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Case report: Communicating bronchopulmonary foregut malformation-type III (esophageal lobe)

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Patent, congenital communications between accessory lung tissue and the esophagus or the stomach are extremely rare and only a few cases have been recorded in literature.[1–5] We present a case of communication between the esophagus and the right lower lobe of the lung -'esophageal lobe.'

Case Report

A 54-year-old man presented with symptoms of retrosternal burning and aspiration when lying down. During his childhood and early adulthood he had not had any significant respiratory or gastrointestinal symptoms. On clinical examination, he was a well-built individual and had normal blood pressure, pulse, respiratory rate, and temperature. He had a few rales and rhonchi in the base of the right lung. A plain radiograph of the chest [Figure 1] showed a patchy opacity confined to the right lower lobe. He was treated with antibiotics and improved clinically, but the right lower lobe lesion persisted.

Gastrointestinal reflux disease (GERD) was suspected and a barium swallow study was performed in the erect and gravity-neutral positions. It showed a communication between the retrocardiac segment of the esophagus and the right lower lobe, which ended in a bronchiectasis-like picture [Figure 2]. There was free gastroesophageal reflux as well. When CT was performed after the barium swallow, barium was seen in the bronchiectatic segments of the right lower lobe [Figure 3] and in the communication [Figure 4]. An esophagoscopy was then performed and a fenestrated orifice was noticed at 35 cm from the incisor teeth [Figure 5]. Bronchoscopy showed that the right lower lobe bronchus was absent. A bronchogram performed with isosmolar contrast revealed normal upper and middle lobe bronchi and absence of the right lower lobe bronchus. No communication was seen with the bronchiectatic segment in the right lower lobe. An aortogram was performed, which showed two well-developed systemic arteries arising from the right side of the aorta [Figure 6]; these were embolized subsequently.[6,7]

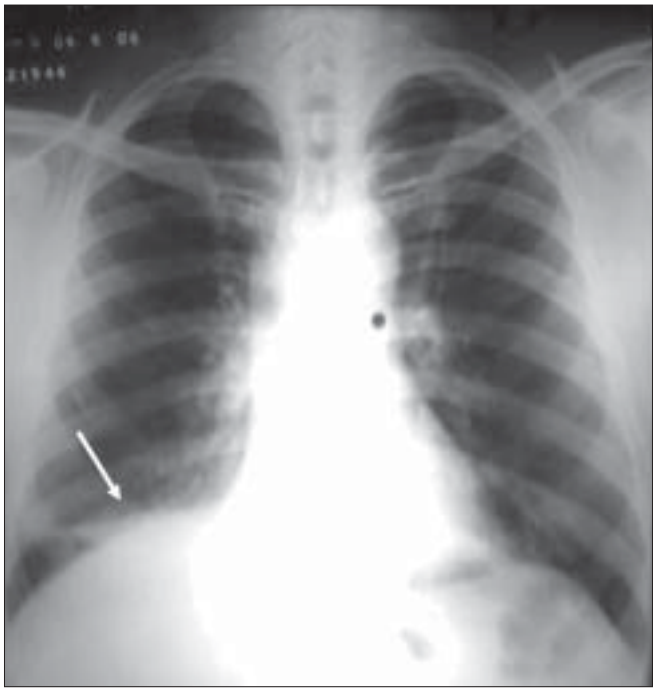

Figure 1: Plain radiograph of the chest shows an ill-defined opacity (arrow) in the right paracardiac region

At surgery, the right lower lobe was removed and the connection between the esophagus and the lung was excised. Histopathology [Figure 7] revealed squamous epithelium on the esophageal side of the communication and pseudostratified ciliated columnar epithelium on the pulmonary side. The fistulous tract was made of cartilage. The whole specimen showed malformed lung tissue with no clear cut bronchus, bronchioles, or alveoli.

Discussion

The term 'bronchopulmonary foregut malformation'

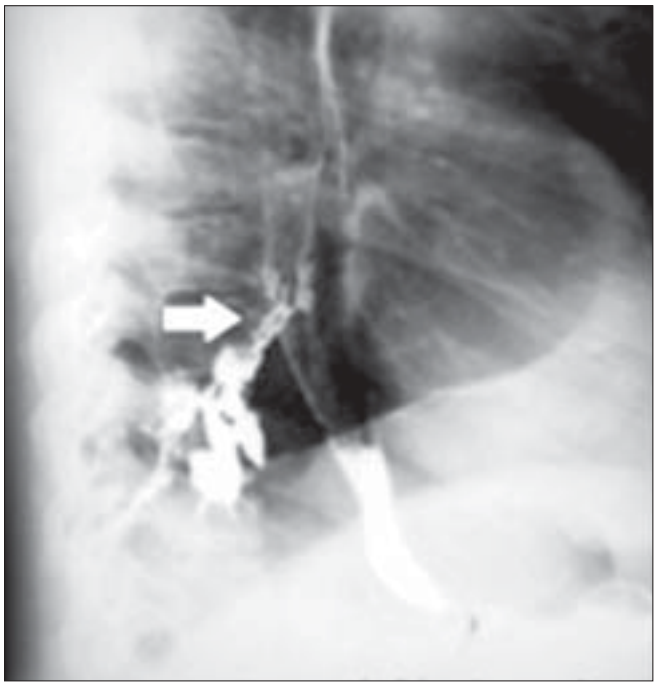

Figure 2: Esophagogram shows a communication between the esophagus and the right lower lobe (arrow)

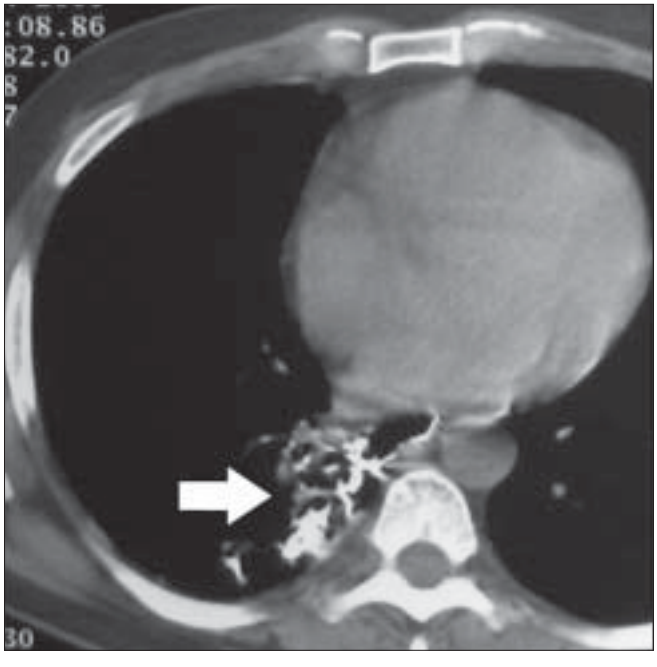

Figure 3: CT of the chest shows a bronchiectatic segment in the right lower lobe with retained barium (arrow) from the esophagogram

was coined by Gerle *et al.* in 1968.[2] It includes a variety of congenital malformations, including pulmonary sequestration, foregut duplication cysts, and esophageal diverticula. It was left to Heithoff et al.[1] to emphasize that these rare anomalies provide evidence of a common pathogenesis for the whole gamut of bronchopulmonary foregut malformations, which includes intralobar and

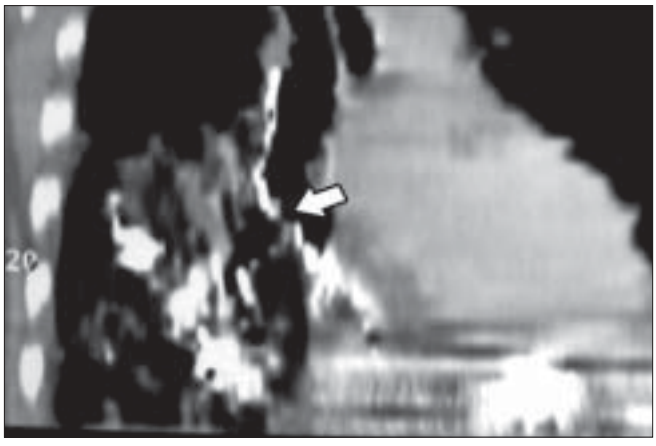

Figure 4: Reconstructed CT images show the communication (arrow) between the esophagus and the right lower lobe

extralobar sequestrations with or without communications with the alimentary tract, foregut duplication cysts, and esophageal diverticula. Srikanth et al. proposed an anatomical classification for bronchopulmonary foregut malformations and introduced the term 'communicating bronchopulmonary foregut malformations.'[3] Our case can be classified as a type III communicating bronchopulmonary foregut malformation.

These communications are explained by the presence of an abnormal collection of pluripotent cells with respiratory potential, arising from the dorsal side of the developing esophagus, distal to the normal lung bud, which develop into lung tissue, retaining the communication with the esophagus (foregut). The term 'esophageal lung' is used when the whole lung is connected to the esophagus through a main bronchus.[4] These anomalies have their own arterial supply, venous drainage, and pleural investment. In our patient, the communication arose from the lower end of the esophagus on the right posterolateral aspect, ending in lung tissue, involving only the right lower lobe. The lower esophagus and gastroesophageal junction are common sites of communication, though communication with the fundus of the stomach as well as the mid- and upper esophagus, has been described.[1]

Angiography, in our patient, demonstrated two large aberrant vessels arising from the mid-thoracic aorta. Arterial supply can also arise from the carotid and the subclavian arteries.[1] The two large aberrant vessels were embolized to get an avascular field during surgery.[6,7]

The treatment varies for the different anomalies. After evaluation of the anomalous pulmonary tissue, if it appears destroyed due to atelectasis, bullae, or abscess, or if only one lobe is involved (Srikanth groups I and III; Table 1), the treatment is resection of the tissue, as was done in our case. If the lung tissue appears to be normal, then angiography is performed to assess the vascular supply along with

Table 1: Classification of communicating bronchopulmonary foregut malformations by Srikanth *et al*.[3]

Group	Description
1A	Total sequestered lung communicating with the foregut, associated with esophageal atresia and tracheoesophageal fistula to the distal pouch.
1B	Sequestered anatomic lobe or segment communicating with the foregut, associated with esophageal atresia and tracheoesophageal fistula to the distal pouch.
II	Total sequestered lung communicating with the lower esophagus; absent ipsilateral mainstem bronchus.
III	Isolated anatomic lobe or segment communicating with the foregut.
IV	Portion of normal bronchial system communicating with the esophagus.

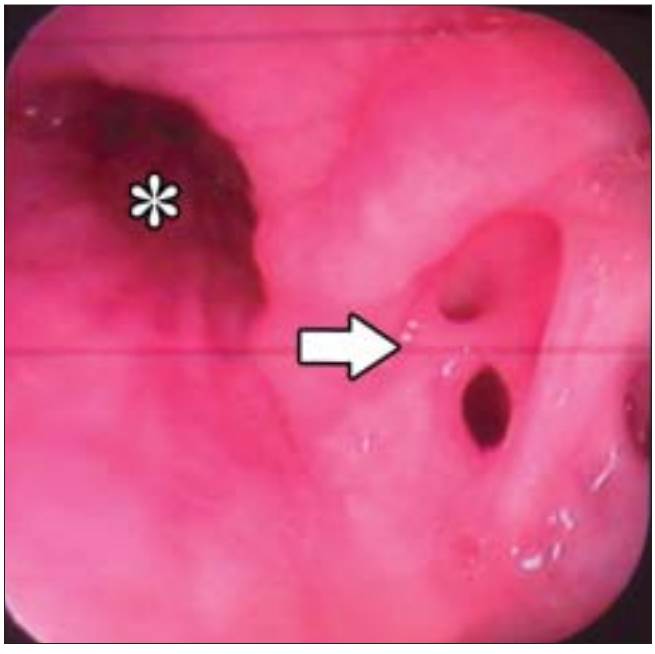

Figure 5: Endoscopy image shows the fenestrated opening (arrow) in the right posterolateral aspect of the esophagus

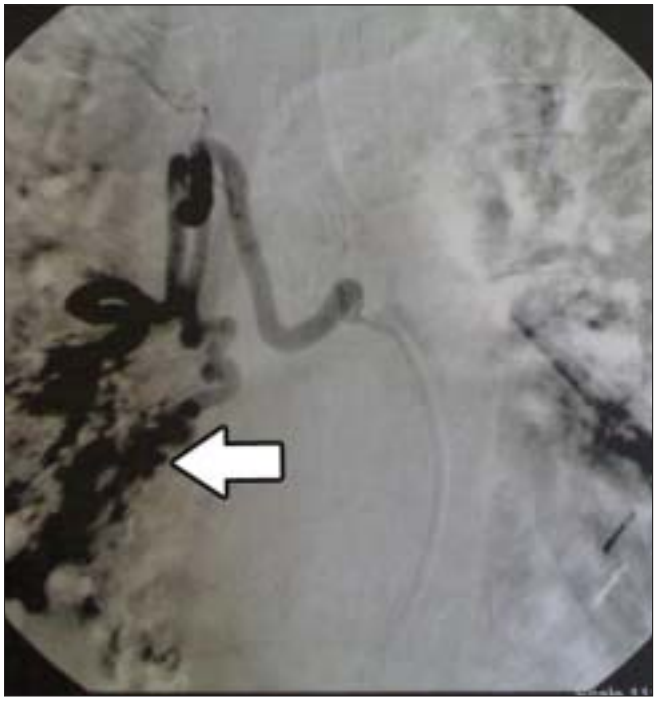

Figure 6: Selective angiogram shows the systemic arterial supply to the abnormal lobe (arrow)

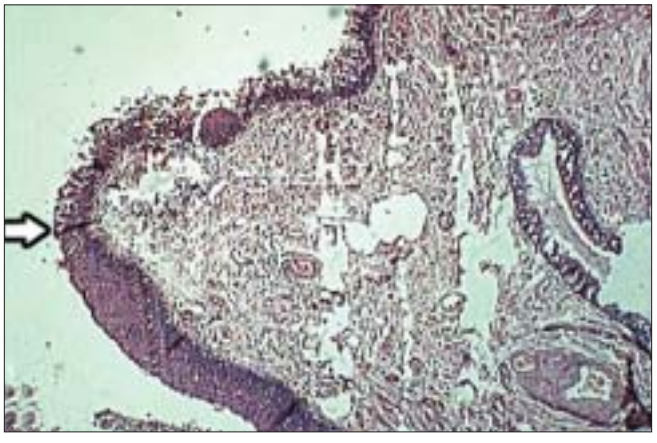

Figure 7: Histopathology specimen shows the transition of the epithelium (squamous to pseudostratified ciliated columnar epithelium) from the esophagus to the lung (arrow)

assessment of the oxygenation capacity of the tissue. Today, multi-slice or multi-detector CT scans can be used instead of diagnostic catheter angiography for assessing the abnormal vascular supply. If the vascular supply appears normal and adequate, bronchial reconstruction can be done.[5]

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