https://doi.org/10.1007/s00383-016-3873-6>.
- [24] N.E. Wiseman, Endoscopic closure of recurrent tracheoesophageal fistula using Tisseel, *J. Pediatr. Surg.* 30 (8) (1995) 1236–1237, [https://doi.org/10.1016/0022-3468\(95\)90031-4](https://doi.org/10.1016/0022-3468(95)90031-4).
- [25] P. Fayoux, M. Morisse, R. Sfeir, L. Michaud, S. Daniel, Laryngotracheal anomalies associated with esophageal atresia: importance of early diagnosis, *Eur. Arch. Oto-Rhino-Laryngol.* 275 (2) (2018) 477–481, <https://doi.org/10.1007/s00405-017-4856-5>.