CASE REPORT

Acquired tracheoesophageal fistula status post laryngeal neoplasm resection

Sarah Luber, DO¹* and Richard Alweis, MD FACP¹,²

¹Department of Internal Medicine, Reading Health System, Reading, PA, USA; ²Department of Internal Medicine, Jefferson Medical College, Philadelphia, PA, USA

A tracheoesophageal fistula (TEF), albeit rare, can be a life-threatening condition that requires prompt identification and treatment. Pulmonary contamination and restriction of proper nutrition are common, unfortunate consequences of untreated TEFs and are often the causes of mortality in this population. In our patient, a history of laryngeal malignancy along with symptoms of chest pain and cough with ingestion of liquids, even without evidence of aspiration pneumonia, appropriately prompted investigation for potential TEF. Initial imaging through barium swallow identified the TEF, and the patient underwent treatment with endoclips by endoscopy with bronchoscopic assistance.

Keywords: TEF Fistula; Endoclips; Laryngeal cancer

*Correspondence to: Sarah Luber, Reading Health System, Department of Internal Medicine, 6th and Spruce Street, Reading, PA 19611, USA, Email: SL-SarahDO.Luber@readinghealth.org

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The patient was a 78-year-old male with a history of laryngeal chondrosarcoma 14 years prior, status post laryngectomy, and tracheal stomoplasty without radiation therapy who presented for evaluation of chronic, progressive, cough, and chest pain. He denied associated dysphagia, but stated that his cough was worse with drinking liquids. He had been using self-prescribed homeopathic options for treatment initially, including swallowing apple cider vinegar. The patient had an esophagogastroduodenoscopy (EGD) 6 years prior that was significant only for a small hiatal hernia. Initial evaluation by barium swallow uncovered a small tract of contrast extending from the anterior esophagus into the trachea at the level of T3, suggestive of tracheoesophageal fistula (TEF). A high-resolution CT scan of the chest was subsequently performed but failed to show definitive gas or contrast-filled tract between the esophagus and trachea. There was also no evidence for metastatic disease of the chest or findings suggestive of acute or chronic aspiration. The patient was evaluated by an otolaryngologist who performed an esophagoscopy, which was again consistent with possible TEF. The tracheostomy stoma evaluation showed pearly mucosa with appropriate secretions. There was no adenopathy or palpable neck masses.

The patient subsequently had an EGD performed, which succeeded in identifying the TEF at approximately 15 cm from the incisors. The procedure was completed with endoclip placement; however, it was complicated by redundant tissue in the region and ultimately unsuccessful. A repeat barium swallow was completed 3 weeks later, showing persistence of the TEF approximately 5.5 cm below the placement of the clip. The patient underwent a second endoscopy with bronchoscopic assistance to better visualize the fistula and two additional endoclips were placed at the fistula tract opening. Despite absence of air bubbles during the procedure, a follow-up barium swallow again showed failure of closure and the patient had persistence of symptoms. He was then referred for potential Ovesco over the scope clip therapy. However, prior to intervention, the patient died of suspected sudden cardiac arrest.

Discussion

TEFs can be categorized into congenital and acquired conditions. The congenital origin is most common, showing predominance in the pediatric population, and the acquired from being significantly rarer and more common among adults (1). Acquired origins can be broken down further into benign or malignant forms. Among the benign conditions, prolonged mechanical ventilation is the most common (47%), followed by trauma (17%), prior laryngectomy (17%), and prior esophagectomy (17%) (2). Within the malignant category of TEF, esophageal tumors accounted for three-fourths of total cases. Secondarily, lung, tracheal, laryngeal, and thyroid tumors combined account for less than 10% of all acquired cases (2).
Malignant acquired TEFs are often associated with poor prognoses as the fistula matures, due to associated aspiration pneumonias (1). In the above case, the patient presented with an acquired benign TEF. It is suspected that the underlying cause was secondary to esophageal puncture with tracheal mucosa biopsy during prior diagnosis of his laryngeal neoplasm. There was no evidence of recurrence of the malignancy on imaging studies or direct visualization. Although there have not been prior reports of vinegar-induced TEFs, caustic ingestions of other chemical agents have been documented (3).

The most common symptoms associated with TEFs include cough during meals and drinking liquids and chest pain, both of which were appreciated in our patient. Patients may also present with dysphagia, increased tracheal secretions, hemoptysis, fever, and recurrent aspiration pneumonias (1, 3).

Imaging studies may demonstrate air-dilated esophagus on chest X-ray, leakage of contrast into trachea on barium swallow, or gas/contrast filled tract between the esophagus and trachea on high-resolution CT (HRCT) of the chest (1, 3). The TEF in our patient was identified only on barium swallow (Fig. 1), not on HRCT (Fig. 2) or chest X-ray. Further identification of TEF location may be completed with endoscopy. In cases of smaller fistulas or redundant tissue, as appreciated in our patient, combined bronchoscopy and endoscopy with utilization of methylene blue dye can identify the precise location of the TEF for intervention (1, 3).

There are several surgical treatment options available, including direct suturing of the esophageal defect, esophageal bypass, esophageal exclusion, and fistula resection. These are complicated and tedious procedures with a significant rate of fistula recurrence (2, 4). Additionally, fibrin sealants or glues have been utilized in small-opening fistulas (1). In cases of advanced malignancies, palliative measures are usually used, including esophageal stenting and chemotherapy/radiation to address dysphagia and risk for pulmonary infections. However, with procedures such as stenting, there is a high incidence of esophageal perforation due to frequent need of tumor dilation (1, 3). Although surgery remains the mainstay of treatment, endoclips have been popularized as a non-surgical alternative for not only perforations and fistulas but also gastrointestinal (GI) bleeding and securing tubes or stents to GI walls (5, 6). However, due to the rarity of disease, there have been no randomized control trials comparing surgical techniques to endoclip use (5, 6). A pooled analysis, including 17 case reports of esophageal perforations in various stages, ranging from acute to chronic, demonstrated a mean perforation closure at 18 days post clipping (7). Acute perforations had up to 98% closure rates as reported in one study (7). Whereas increased chronicity of perforations lead to higher failures of closure resulting in postsurgical ablation along with endoclipping of these fistulas for successful closure. It is suspected that due to the chronicity of this patient’s perforation/fistula, the smaller endoclips were insufficient for complete healing. Alternative options after unsuccessful placement of the initial endoclips were discussed with the patient, including abrasion of the fistula followed by tight clipping with a larger device and surgical suture closure through endoscopy. Unfortunately, the patient died of unrelated causes prior to completion of treatment.

**Conclusion**

A TEF should be considered a potentially life-threatening condition due to potentially severe pulmonary and nutrition consequences, therefore early identification and treatment is essential. A history of malignancy of the esophagus/
surrounding structures or prolonged intubation along with symptoms of chest pain and cough with meals or liquids, with or without evidence of aspiration pneumonias, should prompt investigation of potential TEF. Initial imaging should include barium swallow with further fistula location identification by endoscopy with or without bronchoscopy. Treatment options may include surgical repair or resection, endoclip application, esophageal stenting, or fibrin sealant injections, depending on fistula size and underlying etiology.

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