



## Esophageal lung – A rare bronchopulmonary foregut malformation<sup>☆</sup>



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### ABSTRACT

Esophageal lung is a rare variety of communicating bronchopulmonary foregut malformation characterized by a fistula between an isolated portion of respiratory tissue and esophagus or stomach. It may involve the entire lung or one of the pulmonary lobes. Only 20 cases have been reviewed in 2011. Fifty percent of cases are associated with a tracheoesophageal fistula. We report a case of a 6 month old girl who was previously operated for TEF repair, with esophageal lobe which was successfully excised. The relevant literature is reviewed.

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Bronchopulmonary foregut malformations (BPFM) are rare congenital malformations, which usually present in early neonatal age. The spectrum ranges from congenital cystic adenomatoid malformation (CCAM), intralobar and extralobar pulmonary sequestration (EPS) with associated foregut diverticulae, duplication cysts, tracheoesophageal fistulae (TEF), and bronchoesophageal fistulae [1]. Esophageal lung is a type of rare communicating bronchopulmonary foregut malformation (CBPFM) characterized by a fistula between an isolated portion of respiratory tissue and esophagus or stomach [2]. 20 such cases have been reported so far in literature. We report one such case associated with tracheoesophageal fistula (TEF).

### 1. Case report

A six month old girl, who was operated for an extrapleural repair of TEF type C on day 2 of life, came with complaints of persistent cough and lower respiratory tract infections since past 2 months. 2D echocardiography showed a minor cardiac anomaly (tiny patent foramen ovale), which did not require any cardiological intervention. She underwent an upper GI contrast study (Fig. 1A) that showed an outpouching from the lower third of esophagus.

However, the outpouching seemed to be much lower down than the expected site of esophageal anastomosis. Hence, a diagnosis of recurrent TEF seemed less likely. Another possibility of a second lower pouch fistula, which could have been missed at the time of first surgery, was also thought of. She underwent upper GI scopy where the suspected fistula was visualized (Fig. 2A, B). An attempt was made to tackle the fistula endoscopically and ligate it with vascular clips. However, the symptoms persisted and a repeat dye study showed persistent outpouching (Fig. 1B). A contrast enhanced computed tomogram (CT scan) showed a collapsed lower lobe communicating with the esophagus (Fig. 3). It had no communication with the trachea-bronchial tree and hence a diagnosis of esophageal lobe was made. A right posterolateral thoracotomy with lower lobectomy (excision of esophageal lobe) was planned. The lower lobe bronchus was found to communicate with the esophagus. There was no communication with the parent tracheobronchial tree. The lobe received its blood supply from the pulmonary vessels (Fig. 4). The lobe was excised and the esophagus was closed (Fig. 5). The histopathology report showed lung tissue with lymphocytic infiltrate and the communicating bronchus showed presence of cartilage (Fig. 6). Postoperative dye study was normal. Patient is asymptomatic at follow up of 2 months with weight gain of 800 g.

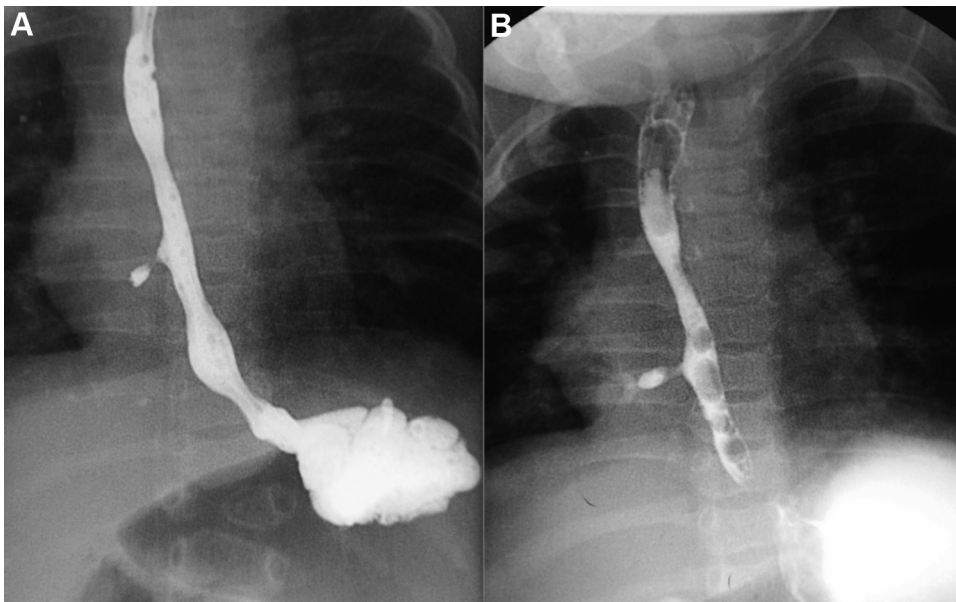
### 2. Discussion

Esophageal lung is an extremely rare type of BPFM in which the mainstem bronchus is connected to the esophagus by a cartilage

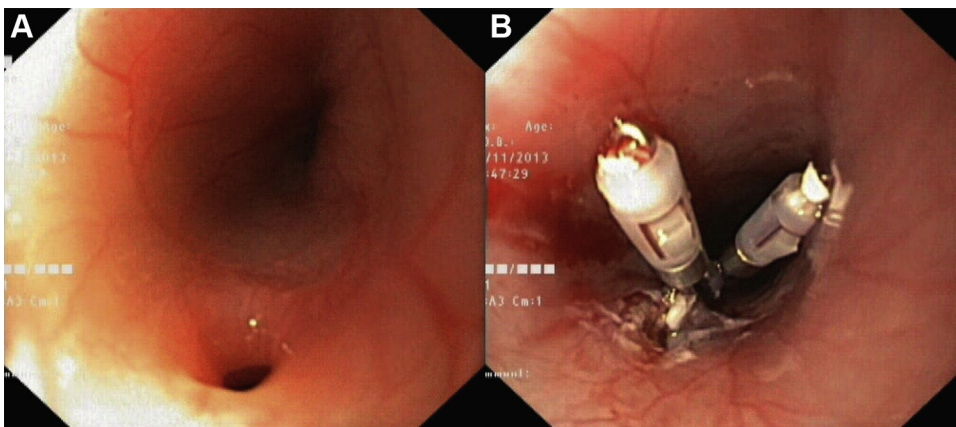
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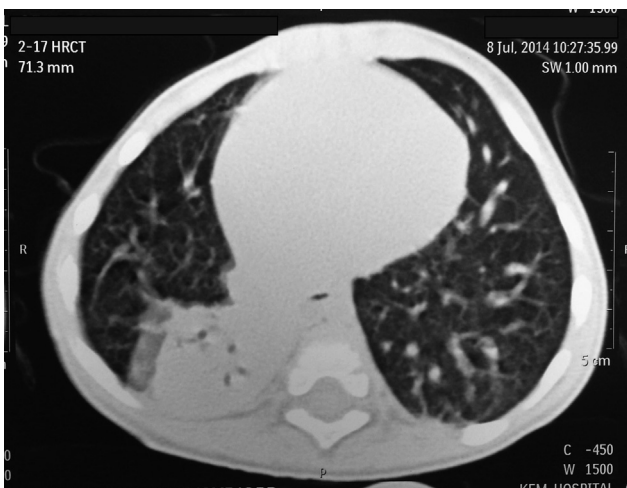
E-mail address: [sandeshparelkar@kem.edu](mailto:sandeshparelkar@kem.edu) (S.V. Parelkar).



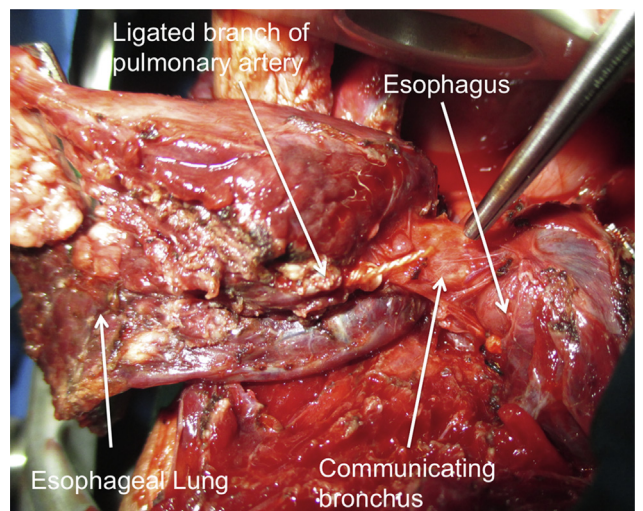
**Fig. 1.** A – Dye study showing lower esophageal-bronchial communication. B – Repeat dye study showing persistent communication.



**Fig. 2.** A – Upper GI endoscopy showing mucosa lined fistulous tract. B – Ligation of the fistula with vascular clips.



**Fig. 3.** CT scan showing non-expanding right lower lobe communicating with the esophagus.



**Fig. 4.** Intra-operative picture showing the dissected lower lobe communicating with esophagus.

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