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ORIGINAL ARTICLE

Secondary Voice Restoration After Laryngotracheal Separation (LTS) for Dysphagia with Intractable Aspiration

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Abstract Intractable aspiration is a serious, often life-threatening condition due to its potential impact on pulmonary function. Aspiration requires therapeutic measures, starting with conservative management but often necessitating surgical treatment. The basic surgical principle is to separate the alimentary and respiratory tracts through a variety of procedures which, unfortunately, nearly all result in the loss of phonation, with the exception of total laryngectomy (TL) which includes the placement of an indwelling voice prosthesis. In this study, we present a modified laryngotracheal separation (LTS) technique that, we believe, offers multiple advantages compared to standard TL. After reviewing the medical records of 35 patients with intractable aspiration who have undergone LTS, we describe the surgical technique and present the postoperative result. In a second surgical procedure about two months following LTS, we aimed to achieve voice restoration by placement of an indwelling voice prosthesis. Intractable aspiration was successfully treated in all patients. Placement of an indwelling voice prosthesis during a second operation was successful in 15 patients, representing the largest reported cohort thus far. LTS is a reliable surgical technique to treat intractable aspiration, with restoration of oral intake, thereby improving the general condition and quality of life of these unfortunate patients. Furthermore, voice restoration can be achieved in selected patients, by placement of a voice prosthesis.

Keywords Intractable aspiration · Surgical management · Laryngotracheal separation · Deglutition · Deglutition disorders

Introduction

Intractable swallowing disorders associated with aspiration are a challenging medical problem that can precipitate life-threatening pulmonary disorders such as pneumonia, bronchiectasis, and interstitial lung disease [1]. Conservative medical treatment, speech-language pathologist therapy, and protective tracheotomy are often insufficient to control chronic aspiration. More invasive surgical procedures designed to permanently treat intractable aspiration have been described [2, 3]. These techniques completely separate the alimentary and respiratory tracts. Some rely on tracheotomy with glottic closure or supraglottic laryngeal inlet closure. Others more drastically interfere with the normal anatomy: total laryngectomy or laryngotracheal separation (LTS). In 1976, Lindeman described the use of tracheo-esophageal diversion for the management of intractable aspiration [4], a procedure in which the trachea is divided and the upper tracheal segment is anastomosed to the esophagus through its anterior wall. The distal tracheal end is brought out to the skin to form a permanent tracheostoma. One year later, he simplified his procedure, by closing the proximal tracheal stump as a pouch instead of diverting it into the esophagus [5, 6]. Interestingly, spontaneous evacuation of the contents of this pouch into

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the natural esophageal inlet has been shown [7]. It is therefore preferable to leave the esophagus untouched in LTS, as the safety and reliability of this procedure has been studied extensively and many have confirmed the validity and usefulness of this technique for the treatment of intractable aspiration. TL is more likely to add a mechanical component to the existing dysphagia due to reduction of the diameter of the pharynx when closing the pharynx on its own after the removal of the larynx. All procedures result in loss of speech production. In TL patients, there is abundant experience with the use of voice prostheses to restore speech while in LTS, there have only been sporadic reports. The choice of the surgical procedure is primarily determined by the general condition of the patient, which is usually poor in case of intractable aspiration. A simple, reliable, and easy to perform surgical intervention with little complication risk is preferable to help these fragile patients. Therefore, we have chosen to treat our patients with the LTS technique, adding some variation in closure of the mucoperichondrial pouch and using voice prosthesis insertion in selected cases. In this article, we describe the results of an additional modification to the LTS procedure and our experience with the insertion of voice prostheses in these patients.

Patients and Methods

Inclusion Criteria

From 2006 to 2013, 35 patients with intractable aspiration underwent LTS surgery in the Department of Head, Neck & Maxillofacial Surgery at Ghent University Hospital. After local ethics committee approval, a retrospective analysis of our hospital records was performed. Aspiration was confirmed by clinical evaluation, fiber optic endoscopic assessment of swallowing, and preoperative videofluoroscopy if the patient's condition made it feasible. Indications for surgery were one or more of the following: (1) a long standing history of aspiration (2) aspiration which could not be controlled despite swallowing rehabilitation by an experienced speech therapist, or (3) an aspiration score higher than 2 on videofluoroscopy (see "Clinical Data"). Patients with medical co morbidities resulting in poor prognosis were excluded, along with those for whom general anesthesia was contraindicated. Information regarding type of diet and feeding method pre- and postoperatively was collected.

Surgical Procedure

Two senior surgeons performed all LTS operations under general anesthesia with ventilation through an orotracheal tube whenever possible, even in the presence of a pre-existing tracheotomy. A nasogastric tube was placed unless a jejunostomy was already present. A horizontal neck skin incision was made, approximately 1 cm above the manubrium sterni. The skin and platysma flap was dissected up to the superior border of the thyroid cartilage. The sternohyoid and sternothyroid muscles were split in the midline and retracted laterally. The underlying thyroid gland was split at the isthmus and each lobe lateralized. A horizontal incision was made in the trachea between the second and third ring or at the level of the pre-existing tracheotomy (Fig. 1a). The distal part of the trachea was then sutured to the skin, creating the lower border of the later tracheostomy opening. The orotracheal tube was then replaced by a tracheal ventilation tube. Next, the cartilage of the anterior part of the cricoid and the adjacent tracheal rings of the proximal tracheal stump were removed, preserving the mucoperichondrium. The subglottic mucoperichondrium is thicker than the tracheal one, facilitating the creation of a more robust closure of the pouch (Fig. 1b). The strap muscles and one thyroid lobe pedicled on its superior vascular pole were used as vascularized tissue flaps to reinforce the subglottic mucoperichondrial pouch suture. The sternal heads of the sternocleidomastoid muscles were released from the bone optimizing the position of the tracheostomy for potential future speech valve rehabilitation. After skin and subcutis closure (Fig. 2), a low-pressure cuffed Shiley^{TM} cannula was placed to protect the lower airway during the first postoperative days (Fig. 2).

Postoperative care consisted of general wound and cannula care. Aerosol therapy with saline solution was

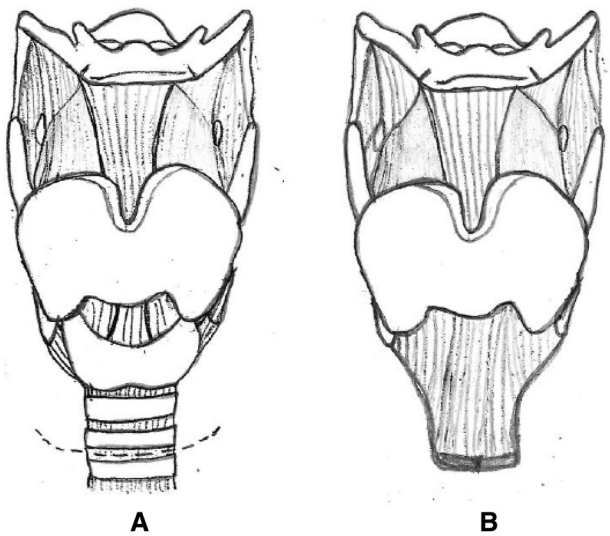

Fig. 1 The tracheal incision is made between the second and third tracheal ring (**a**). Proximal tracheal pouch, after removal of the anterior part of the cricoid and upper two tracheal rings (**b**)

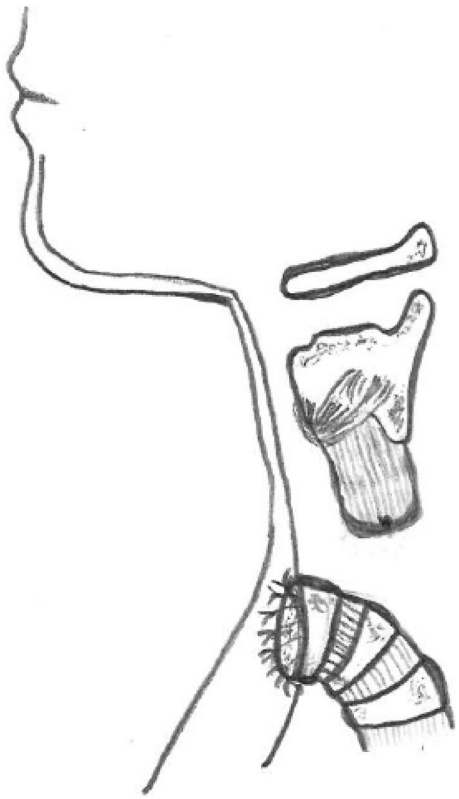

Fig. 2 Permanent tracheostoma and proximal pouch after modified laryngotracheal separation at the Ghent University Hospital

administered to prevent mucus impaction in the trachea. The Shiley cannula was replaced by a silicone Larytube™ on the second postoperative day. Prophylactic antibiotic treatment (Amoxicillin/Clavulanic Acid, 875/125 mg three times daily for 10 days) was initiated only if the patient had a history of radiation therapy. Skin and stomal sutures were removed after 8 and 10 days, respectively. On the second postoperative day, the patient was allowed to drink water. If there were no signs of leakage, oral feeding was started. However, for patients with dysphagia and aspiration after radiation therapy, oral intake was not started until 8 days after surgery. Two months after surgery, an insufflation test was performed using a thin silicone tube positioned into the upper esophagus through the nose. Air is then introduced into the upper esophagus at the approximate level of the future voice prosthesis. If the air flows gently upward and produces a basic sound, then, patient will most likely be able to speak with a voice prosthesis. The following selection criteria were used for secondary voice prosthesis placement: (a) ability to produce functional speech pre-operatively without aphasia or other speech-language deficiencies; (b) absence of neurological deterioration; (c) absence of upper extremity disabilities which would make manual closure of the tracheostoma impossible; (d) absence of severe fibrosis in the neck with potential impaired tissue healing after placement of the voice prosthesis; (e) positive insufflation test after healing of the LTS procedure.

The placement of the voice prosthesis was performed under general anesthesia, using jet-ventilation via the tracheostoma. We first placed a rigid esophagoscope transorally, under direct vision using a 0° endoscope. The esophagoscope was inserted up to the level of the tracheostoma, where the tracheo-esophageal puncture (TEP) was performed. We used a 2.5 mm sterile puncture needle for the TEP, and placed a guide wire through the puncture needle with the endoscope still in place. The guide wire was brought out through the mouth while retracting the esophagoscope. After removal of the puncture needle, the guide wire was attached to the voice prosthesis, allowing a retrograde insertion.

Statistical Analysis

IBM SPSS statistics 20 was used to analyze data. All data are expressed as mean ± standard error of the mean. A significance level of $P < 0{,}05$ was used.

Results

Patient Characteristics

Among the 35 patients studied, 11 had a history of a previous head and neck tumor. Ten of these 11 patients were treated with radiation therapy alone or in combination with other treatment modalities. One patient with a supraglottic tumor had a hemi-laryngectomy and bilateral neck dissection. The other 22 patients suffered a causal neurological disorder (Table 1).

Clinical Data

Mean patient age at time of surgery was 63.2 ± 9.6 years; 9 out of 35 (25.7 %) patients were 70 or older (Table 2). There was a male predominance (77.1 %).

To assess the severity of aspiration, if possible a video fluoroscopic examination was done, and scored according to the classic scale: 0 = normal; 1 = stasis in the vallecula or in the piriform sinus; 2 = penetration of contrast fluid in the larynx; 3 = aspiration of contrast fluid in the airway. The majority (88 %) had a score of 3, indicating severe aspiration. In 10 patients, videofluoroscopy was not possible because of severe disability. For those patients, the severity of aspiration was assessed by fiber optic evaluation alone.

Pre-operatively, 20 patients were fed through a gastrostomy, 5 patients through a nasogastric tube, and 2

Table 1 Underlying disease in 35 patients treated with LTS

Underlying disease	*N*	Total percentage
Therapy for head and neck tumor	11	31.6
Resection + radiation therapy	1	2.9
Neck dissection + radiation therapy	3	8.6
Neck dissection + chemoradiation therapy	1	2.9
Radiation therapy	3	8.6
Chemoradiation therapy	2	5.7
Hemilaryngectomy + neck dissection	1	2.9
Neurological disease	22	63.1
Cerebrovascular accident	5	14.3
Multiple sclerosis	1	2.9
Parkinson's disease	3	8.6
Amyotrophic lateral sclerosis	1	2.9
Cerebral palsy after meningo-encephalitis (TB)	1	2.9
Congenital obstructive hydrocephaly	1	2.9
Head injury	9	25.7
Brainstem tumor	1	2.9
Alcoholic polyneuropathy	1	2.9
B Cell lymphoma of the stomach treated with chemotherapy and stem cell transplantation, general weakness	1	2.9
Total	35	100

Table 2 Summary of patient characteristics (N = 35)

Characteristic	*N*	Total percentage
Sex		
Male	27	77.1
Female	8	22.9
Age		
<70 years	26	74.1
>70 years	9	25.9
Feeding condition preoperative		
Oral	7	20.0
TPN	1	2.9
Nasogastric	5	14.3
Gastrostomy	20	57.1
Jejunostomy	2	5.7
Tracheotomy preoperative		
Yes	22	62.9
No	13	37.1
Aspiration score		
0: Normal	0	0
1: Stasis vallecula/piriform sinus	2	8
2: Penetration	1	4
3: Aspiration	22	88
Missing data	10	

patients through a jejunostomy tube. Seven patients did not require enteral feeding, achieving their caloric needs through oral intake with dietary adjustment (thickening of liquids or blending of solid food). One patient received total parenteral nutrition.

The mean operative time of the modified LTS procedure as described above was 162 ± 34 min. In four patients, the operation was initiated as a LTS, but was converted to a TL due to poor tissue quality and concern for subsequent healing difficulties. These patients were excluded from this study.

After LTS, all patients were free of aspiration, as was clinically evaluated during swallowing rehabilitation. At discharge, more than half of the patients (53.1 %) regained sufficient swallowing function to enable adequate oral intake (Table 3). The remaining patients were discharged with additional nutritional support through a jejunal feeding tube to ensure adequate caloric intake.

A summary of postoperative complications is presented in Table 4. Twenty of the 35 (57 %) patients who underwent LTS had no complications. Tracheocutaneous fistulas developed in 8 of 35 (23 %) patients; 3 of these 8 patients had previous radiation therapy. Five fistulae were healed with conservative local care. One fistula required a major pectoral muscle flap 23 days after a first attempt with simple closure. In the 2 remaining patients with fistulae, conversion from LTS to TL was performed after 3 and 6 weeks. Both patients had not received radiation therapy prior to LTS. However, both experienced poor wound healing postoperatively, attributed to a catabolic state. Patients with previous radiation therapy or pre-existing tracheotomy, however, were not significantly more prone to develop fistulas or other complications compared to the other subgroups (Fisher exact test $P = 0.317$).

An indwelling voice prosthesis was successfully placed during a second procedure in 15 of 35 patients (43 %). Ten of these 15 patients were able to recover speech function by using the voice prosthesis. Five patients were unable to produce speech through the valve due to progressive neurological deterioration. In two of these five patients, this progressive disease leads to insufficient air volume production. The situation was different for the remaining three patients: although the insufflation test had been positive, they were unfortunately incapable of producing sound at the pharyngo-esophageal level. Attempts have been made to solve the lack of sound production in these 3 patients: in one patient dilations of the cricopharyngeal sphincter were done in clinic, in the other two patients a surgical myotomy of the cricopharyngeal sphincter was performed. Unfortunately, none of these three patients were able to achieve speech through the valve. The two myotomy patients currently successfully use an elektrolarynx. The third patient could not use the elektrolarynx due to apraxia of the upper limbs.

Table 3 Feeding at discharge after LTS

Feeding at discharge	*N*	Total percentage
Oral	17	53.1
Nasogastric	2	6.2
Gastrostomy + liquid diet	2	6.2
Gastrostomy + mixed diet	2	6.2
Gastrostomy + conventional diet	1	3.2
Jejunostomy	2	6.2
Gastrostomy only	6	18.8
Missing data	1	
Laryngectomy after fistulization	2	
Total	35	100

Table 4 Postoperative complications in patients who underwent LTS

Complication	*N*	Total percentage
None	20	57.1
Fistula	8	22.9
Other wound problems	7	20
Total	35	100

Discussion

Intractable aspiration is a serious and life-threatening problem with high morbidity and mortality due to chronic aspiration pneumonia. In this study, we present two additional modifications to the LTS technique described by Lindeman, a reliable life-saving treatment with excellent results to recover oral intake. First, we remove the cricoid cartilage to obtain a stronger closure of the mucoperichondrium of the subglottic area; second we add the possibility of speech restoration by an indwelling tracheaesophageal voice prosthesis, which could be achieved in 38 % (10/35) of our patients.

In the surgical management of intractable aspiration, we prefer LTS over TL. In our opinion, more pharyngeal mucosa can be preserved in LTS, which we consider important in trying to prevent postoperative swallowing difficulties. As the larynx forms the anterior wall of the hypopharyngeal space, TL or even narrow-field total laryngectomy may cause a certain degree of narrowing of this area, while closing the mucosa to reconstruct the neopharynx. This problem is particularly relevant in patients with a previous history of radiation and/or chemotherapy which causes significant fibrosis and swallowing difficulties.

Another strength of our technical modification, is the preservation of more and stronger mucoperichondrium at the level of the cricoid cartilage. As described above, the anterior part of the cricoid cartilage is removed together with the proximal tracheal cartilage during the creation of the proximal tracheal stump. As the subglottic mucoperichondrium is thicker than the tracheal one, this facilitates creation of a more robust pouch closure.

In our study, the occurrence of postoperative complications appeared to be independent of pre-existing conditions. Looking at tracheocutaneous fistulas, our overall complication rate was 23 %, comparable to the reported fistula rate of 18–38 % in LTS, [8–10] and 14–34 % in TL [11].

Contrary to previous reports [12], we could not demonstrate a significant difference in fistulization rates when we compared patients with or without preoperative tracheotomy (Fisher exact test $P = 0.317$). It is known that the presence of a preoperative tracheotomy, with the associated local tissue reaction and fibrosis, critically influences tissue quality in patients suffering from intractable aspiration [13]. Scar tissue increases dissection difficulty, often resulting in a shorter proximal tracheal stump and thereby compromising mucoperichondrial closure without tension [12]. We hypothesize that the creation of a more robust mucoperichondrial pouch might reduce the risk of tracheocutaneous fistulas in LTS patients with a preexisting tracheotomy.

Patients with a history of head and neck tumor treated with radiotherapy and/or chemotherapy experienced complication rates similar to those suffering from a neurological disorder (Chi squared test $P = 0.313$). Interestingly, in patients previously treated with radiation therapy, we observed that aspiration problems typically began approximately 10 years after therapy. This is likely due to tissue immobilization by fibrosis, edema, and xerostomia [14, 15]. Moreover, we believe dysphagia in this last group of patients is further provoked by reduced laryngeal elevation during swallowing combined with the loss of pharyngeal sensitivity, both post radiation therapy effects. LTS proves to be a reliable technique with low morbidity to deal with intractable aspiration in these patients.

Although some authors stress the potential reversibility of LTS as an advantage compared to TL [4, 16, 17], reversibility of the surgical procedure in our patients was of minor importance due to the progressive degenerative nature of their underlying disease. During the decision-making process prior to LTS surgery, the goal of the intervention—the restoration of oral intake—was extensively discussed with patients and their families. Patients and their families were aware that after LTS, possible speech recovery by tracheal restoration would be irreversibly lost and would only be possible with speech valve placement in a permanent tracheostoma, pending postoperative evaluation. All patients accepted the uncertainty of

eligibility for valve placement and the uncertain predictability of successful speech production after valve insertion. All patients preferred restoration of oral intake above speech.

It is clear that aphonia has a negative impact on quality of life, which has already been shown in patients after TL [18]. Reversal of the classic LTS procedure is rarely described, and the complication rate of this procedure seems to be significant [19, 20]. Therefore, we have chosen the use of a voice prosthesis to restore speech. This was done in 15 out of 35 patients, which represents the largest series described in the literature so far. Although we used specific criteria to select patients for a subsequent voice prosthesis placement, accurate prediction of functional speech after prosthesis placement proved difficult. This is shown by the fact that 5 of these 15 patients did not manage to produce speech through a patent speech valve. Two patients were not able to produce speech because they could not produce enough air volume after further neurological deterioration unrelated to the LTS. In three other patients, pharyngo-esophageal vibration was insufficient to produce sound. Ten of these prostheses were functional and restored speech (38 %–10/35 patients).

Detailed analysis of medical records allowed us to observe a clear improvement in the general condition of patients after surgery. Patients pulmonary problems recovered swiftly after LTS, and they could be quickly transferred from the intensive care unit to the regular nursing care units. Patients made fast progress in their rehabilitation program afterward.

Conclusion

This study summarizes our experience with a modified LTS technique to treat intractable aspiration. The complication rate was acceptable, complications could be resolved, except in 2 cases where a laryngectomy needed to be done. Recurrence of aspiration was not observed. In the majority of cases, patients recovered the ability for oral intake which improved their general quality of life. Moreover, one third of our patients recovered speech after voice prosthesis placement, offering them an additional increase in their quality of life.

Author contributions KB and HV developed and performed all surgical procedures. KB, MDL,WH, and HV, gathered and analyzed the patient data, drafted, and finalized the manuscript. AV was in charge of the preoperative assessment and the postoperative rehabilitation of all LTS patients. FD, PD helped to interpret the data and critically revised the manuscript. All authors read and approved of the final version of the manuscript.

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Compliance with Ethical Standards

Conflict of interest The authors declare that they have no financial interests related to the subject of this manuscript.

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