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CASE REPORT

Case report: Acquired esophago-pulmonary fistula

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Abstract Acquired esophago-respiratory fistulae are usually esophago-tracheal or esophago-bronchial. Esophago-pulmonary fistulae have been rarely described. We report a case of 17-year-old girl with esophago-pulmonary fistula. She experienced a long history of cough and expectoration. CT scans of the chest demonstrated right lower and middle lung lobes multiple cavitary lesions with marked dilatation of the esophagus and a communicating fistula between the esophagus and parenchyma of the right lower lobe of the lung. Excision of the fistula and right bilobectomy were done via right thoracotomy. The patient was markedly improved after surgery in terms of clinical symptoms and radiologic findings and discharged 18 days after surgery. Although quite rare, brocho-esophageal fistula can occur as a complication for long standing suppurative lung disease. It should be considered in cases where combined symptoms from suppurative lung diseases and upper GIT symptoms co exist.

Keywords Esophagus · Fistula · Surgery · Bronchi

Introduction

The term "esophago-respiratory fistula" is used to describe all fistulae between the bronchial tree and esophagus, the site is esophago-tracheal in 52–57 % of patients and esophago-bronchial in 37–40 % while in the remaining (3–11 %) communication is established peripherally through the lung parenchyma, forming an esophago-pulmonary fistula [1–3].

Congenital esophago-respiratory fistulae are usually diagnosed in neonatal period, but in few cases, they may remain silent till adulthood. Esophago-respiratory fistulae in adults are less commonly reported in the literature [4], when they occur they are mostly acquired in nature. Acquired causes of broncho-esophageal fistula include malignancies involving esophagus or adjacent structures. Benign conditions causing fistula are less common and consist of infections [5].

Case presentation

We report a 17-year-old girl with a history of cough, expectoration and recurrent attacks of fever for a long time. Her mother said she has had chest problems since early childhood. The mother described an uncertain history of foreign body inhalation at the age of 2 years. Medical treatment was repeatedly prescribed without significant improvement and her chest complaints have never stopped since then.

The condition of the patient was fluctuating over the last 15 years. However, fever, cough and expectoration increased over the last 2 years and she reported occasional coughing of some food particles. She was admitted to the chest department 2 years ago where CT chest revealed multiple cavitary lesions in the right lower lobe with mild esophageal dilatation. Intravenous antibiotics were administered and pulmonary resection was decided but the patient refused surgery.

Recently the patient asked for surgical consultation after exaggeration of her symptoms. Re-evaluation CT chest

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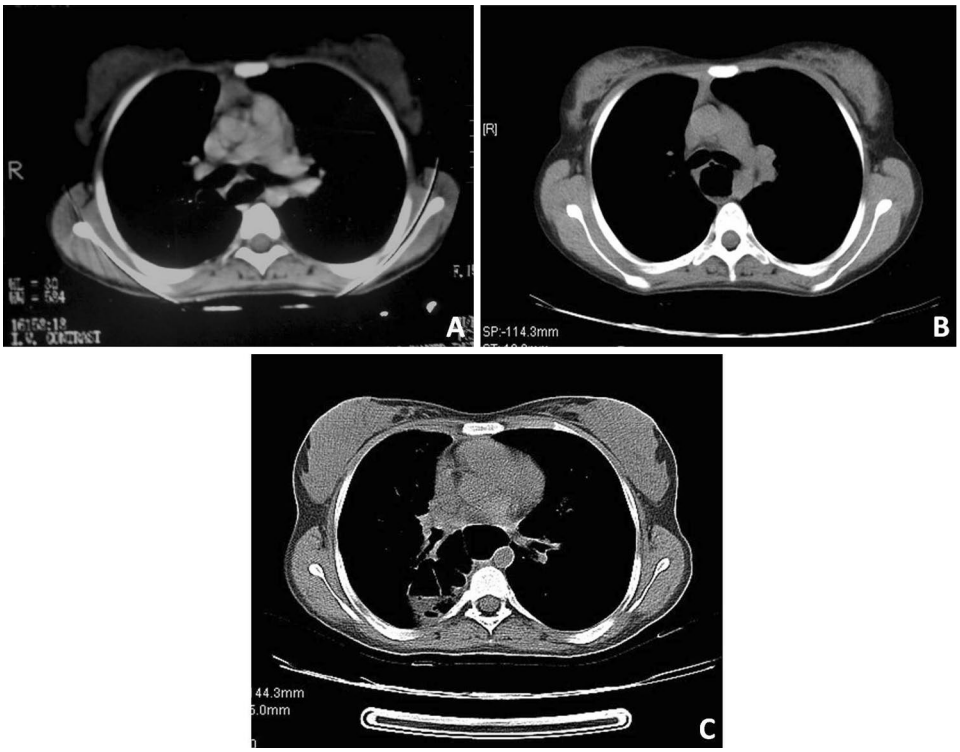

Fig. 1 CT chest. **a**, **b** Progressive dilatation of the esophagus as the cuts are going down. **c** Lower chest cut show fistulous communication between esophagus and right lower lobe with multiple RLL cavitary lesions (Power Point 2007 was used)

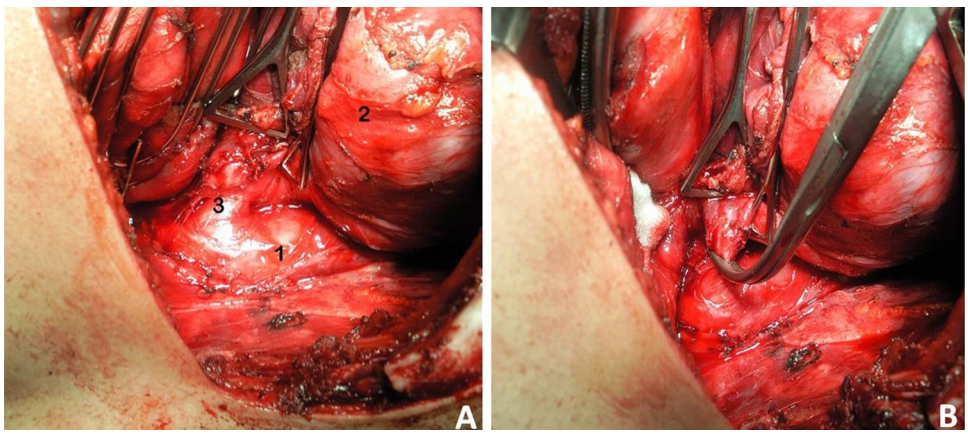

Fig. 2 Operative views. **a** Intra-operative view before excision of the fistula showing (*1*) dilated lower esophagus, (*2*) right lung lower lobe, (*3*) fistula. **b** Clamping of the fistula was done before excision to control repeated de-saturation (Power Point 2007 was used)

revealed marked dilatation of the esophagus with multiple cavitary lesions involving right middle and lower lung lobes and suspicious communication between right lower lobe parenchyma and the esophagus (Fig. 1). Virtual bronchoscopy turned out normal and other routine preoperative investigations revealed no abnormal findings apart from leucocytosis and anemia.

She was scheduled for pulmonary resection.

Unfortunately selective ventilation was not possible because of small size of the patient so anesthesia was conducted via single lumen endo-tracheal tube.

Preoperative rigid esophagoscopy was done after induction of anesthesia and intubation of the patient for a planned resection of the destroyed right lower lobe; it revealed hyperemic indurated area in the lower third esophagus with no definite hole. Upper endoscopy was terminated and we proceeded to lung resection surgery.

The approach was classic postro-lateral thoracotomy. The chest was accessed through the sixth space. Extensive adhesions around the lung were dissected. Meanwhile, multiple episodes of de-saturation happened and were managed by frequent jet ventilation. A 3-cm long and 2-cm wide fistula between the dilated esophagus and parenchyma of the right lower lung lobe was identified and clamped after which the patient oxygenation returned to normal (Fig. 2). The fistula between esophagus and lower lung lobe was excised and the esophageal wall was sutured in two layers with Vicryle 4-0. Two tubes were left and the chest was closed in layers.

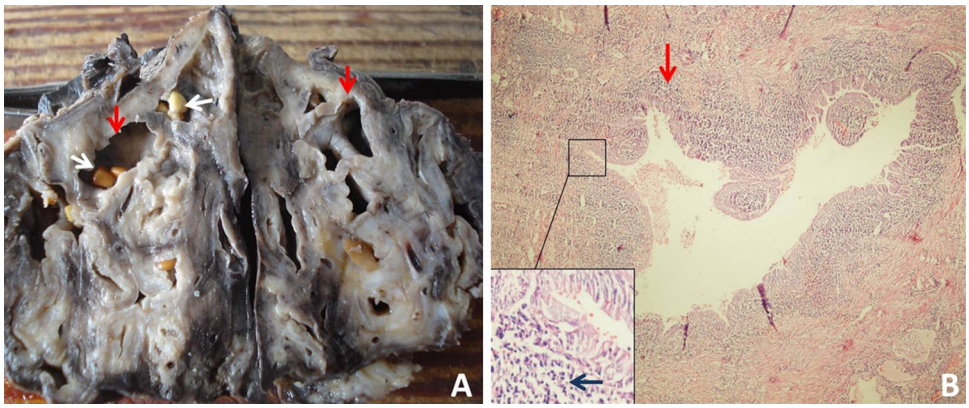

Fig. 3 Pathological features of resected lung lobes. Naked eye examination, **a** showed consolidation of the lung tissue with dilated bronchi (*red arrows*) containing food seeds (*white arrows*) on cut section. Histological evaluation, **b** showed dilated bronchi (main slide, *red arrow*) with infiltration of bronchial wall and surrounding lung tissues by acute and chronic inflammatory cells (inset, *blue arrow*). Magnification is ×40 for the main slide and ×200 for the inset

After surgery the patient was transferred to the ICU where she was on mechanical ventilation for delayed recovery from anesthesia. One day later, the patient was extubated and transferred to the ward. Postoperative X-ray revealed expansion of both lungs.

Nothing per mouth regimen was applied for 5 days and she had been given IV fluids. Oral feeding was allowed on the sixth-postoperative day; however, a small amount of food particles leaked into the chest tube. Gastrographin esophagography was done and revealed leakage of dye into the mediastinum with normal gastric filling. Postoperative oesophago-pleural fistula was confirmed and managed conservatively by stoppage of oral feeding, returning back to TPN, correction of hypo-albuminemia and meticulous follow-up of the chest drains.

Over the next 10 days, the amount of chest tube drainage decreased gradually and chest tubes became cleaner. The general condition of the patient markedly improved. She was gradually allowed to resume oral feeding and chest drains were removed on postoperative day 16 after no more leakage was observed with oral feeding. She was discharged on POD 17.

2 months later another CT scan of the chest was done and revealed reduction in esophageal dilatation with marked improvement of patient's general condition.

Pathological evaluation of the resected lung lobes showed diffuse consolidation of the lung parenchyma. On gross examination (Fig. 3a), there were multiple variable-sized dilatations of the large- and medium-sized bronchi, of which some have saccular appearance. The dilated bronchi contained fruit seeds and food particles. The covering pleura were thick, opaque and adherent. Microscopically, features of bronchiectasis were prominent (Fig. 3b). There were dilatations of bronchial lumina and thickening of bronchial walls as were the walls of surrounding blood vessels. The bronchial walls and surrounding lung parenchyma were heavily infiltrated by a mixture of acute and chronic inflammatory cells, predominantly lymphocytes, plasma cells and polymorphnuclear leucocytes. Hyperplasia, squamous metaplasia and mild to moderate squamous dysplasia of the bronchial epithelium were evident.

Discussion

Acquired esophago-pulmonary fistula has very rarely been reported in literature.

The course of the disease in this patient may be related to neglected foreign body inhalation complicated with bronchiectasis and chronic lung abscess. The infection and inflammation have extended to other segments and finally the whole lobe has become involved. Negligence of the patient's condition has led to extra-pulmonary extension of the inflammatory process so that the covering pleura and the nearby esophagus have become involved. Probably 2 or 3 years ago, a fistula between the esophagus and one of the cavities of the right lower lobe developed. This scenario explains simultaneous deterioration of the patient`s condition and appearance of new symptoms such as coughing of food particles and vomiting, with progressive dilatation of the esophagus.

Other scenarios cannot be excluded. A congenital esophago-pulmonary fistula with continuous passage of food particles to the right lower lung lobe is one possibility. However, the delayed presentation of GIT symptoms and the extreme rarity of such congenital anomaly would argue against it. Another possibility is the presence of oesophageal diverticulum that can easily fistulate into adjacent lung parenchyma assisted by inflammatory process in any of them. The long clinical history of pulmonary manifestation

and absence of any initial gastro-esophageal symptoms would make this scenario remote.

Taken collectively and considering the available clinical history and investigations, we are convinced that the condition is primarily a suppurative lung disease complicated with esophago-pulmonary fistula.

Treatment options were confined to operations on the pulmonary side, looking to the fact that a diseased lobe has to be removed. Placement of esophageal stents or bypass surgery would be a good option if the patient was not candidate for lung resection.

Conclusion

Although pulmonary esophageal fistula is extremely rare it should be suspected whenever combined pulmonary and gastrointestinal symptoms develop. Early surgical management always provides the best chance for the patients.

Ethical Statement Our work conforms to the guidelines set forth in the Helsinki Declaration of 1975, as revised in 2000, concerning Human and Animal Rights. Informed consent was obtained from the patient included in this study.

Conflict of interest Author Abdelrahman Adel Abdelaziz Elsayed, Author Essam Elbadry Hashim Mohamed and Author Ahmed R. H. Ahmed have no conflict of interests.

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