

Oesophageal duplication cyst: an unusual cause of retrosternal pain and dysphagia in an adult

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ABSTRACT

Oesophageal duplication cysts in adults are a rare entity and are mostly asymptomatic. We describe the imaging findings in a rare case of oesophageal duplication cyst simulating cold abscess, causing retrosternal pain and dysphagia in a 25-year-old man.

Keywords: congenital oesophageal anomaly, duplication cyst, oesophageal duplication cyst, oesophagus

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INTRODUCTION

Oesophageal duplication cysts are rare congenital oesophageal anomalies, particularly in adults. Diagnosis of an oesophageal duplication cyst is usually made in infancy and childhood while investigating for respiratory distress or feeding difficulty. Complications are known to occur during the natural course of the disease due to bleeding, infection and mass effect. In adults, oesophageal duplication cysts are usually asymptomatic and the diagnosis is made most often from an incidental finding on the chest radiograph. Complete surgical excision is the standard treatment for symptomatic oesophageal duplication cyst. Endoscopic ultrasonography is a reliable means of accurate preoperative diagnosis of oesophageal duplication cyst. We describe the imaging findings of a rare case of oesophageal duplication cyst simulating cold abscess in a young man.

CASE REPORT

A 25-year-old man, previously asymptomatic, presented with progressive dysphagia, retrosternal pain, intermittent fever and left-sided pleuritic chest pain and weight loss of approximately 5 kg over a period of the past two months. His past history was not significant. The initial chest radiograph revealed pleural effusion on the left side, with passive collapse/consolidation of the underlying lung. Retrocardiac opacity with convex right border was also seen in the paraspinal region. The adjacent vertebral bodies and the intervertebral disc spaces were unremarkable

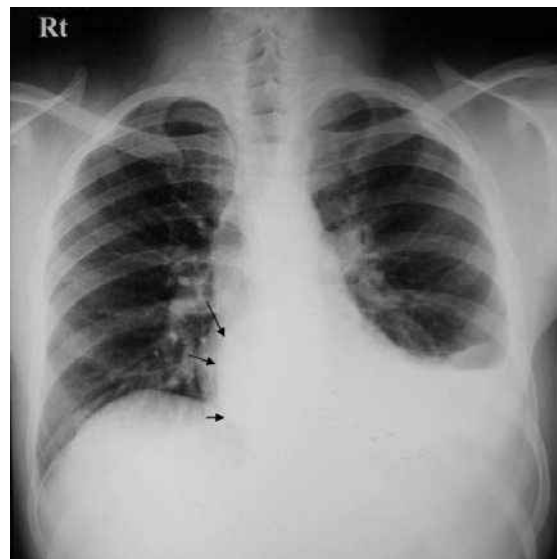


Fig. 1 Frontal chest radiograph shows a left-sided pleural effusion. There is a retrocardiac curved density (arrows) projected near the right heart border.

(Fig. 1). Aspiration of straw-coloured pleural fluid demonstrated dominance of lymphocytes. The pleural fluid adenine deaminase level unit was 76 U/L. At this point, a diagnosis of tuberculous pleural effusion with right paravertebral abscess was suggested. A barium swallow study was also performed to evaluate his dysphagia, which revealed a large extrinsic impression along the right side of the distal oesophagus. The distal oesophagus was stretched around this extrinsic impression (Fig. 2).

Contrast-enhanced computed tomography (CECT) and magnetic resonance (MR) imaging were performed to further characterise the mass and to rule out vertebral involvement. CECT showed a well-circumscribed pre-vertebral thin-wall cystic mass measuring 45 mm × 75 mm × 56 mm (Fig. 3), with pleural effusion and underlying atelectasis on the left side. The attenuation of the cyst fluid was 24 Hounsfield units (HU). No appreciable contrast enhancement of the cyst wall was seen. No vertebral involvement was seen on MR imaging. The mass was hypointense on T1-weighted images and brightly hyperintense on T2-weighted images (Fig. 4). Based on the imaging findings, the diagnosis was reviewed and

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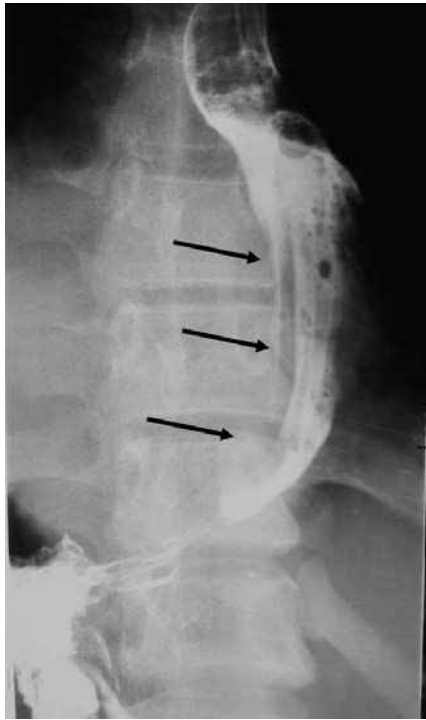


Fig. 2 Barium swallow shows a large extrinsic impression on the right side of the oesophagus. The visualised oesophageal mucosa and adjacent vertebrae, including intervertebral disc spaces, appear normal.

a final diagnosis of tuberculous pleural effusion and oesophageal duplication cyst was made. The patient was treated with anti-tuberculous therapy (ATT). The cystic lesion was completely excised via thoracotomy after two months of starting ATT. Histopathology of the specimen confirmed the diagnosis of oesophageal duplication cyst, with lymphocytes infiltration in its wall, likely due to secondary infection. Hence, a final diagnosis of secondarily-infected oesophageal duplication cyst with left-sided pleural effusion was made. His postoperative period was unremarkable and he was discharged on the tenth postoperative day. ATT was continued six months postoperatively, although there was no histological evidence of tuberculosis. He was doing well at eight months follow-up.

DISCUSSION

Oesophageal duplication cyst in adults is a rare clinicopathological entity. Oesophageal duplication cyst results from an abnormal vacuolisation process that produces the oesophageal lumen in the fifth to eighth week of embryonic life.⁽¹⁾ Oesophageal duplication cyst usually involves the thoracic oesophagus, and although rarely, it can involve the cervical oesophagus or even be intrabdominal.^(2,3) This lesion usually present alone; rarely, they may occur in association with other abnormalities like bronchogenic duplication cyst and other tracheoesophageal / pulmonary malformations.^(4,5)

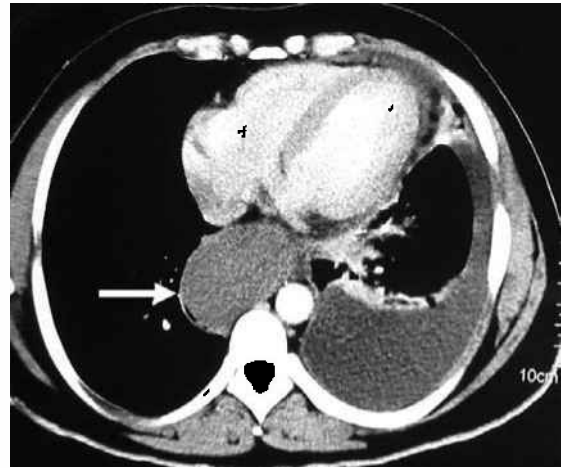


Fig. 3 Axial CECT image shows a fluid-attenuating cystic structure in the retrocardiac region (arrow). There is a left-sided pleural effusion and associated passive atelectasis of adjacent lung parenchyma.



Fig. 4 (a) Sagittal and (b) coronal T2-W inversion recovery MR images show a hyperintense posterior mediastinal lesion. There is a hyperintense left-sided pleural effusion.

Uncomplicated oesophageal duplication cysts are mostly asymptomatic and are often detected incidentally on chest radiographs. Infection, bleeding and mass effects are the most common complications associated with it. Some of the rare complications reported in the literature are acute rupture, rapid enlargement and malignant transformation of oesophageal duplication cyst.⁽⁶⁻⁸⁾ Oesophageal duplication cyst at times may pose a real diagnostic challenge for the radiologists. With the help of endoscopic ultrasonography, accurate preoperative diagnosis of oesophageal duplication cyst is now possible.⁽⁹⁾ When the imaging findings are equivocal, the diagnosis of oesophageal duplication cyst can only be made on histopathology. Traditionally, oesophageal duplication cysts are treated with complete surgical resection via a thoracotomy. In expert hands, successful laparoscopic resection of oesophageal duplication cyst is also possible.⁽¹⁰⁾

The unusual feature in our case was its presentation. History of cough, fever, weight loss along with

lymphocyte predominant pleural effusion in our patient was considered to be due to tuberculosis. Finding of a paraspinal lesion with the above background was thought to possibly represent a cold abscess, especially in a country like India where tuberculosis is prevalent. Kumar et al described a similar case of oesophageal duplication cyst simulating an empyema.⁽¹¹⁾ There is scanty literature about the MR imaging features of oesophageal duplication cyst.⁽¹²⁾ We described the comprehensive imaging findings, including MR imaging findings, in our case. Our case demonstrates unusual clinical as well as radiological features in an infected oesophageal duplication cyst and highlights the need for a strong index of suspicion if one has to diagnose oesophageal duplication cyst preoperatively, even in the presence of a second pathology which can masquerade it.

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