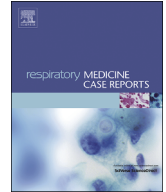


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Case Report

Novel endoscopic approach for treatment of malignant tracheoesophageal fistula (TEF): A case report

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ABSTRACT

Cardiac septal occluder (CSO) can be used endoscopically in treatment of tracheoesophageal fistula (TEF). We present a case of metastatic cervical cancer with direct tracheal invasion resulting in malignant TEF treated previously with radiotherapy and tracheal stenting twice. This TEF was closed by CSO - patent foramen ovale (PFO) occluder as palliative management to prevent recurrent aspiration pneumonia, decrease morbidity and improve quality of life.

1. Introduction

Malignant tracheoesophageal fistula (TEF) can be caused by primary malignancy or metastatic disease. In patients with advanced malignancy causing TEF, surgical treatment is mostly deferred, and endoscopic stenting is the treatment of choice [1]. With advent of new modalities for treatment of advanced malignancies, survival in such patients has increased [2]. As a result, TEF despite endoscopic tracheal stenting might persist and can cause significant morbidity in form of recurrent aspiration pneumonias. Off label use of patent foramen ovale (PFO) occluder [3] can be done in such patients as a palliative option for management of TEF to improve quality of life. Our patient is 60-year-old woman with metastatic cervical squamous cell carcinoma causing TEF. She was treated with tracheal stenting twice for TEF but continued to develop recurrent aspiration pneumonias due to persistent TEF. PFO occluder was used for palliative management of TEF to improve quality of her life and prevent aspiration pneumonias.

2. Case report

A 60-year-old woman with a history of metastatic cervical squamous cell cancer with known prior invasion of the trachea presented with complaint of having progressively worsening cough and shortness of breath. She was treated four times in the past six months for recurrent pneumonia. After her initial diagnosis of cervical cancer almost 2 years back, she was treated with chemotherapy, radiation therapy and immunotherapy with poor response and developed right upper lobe pulmonary metastasis with direct tracheal invasion (Fig. 1A.) and extensive hilar lymphadenopathy. This tracheal stenosis was managed with endoluminal stenting (with mid tracheal biopsy indicating squamous cell carcinoma as shown in Fig. 1B, C and D) on two occasions previously to maintain airway patency along with repeat radiotherapy for right upper lobe pulmonary mass. Computed tomography (CT) of chest during this admission showed patent tracheal stent with endoluminal aspirated debris along with interval decrease in right upper lobe pulmonary mass and lymphadenopathy as shown in Fig. 2. A.

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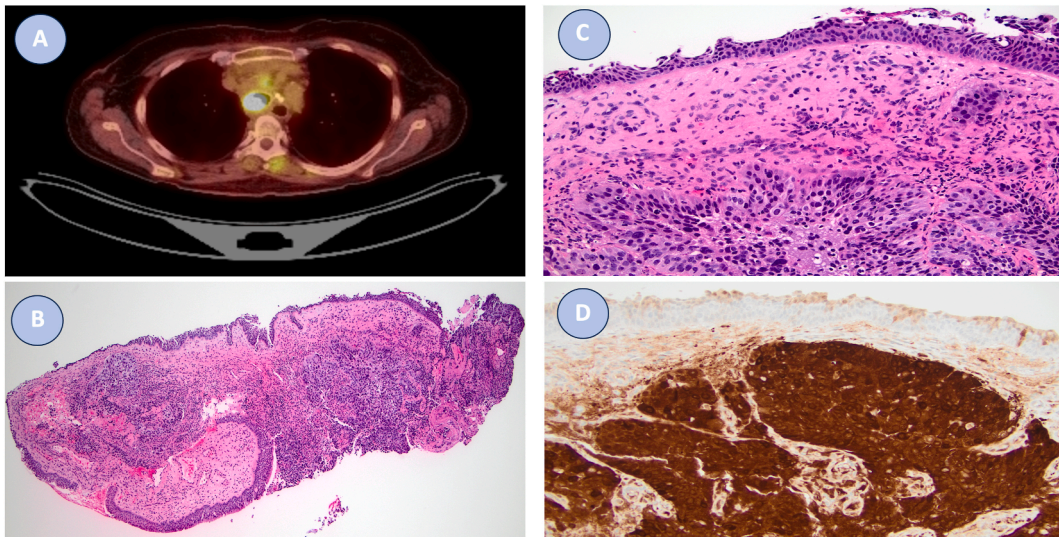


Fig. 1. A) PET/CT scan showing direct invasion of trachea by FDG avid metastatic deposit. B) hematoxylin and eosin staining of tracheal biopsy showing squamous cell carcinoma (SCC) at 20 times magnification C) at 200 times magnification D) p16 staining showing (SCC).

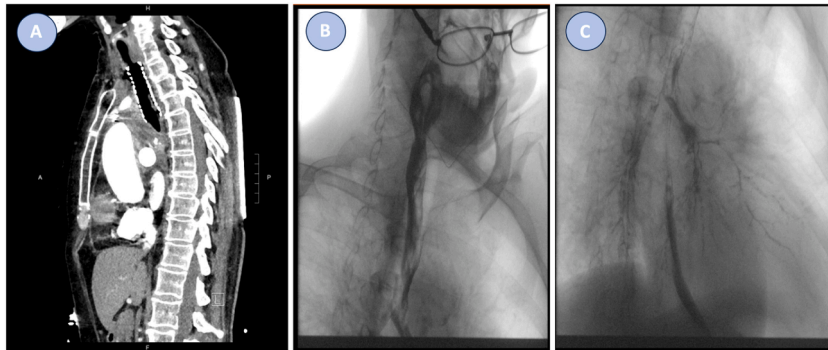


Fig. 2. A) CT chest sagittal view showing food particles in tracheal stent. B) & C) Barium swallow showing aspiration of contrast from esophagus into trachea indicating tracheoesophageal fistula (TEF).

Barium swallow was performed which was suggestive of tracheoesophageal fistula with aspiration (Fig. 2B and C). Percutaneous endoscopic gastrostomy (PEG) tube placement was done to provide nutrition to patient by alternative means. Considering extent of her underlying disease, patient was deemed not a surgical candidate for treatment of malignant TEF. However, patient expressed continued desire to eat and after collective decision, it was opted to place a patent foramen ovale (PFO) occluding device for treatment of TEF.

In collaboration of interventional pulmonology, interventional cardiology and advanced gastroenterology teams, we proceeded with general anesthesia with laryngeal mask ventilation. Bronchoscopy demonstrated tumor in the mid trachea with a TEF approximately 2.5 cm from the cricoid. A guidewire was passed from trachea through the fistula into esophagus and the position of guidewire in esophagus was determined with direct visualization with esophagoscopy. Interventional cardiologist deployed a PFO occluder over the guidewire through bronchoscope but positioning was difficult (Fig. 3 A-F).

The guidewire was then passed through the esophagus into the trachea through TEF using esophagoscope. First part of PFO occluder was deployed over the guidewire into the trachea, which provided occlusion of tracheal surface of TEF. This was followed by deployment of second part of closure device approximated on esophageal side of the TEF (Fig. 4A–D). Simultaneous esophageal and tracheal balloon dilations were done to compress PFO occluder in position. The PFO occluder was noted in good position with patency maintained in both tracheal and esophageal lumen at end of procedure.

The patient exhibited a stable immediate post-procedural course, promptly returning to her baseline condition. She remained hemodynamically stable for three days following the procedure. She continued to be on her baseline oxygen requirement of 3 L/min and tolerated tube feeds without any complications. Our initial plan was to perform a repeat barium swallow study before resuming oral nutrition. However, on the third day post-procedure, she experienced worsening oxygen requirements. Chest X ray showed worsening patchy bilateral opacities, more pronounced than those observed prior to the procedure. Despite treatment with broad spectrum antibiotics, her condition continued to decline, which we suspect was due to the progression of aspiration pneumonia or pneumonitis that predated the procedure. After careful consideration, the patient and her family chose hospice care, and she passed away on the

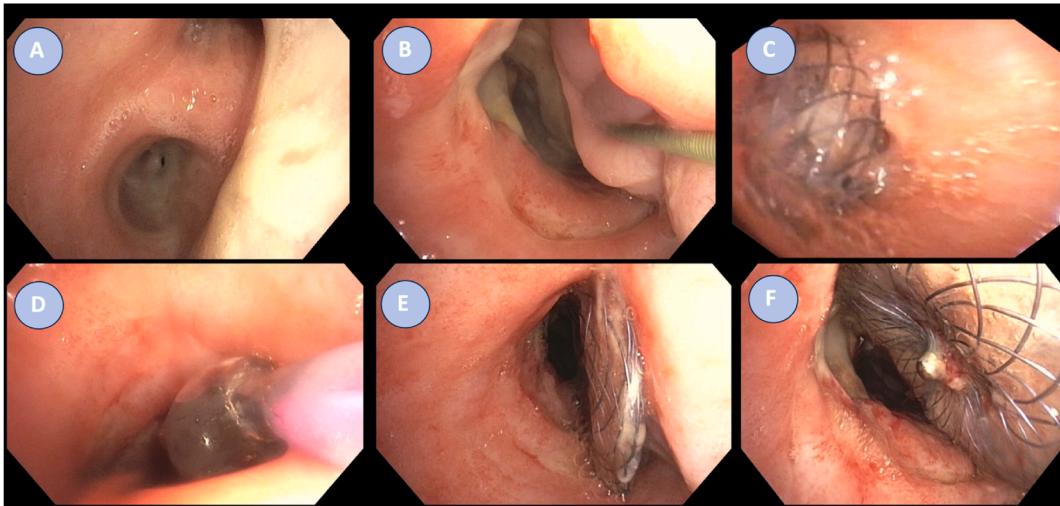


Fig. 3. Bronchoscopic images showing A) tracheoesophageal fistula(TEF) B) guidewire in TEF C) failed attempt with PFO occluder from tracheal side D) Dilatation of PFO occluder with second attempt from esophageal side E) Opening of second part of PFO occluder after placement from esophageal side F) PFO occluder closing TEF.

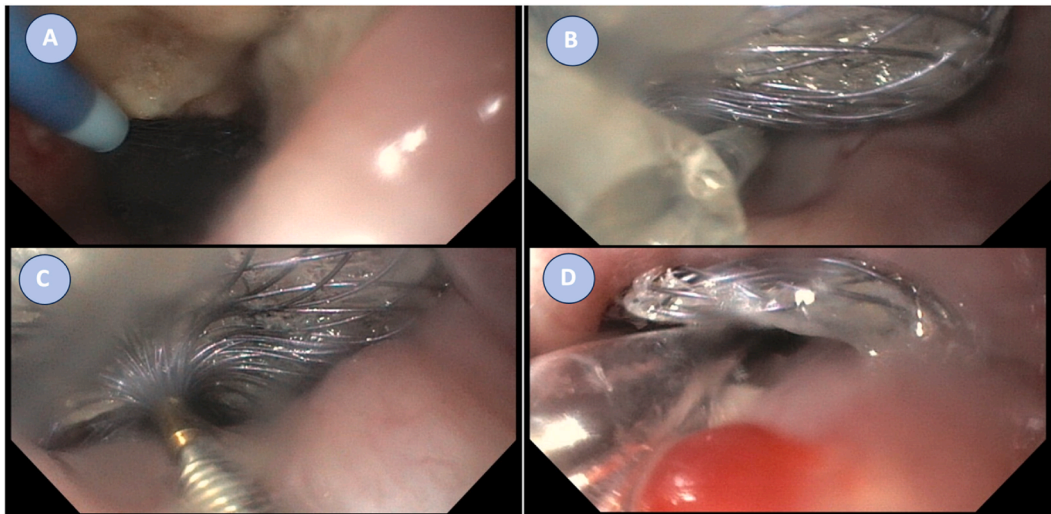


Fig. 4. Gastroscopic images showing A) Deployment of PFO occluder near tracheoesophageal fistula(TEF) B) First balloon dilation near TEF C) PFO occluder in TEF D) Sequential dilation near TEF.

fifth day post-procedure. While the patient could not be transitioned to oral nutrition due to her decline from pneumonia, we regard the procedure as a technical success. The tracheoesophageal fistula was successfully repaired using a minimally invasive approach, without any procedure-related complications.

3. Discussion

Tracheoesophageal fistula is a pathological connection in one or more places between trachea and esophagus [4]. It has two types: congenital and acquired. Acquired TEF is mostly seen in adults and can be benign and malignant [1]. Benign TEF are mostly caused by trauma, granulomatous inflammation, foreign bodies, post-intubation injuries or after surgical procedures [5]. Malignant TEF occurs due to primary cancers of trachea, lungs, larynx, esophagus, thyroid or secondary metastatic disease [6] (similar to our patient).

Management of tracheoesophageal fistula includes surgical approach versus endoscopic management. For benign TEF with size of fistula greater than 5 cm, surgical intervention is preferred [1]. However, patients who are poor surgical candidates, endoscopic stenting can be offered. Type of stenting mainly depends on location of TEF. For proximal TEF, dual tracheal and esophageal stenting can be offered to patient while for distal TEF, only esophageal stenting is preferred [1]. For malignant TEF, endoscopic stenting is treatment of choice with same parameters for dual and esophageal stenting as for benign TEF [1]. Complications of endoscopic stenting for TEF includes retrosternal chest pain, hemorrhage, stent migration, perforation, airway compromise and formation of new TEF [7].

Our patient had malignant TEF in setting of metastatic cervical malignancy with direct tracheal involvement treated with endoluminal stenting twice previously. This malignant TEF was approximately 2.5 cm from cricoid. As she continued to express desire to eat and was having recurrent aspiration pneumonias, she was offered closure of TEF by PFO occluder. Long term complication of TEF closure by PFO occluder could not be assessed as our patient passed away unfortunately.

Off-label use of PFO occluders for treatment of fistulae have been used documented previously [8–10]. PFO occluders like tracheal or esophageal stents have been associated with complications including migration, airway occlusion and worsening of TEF [11]. However, in those subsets of patients in which tracheal stents fail, are contraindicated or cause complications, PFO occluders might offer an alternative option. More clinical evidence is required to ensure safe use of PFO occluders, a palliative option for management of malignant TEF.

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CRediT authorship contribution statement

Haris Zia: Writing – review & editing, Writing – original draft, Visualization, Conceptualization. **Matthew D. McCoy:** Writing – review & editing. **Jamie Sturgill:** Writing – review & editing, Writing – original draft, Conceptualization. **Walid Hadid:** Writing – review & editing, Writing – original draft. **Ashish P. Maskey:** Writing – review & editing, Writing – original draft.

Declaration of competing interest

The author(s) declare no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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