J Med Sci 2014;34(1):40-43

DOI: 10.4103/1011-4564.129392

CASE REPORT

A Chronic Traumatic Tracheoesophageal Fistula Functioning as a Respirator and a Phonator Simultaneously

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Acquired benign tracheoesophageal fistula (TEF) is an infrequent complication of prolonged intubation or chest blunt injury. Controversy exists as to whether this should be repaired in a single-stage or in a two-stage procedure. To understand the advantage of one-stage surgery on this complicated injury, and vocalization after reconstruction, we will present a case that had a chronic traumatic TEF, compounded with total laryngotracheal obstruction and an existing unilateral vocal fixation. A 28-year-old female sustained a laryngotracheal injury in a car accident eight years ago and underwent a temporary laryngotracheal stent placement after reconstructive surgery, for one year, in another hospital. Relapsing aspiration pneumonia had developed since then. Video laryngoscopy revealed a mobile right vocal fold, a completely obstructed glottic lumen by granulomatous tissue, and a TEF. This chronic fistula functioned as a respirator without any assistance from the ventilator tube placement, as also a phonator, offering a socially acceptable voice simultaneously, as the larynges were totally obstructed by the scarring granulation tissue. This surrogate glottis enabled survival without a tracheostoma and challenged the justification of any further reconstruction in this patient. Eventually, TEF repair and reconstruction of the laryngotracheal airway were conducted in one stage. Subsequently, the insufficient glottis was corrected by medialized laryngoplasty, to complete the entire reconstruction work.

Key words: Traumatic tracheoesophageal fistula, repair of fistula, laryngotracheal stenosis, laryngotracheal reconstruction, medialized laryngoplasty

INTRODUCTION

The characteristics of tracheoesophageal fistulae (TEF) and their surgical management differ from benign to malignant, congenital to acquired, acute to chronic, and cervical to thoracic fistulae, and from those with or without concurrent tracheal stenosis. Granulomatous infection, foreign bodies, and trauma used to be the most common causes of benign acquired TEF. With the widespread use of cuffed tubes for ventilation, post-intubation fistulae became predominant. By 1973, Thomas had collected 46 TEF related to cuffs (30 fully documented), including seven of his own.[1] Although the use of low-pressure, large-volume cuffs has reduced the incidence, fistulae from this source remain the most common.

Received: April 2, 2013; Revised: July 26, 2013; Accepted: September 30, 2013
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Tracheal injuries, when present, should be treated simultaneously with the closure of the fistula.[2] An attempt to close a post-intubation fistula in a patient who is still on a ventilator is almost certain to fail. Not complicated by a respirator or even a ventilation tube in this case, a chronic posttraumatic fistula simplified the management and enabled a combined repair. However, the presenting case, with a totally occluded glottis, used the TEF as a surrogate glottis to breathe and phonate, with a socially acceptable voice. This condition complicated further management of the airway. Refinement of the surgical approaches to TEF became crucial to allow adequate laryngeal airway reconstruction with minimal negative impact on voice quality. Vocalization after repair of the TEF and reconstruction of the tracheal stenosis was rarely investigated, because the recurrent laryngeal nerve was usually well preserved. In this case, it became even more crucial because of an existing unilateral vocal cord fixation.

CASE REPORT

A 28-year-old female sustained a laryngotracheal injury in a car accident eight years ago and underwent a temporary laryngotracheal stent placement after reconstructive surgery,

for one year, in another hospital. Relapsing aspiration pneumonia had developed since then. Pulmonary function testing revealed a forced expiratory volume in the first second, of 1.77 L (63% of the predicted volume), forced vital capacity of 2.6 L (75% of the predicted capacity), and a forced expiratory volume in the first second/forced vital capacity ratio of 68%, which indicated mild obstructive ventilatory impairment. Meanwhile, the ratio of the maximum expiratory to inspiratory flow, at 50% of the forced vital capacity (FEF50/FIF50), was 1.1 (2.42 / 2.2), which represented the condition of the upper airway obstruction. Video laryngoscopy [Figure 1a] revealed a mobile right vocal fold, extensive granulomatous tissue formation involving the full length of the left vocal cord, obstructing the whole glottic lumen in the inspiratory phase, and an opene cricopharyngeal lumen [Figure 1a], both in the inspiratory and expiratory phases, through which a TEF (2 cm on its esophageal side) was revealed by flexible endoscopy [Figure 1b]. No ventilation tube being required, the patient could breathe totally via the TEF. The voiced sound could be perceived and appeared to arise from the vibration of the margin of the fistula. Voice analysis was performed (jitter = 0.62% and shimmer = 5.32%) and the maximal phonation time was five seconds.

A surgical plan including repair of the fistula, and reconstruction of the laryngotracheal airway and phonatory apparatus (larynx) was determined. A tracheostomy was started initially, followed by gastrostomy and jejunostomy. The transverse separation of the trachea at the subcricoid level was used to explore the aerodigestive lesions and enhance access to the fistula. The TEF, located 5 mm distal to the lower cricoid margin, was repaired with double sutures after trimming the edges of the fistula. An inferiorly based sternohyoid muscle was interpositioned to re-enforce the closure. After the granulation tissue in the glottis was removed and the deformed cricotracheal cartilage was tailored, a T-tube stent, with its upper edge positioned above the vocal cords, was placed *in situ*. The cricotracheal defect was patched with a sternocleidomastoid myoperiosteal flap.

The T-tube, which was regularly replaced monthly, was removed 18 months after surgery and no aspiration developed for six years. Smooth tracheal lumen was revealed in the follow-up endoscopy [Figure 2]. Glottic insufficiency resulting from the former surgical intervention was corrected by silicone medialization laryngoplasty [Figure 3]. The results of voice assessment performed 21 months after initial surgery showed satisfactory voice quality with a jitter of 0.59% and shimmer of 4.21%. The maximal phonation time was improved to 10 seconds. Pulmonary function testing performed 21 months after initial surgery revealed forced expiratory volume in the first second, of 2.1 L (74% of the predicted volume), forced

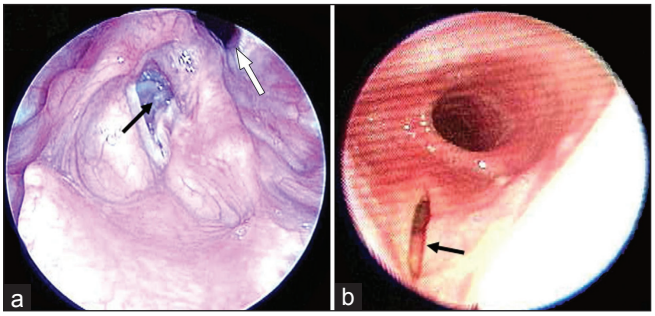

Figure 1. (a) The video laryngoscopic view demonstrated a glottic configuration before reconstruction. Granulation tissues occupied the whole glottic lumen (black arrow). The cricopharyngeal entrance was maintained and opened both in the inspiratory and expiratory phases (white arrow). (b) A flexible endoscopic view in the esophageal lumen before reconstruction showed a tracheoesophageal fistula (black arrow), 2 cm in length, on the esophageal side

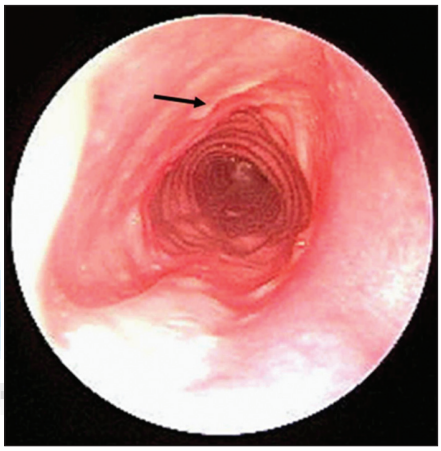

Figure 2. Flexible endoscopic view through the tracheostoma after T-tube removal. The tracheal lumen was smooth, except for the surgical scar (arrow) from the fistula closure, 18 months after surgery

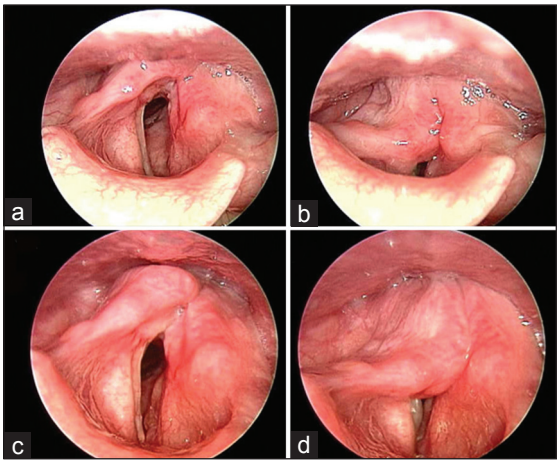

Figure 3. Laryngoscopic views after repair of TEF and glottic stenosis demonstrated an inspiration phase (a) and a phonation phase (b) before medialization laryngoplasty. Anteroposterior approximation of a supraglottic structure was manifested. Inspiration phase (c) and phonation phase (d) after medialization laryngoplasty showed adequate air passage and good vocal fold approximation

vital capacity of 2.76 L (79% of the predicted capacity), and forced expiratory volume in the first second/forced vital capacity ratio of 76%. The FEF50 / FIF50 ratio was improved to 1.03 (3.85 / 3.76).

DISCUSSION

The common causes of acquired TEF include granulomatous infection, foreign body, prolonged endotracheal intubation, and trauma. In fistulae due to granuloma and foreign body, the pathology involves the membranous wall of the trachea and is often limited in extent.[3] Traumatic fistulae may be very extensive and may be accompanied by mediastinal inflammation and infection. A post-intubation fistula results from erosion of the membranous wall of the trachea and the adjacent esophageal wall, and may erode the entire width of the membranous wall and cause a giant fistula.[4,5]

On the basis of the past history reviewed, the case we studied here was recognized as a chronic traumatic TEF, as no long-term cuffed intubation placement was definitely noted. On the contrary, a laryngotracheal stent could gradually irritate an injured and uneven tracheoesophageal wall by its upper end and cause a TEF in one year. In that case, a foreign body–induced fistula would be the first consideration, as the fistula size was small. Otherwise, the TEF could have been caused directly by the original trauma from the car accident and would have become smaller under the protection of the T-tube placement.

If a patient is still on a respirator when the fistula is discovered, the patient must be weaned from it prior to repair. The case we studied here had two advantages in the management:

1. Absence of respirator-dependence and
2. Absence of tracheostomy-dependence.

These connoted a relatively healthy upper and lower airway. As the glottic lumen was totally obstructed by the glottic granulation tissue, respiration was not allowed through this natural conduit. Interestingly, the traumatic TEF not only provided a negative impact as an aspiration conduit, but also a positive one, by providing the service of sufficient air passage for respiration and phonation. On account of this special situation, there were two special considerations concerning the therapeutic plan.

First, utilizing this fistula, the patient still had normal respiration in spite of occasional dyspnea and aspiration, as also a socially acceptable voice, before reconstruction. After closure of this surrogate glottis (fistula), a new glottis would be required for respiratory and phonatory functions. However, the reconstruction of a totally obstructed glottis might not warrant a well-functioning larynx. The possible surgical morbidities included breathy hoarseness and perhaps a resultant aspiration. Furthermore, subsequent restenosis of the airway, after reconstruction, might require an additional tracheostomy. These reconstructions would not be justified once one of these morbidities developed or remained. Therefore, the benefits of the fistula repair had to be weighed against its morbidity and preoperative counseling was important. Second**,** the surgical plan had to determine whether one- or two-stage procedures should be utilized to complete the task. Grillo *et al*.[5] and others recommended correction of the TEF and the trachea in one stage. Others recommended a two-stage procedure for their correction, in case an unsuccessful reconstruction of the larynx complicated the situation.[6,7] The priority sequence of these staged procedures favored the fistula closure first and delayed tracheal reconstruction, until the fistula was well closed. In this study, we favored a one-stage surgery, as an attempt to conduct this in stages demanded two difficult operations, and as the second surgery might be compounded by the surgical reaction to the first intervention.[8] Therefore, a combined repair in one stage was preferred.

There was also a diversity of opinion regarding the optimal surgical management of TEF. Several differing approaches to the management of TEF had been proposed and the optimal management strategy was controversial. Direct suture closure of both the tracheal and esophageal defects, segmental tracheal resection and primary anastomosis with direct esophageal closure, tracheal closure using an esophageal patch, closure of the defects with soft tissue flaps, a combined surgical and endoscopic approach, and a two-stage approach with esophageal diversion, and primary closure of the tracheal defect, had all been advocated. Several series seemed to conclude that the most common operative repair was direct primary suture closure of both the tracheal and esophageal defects, with pedicled soft tissue flap interposition.[9] Whether the fistula could be simply divided and the tracheal and esophageal defects closed primarily or whether segmental tracheal or bronchial resection and reconstruction should be performed was controversial. The published results on the repair of acquired nonmalignant TEF were limited to the experience of only a few institutions. In the series by Mathisen and colleagues, of 41 operations on 38 patients, nine patients (23.7%) were managed by simple division and closure of the fistula, whereas, tracheal resection and reconstruction was performed in 29 patients (76.3%).[7] In our case, the high TEF close to the lower cricoid margin was difficult to access without separation of the anteriorly located airway. Therefore, a preceding cricotracheal resection, before the fistula repair, and direct primary suture closure of both the tracheal and esophageal defect, with pedicled soft tissue flap interposition in one stage, seemed to be optimal for her.

Although there was no optimal duration of stenting following laryngotracheal reconstruction, it was recommended to place the T-tube for three to six months at least. Stern *et al.* reported a duration of stenting ranging from two weeks to 23 months in pediatric patients, with a mean of 7.4 months.[10] Cui *et al.* suggested a prolonged placement (from 12 to 22 months in their series), in order to hold the cartilage and skin grafts in position and to prevent scar contracture.[11] One of their patients failed to decannulate after nine months of stenting, and required a second laryngotracheal reconstruction procedure and stenting for a further 13 months before ultimate decannulation. A recent animal study with a canine showed that six months of T-tube placement was probably sufficient; while three months of placement might not be adequate.[12] In my personal experience, the T-tube should remain in place for a relatively long time, especially for patients with idiopathic laryngotracheal stenosis or severe traumatic cases with extensive framework distortion. Therefore, in this presenting case, prolonged stent placement was performed *in situ* and the outcome was optimal.

Voice reconstruction may encounter two disadvantages in the surgical technique compared with that from merely recurrent laryngeal nerve injury. This glottic insufficiency, resultant from the surgical defect and surrounded by scarring tissue, mimicked that from partial vertical laryngectomy and demanded meticulous elevation of the paraglottic soft tissue from the thyroid lamina. Moreover, the old laryngotracheal injury could cause disruption of a surgical landmark on the thyroid lamina for identification of the vocal cord height. Nevertheless, a successfully medialized vocal cord, coupled with the normally functioning contralateral vocal cord, offered sufficient vocal endurance and eliminated muscle tension dysphonia.[13]

In conclusion, these combined repairs successfully eliminated the aspiration conduit and provided a natural respiratory tract. To complete an intact reconstructive work, the subsequent glottic correction offered this patient a real vibrator and further resulted in a satisfactory voice. This therapeutic algorithm, including the initial combined repair of the aerodigestive tract and a subsequent correction of the glottic insufficiency, seemed optimal in this unusual situation.

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