Türk Göğüs Kalp Damar Cerrahisi Dergisi 2012;20(3):645-648
doi: 10.5606/tgkdc.dergisi.2012.127

*Case report / Olgu Sunumu*

# An incidentally diagnosed bronchoesophageal fistula in an adult

*Erişkin bir hastada tesadüfen saptanan bronkoözofageal fistül*

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Congenital bronchoesophageal fistulas (BEFs) are usually diagnosed during the neonatal period. Only simple types (type 2) may remain undiagnosed until adulthood. Congenital BEFs may remain hidden if the orifice of the fistula is closed with a permanent membrane, and/or the esophageal orifice is below the level of the bronchial orifice. Bronchoesophageal fistulas should be treated as soon as the diagnosis is confirmed with early surgical intervention. Despite their benign nature, congenital BEFs may lead to fatal complications, for example massive hemoptysis. In this article, we present a 55-year-old female patient with a congenital BEF. The fistula was first suspected during an inguinal hernia operation. Her mild symptoms became worse following the diagnosis, and hemoptysis developed. A fistulectomy using a right thoracotomy and right lower lobectomy were then performed.

*Key words:* Bronchoesophageal fistulas; hemoptysis; lobectomy; thoracotomy.

Doğuştan bronkoözofageal fistüller (BEF) genellikle yenidoğan döneminde tespit edilir. Sadece basit tip (tip 2) fistüller erişkin çağa kadar tanı konulmadan kalabilir. Doğuştan BEF'ler, fistül ağzının membran ile kapalı olduğunda ya da bronşiyal orifis seviyesi özofageal orifisin üzerinde olması gibi durumlarda uzun zaman gizli kalabilir. Bronkoözofageal fistüller tanı konulduktan sonra derhal cerrahi girişim ile tedavi edilmelidir. Doğuştan BEF'ler, iyi huylu olsalar da masif hemoptizi gibi hayatı tehdit eden komplikasyonlara da yol açabilirler. Bu yazıda 55 yaşında, doğuştan bronkoözofageal fistülü olan bir kadın hasta sunuldu. Bronkoözofageal fistül şüphesi, ilk kez kasık fıtığı ameliyatı sırasında gelişti. Hastanın önceden hafif olan semptomları tanısal işlemler sonrasında kötüleşmeye başladı. Daha sonra sağ torakotomi ile fistülektomi ve sağ alt lobektomi ameliyatı yapıldı.

*Anahtar sözcükler:* Bronkoözefageal fistül; hemoptizi; lobektomi; torakotomi.

Bronchoesophageal fistulas (BEFs) may be congenital or acquired. Congenital lesions always clinically manifest in early infancy. Braimbridge and Keith[1] classified congenital BEFs into four types (Table 1), and they may remain occult until adulthood if the fistula between the respiratory track and esophagus is simple (type 2). These types of BEF are always associated with repeated mild respiratory infections and coughing episodes after drinking or eating (Ohno's sign). In some cases, BEFs can be diagnosed incidentally in adulthood, as was the case with this patient.

## CASE REPORT

A 55-year-old female patient underwent an inguinal hernia operation and was suspected of having a BEF during the anesthesia. The operation was terminated, and the patient was extubated urgently because of hypoxia and swelling of the stomach due to the mechanical ventilation. A bronchoscopy revealed inflammation in the right lower lobe bronchus, but no fistula was detected. An endoscopic examination revealed a fistula orifice at the 22$^{nd}$ cm of the esophagus. Barium swallow esophagography showed a fistula between the

Available online at
www.tgkdc.dergisi.org
doi: 10.5606/tgkdc.dergisi.2012.127
QR (Quick Response) Code

*Received:* July 15, 2009 *Accepted:* February 23, 2010

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**Table 1. Braimbridge and Keith's classification of bronchoesophageal fistula**

| | |
|---|---|
| Type 1 | Congenital bronchoesophageal fistula associated with oesophageal diverticulum |
| Type 2 | Simple bronchoesophageal fistula |
| Type 3 | Bronchoesophageal fistula with an intralobar cyst |
| Type 4 | Bronchoesophageal fistula communicating with a pulmonary sequestration |

inferior part of the esophagus and the right lower lobe (Figure 1a). The patient rejected immediate surgical treatment for the BEF as she had no serious complaints. However, during the diagnostic process, coughing bouts accompanied by severe hemoptysis were observed. Her physical exam, chest X-ray, and blood tests, which showed normal values at the beginning, got worse, and submassive hemoptysis (200 cc/per day) occurred (Figure 2). Three months after the diagnosis, the patient underwent a right posterolateral thoracotomy. Subsequently, a fistulectomy and primary closure of the esophageal tract with interrupted 4/0 polyglactin along with a right lower lobectomy were performed. Almost the entire inferior lobe was destroyed, and the pleural thickenings on the diaphragmatic and parietal surface of the pleura were detected. The patient was monitored in the intensive care unit (ICU) for two days. Oral feeding was started on the fifth day following a barium esophagoraphy performed for surveillance (Figure 1b), and the patient was discharged on the seventh day following the surgery. A pathological examination of the resected parenchyma and fistula revealed no evidence of malignancy, tuberculosis, or other infections. Her postoperative course was symptom-free and uneventful during the 18 months of follow-up.

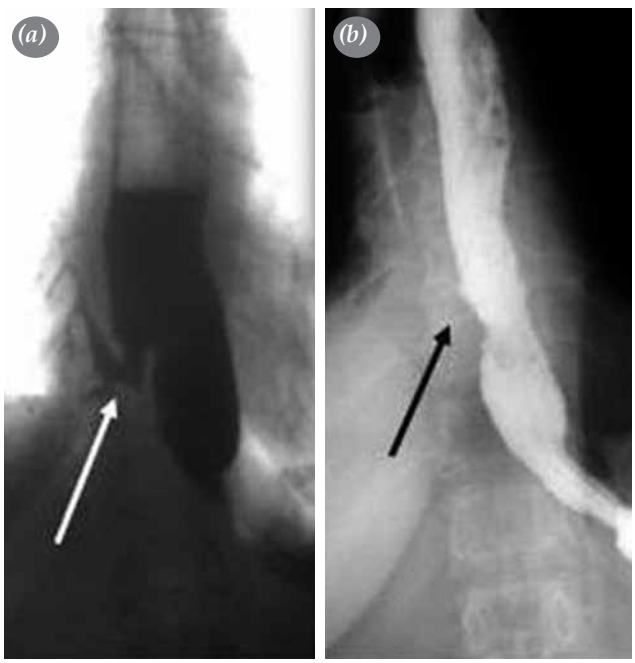


**Figure 1. (a)** Preoperative barium swallow esophagography revealed a fistula directed downward between the inferior part of the esophagus and right lower lobe. **(b)** Postoperative barium esophagography of the patient showed that the esophagus was functioning normally.

## DISCUSSION

While BEFs in adults usually have a malignant origin, the majority of benign cases are generally linked to trauma and infectious causes. On the other hand, congenital BEFs are very rare.[2-3] Type 2 BEF (simple type), which may remain occult until adulthood, usually consists of a short tract running directly from the esophagus to the bronchus.[1] This type happens to be the most prevalent and comprises almost 90% of the cases.[4] Fistulas are three times more common on the right side.[3] The most frequent localization of the simple type is a connection between the right lower lobe and the lower third of the esophagus. Male and female patients are equally affected with the highest incidence occurring in the third decade of life.[4]

Our patient had a type 2 BEF between the right lower lobe and lower third of the esophagus with typical localization that was hidden until the fistula was triggered by mechanical ventilation. Pathologically, congenital BEF is suggested by the absence of surrounding inflammation[3] and adherent lymph nodes along with the presence of a mucosa and a definitive muscularis mucosa within the fistulous tract.[4] Surgically, uncomplicated and easy dissection of the surrounding area of the fistula by the absence of inflammation suggests a congenital fistula.[5] The pathological and surgical aspect of this case favored a congenital BEF because no adherent lymph nodes or inflammation were detected.

Presenting symptoms detailed in a review by Azoulay[6] included coughing (96%), whereas frequent respiratory infections and coughing when swallowing liquids (Ohno's sign) were reported to be pathognomonic for this condition and present in 65% of the cases. Hemoptysis is an infrequent, but serious complaint which leads to urgent treatment. In our patient's history, mild respiratory infections and Ohno's sign had been present for an extended period. However, hemoptysis had only recently occurred as a life-threatening complication of the BEF.

In English literature, about 100 cases of congenital BEF have been reported, and most of these were case reports. The largest review of the literature was done by Risher[4] and included 100 cases, with 25% of these

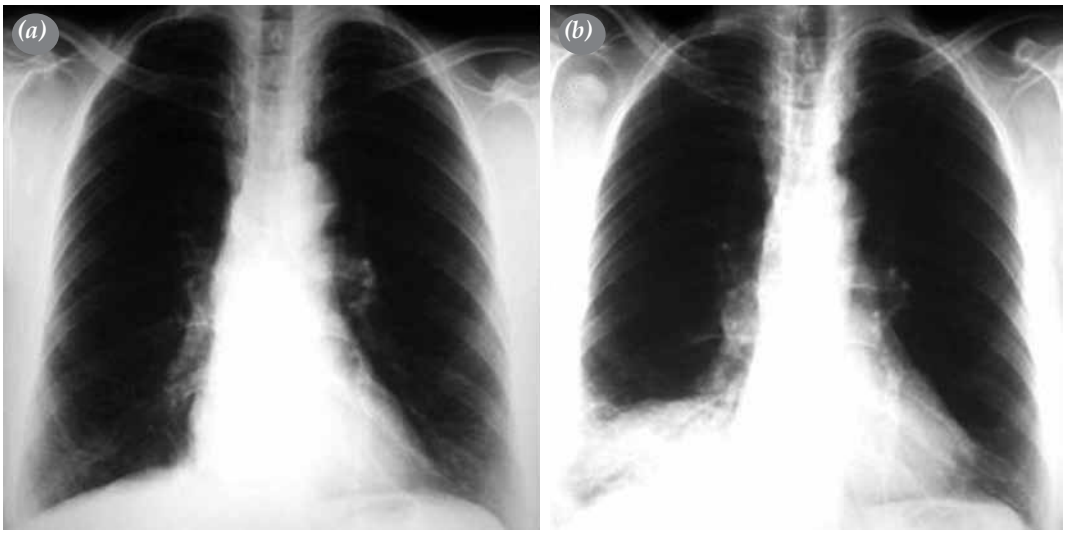


**Figure 2. (a)** A chest X-ray of the patient before the diagnosis taken prior to surgery for an inguinal hernia is shown. **(b)** A chest X-ray of the patient three months after the diagnosis (before the right thoracotomy) is shown.

being children. The current literature mainly consists of individual cases. We report our case to contribute to the literature about this rare topic.

Normally, the treatment of a congenital BEF includes the division of the fistula and repair of the esophageal defect. The decision on whether to resort to a surgical approach for bronchial repair or lung resection depends on the coexistence of pulmonary disease.[7,8] Obliteration of the fistula with synthetic materials such as BioGlue or silver nitrate may be an alternative treatment in patients whom the prognosis for surgery is poor.[9] The surgical treatment via a thoracotomy yields excellent results.[2-8] To prevent refistulization, reinforcement of the suture line with a pleural or muscular flap is usually recommended.[5,10] In our case, a right lower lobectomy was performed because of a destroyed lung, and a pleural flap was used to support the esophageal orifice of the BEF. In our opinion, an anatomic resection should be used to prevent refistulization and infection of the pleural cavity.

One possible cause for a delay in the onset of symptoms and diagnosis is the presence of a membrane closing the fistula's orifice that may subsequently rupture during mechanical ventilation. This is similar to what we observed in our case. In addition, a spasm of the smooth muscle in the wall of the fistula or closing of the esophageal orifice during swallowing may mask the presentation.[1,4] Another hypothesis is that the level of the bronchial orifice of the fistula is over the esophageal orifice, but this was not the case for our patient as her BEF showed a downward direction from the esophagus to the bronchus.

Diagnosing a fistula may be difficult when the typical signs and symptoms are not present. A barium esophagram usually confirms the diagnosis and should be chosen as the initial study of choice. An esophagoscopy or bronchoscopy may not always demonstrate the fistulous orifice. Computed tomography can be utilized to rule out the presence of lymphadenopathy and/or a malign lesion while also defining the extent of coexisting pulmonary disease, which may require resection.

Bronchoesophageal fistulas must be treated surgically following their diagnosis. Even if the lesion is benign and asymptomatic, it may lead to serious complications such as hemoptysis and a destroyed lung. Treatment via a thoracotomy, a fistulectomy, or, when needed, a lung resection offer excellent results.

### Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

### Funding

The authors received no financial support for the research and/or authorship of this article.

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