CASE REPORT

Acquired esophagobronchial fistula without Ono's sign and with unusual cause

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SUMMARY

A 60-year-old woman presented with dyspnoea and respirophasic chest discomfort, as well as a history of idiopathic oesophageal diverticulum. Physical examinations showed no evidence of Ono's sign, fever and weight loss. Chest radiograph revealed a right-sided transudative pleural effusion. Barium oesophagogram made a diagnosis of acquired esophagobronchial fistula communicating between oesophagus and bronchus. The oesophagobronchial fistula, causing pleural effusions, was very small and could be caused by idiopathic oesophageal diverticulum. The pleural effusion was removed by thoracentesis, which improved her symptoms. Surgical therapy or covered oesophageal stenting was advised, but she declined. She is followed-up regularly on an outpatient basis.

BACKGROUND

The majority of acquired esophagobronchial fistulas (EBFs) are due to chest or oesophageal carcinoma. There are also a variety of non-malignant causes of acquired EBFs. However, a case of a recurrent pleural effusion had no evidence of these causes and common EBF's symptoms, making it very difficult to diagnose EBFs. This case shows that although there are no common symptoms and causes of EBF, barium oesophagogram is needed for the exact and fast diagnosis of EBF in the patient with dyspnoea and respirophasic chest discomfort due to the pleural effusion.

CASE PRESENTATION

In September 2009, a 60-year-old woman was presented with dyspnoea and respirophasic chest discomfort. She had a history of idiopathic oesophageal diverticulum diagnosed by annual medical check in May 2003. Physical examinations showed no paroxysms of Ono's sign (coughing caused by swallowing fluids), fever and weight loss. The chest radiograph revealed small amount of fluid accumulation on the right side of the pleural cavity. Seven days after a diuretic treatment, the radiograph showed no accumulation fluid in the pleural cavity. She improved with the diuretic therapy and was discharged.

In October 2010, the patient was readmitted with continued respirophasic chest discomfort and a history of 2-week dyspnoea. Her pulse (100 bpm) and body temperature (36.9°C) were normal. The chest radiograph showed a right-sided pleural effusion (transudate defined by aspirated fluid analysis; figure 1) but no evidence of other abnormalities. After the diuretic therapy, the chest radiograph

Figure 1 Posteroanterior chest radiograph before thoracentesis showing a right-sided transudative pleural effusion (arrow).

still showed the transudative pleural effusion. Thoracentesis was performed and 700 mL transudative fluid was removed (figure 2). She improved after thoracentesis and was discharged. However, the cause of the transudative pleural effusion was unknown at that time and she was readmitted with dyspnoea and pleural effusion in May 2011.

Figure 2 Posteroanterior chest radiograph after thoracentesis showing that the transudative pleural fluid was removed (arrow).

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INVESTIGATIONS

Frequently reported symptoms of the transudative pleural effusion include dyspnoea and respirophasic chest discomfort.¹ Liver cirrhosis, heart failure, nephrotic syndrome or urinothorax was suspected because the transudative pleural effusion can be typically caused by these diseases.¹ Laboratory test results and echocardiograms revealed no sign of these diseases and infections by bacteria, fungi and viruses. Sputum cytology for lung cancer screening and blood cancer markers were negative. There was no cavity visible in the chest radiograph and tuberculosis diagnosis tests were negative. Leucocytes ($6.84 \times 10^9/L$), erythrocyte sedimentation rate (11 mm/h), and C reactive protein level (1.5 mg/L) were normal. These results indicated that there was no carcinoma, inflammation or infection.

DIFFERENTIAL DIAGNOSIS

The only key to make the diagnosis of EBF is the history of idiopathic oesophageal diverticulum diagnosed by annual medical check in May 2003. Therefore, a barium oesophagogram was obtained. The barium oesophagogram revealed only an acquired fistula (EBF; figure 3) communicating between oesophagus and bronchus. Barium was filling the right bronchial tree from the oesophagus (figure 4). There was no evidence of the idiopathic oesophageal diverticulum diagnosed in 2003 and other fistulas (eg, oesophagopleural and bronchopleural fistulas causing pleural effusions^{2 3}).

OUTCOME AND FOLLOW-UP

The transudative pleural effusion was removed by thoracentesis, which dramatically improved her symptoms. Surgical therapy or covered oesophageal stenting was planned, but she declined. She is followed up regularly on an outpatient basis.

DISCUSSION

The common symptoms of EBF are coughing (Ono's sign), fever and/or weight loss.^{4 5} However, the patient did not show these symptoms. Therefore, we did not suspect EBF at first. This

Figure 3 Barium oesophagogram showing an esophagobronchial fistula (arrow) from the oesophagus.

Figure 4 Barium oesophagogram showing the fistula from the oesophagus (arrow) and that barium was filling the right bronchial tree (arrowhead), showing the communication of the oesophagus with the right bronchus.

acquired EBF might be associated with the transudative pleural effusion because there are no common causes of the effusion and a similar result was previously reported.⁶ The EBF is so small (<0.3 cm in diameter) that there may be no sign of inflammations or infections, as previously reported.⁵ Acquired EBF is a rare disorder with a variety of causes. The majority (approximately 80%) acquired EBFs are due to chest or oesophageal carcinoma.⁴ Non-malignant causes of acquired EBFs include chronic infections (eg, tuberculosis), radiation injury, postsurgical lesions, local injury (trauma or poisoning), broncholithiasis and Barrett's ulcer.⁴ However, the patient had no evidence of these causes. In this case, only one possible cause of the EBF could be the oesophageal diverticulum because (1) the oesophageal diverticulum could cause acquired EBFs in a few reports^{4 5 7} and (2) the EBF was detected at the place where the previously diagnosed oesophageal diverticulum had been disappeared. The chronic nature of the process whereby the small EBF remains localised might partially explain why the patient had no Ono's sign. Other reports support this notion.^{8 9}

Although a surgical cure of EBF was considered, the patient's emphatic refusal of it prevented this practice. She was discharged due to her symptomatic relief by thoracentesis. As no non-surgical therapy is available for the cure of the EBF,¹⁰ these

Learning points

- ▶ Our patient with recurrent pleural effusion had no evidence of common esophagobronchial fistula's (EBF's) symptoms (eg, Ono's sign, fever and/or weight loss), making it very difficult to diagnose EBFs.
- ▶ Further evaluation elucidated that only one possible cause of the EBF could be the oesophageal diverticulum, which is not a common cause of EBF.
- ▶ Although there are no common symptoms and causes of EBF, barium oesophagogram is needed for the exact and fast diagnosis of EBF in patients having dyspnoea and respirophasic chest discomfort due to the pleural effusion.

continued drainages should be replaced with a surgical remediation to treat the EBF causing her recurrent pleural effusion. Although the patient outcome cannot be addressed at this time, she has been encouraged to take (1) the surgical therapy (oesophageal resection and fistula closure) for achieving definitive repair of the EBF or (2) the covered oesophageal stenting for the treatment of this condition in the patient refusing surgery.

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