

Benign Broncho-oesophageal Fistula in an Adult

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Abstract

Presentation

We describe the case of an elderly lady who was diagnosed with a BOF after presenting in type 1 respiratory failure, with no risk factors or malignancy. A history of recurrent pneumonia with multi-drug resistant pathogens led to radiological investigation into an abnormal connection between the GI and respiratory tracts.

Diagnosis

A fluoroscopic barium swallow study is the radiological investigation of choice. Our case illustrates that CT may also be useful in identifying and characterizing a BOF but requires a high index of suspicion as they are usually subtle.

Treatment

A non-surgical approach was employed due to the frailty of this patient. An excellent outcome was achieved, with full resolution of symptoms and prevention of further complications.

Discussion

There are few cases described of benign broncho-oesophageal Fistulas (BOF) in adults. Greater than fifty per cent of cases are caused by oesophageal or lung malignancies. The majority of benign cases are iatrogenic.

Introduction

An 88-year-old lady presented to the Emergency Department with worsening dyspnoea and a non-productive cough.

Her medical history includes recurrent episodes of pneumonia requiring hospital admission, with multi-drug resistant and opportunistic pathogens on a background of bronchiectasis.

Case Report

The patient had a history of a previous right middle lobectomy for reduction of frequent acute exacerbations of bronchiectasis due to localized disease. Her surgical history also includes right lumpectomy and radiotherapy for triple negative breast cancer in 2013 and an oesophageal diverticulum.

Chest radiograph performed on admission demonstrated new consolidation in the right and left lower zones correlating with clinical findings.

A CT Thorax performed during a recent prior admission was reviewed, at the multidisciplinary respiratory conference, and demonstrated extensive airway wall thickening, ground-glass and lobular consolidation in the right lung particularly the right lower lobe with cylindrical bronchiectasis. (Figure 1) A tract was identified communicating between the right mainstem bronchus and anterior aspect of the oesophagus, compatible with a broncho-oesophageal fistula.

A barium swallow was then performed, confirming the presence of a fistula between the mid-thoracic oesophagus and the proximal aspect of the right main bronchus approximately 1 cm distal to the carina, with aspiration of barium into bronchiectatic airways in the right lung base.

Definitive treatment of the BOF was achieved using OGD. The oesophageal portion of the BOF was identified and successfully clipped using an over-the-scope device.

Figure 1

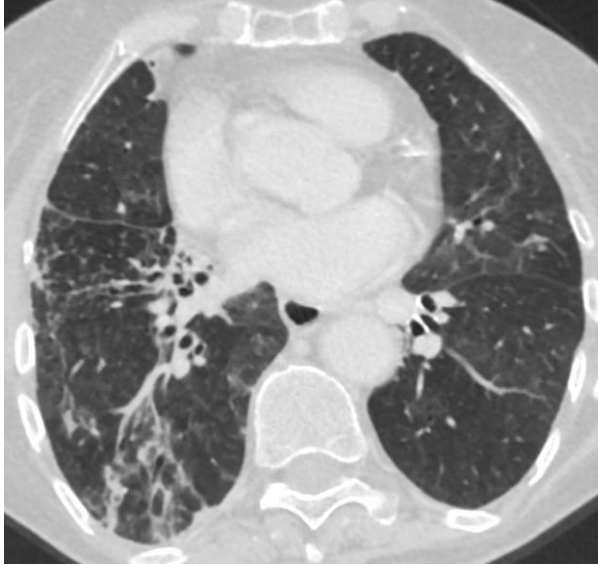


Figure 1a

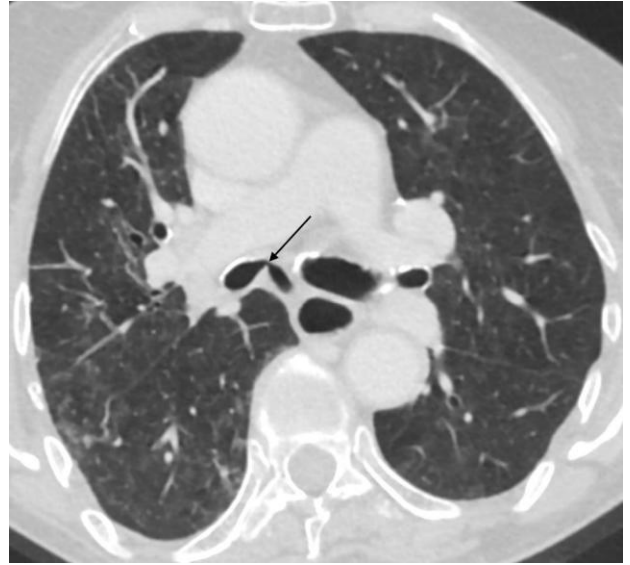


Figure 1b

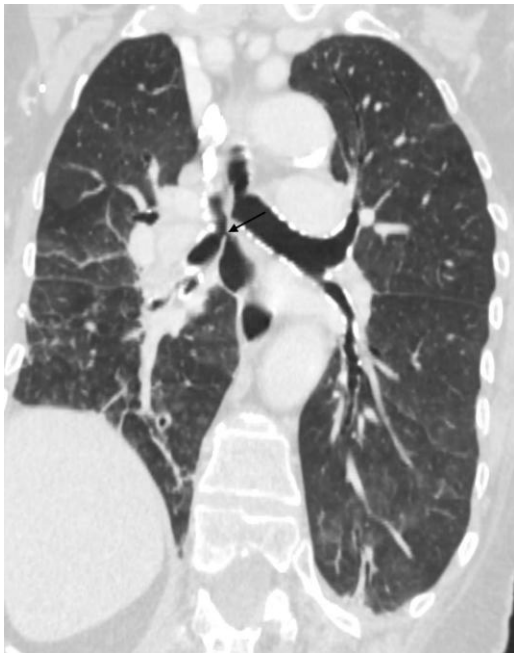


Figure 1c

Figure 1. Axial CT thorax in the lower thorax (1A) shows extensive cylindrical bronchiectasis, bronchial wall thickening and ground glass in the right lower lobe. At the level of the carina (1B), there is an abnormal communication between the oesophagus and right main bronchus (arrow), well demonstrated on a coronal oblique multiplanar reformat (1C) consistent with a tracheoesophageal fistula. Review of a prior CT from 7 years previous shows extensive interval progression in the right lower lobe bronchiectasis. At that stage, there was an oesophageal diverticulum abutting the right main bronchus, but no visible fistula.

Figure 2

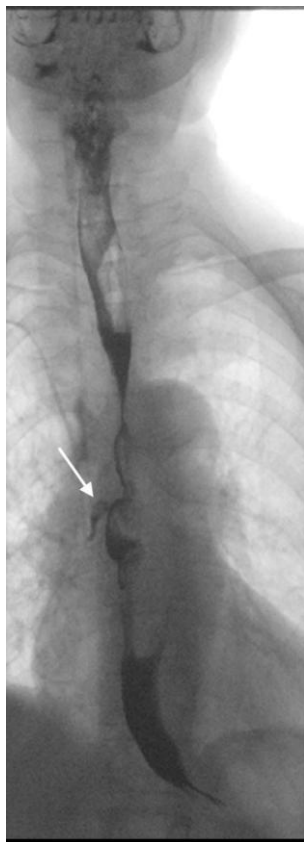


Figure 2a

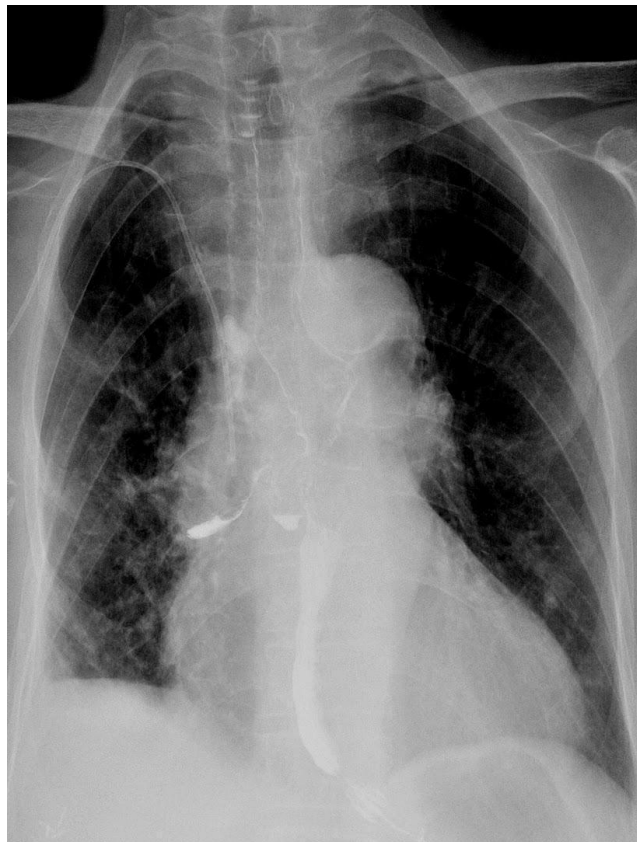


Figure 2b



Figure 2c

Figure 2. Barium swallow (2A) performed following the CT demonstrates contrast extending from a mid-oesophageal diverticulum in the right main bronchus (arrow). A delayed exposure (2B) from the same study shows extensive accumulation of barium in the right main bronchus and bronchus intermedius. Subsequent swallow post closure (2C) shows the closure device in the vicinity of the fistula, with no residual communication between the oesophagus and bronchus.

Discussion

Broncho-oesophageal fistulas are pathological connections between the oesophagus and a main bronchus. Congenital BOF is more common, typically presenting in infancy.

Acquired BOF are divided into malignant and benign causes with management varying depending on the cause. Oesophageal cancer is the most common type of cancer associated with malignant BOF ¹. This can occur either due to local tumour extent, or as a treatment-related complication, particularly following radiotherapy and/or oesophageal stenting ¹.

Greater than three quarters of benign acquired BOF are iatrogenic ². Prolonged mechanical ventilation, endotracheal tube insertion and tracheostomy can create abnormal connections due to pressure necrosis from the inflated cuff ². Granulomatous infection was historically the most common cause of benign BOF due to tuberculosis ¹.

The majority of the airway component of BOFs are located in the trachea and left main bronchus, due to the posteriorly lying upper and middle oesophagus ³. In this case, the BOF connected an oesophageal diverticulum and proximal right main bronchus.

Oesophageal diverticulum typically occur in the mid-third of the oesophagus, often on the right side due to inflamed subcarinal lymph nodes located at the right anterior oesophagus ⁴.

A high index of suspicion is required to diagnose this rare condition. BOF should be considered in patients with risk factors outlined above, who describe worsening cough after ingestion, recurrent aspiration pneumonia and malnutrition ². One study described Ono's sign, paroxysmal coughing after swallowing solids or liquids, being present in approximately 81% of patients with TOF ⁵.

Local surgical therapies are preferred for smaller fistulas < 5mm, as the risk of stenting outweighs the benefits. Endoscopic over-the-scope-clipping is a new technique that is fast and ensures persistent sealing of the fistula, compared to occlusive therapies, with fewer major complications ⁶. The clip is applied on the gastrointestinal side of the fistula as it requires soft, expansile tissue ⁷.

Declarations of Conflicts of Interest:

None declared.

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Informed Consent Statement:

Informed consent was obtained by both patient and next-of-kin on 27/08/2022.

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