

## Case Report

# Successful Closure of Pharyngo-cutaneous and Pharyngo-tracheal Fistulas using Removable Hypopharyngeal Stent after Laryngectomy for Laryngeal Carcinoma

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## ABSTRACT

Placement of removable stents to close pharyngo-cutaneous and tracheo-pharyngeal fistulas after laryngectomy has not been reported before. This case presents the feasibility of removable esophageal stent in closing pharyngo-cutaneous and tracheo-pharyngeal fistulas after laryngectomy for laryngeal cancer. Consecutive patients who underwent placement of removable esophageal stent for closing pharyngo-cutaneous and tracheo-pharyngeal fistulas after laryngectomy for laryngeal cancer. Three patients underwent successful stent placement in the hypopharynx. The stents were well tolerated. Patient one had the stent for 14 months, leading to complete healing of the fistula. Removal was successful. The second patient was palliated but died 8 weeks after stent placement. The third patient has successful palliation of his tracheo-esophageal fistula and the stent is being exchanged every 3-4 months to palliate his fistula. Closure of pharyngo-cutaneous and tracheo-esophageal fistulas is feasible with esophageal removable stents. These stents provide alternative options when dealing with these challenging problems.

**Key Words:** Laryngeal cancer, leaks, removable esophageal stents, stricture

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The combination of radiation and chemotherapy is the mainstay for patients with locally advanced laryngeal cancer, since this regimen may improve local tumor control and survival.<sup>[1,2]</sup> Unfortunately, patients with head and neck cancer who receive combined therapy are prone to esophageal and hypopharyngeal stenosis due to the development of fibrosis.<sup>[3,4]</sup> Severe dysphagia has been reported in 45% of patients with head and neck cancer after they undergo combined therapy.<sup>[3]</sup> Strictures in the proximal esophagus occurred in 10-58% of these patients<sup>[5]</sup> and complete esophageal obstruction has been reported in 23%.<sup>[6]</sup> Serial esophageal dilation is usually employed to alleviate dysphagia due to upper or lower fixed strictures. In addition to this common practice, one report described utilization of a

self-expanding stent for palliation of dysphagia secondary to benign hypopharyngeal strictures with varied success.<sup>[7]</sup>

A few reports suggest that esophago-cutaneous fistulas also may occur as a result of such treatments.<sup>[8]</sup> Authors from Greece employed non-removable esophageal stents to palliate two patients with hypopharyngeal cutaneous fistula.<sup>[8]</sup> In general, stents are reserved for refractory esophageal strictures and or co-existing leaks,<sup>[8]</sup> that in the last decade, have been of the non-removable variety. Most commonly, these non-removable stents were also used to palliate patients with proximal cervical esophageal or hypopharyngeal cancer.<sup>[9,10]</sup> With recent innovations in technology, removable esophageal stents are now available.<sup>[11,12]</sup> We report here three cases of pharyngo-cutaneous and/or tracheo-esophageal fistulas that developed after treatment for laryngeal cancer and were treated successfully with removable esophageal stents.

## CASE REPORTS

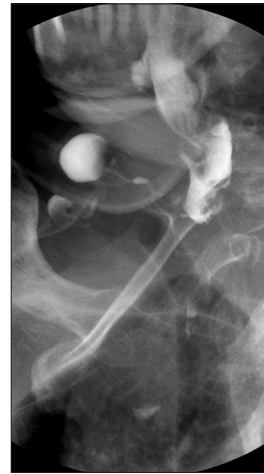
### Case 1

A 70-year-old African-American male underwent total laryngectomy for laryngeal cancer in October 2004. He suffered

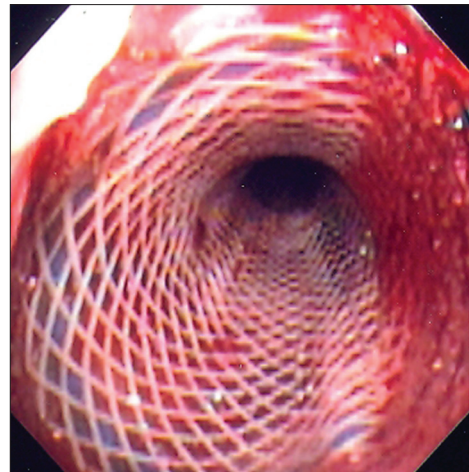
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a recurrence in January 2006 for which he received radiation and chemotherapy. He presented to the ENT clinic in March 2006, with saliva leaking out of the lower cervical region, as well as through the former tracheoesophageal puncture site. He could not swallow anything solid or liquid, because when he tried, it drained directly out of the pharyngo-cutaneous and esophagotracheal fistulas. As a result, with attempts to drink or eat, he suffered choking and coughing spells. Under general anesthesia, the hypopharynx and pharynx were inspected, first with a laryngoscope and subsequently with an 8-mm cervical esophagoscope. There was significant acute radiation-induced toxicity and esophagitis. The esophagoscope was carefully advanced into the pharynx and came to a blind end at a level immediately superior to the tracheotomy site. The distal portion of the pharynx was completely obliterated and the endoscope could not be advanced to the esophagus. The patient underwent transgastric retrograde esophagoscopy with antegrade dilation (TREAD) and esophageal lumen restoration as previously described.<sup>[13,14]</sup> Repeat barium swallow showed a tight stricture of pharyngo-esophageal junction and extravasation from the upper end of the stricture. In addition, separate tracheoesophageal and pharyngo-cutaneous fistulae were demonstrated [Figure 1a]. He subsequently underwent gradual dilation of the stricture at the pharyngo-esophageal junction, requiring eight endoscopy sessions leading to final dilation to 42 fr.

A removable stent placement was employed as a potential method to seal the leak as well as maintain patency of the esophageal lumen. Upper endoscopy with a thin (4.9 mm, Olympus GIF N180) endoscope revealed the neopharynx/pharynx at 13 cm from the incisors. One stricture started at 17 cm from the incisors and another stricture at 20 cm from the incisors. The esophagus distal to the stricture appeared normal. The stomach had a feeding tube that has been placed initially for enteral feeding. A Savary wire was placed under direct vision in the stomach and dilation was carried out with 38 and 42 fr Savary-Gilliard dilators. After adequate marking of the stricture level using radio-opaque arrows placed on the body surface during fluoroscopic localization of the endoscope, a silicone stent was deployed successfully (Polyflex stent 9-cm long and 16 mm in diameter, Boston Scientific, Natick, MA). The stent was deployed 14-23 cm from the incisors [Figure 1b]. Repeat barium swallow showed preferential drainage into the esophagus, with minimal extravasation into the tracheostomy site. Repeat barium swallow at 3 and 7 months showed a patent stent with no leak or migration [Figure 1c]. At this time, an attempted stent removal was unsuccessful due to difficulty with sedation and patient intolerance of the procedure. Because a repeat positron emission tomography (PET) scan suggested local recurrence, the patient underwent further treatment with chemotherapy and radiation. He returned 14 months later; repeat endoscopy was performed under general anesthesia



**Figure 1a:** Right posterior oblique spot radiograph during esophogram shows obliterating stricture at the pharyngo-esophageal junction. Contrast material extravasated into the soft tissues of the anterior right neck and through the tracheostomy stoma

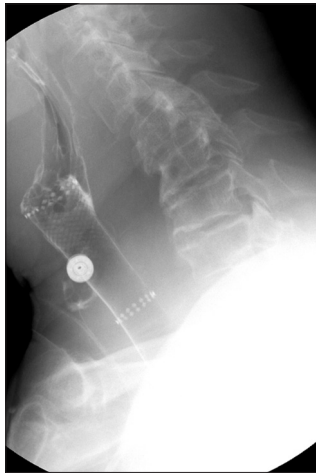


**Figure 1b:** Endoscopic view of the stent at the level of hypopharynx

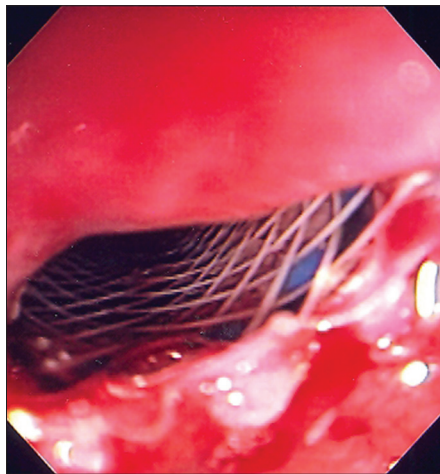
and the stent removed successfully using the double-channel therapeutic endoscope and a rat tooth forceps [Figure 1d]. Both strictures of the hypopharynx and the pharyngo-cutaneous fistula healed without sequelae. Repeat barium swallow confirmed the absence of leakage [Figure 1e], and the patient tolerated oral intake without dysphagia, choking, or leakage through the cervical soft tissues. He is doing well 14 months after closure of the fistula and opening of the near occlusive esophageal stricture. Unfortunately, he had a recurrence of his stricture 3 months after stent removal but no leakage was noted. He now requires frequent dilations of the hypopharyngeal stricture to palliate his dysphagia.

## Case 2

A 57-year-old white man had a history of locally advanced recurrent squamous cell carcinoma (SCC) of the larynx, following total laryngectomy in 2004. He underwent radiation



**Figure 1c:** Repeat esophagogram in right lateral position shows closure of leak and bridging the pharyngo-esophageal stricture. Contrast seen anterior to the stent in this projection is within the neopharynx pouch



**Figure 1d:** Slight embedding of the stent along the posterior wall of the hypopharynx. The stent was easily removable with the rat tooth forceps

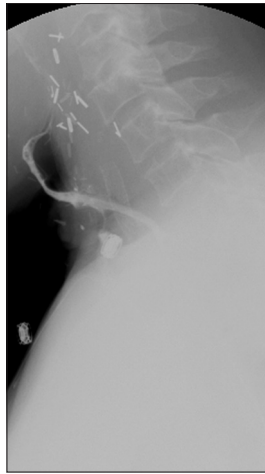


**Figure 1e:** Repeat esophagogram 14 months later (one day after stent removal) confirms closure of pharyngo-cutaneous fistula and patent esophagus

and chemotherapy for parastomal recurrence, in the fall of 2005. Repeat computed tomography (CT) in March 2006 showed a larger recurrent stomal mass, with invasion of the right sterno-cleidomastoid muscle in addition to encasement of the right internal jugular vein and partial encasement of the right common carotid artery. The patient received additional chemotherapy for local control. In the fall of 2006, he presented with incessant cough during swallowing, dysphagia, and fluid passing out through his tracheal stoma after drinking. Barium swallow showed a pharyngo-tracheal fistula [Figure 2a]. Upper endoscopy was performed with a small caliber 4.9-mm endoscope. There was a pharyngo-tracheal fistula seen at 18 cm from the incisors. Under fluoroscopic guidance, a fully covered removable esophageal stent (18x120 mm ALIMAXX-E Alveolus, Inc., Charlotte, NC) was placed. Repeat endoscopy and contrast esophagogram showed proximal stent margin located at the base of the tongue [Figures 2b and 2c] and the fistula sealed completely. No foreign body sensation or chest pain was reported by the patient. Repeat barium swallow showed complete occlusion of the fistula [Figure 2d]. The patient was able to tolerate liquids well and experienced no extravasation through the stoma. Clinical follow-up at 6 weeks suggested that the stent was patent and in place. The patient succumbed to his disease 8 weeks after stent placement.

### Case 3

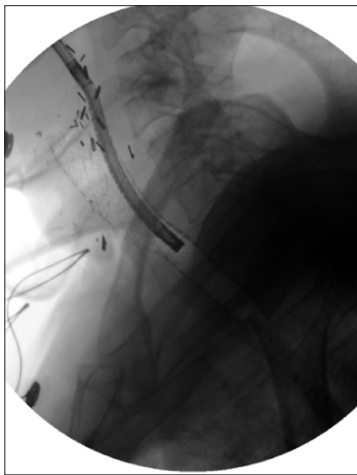
A 63-year-old white man received primary radiation therapy for a T2N0 SCC of the supraglottic larynx in October 2006. He suffered a recurrence and underwent a total laryngectomy and neck dissection in February 2007. He was staged as a recurrent T3N0 SCC of the larynx. He underwent anterior lateral thigh flap reconstruction to close the laryngo-pharyngeal defect. He had partial loss of the flap and development of a pharyngo-cutaneous fistula. He had multiple secondary procedures to cover the carotid artery and local flap procedures to close the fistula. He was gastrostomy tube dependent and aspirated secretions that led to recurrent and frequent hospitalizations. Repeat barium swallow confirmed a pharyngo-tracheal fistula. Since he failed multiple surgeries to primarily close the defect, including placement of a salivary bypass tube, an esophageal removable stent was requested in an attempt to close the fistula. Upper endoscopy was performed with a 4.9-mm thin endoscope, which revealed a skin flap with hair starting at approximately 13 cm from the incisors. At above 18 cm from the incisors, a fistula was seen, containing some granulation tissue. The remaining part of the esophagus was normal distal to that area. Under fluoroscopic guidance, a fully covered removable esophageal stent (18 × 100 mm ALIMAXX-E Alveolus, Inc., Charlotte, NC) was placed. The stent was deployed over Savary wire under fluoroscopy guidance. Four centimeters of the stent was proximal to the leak and the 6 cm were distal. Repeat endoscopy showed the stent in a proper position covering the fistula and extending 14-24 cm



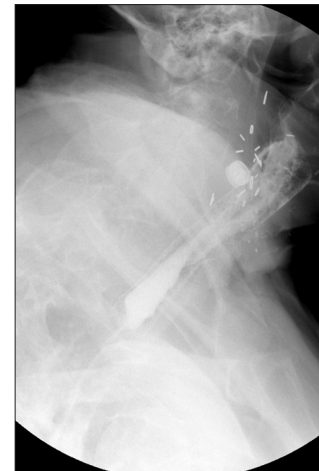
**Figure 2a:** Right lateral spot radiograph during esophagogram shows contrast material extravasated into the anterior cervical soft tissues toward the tracheostomy stoma



**Figure 2b:** Endoscopic view of the stent at the base of the tongue



**Figure 2c:** Spot radiograph obtained while stent placement confirmed with combined endoscopy and fluoroscopy



**Figure 2d:** Repeat esophagogram in the left lateral position shows patency of the stent lumen and closure of leak

from the incisors. Contrast readily passed through stent into the intrathoracic esophagus. There was no filling and thus complete closure of previously identified tracheoesophageal fistula occurred. No other leaks were observed. The patient tolerated liquid challenge very well prior to being discharged home after the same-day procedure. The patient, however, complained of “sore throat” upon cervical flexion, which was treated symptomatically with narcotics. The stent was replaced twice at 3-4 month intervals. While the fistula is reducing in size, it remains patent. The patient reports symptomatic satisfaction with his liquid diet and is able to tolerate the stent well.

## DISCUSSION

Patients undergoing salvage surgery for recurrent laryngeal cancer or combined modality treatment for advanced

laryngeal cancer are prone to significant complications from treatment, including esophageal and hypopharyngeal stenoses, aspiration pneumonia, and fistula formation.<sup>[3,4]</sup> Esophageal prostheses have been utilized when dilation fails to palliate dysphagia. Although palliative stents have been used routinely and are well tolerated when located in the thoracic esophagus, placing endoprotheses at the pharyngo-esophageal level has been met with technical challenges and limitations.<sup>[8]</sup> Moreover, patient tolerance of these stents is lower compared to that for thoracic esophageal stents. The proximity of the cricopharyngeal sphincter has been regarded as a relative contraindication to proximal stent placement because of the potential problems with persistent foreign body sensation, pain, odynophagia, compression of the trachea, or proximal (retrograde) migration of the prosthesis.<sup>[15,16]</sup>

Profili and colleagues have reported successful results with self-expandable metal stents in 10 patients with malignant cervical

and hypopharyngeal strictures.<sup>[10]</sup> Seven of the 10 patients demonstrated rapid improvement in their dysphagia. Two patients had stent position that was very proximal and interfered with swallowing. One stent was shortened under laryngoscopic guidance and the other removed since it could not be shortened. One patient experienced stent twisting after deployment, which required immediate balloon dilation. Another patient had distal misplacement of the stent that required a second partially overlapping co-axial stent. All patients in this study complained of foreign body sensation and mild pain for a week; however, these symptoms resolved without intervention. On long-term follow-up, 50% of the patients developed recurrent dysphagia that required endoscopic interventions. Recurrent dysphagia was due to tumor in-growth in four of nine patients with malignancy, and to granulation tissue in a patient with benign stricture. Clinical success was achieved in 80% of the patients.<sup>[10]</sup>

Over a span of 10 years, Eleftheriadis and colleagues had treated 19 patients with stents placed at the pharyngo-esophageal level.<sup>[8]</sup> Eleven patients had stents for malignant tumors: Seven causing obstruction, while four complicated by an esophagotracheal or esophagocutaneous fistula. Eight patients suffered from an acquired benign tracheo-esophageal fistula due to prolonged intubation. The covered Ultra flex stent (Boston Scientific, Natick, MA) was used in all cases, except two; the Flamingo Wall (not FDA approved, Boston Scientific) stent was used in two patients with esophago-cutaneous fistula following laryngectomy. Prosthesis implantation was technically successful in all patients. Dysphagia score was improved from a median value of 3 (range, 2-4) to a value of 2 (range, 1-3) in patients with esophageal stenoses. Fistula sealing was achieved in all cases, both benign and malignant.

With regard to foreign body sensation, no patients except two with esophago-cutaneous fistula stented by Flamingo tubes, experienced a foreign body sensation. In these two patients, when the neck was flexed, the proximal end of the stent was at the level of the mesopharynx, easily visible through an open mouth. Both stents remained in place for three months until the fistula was totally closed; they were then removed by grasping with retrieval forceps and pulled out without difficulty.<sup>[8]</sup> More recently, Conio and Colleagues have reported successful use of a modified self-expanding stent for benign hypopharyngeal stricture in seven patients.<sup>[7]</sup> After placement of the first stent, dysphagia improved in all patients. Six of seven patients developed stent migration and/or granulomatous tissue in-growth or overgrowth. Additional stents were placed in all patients at a median of 3 months after the initial stent placement. One patient developed an esophago-respiratory fistula caused by a Polyflex stent. Two patients died of causes unrelated to the stent. The remaining five patients remained alive and asymptomatic after a median follow-up of 10 months. The authors concluded that the use

of the modified Niti-S stent helps avoid both the enteral nutrition requirements and the need for periodic bougienage in patients with difficult-to-treat benign hypopharyngeal strictures.<sup>[7]</sup>

Our report confirms that the use of removable stents is a good approach to palliating pharyngo-cutaneous fistula complicating chemoradiation therapy in patients with laryngeal cancer. We are aware of a report that used the Flamingo stent to palliate two laryngeal cancer patients who had experienced similar post-treatment complications.<sup>[8]</sup> Ours is the first report that utilized stents that are designed to be removed in the efforts to palliate pharyngo-cutaneous fistulas after combined modality treatment for laryngeal cancer. Despite very proximal placement in the hypopharynx, our patients tolerated the stents very well and had improvement in dysphagia during closure of their fistulas, without any foreign body sensation or complications related to the stent. While in our first patient, the stent remained for a long period of time, it is advisable to remove these stents within six months to avoid stent embedding and stent-related complications.<sup>[7,12]</sup> In the event that the stent is desired to be in place for a longer period of time, frequent endoscopic or radiographic surveillance studies may be beneficial.

In summary, this report suggests that closure of pharyngo-cutaneous fistula and palliation of dysphagia is feasible with removable stents. Removable stents offer the endoscopist and the patient flexibility in handling these stents if they are not tolerated by the patient, misplaced at the time of insertion or if migration occurs, or if tissue in-growth/complications occur. More data are needed to support our conclusions. Working closely with colleagues in radiology and surgery provides these patients with a multidisciplinary approach to dealing with difficult problems.

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