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# Broncho-Oesophageal Fistula Complicating Carcinoma of the Oesophagus

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### ABSTRACT

Broncho-oesophageal fistula refers to an abnormal communication between a bronchus and the oesophagus. Broncho-oesophageal fistula is rare and can be either congenital or acquired. Congenital fistulas are usually diagnosed in the neonatal period as they present with respiratory distress and cyanosis during feeding. Whereas developmental anomaly is the commonest cause in infancy and childhood, the aetiology in adults is most frequently secondary to an oesophageal malignancy<sup>1</sup>. Non-malignant causes of fistulae are rare. Diagnosis of a broncho-oesophageal fistula may sometimes be difficult because of an insidious and non-specific clinical course<sup>2</sup>.

A rare case of broncho-oesophageal fistula due to carcinoma of the oesophagus, which was discovered incidentally during a barium swallow study is presented.

### SUMMARY

A 68-year-old man with dysphagia who was diagnosed to have carcinoma of the oesophagus, which was complicated by broncho-oesophageal fistula is presented. The role of radiology in the management of this condition is discussed.

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## CASE REPORT

M. E. is a 68-year-old retired teacher who presented at the University of Port Harcourt Teaching Hospital (UPTH) with a 6 month history of dysphagia. The dysphagia was progressive, at first to solids, then semi-solids and liquids. There was associated history of chest pain, loss of appetite and weight loss. There was no history of odynophagia or trauma. There was also history of cough which was said to have started about a week prior to presentation. The cough was non-productive and observed to be worse upon ingestion of liquids. There was no history of fever, night sweats, nausea or vomiting.

For these complaints, he had been treated in a hospital in his village but there was no significant improvement. Patient then resorted to traditional medicine men who gave him some local herbs which he used for about a month, but his symptoms worsened. The patient sought further help and was directed to UPTH, but did not present earlier due to lack of funds.

Past medical history was not contributory. Patient had a 25 year history of smoking, which was stopped about 10 years ago. He also consumed alcohol occasionally. He was not a known hypertensive or diabetic.

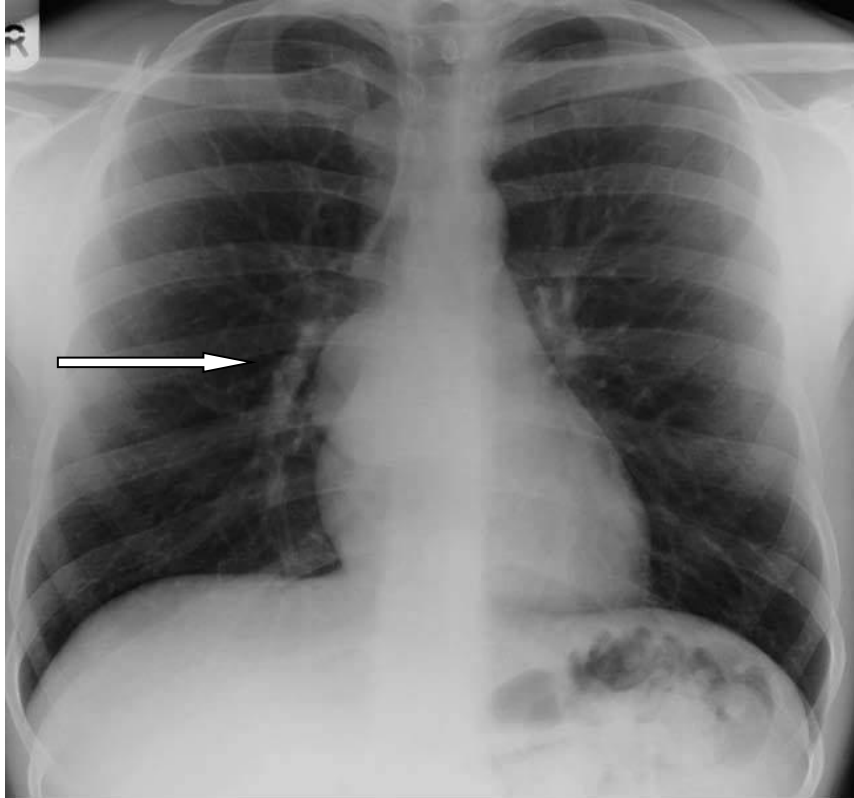
Physical examination revealed an anxious, weak and asthenic elderly man in no obvious respiratory distress. He was pale, but neither icteric nor cyanosed. Blood pressure was 110/70mmHg and pulse rate was 88 beats per minute. The chest and abdominal examinations were not contributory.

The patient was anaemic with haemoglobin of 7.8 g/dl (Normal: 12.5-15.0g/dl). Other laboratory investigations were normal: WBC  $7.6 \times 10^9/L$  (Normal:  $4-10 \times 10^9/L$ ), neutrophils 68% (Normal: 40-75%), lymphocytes 28% (Normal: 21-40%). A provisional diagnosis of oesophageal carcinoma with ? metastasis to the lungs was made. He was then sent to the Radiology Department for a chest radiograph, abdominal ultrasound and barium swallow

examination. The chest radiograph demonstrated a mediastinal mass of soft tissue density with a convex outer border projecting into the right hilar region. No calcifications or lucencies were seen within it. The ipsilateral cardiac border could be seen through it and the lung fields were clear (Fig. 1). The barium swallow demonstrated a long segment stricture and mucosal irregularity of the middle third of the oesophagus, giving an 'apple-core deformity', with extravasation of contrast into the left main bronchus (Fig 2 and 3). Diagnosis of carcinoma of the oesophagus with left broncho-oesophageal fistula was made. Upper gastrointestinal endoscopy and biopsy was carried out which revealed a circular ulceroproliferative growth in the oesophagus. Patient was scheduled to have a chest computed tomography (CT), but his condition continued to deteriorate and he died two weeks after presentation. The biopsy later revealed the mass to be squamous cell carcinoma of the oesophagus.

## DISCUSSION

Broncho-oesophageal fistulae are uncommon and difficult to diagnose. In the elderly, they are most frequently seen with an intrathoracic malignancy and are most commonly associated with malignancy of the oesophagus<sup>1</sup>. The malignant tissue spreads to involve the tracheal or bronchial wall. Subsequent ulceration and necrosis of the malignant tissue leads to tissue breakdown and fistula formation<sup>3</sup>. Though a majority of acquired broncho-oesophageal fistulae result from malignancy of the oesophagus, they can also result from tumours of the lung, trachea and lymph nodes<sup>4</sup>. Malignant causes of broncho-oesophageal fistula accounted for about 5-15% of cases<sup>5</sup>, while non-malignant causes were found to be responsible for only 5-6% of such cases<sup>6</sup>. Non-malignant causes of acquired broncho-oesophageal fistula are radiotherapy, ingestion of corrosives, blunt and penetrating chest trauma, granulomatous mediastinal infections, prior surgery on oesophagus and



**Fig. 1.** Postero-anterior chest radiograph demonstrating the mediastinal mass of soft tissue density with a convex outer border projecting into the right hilar region (white arrow). The ipsilateral cardiac border can be seen through it.



**Fig. 2.** Single contrast barium swallow demonstrating a long segment stricture and mucosal irregularity of the middle third of the oesophagus, giving an 'apple-core deformity'(black arrow), with contrast filling of the left main bronchus (white arrow). There is pre-stenotic dilatation of the proximal oesophagus.



**Fig. 3.** Single contrast barium swallow demonstrating a long segment stricture and mucosal irregularity of the middle third of the oesophagus, giving an 'apple-core deformity'(black arrow), with contrast filling of the left bronchial tree (white arrow).

tracheobronchial tree, iatrogenic injuries and indwelling stents<sup>4</sup>. Other conditions known to cause acquired broncho-oesophageal fistula include inflammatory conditions like Crohn's disease, Behect's disease and broncholithiasis<sup>7</sup>.

Acquired broncho-oesophageal fistulae are frequently misdiagnosed. The most common symptoms associated with broncho-oesophageal fistula include cough during meals and drinking liquids and chest pain, both of which were present in the index patient. Bouts of coughing while eating or drinking is known as the Ohno's sign<sup>7</sup>. With the background provisional diagnosis of oesophageal carcinoma, this sign should have raised the suspicion of a fistula complicating the carcinoma in this patient, rather than lung metastasis. Delay in diagnosis may be complicated by pneumonia, life-threatening hemoptysis, and respiratory failure<sup>7</sup>.

The barium oesophagogram is an important study in the diagnosis of a broncho-oesophageal fistula at any age. Barium oesophagogram is useful in outlining the characteristics of the fistula and may be the investigation of choice in the majority of cases. It is considered to be the most sensitive test for diagnosing broncho-oesophageal fistula. This investigation provides a definitive diagnosis in 78% of cases<sup>7</sup>. Contrast may be seen passing through the fistula from the oesophagus to the lumen of the bronchus. In the present patient, the barium study demonstrated contrast in both the oesophagus and the left main bronchus. Being a luminal study, barium oesophagogram would fail to show changes in the wall of the esophagus and in the mediastinum that are shown accurately by CT. CT scan of the thorax allows evaluation for neoplasm, lymphadenopathy, or other associated anomalies and it may also demonstrate the fistula<sup>8</sup>. A routine CT study could miss the fistula if the fistula tract is collapsed. This

drawback can be overcome by performing a CT oral contrast swallow study<sup>3</sup>. The current patient died before a CT scan could be done. Endoscopy is the best diagnostic method, as it allows for direct visualization. In this way, biopsies of the mucosa may be obtained, ultimately helping to determine the aetiology of the fistula. This patient had an upper gastrointestinal endoscopy which confirmed the diagnosis of squamous cell carcinoma.

Regardless of the aetiology, the important life-threatening complications from broncho-oesophageal fistulae are pulmonary infections and poor nutrition<sup>3</sup>. Once these are diagnosed, urgent treatment is warranted, the delay of which results in catastrophic pulmonary complications. Surgical resection or ligation/suturing of the fistulous tract of non-malignant broncho-oesophageal fistulae gives good results and the recurrences are uncommon<sup>9</sup>. There is no cure for malignant broncho-oesophageal fistulae and palliative procedures like oesophageal stenting, oesophageal exclusion, oesophageal bypass, fistula resection and repair may prolong survival<sup>4, 10</sup>. Insertion of a coated self-expandable metal stent is the treatment of choice for an individual with malignant broncho-oesophageal fistula<sup>11</sup>. The patient did not live long enough to benefit from any of these procedures.

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