

ACQUIRED BENIGN TRACHEOESOPHAGEAL FISTULA. A CASE REPORT AND REVIEW OF THE LITERATURE.

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Summary

A case of a 35 year old man with an acquired benign tracheoesophageal fistula (TEF) is reported. He had an impacted denture in the mid oesophagus for 15 years with intermittent odynophagia and dysphagia. Dysphagia became more persistent and associated with cough after taking solid and liquid food. Gastrografin swallow studies showed a tracheoesophageal fistula. He had a right thoracotomy and removal of the denture. There was a defect and stricture of the oesophagus and a defect in the trachea at the site of impaction. The trachea was repaired. Primary repair of the oesophagus was not possible. Oesophagectomy, cervical oesophagostomy and feeding gastrostomy were carried out. Colon replacement of the oesophagus was effected six weeks after the oesophagectomy.

Key words: Tracheoesophageal fistula- oesophagus- trachea -foreign body-feeding gastrostomy- oesophagectomy -cervical oesophagostomy

Introduction

Fistulas between the airway and the oesophagus can be congenital or acquired. Congenital TEF is a fairly common disorder that has to be considered in a neonate who develops feeding difficulties and respiratory distress in the first few days of life. Currently acquired benign TEFs are mainly iatrogenic occurring after tracheal intubation, surgical trauma and blunt injuries^{1,2,3}. It is also known to occur in approximately 0.5% of patients undergoing tracheostomy^{4,5}. Burrington and Raffensperger reported several acute TEFs resulting from caustic ingestion in children⁶. Santiago et al also reported a case of TEF in a 16 year old girl after she had attempted suicide by ingesting a caustic substance⁷. Traumatic TEFs occur secondary to either blunt trauma or open avulsion injury to the neck and thorax. In blunt traumatic injuries, the TEF is intrathoracic and is usually located at the carina level¹⁰. TEF may be deliberately constructed with surgery to aid talking in a patient who had laryngectomy⁸. Infective causes of TEF have also been reported and these include tuberculosis, syphilis, and histoplasmosis infecting lymph nodes⁹. Tracheoesophageal fistula

from retained foreign body is very rare. Most patients with a chronically retained foreign body are nonverbal¹⁰.

We report a case of an accidentally swallowed denture which was retained for 15 years with the development of tracheoesophageal fistula, the management and review of the relevant literature.

Case report

A 34 year old university student was referred to the Cardiothoracic Centre with a year history of and a two year history of dysphagia. The cough was productive of offensive sputum and became worse two months prior to admission. Coughing was particularly linked with ingestion of liquid food. He swallowed his denture 15 years ago and after a couple of unsuccessful attempts to remove it by endoscopy, he was told to watch his stool for the denture. Two years before presentation he noticed progressive dysphagia and odynophagia. At the time of presentation he could only swallow liquid food. He admitted to losing significant weight over the six months prior to presentation. He was compelled to defer his course in the university due to his medical condition.

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The results of physical examination and routine haematological investigations were unremarkable. Sputum culture and sensitivity isolated alpha hemolytic streptococci sensitive to erythromycin and flucloxacillin. Chest X-ray did not show any lesion in the lungs. Gastrografin swallow showed oesophageal structure and prestenotic dilatation with fistulous connection at T3, and contrast medium showing the extravasating into both main-stem bronchi (Fig.1 and 2).



Fig. 1. Gastrografin swallow showing the

Fig. 2. Gastrografin swallow showing contrast outlining the trachea, both bronchi, prestenotic dilatation of the oesophagus and the trachea.

Fibreoptic oesophagoscopy showed a tight stricture at 24 cm. The scope could not go through it.

The patient had surgery after one week of antibiotic therapy. He had a right thoracotomy and exploration of the impacted denture. A mass of granulation tissue was found close to the level of the carina. The impacted denture (Fig. 3) was removed from this mass, leaving a stricture and a large defect in the oesophagus and a smaller defect in the trachea close to the carina. The defect in the trachea was closed primarily with a nonabsorbable suture. The structured oesophagus could not be repaired. Oesophagectomy, cervical oesophagostomy and feeding gastrostomy were carried out. Post operative recovery was uneventful. After eight weeks, restoration of the gastrointestinal continuity was achieved by retrosternal colon bypass. He is currently living a normal life and will be returning to complete his course in the university.

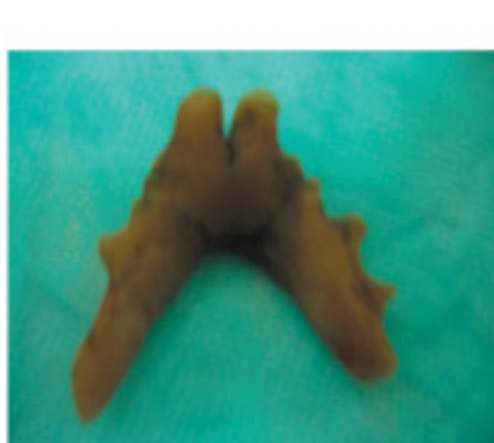


Fig. 3. Impacted denture swallowed for 15 years.

Discussion

Tracheoesophageal fistula from a retained foreign body in an adult is rare. The case presented is unusual and unique, in that the patient was sane and yet kept the swallowed denture for 15 years. The history is pathetic since he was not even told how long he had to watch out for the denture in his stool. After several years of watchful waiting he developed a tracheoesophageal fistula. The oesophagus was destroyed and had to be reconstructed with colon. Acquired TEF has often been reported with 'button battery' ingestion when direct impaction in the oesophagus allows direct corrosion, pressure necrosis, perforation and fistula formation¹¹. TEFs caused by endotracheal tube intubation depend on several factors including prolonged intubation, an irritating or abrasive tube and pressure exerted by the cuff. Pressures exceeding 30 mmHg can significantly reduce mucosal capillary circulation and result in tracheal stenosis⁴. Keeping endotracheal cuff pressures less than 25 mmHg is important in the prevention of TEF.

Foreign bodies in the oesophagus can be classified into four categories¹². The first is the purposeful ingestion of a foreign body either in a child or psychiatric patient. The second is accidental ingestion of a foreign body usually in food. The third is the impaction of a foreign body in the presence of an obstructive pathology in the oesophagus. And the fourth, the so-called steak bouse syndrome, in which a bolus, usually meat, is obstructed at the distal end of the oesophagus. The most common locations of foreign body lodgement are at three areas of normal physiologic oesophageal narrowing. The uppermost narrowing is located at the entrance into the oesophagus and is caused by the cricopharyngeal muscle. It is the narrowest point. The middle narrowing is the result of an indentation of the anterior and left lateral oesophageal wall caused by the crossing of the left main stem bronchus and aortic arch. The lowermost narrowing of the oesophagus is at the hiatus of the diaphragm and is caused by the gastrooesophageal sphincter mechanism.

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The common initial signs of TEF are a sudden cough associated with ingestion of fluids and solids (Ono's sign)¹³. The patient may also present with sputum mixed with food, recurrent pulmonary infections, weight loss and profound weakness⁷. Some reported cases also described persistent history of cough, noisy breathing, stridor and a recent history of vomiting after feeds¹⁴. The case reported had a long history of intermittent dysphagia, cough productive of offensive sputum associated with liquid and solid ingestion, dysphagia, odynophagia and weight loss. The weight loss was due to the dysphagia which became progressive a year before presentation. Patients intubated in the intensive care unit on intermittent positive ventilation for prolonged periods who develop TEF may present differently. Fitzpatrick et al described detection of an air leak through the nasogastric tube via an underwater seal in the expiratory phase¹⁵. Marked gaseous abdominal distension have been described¹⁶. Rampaul et al described a clinical sign in these patients with TEF called the «breathing bag» sign¹⁵. In patients diagnosed with TEF, a chest X-ray can be useful in revealing aspiration pneumonia⁹. The chest X-ray of the case presented did not show any radiological sign of infection. If aspiration pneumonia is diagnosed, it has to be treated before any major surgery is carried out. TEF is usually demonstrated by radiologic contrast study or by direct endoscopic visualization^{17,18,14}. The imaging method of choice for the evaluation of a TEF is an oral contrast study¹⁹. Meglumine diatrizoate (Gastrografin) is used in upper gastrointestinal examinations when perforation is suspected. The positive sign for TEF is the passage of the contrast media from the oesophagus to the trachea with or without delineation of the fistulous tract⁷. The case presented had gastrografin swallow with the passage of the contrast into the trachea and outlining both mainstem bronchi. Barium sulphate can also be used in similar studies⁹. Direct visualization by flexible oesophagoscopy and bronchoscopy is useful depending on the expertise and the experience of the individual. Bronchoscopy helps to locate the exact site of the fistula¹⁶. The flexible oesophagoscopy carried out in the case presented could not go beyond the tight stricture at 24cm. It was not useful in the diagnoses of the TEF in our case. For unstable patients in the intensive care unit, these methods of diagnoses may be cumbersome. Instillation of methylene blue through the nasogastric tube and its appearance in the endotracheal tube is one way of demonstrating TEF at the bed side^{4,14}. Kelvin et al used the analysis of gases from the stomach of patients receiving positive pressure ventilation to diagnose TEF¹⁴. Analysis of gases from the stomach, ventilator and room air suggested that the gastric gases came from the ventilator of the patient with TEF. Computerised Tomography scan has proven to be valuable in the evaluation of the trachea and oesophagus when a TEF is suspected⁷. Thin section helical CT images are used to evaluate the tracheal and oesophageal walls. If there is an anomalous communication between these two structures, the wall defect can be identified.

For TEF, the difficulty in the management results from the need to manage both the consequences of the oesophageal communication and those of the illness responsible for the fistula¹. For patients on mechanical ventilation who developed TEF, a conservative approach is used until patient is weaned from the ventilator⁴. The cuffed endotracheal tube is placed distal to the fistula site in order to prevent reflux of gastric contents into the lungs. A tracheostomy tube is placed distally to the TEF if possible. The head of the bed is elevated and oral secretions frequently suctioned. A gastrostomy tube is placed to minimize gastrooesophageal reflux, and feeding jejunostomy tube is placed to minimize the gastrooesophageal reflux, and for nutritional purposes. If the patient is critically ill and definitive surgery cannot be undertaken, oesophageal diversion, creation of a high salivary fistula and feeding gastrostomy is carried out⁴. In a case report by Angel Luis et al, a conservative approach with temporary airway stenting was successful in stabilizing a patient with a large postintubation tracheal necrosis and weaning her from the ventilator. The fistula healed spontaneously and the stent was removed without complication²⁰.

There are various surgical options available in the initial management of TEF are almost always supportive and followed by definitive surgical correction¹⁷. Primary closure of the oesophageal and tracheal defect with or without interposition of muscle flaps has been reported^{21,17,18,20}. Interposition of radial fascial free flaps has been used to treat recurrent TEFs²¹. Resection of the trachea and re-anastomosis with oesophageal resection and reanastomosis or oesophageal diversion is another option⁴. The use of cervical and thoracic segments of the oesophagus as a patch to close the posterior tracheal wall has been described¹⁹. There have been reported cases of the use of skin grafts, pericardial patching and oesophageal patching of the trachea¹⁶. The case presented had an initial primary repair of the trachea, oesophagectomy, cervical oesophagostomy and feeding gastrostomy. The mass of granulation tissue at the site of impaction, the extent of the oesophageal stricture, and the defect created after removal of the denture made it impossible for primary repair of the oesophagus. Restoration of the gastrointestinal continuity using retrosternal colon bypass was accomplished two months after the first operation.

Stents have been used in combination with antituberculous treatment in patients who develop TEF following pulmonary tuberculosis⁹.

The major postoperative complications are tracheal stenosis and recurrent fistula. Tracheal stenosis occurs in patients who have repair injury to the posterior tracheal wall. Surgical repair of tracheal stenosis may be performed at a later date. Recurrent fistulas may develop in patients who require postoperative intubation⁴.

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