Isolated Oesophageal Cyst Connected with the Bronchus

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ABSTRACT

We report an unusual case of a patient with an oesophageal cyst connected to the bronchus. A 24-year old male with a two-year history of repeated attacks of chest infection and haemoptysis was found to have a cyst of 4×4 cm affecting the anterior and apical segments of the right upper lobe. The cyst was excised in its entirety and the histopathological study of the cyst showed stratified squamous epithelium with submucosal and muscular layer but no cartilage. The pathological diagnosis was an oesophageal cyst. No previous case of isolated oesophageal cyst connected to the bronchus has been reported according to the available literature.

Quiste Esofágico Aislado Conectado a Bronquio

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RESUMEN

Reportamos aquí un raro caso de un paciente con un quiste esofágico conectado a bronquio. A un hombre de 24 años de edad con una historia de dos años de ataques repetidos de infección del pecho y hemoptisis, se le halló un quiste de 4 ×4 cm. que afectaba los segmentos anterior y apical del lóbulo superior derecho. El quiste fue extirpado en su totalidad y el estudio histopatológico del quiste mostró un epitelio escamoso estratificado con capas submucosas y musculares pero sin cartílago. El diagnóstico patológico fue un quiste esofágico. Ningún caso anterior de quiste esofágico aislado conectado a bronquio se reporta en la literatura disponible.

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INTRODUCTION

In 1711, Blasius initially described oesophageal cysts. Evidence from autopsy series suggest an incidence of 1:8200 for oesophageal cyst with a 2:1 male predominance. Most of the cysts were reported to arise in the lower oesophagus with only 23% occurring in the upper third (1–3). Oesophageal cysts are usually located in the para-oesophageal area and have a connection with the proper oesophagus (4, 5). No previous case of an isolated oesophageal cyst connected to bronchus has been reported according to the available literature. In this case report, we describe a case of a pulmonary oesophageal cyst with a bronchial connection.

Case Report

A 24-year old male was referred to Affiliated Hospital of Guangdong Medical College with a two-year history of repeated attacks of chest infection and haemoptysis. Physical examination revealed that the percussion note was dull and air entry was decreased in the right upper zone. The rest of the examination was normal and routine investigation *ie* haemoglobin, serum urea and electrolytes were within normal limits. Barium swallow was normal. Fiberoptic bronchoscopic examination and a bronchial biopsy specimen suggested an inflammatory process. He received several courses of antibiotics and was also investigated for pulmonary tuberculosis because of persistent lung infiltrates on his chest X-ray (Fig. 1). All investigations for tuberculosis were nega-

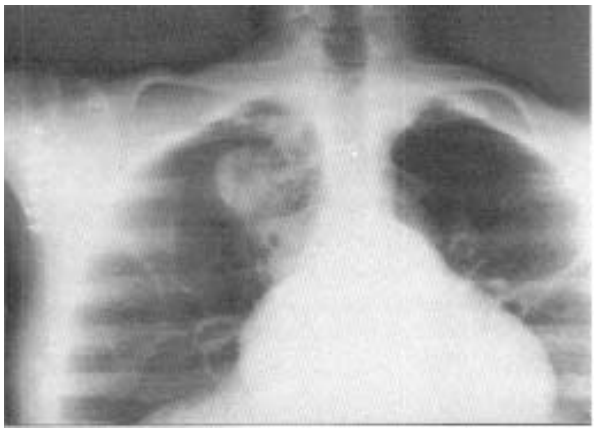

Fig. 1: Kyphotic chest radiograph reveals a 4 x 4 cm mass in the right upper lobe.

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tive, but despite this, he was treated empirically with antituberculous drugs for six months without improvement. He had a Computed Tomography scan (CT) of the chest (Fig. 2)

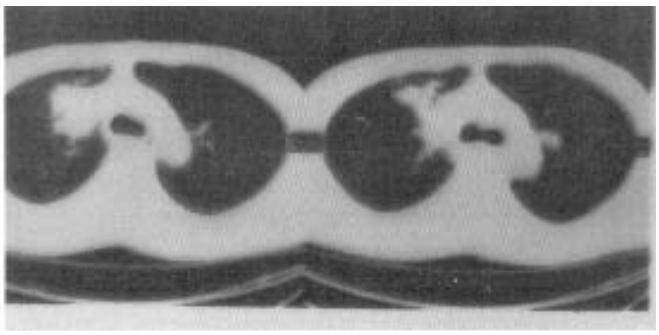

Fig. 2: CT scan of the chest showing a 4 x 4 cm cyst affecting the anterior segment and apical segment of the right upper lobe.

which showed a mass of size 4 x 4 cm affecting the anterior and apical segments of the right upper lobe. He underwent a diagnostic right thoracotomy. The right upper lobe was found to be firm in consistency and contained adhesions to the inner chest wall. Operative findings revealed a 4×4×3 cm sized cyst in the right middle and upper thorax extending into the mediastinum. The cyst was unilocular about 4 mm thick and contained white fluid. The lower end of the cyst was connected to the right upper lobe bronchus. The cyst was excised in its entirety and a chest tube inserted for drainage. After the lung had expanded completely, the intercostal chest tube was removed on the fifth postoperative day. The patient was afebrile and was discharged on the seventh postoperative day. The clinical diagnosis was a bronchial cyst but the histopathological study of the cyst showed stratified squamous epithelium with submucosal and muscular layer with no cartilage. There was chronic inflammatory cell infiltrate in some areas with formation of lymphoid follicles. The pathological diagnosis was an oesophageal cyst (Fig. 3).

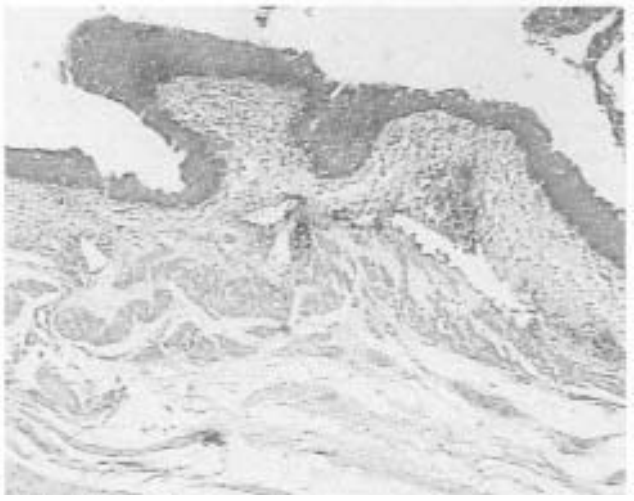

Fig. 3: Microscopy of the specimen shows a stratified squamous epithelium with submucosal and muscular layer.

DISCUSSION

Oesophageal cysts are retention cyst that result from the abnormal budding of the ventral diverticulum of the primitive foregut; it commonly originates near the oesophagus in the middle mediastinum (6). Oesophageal cysts in the para-oesophageal area arise during the fourth week of fetal life, at which point separation of the primitive foregut into oesophagus and trachea occurs. During this separation, if the abnormal budding separated from the primitive foregut, then an oesophageal cyst or a bronchial cyst could occur. Because of the similarity between an oesophageal cyst and a bronchial cyst in terms of origin and radiological and pathological findings, the generic term *foregut cyst* is sometimes used (1, 6). Respiratory epithelium with scattered submucosal glands and islands of cartilage are characteristics of bronchogenic cysts. Immature squamous epithelium and thick smooth muscle coats are compatible with oesophageal cysts. In the index patient, the cyst was lined by stratified squamous epithelium and the wall contained smooth muscles. There was no cartilage. It is sometimes difficult to distinguish oesophageal cyst from bronchogenic cyst. The epithelial lining of an oesophageal cyst may be of respiratory or alimentary tract origin, or rarely, a combination of both (7). Sun Lee *et al* reported a case of extralobe pulmonary sequestration with an associated cyst of mixed bronchogenic and oesophageal type (8). The foregut cyst of mixed bronchogenic and oesophageal type offers evidence of early embryologic insult. Four cases of bronchogenic para-oesophageal cysts with oesophageal communication have been reported (9). Heithoff and Arbona *et al* suggested a unified concept where the foregut cyst arises from abnormal primitive foregut development. The spectrum of foregut cysts includes oesophageal or gastric diverticula, intralobar and extralobar pulmonary sequestration and oesophageal or bronchogenic duplication cysts (10, 11).

Oesophageal cysts become apparent in the first four decades of life. Patients are usually asymptomatic but may present with infections, rupture, haemorrhage or ulceration due to migration of the gastric mucosa. A few cases of malignant transformation have been reported and complete resection is recommended even in asymptomatic patients. There have also been a few case reports of oesophageal cyst complicated with spinal malformation (12). Modern imaging techniques, such as computed tomography, endoscopic ultrasonography and magnetic resonance imaging may help in excluding malignancy and in evaluating the topographic relationship of the mass in order to plan the most appropriate surgical approach (13). The definitive diagnosis of oesophageal cyst requires pathological evaluation. Complete resection of the oesophageal cyst is usually effective.

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REFERENCE

1. Arbona JL, Figueroa-Fazzi JG, Mayoral J. Congenital esophageal cysts: case report and review of literature. Am J Gastroenterol 1984; **79:** 177–82.
2. Woodring JH, Vandiviere HM, Dillon ML. Air filled, multi-locular bronchopulmonary foregut duplication cyst of the mediastinum. Unusual computed tomography appearancce. Clin Imaging 1989; **13:** 44–7.
3. Harvell JD, Macho JR, Klein HZ. Isolated intra-abdominal esophageal cyst. Am J Surg Pathol 1996; **120:** 476–9.
4. Leblanc J, Guttentag AR, Shepard JA, Macloud TC. Imaging of mediastinal foregut cysts. Can Assoc Radiol J 1994; **45:** 381–6.
5. Nawaz A, Matta H, Jacobsz A, Ansari F, Al-Khouder G, Al-Salem AH. Interthoracic foregut duplication cysts in neonates. Ann Saudi Med 2001; **21:** 206–9.
6. Strollo DC, Rosado-de-Christenson ML, Jett JR. Primary mediastinal tumours, Part II. Tumours of the middle and posterior mediastinum.Chest 1997; **112:** 1344–57.
7. Diane CS, William RS, Joseph CE. Benign development cyst of the mediastinum. Arch Pathol Lab Med 1997; **101:** 136–8.
8. Sun L, Jae HP, Youn WH, Moon HY, Dong W. Extralobar pulmonary sequestration with an associated cyst of mixed bronchogenic and esophageal type. J Korean Med Sci 1997; **12:** 567–9.
9. Kne eviæ J, Radovanoviæ N, Simiæ A, Micev M, Radojkoviæ S, Gerziæ Z. A paraesophageal bronchogenic cyst with esophageal communication. Dis Esophagus1999; **12:** 321–3.
10. Heithoff KB, Sane SM, Williams HJ, Jarvis CJ, Carter J, Kane P, et al. Bronchopulmonary foregut malformations, a unifying etiological concept. AJR Am J Roentgenol 1976; **126:** 46–55.
11. Arbona JL, Fazzi JG, Mayoral J. Congenital esophageal cyst: case report and review of literature. Am J Gastroenterol 1984; **79:** 177–82.
12. Laraja RD, Rothenberg RE, Chapman J, Imran-ul-Haq, Sabatini MT. Foregut duplication cyst: a report of a case. Am Surg 1995; **61:** 840–1.
13. Geller A, Wang KK, Dimango EP. Diagnosis of foregut duplication cysts by endoscopic ultrasonography. Gastroenterology 1995; **109:** 838–42.