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Voice and swallowing outcomes for adults undergoing reconstructive surgery for laryngotracheal stenosis

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Abstract

Background and purpose:
Adult laryngotracheal stenosis is a rare, multifactorial condition which carries a significant physical and psychosocial burden. Surgical approaches have developed in recent years, however, voice and swallowing function can be affected prior to treatment, in the immediate post-operative phase and as an ongoing consequence of the condition and surgical intervention. In this paper we discuss: 1) the nature of the problem; 2) surgical interventions to address airway disorders; 3) optimal patterns of care to maximise voice and swallowing outcomes.

Recent findings:
Studies in this field are limited and focused on surgical outcomes and airway status with voice and swallowing a secondary consideration. Retrospective studies of swallowing have focused on factors such as the duration of dysphagia symptoms following airway surgery and made comparisons between type of surgery, use of stent and length of swallowing problems. The literature suggests that patients are likely to return to their pre-operative diet. There has been a focus on voice outcomes following cricotracheal resection (CTR) which results in a post-operative decrease in the fundamental frequency. However, study comparisons are limited by the use of inconsistent outcome measures (for both voice and swallowing) which are often not validated, with heterogenous groups and varying surgical techniques.

Summary:
The limited literature suggests that swallowing function is more likely to recover to pre-surgical status than voice function. Further prospective studies incorporating consistent instrumental, clinician and patient-reported outcome measurement are required to understand the nature and extent of dysphagia and dysphonia resulting from this condition and its treatment.
Keywords
Laryngotracheal stenosis – laryngotracheal reconstruction – cricotracheal reconstruction – swallowing – voice

Introduction
Laryngotracheal stenosis (LTS) is defined as a narrowing of the airway at any point between the supraglottis and the carina. LTS can be a result of a variety of conditions with the majority (80%) due to acquired causes. The remaining 20% are due to congenital causes which present at birth and are often associated with other syndromes and conditions. This review will focus on LTS in adults.

The commonest acquired cause is from trauma and this includes intubation-related injuries. In our unit 49.9% of the patients being treated for LTS are due to post-intubation injuries. Endotracheal tubes apply the greatest pressure on the subglottis and posterior glottis. This pressure ultimately results in oedema, ischaemia and pressure necrosis (1). The circumferential wound heals with fibrosis and contracture resulting in stenosis. In idiopathic subglottic stenosis, the patient presents with progressive dyspnoea on exertion and noisy breathing with no history of endotracheal intubation. The condition is often misdiagnosed as asthma that has failed to respond to treatment. It most commonly occurs in women of a white, European background, aged between the ages of 40 and 60 and results from a progressive fibro-inflammatory process (2). Other causes of LTS include auto-immune inflammatory conditions such as granulomatosis with polyangiitis and sarcoidosis. These rare causes often present with other systemic manifestations of the disease which may include multiple levels of airway stenosis or glottic involvement. There are also malignant causes of LTS which can be due to intrinsic narrowing or extrinsic compression. Thyroid, lung or oesophageal cancer can all involve the airway. In thyroid surgery, the recurrent laryngeal nerve (RLN) may be injured impairing vocal fold function. Following chemo-radiotherapy, there can be significant effects on the larynx resulting in LTS thus affecting both voice and swallow.
**Patient assessment**

On presentation with LTS, patients will require a comprehensive history and examination including assessment of vocal fold movement, even where there is an established stenosis elsewhere. The glottis is the narrowest part of the adult airway and any impaired mobility of the vocal folds can cause significant restriction in airflow. Nasoendoscopy is used to examine vocal fold mobility and laryngeal anatomy. Any evidence of pooling or aspiration of saliva should signpost clinicians to request further swallowing investigations. A number of patients referred to a tertiary airway centre will already have a tracheostomy tube in situ which needs to be considered as part of a comprehensive evaluation of swallowing. If there is no obvious cause for the LTS, it is vital to rule out systemic diseases. A full range of investigations include blood tests for vasculitis and autoimmune conditions. Lung function tests are undertaken including peak flow and spirometry to help confirm the diagnosis and assess for other airway pathology. CT imaging of the neck and thorax is also performed to rule out any other causes (e.g. external compression) and assess for other airway pathology. An assessment by the speech and language therapist (SLT) to assess voice and swallowing function is crucial before considering any reconstructive surgery, particularly given the often long-standing nature of the constriction and potentially, the number of surgical procedures patients have undergone.

Prior to any reconstruction, an assessment of the airway is performed under general anaesthesia. This enables an assessment of the airway, the length and extent of stenosis and an anaesthetic evaluation. Suspension laryngoscopy is performed under general anaesthesia usually with jet ventilation. A 4mm Hopkins rod endoscope is passed through the laryngoscope to visualize the larynx, trachea and proximal main bronchi. The extent of the LTS is measured and if required, dilatation of the stenosis is performed. In our unit, we will make radial cuts with a CO₂ laser and prior to dilatation of the stenosed area. If there is an inflammatory component to the stenosis we
will first inject with up to 3ml of depomederone\textsuperscript{TM} (40mg/ml). A biopsy is routinely taken of the stenosis to aid or confirm diagnosis.

**Surgical approaches**

Endoscopic dilatation for LTS has a high rate of recurrence but low morbidity for the patients (3).

The decision to perform a definitive procedure is considered after a multi-disciplinary discussion and matching this to the patient’s expectations. For patients with tracheostomy tubes there is an understandable strong patient drive for decannulation. Candidacy for the procedure must be carefully considered and the patient should be counselled for a variety of potential outcomes, including only partial improvement of the airway and changes to voice and swallow function.

There are several definitive procedures available depending on the site of the LTS, extent of stenosis and function of the vocal folds. For subglottic stenosis (SGS) the main open reconstructive techniques include laryngotracheal reconstruction (LTR) and tracheal or cricotracheal resection (CTR). Extended procedures can be performed for glottic involvement with a posterior cricoid split and placement of a cartilage graft.

An LTR would normally require a laryngofissure to access the stenosis and provide room to insert grafts. During laryngofissure it is important that the vocal folds are not damaged and the endoscope can be used to correctly place the incision (4). When the thyroid laminae are re-approximated it is important that the anterior commissure is realigned to preserve voice. Accurate realignment prevents anterior webbing and misaligned vocal folds which would result in a poor voice outcome.
Endoscopic LTR has developed over recent years and is minimally invasive and potentially less traumatic. The endoscopic technique has been developed for SGS and posterior glottic stenosis. The posterior cricoid is split endoscopically and graft inserted with successful series in children (5). This is more challenging in adults due to the calcification of the cricoid (6). However, with the correct endoscopic equipment it has been proven to be a feasible technique which prevents laryngofissure and help preserve voice.

CTR is a well-established treatment for SGS in children and adults with successful decannulation at more than 90% (7). Pre-operative vocal fold dysfunction is a significant risk for failure of the procedure. White et al (7) found that pre-operative vocal fold dysfunction (unilateral or bilateral palsy/fixation) resulted in 57% (12 out of 21) of patients requiring a second open procedure. If the posterior glottis is involved then an extended CTR is required with a posterior cricoid split and cartilage graft. CTR is also used in a single stage procedure for idiopathic SGS (8). Wang et al (8) reported success of 91% in 263 patients (252 females) with idiopathic SGS treated with a ‘tailored’ CTR. A 10-point questionnaire was sent out to all patients with 180 replies. It included a question on voice and 99 (55%) patients reported voice changes whilst 81 (45%) reported a normal voice. Impaired voice projection was reported in 121 (67.2%) patients.

Tracheal stenosis that does not involve the larynx is less likely to affect swallow or voice. A tracheal resection with primary anastomosis is unlikely to affect swallow unless a supra/infra hyoid release is performed.

**Voice and swallowing outcomes – issues and clinical management**

Patients can experience voice and swallowing difficulties before, during and after these procedures. As a chronic-progressive condition, patients may re-present over time with issues. Surgeons and
SLTs should work with the wider multidisciplinary team to ensure patients are appropriately counselled for the immediate and potential longer term effects of their surgery on voice and swallowing. Preparation needs to take into account both the cause of the stenosis, surgical procedure chosen and patient’s voice and swallowing baseline. The role of SLT is to provide pre-operative counselling and preparation (including voice and swallowing therapy if appropriate), and to provide post-operative rehabilitation as required.

**Voice**

The involvement of the fragile structures of the supraglottis, larynx and subglottis in laryngotracheal stenosis and the complexity of resection and reconstructive surgeries mean that voice and swallowing problems in this patient group are frequently observed (2, 9, 10)

In the immediate post-operative phase, patients will present with voice changes and communication is a particular issue for those whose procedure has required a closed stent with a tracheostomy, thus eliminating glottic airflow and the ability to voice. Patients should be appropriately counselled for this, particularly as they will need to remain in the hospital setting for 2 weeks and this may contribute a sense of social isolation.

Voice outcomes following endoscopic dilatation depend on the level of the stenosis. In a retrospective study by Hatcher et al, the voice outcomes of 44 patients treated with endoscopic dilatation for LTS were analysed (11). The study included 30 patients with subglottic or tracheal stenosis and 14 with posterior glottis stenosis. In the posterior glottis stenosis group, 78.6% of patients had dysphonia. This was measured by the condensed voice questionnaire VHI-10. There was a significant difference (p=0.004) between the groups with the average VHI-10 score for posterior glottis stenosis being 22.4 compared to 10.9 for the subglottic/tracheal stenosis group.
Common voice problems post-surgery include lower pitch, huskiness, difficulties with projection, vocal fatigue and inability to sing (10, 12). Cricotracheal resection, in particular, is understood to alter female voices and can bring their vocal profile into a male range. Consensus of opinion from the limited body of literature is that changes to voice should form part of pre-surgical counselling and in certain cases, vocal demands should be taken into account prior to any surgical decisions, for example whether to complete a major reconstructive procedure or continue with dilatation.

CTR causes disruption of the cricoid ring and partial resection of the cricoid. This can affect voice and swallow due to numerous factors. The attachment of the cricothyroid muscle is disrupted and therefore affecting the lengthening and tensing of the vocal folds. This is important as the majority of patients with idiopathic SGS are female and as a result have a significant lowering of pitch. Smith et al in a study of 14 females who underwent CTR found the fundamental frequency dropping by 21 Herz (Hz) (p=0.03) in the post-operative period (12). However, the post-operative recordings were taken at different intervals (2.4-5.2 months) and the patients had a mixture of CTR techniques with varying degree of cricoid resection. There is also a risk to the recurrent laryngeal nerves with injuries reported in 2% of patients (7) and the cricothyroid joint may be affected.

Bryans et al (10) compared voice outcomes in 11 females following CTR with 12 females post dilatation. The CTR was performed with the anterior 25-33% of the cricoid cartilage and stenotic trachea being removed with complete release of the cricothyroid muscle. Two patients in the CTR group required suprahynoid musculature release. Voice evaluation included the 30-item VHI, perceptual rating of voice with CAPE-V, acoustic measurements and stroboscopy. Voice outcomes were captured at a wide range post treatment. In the CTR group the average time since treatment was 33.2 months (SD, 19.4 months; range 11-70 months) and in the dilatation group the average
was 12.8 weeks (SD, 11.5 weeks; range 3 to 36 weeks). VHI-30 scores were significantly higher in the CTR group 36.9 +/- 30.66 compared to the dilatation group 6.29 +/- 6.79 (p<0.01). This was consistent in all the VHI subscales with CTR scores higher than the dilatation group. The perceptual rating of voice using the CAPE-V scale revealed worse scores in all the subscales in the CTR group compared to the dilatation group. The overall CAPE-V scores were 19.78 +/- 24.67 in the CTR compared to 6.67 +/- 7.77 in the dilatation group (p=0.15). This would correlate with a mild-moderate dysphonia. The CTR group had a significantly lower mean fundamental frequency 188.13Hz compared to the dilatation group 214.05Hz. The stroboscopy assessment revealed one post-operative cord paresis in the CTR group.

Houlton et al (13) assessed 16 patients post CTR with post-operative data on 13 patients. The mean overall CAPE-V score before CTR was 12 and after 61.5 (p=0.28). Only seven patients underwent pre- and post-operative evaluations with the mean fundamental frequency significantly reduced post-operatively (206.5Hz before and 151.1Hz after CTR (p=0.28)).

A retrospective review of 31 patients with SGS was performed and included 22 females and nine males (14). There was a range of treatments for the SGS and 13 patients had additional levels of stenosis. The GRBAS scores significantly worsened for patients with multiple levels of stenosis as well as those with vocal fold motion impairment.

Tanner et al (15) assessed the voice in 11 women with idiopathic SGS pre- and post CTR. The CTR was a revised technique and termed ‘voice-sparing CTR’. The fundamental frequency still decreased post CTR (215Hz SD40Hz, to 201Hz SD65Hz) but was not as great a reduction as some of the previous studies.
Swallowing

Prior to surgery, an instrumental evaluation of swallowing may be required to inform surgical planning. Following surgery, aspiration may be an issue (9). It is also possible for patients with laryngotracheal stenosis to experience swallowing problems before surgery. Impairments include reduced hyolaryngeal excursion leading to reduced airway protection during the swallow and reduced upper esophageal sphincter opening leading to a build-up of residue in the pharynx. During any resection procedure, it may be necessary to release the suprahyoid musculature from the hyoid bone to aid mobility of the larynx for re-anastomosis. This can affect swallow in the post-operative period.

For those patients undergoing a major reconstructive procedure, in the immediate period post-operatively they will be fed via nasogastric tube. Dependent on the results of any swallowing evaluation or surgical complications, the nasogastric feeding is usually necessary for a few days and on occasion longer. Similarly, even if patients are able to swallow safely post-surgery, oedema, structural changes and pain may require them to take a modified diet for a period of time after their operation.

The ideal clinical management of these patients requires detailed assessment, including instrumental assess of their swallowing prior to surgery and following intervention, particularly when complex surgical reconstruction is required. If the patient presents with significant dysphagia this can result in the surgery being cancelled or postponed due to the risk of negatively impacting the swallow. In our experience, this is often the case for patients with airway compromise as a late radiation treatment effect. In these cases, patients may be offered a rehabilitation programme, however, this
needs to be with the caveat that late radiation associated dysphagia may be refractory to swallowing therapy (16).

For those who are judged to have a functional swallow at baseline and progress to surgery we aim to complete instrumental evaluation of their swallowing (Fibreoptic Endoscopic Evaluation of Swallowing (17)) on the day after their surgery, and then when the stent is removed (if applicable). Management of any dysphagia will encompass the usual range of therapeutic manoeuvres, dietary modifications and exercise programs as appropriate. Wherever possible, we avoid keeping people 'nil by mouth' completely in order to minimise the risk of additional disuse-dysfunction. While the majority of patients may progress well following surgery, others will require ongoing swallowing evaluation and rehabilitation.

In our experience, reflux has also been noted to be a significant problem in this caseload with an impact on both voice and swallowing parameters and part of the SLT care package is to liaise closely with surgical colleagues to make sure that the dual approach of medications management and lifestyle changes are implemented.

**Conclusion**

Laryngotraceal stenosis is a rare condition that can persist from childhood as well as having an onset in adulthood. Both voice and/or swallowing can be affected to varying degrees at different stages of their care pathway and this is both patient and procedure dependent. Given the chronic and in certain cases, recurrent presentation of this condition, patients may re-present with difficulties over time. Given the relatively small numbers of patients with this condition, it is best managed in specialist centres with a dedicated MDT. There is little evidence to guide optimal service delivery, however, patients have often had multiple procedures and are experienced and valued partners to
this end. At our own centre, we have engaged with quality improvement specialists and patients to review and co-design how we deliver care (18).

Optimal outcomes are dependent on a strong partnership between surgeons, nursing staff, allied health professionals and patients. As surgical techniques become more advanced, detailed prospective evaluation of voice and swallowing outcomes are required to inform practice and ensure that patients have best possible health-related quality of life.

Conflicts of interest
None declared.

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