CASE REPORT

Tuberculosis presenting as broncho-oesophageal fistula in a young healthy man

Ahmed Sayeed,¹ Eid Humaid Alqurashi,² Adnan B Alzanbagi,³
Nabil Abdulwadod Badr Ghaleb¹

¹Department of Pulmonology,
King Abdullah Medical City,
Mecca, Saudi Arabia

²Department of Pulmonology,
King Abdullah Medical City,
Mecca, Saudi Arabia

³Department of
Gastroenterology, King Abdullah
Medical City, Mecca, Saudi
Arabia

Correspondence to

Dr Ahmed Sayeed,
sayeedheena@gmail.com,
sayeedmodi@yahoo.co.in

Accepted 15 July 2017

SUMMARY

A 21-year-old Saudi man presented with a history of dysphagia and choking. CT scan of the chest showed clear evidence of chronic recurrent aspiration pneumonia in the left lung. It also showed a fistula connecting the left main bronchus to the oesophagus. Endoscopy showed clear opening on the oesophageal side. Bronchoscopy also confirmed the presence of a broncho-oesophageal fistula on the left bronchial side with the presence of secretions on swallowing. Bronchoalveolar lavage (BAL) was done and sent for mycobacterial tuberculosis culture. The fistula was closed with clips under endoscopic guidance, which alleviated his symptoms of dysphagia and choking. The BAL culture grew mycobacterial tubercle bacilli. The patient showed marked improvement after starting antitubercular therapy and was discharged to be followed up in the clinic.

individuals. Hence, we present this as the case report.

CASE PRESENTATION

A 21-year-old Saudi young man presented to us with a history of dysphagia, choking and shortness of breath since 1 month. The dysphagia was initially to solids and liquids and was associated with odynophagia. Later he developed dysphagia to only liquids associated with choking sensation on swallowing any liquids. At the age of 9 years, he had a history of swallowing a foreign body, but it was passed spontaneously in stools. He had a history of significant weight loss of 5 kg in 1 month and also a positive history of contact with a patient with tuberculosis. On examination, his vital signs were stable and chest was clear.

INVESTIGATIONS

The fistula was suspected based on history and some findings of aspiration pneumonia in chest X-ray. CT scan of the chest confirmed the findings of recurrent aspiration pneumonia in the form of chronic aspiration changes noted at the basal segments of the left lower lobe in a form of mild bronchiectasis and sub-segmental atelectatic plates with tiny nodules also noted at the posterior basal segment of the left lower lobe. There were tree-in-bud air space opacities seen in the base of the left upper lung lobe, superior segment left lower lobe and outer aspect of the lingula. There are also enlarged subcarinal lymph nodes that were matted together, the largest nodes are showing central necrosis (caseation), and the largest one measures about 22 mm in diameter (figure 1A). Also noted were enlarged small mediastinal lymph nodes in the right paratracheal and superior mediastinum showing central necrosis with a diameter of 12 mm (figure 1B).

There was evident oesophagobronchial fistula communicating the oesophagus to the origin of the left main bronchus. The fistula was located at the mid-third oesophagus 11 cm proximal to the gastro-oesophageal junction and at level opposite to D5 vertebral body (figure 1C,D).

Based on the CT findings of multiple enlarged matted lymph node with central caseation and also the presence of fistula, the radiologist gave an opinion that the overall picture is suggestive of pulmonary tuberculosis with mediastinal lymph nodes complicated by oesophagobronchial fistula.

BACKGROUND

The most common cause of oesophagorespiratory fistula is malignancy and trauma in adults. Tracheo-oesophageal fistula is more common especially in immunocompromised patients. Oesophagobronchial fistula due to tuberculosis is rare and even rarer in young healthy immunocompetent

Figure 1 (A, B) Multiple enlarged, matted subcarinal lymph nodes with central caseation and (C, D) broncho-oesophageal fistula communicating the oesophagus to the origin of the left main bronchus.

CrossMark

To cite: Sayeed A, Alqurashi EH, Alzanbagi AB, et al. *BMJ Case Rep* Published Online First: [please include Day Month Year]. doi:10.1136/bcr-2017-220821

Figure 2 Presence of frothy secretions (A), small opening in the left main bronchus (B), fistula on the oesophageal side (arrow in C) and clipped fistula (D).

Based on these findings, bronchoscopy was done, which showed a small opening in the left main bronchus (figure 2B) and the presence of frothy secretions on swallowing (figure 2A). There was also a bulge at the beginning of the right main bronchus with whitish mucosa covering its surface. Bronchoalveolar lavage was done and sent for tuberculosis culture.

CT scan of the chest showed lesions consistent with tuberculosis. Therefore, we had already achieved reasonable diagnosis. Hence, we did not feel it was necessary to take another biopsy as it might worsen the fistula, create a new fistula or may cause pneumothorax as the mucosa was very friable. He later underwent upper gastrointestinal endoscopy, which confirmed the presence of fistula on the oesophageal side about 25 cm from incisor in the oesophagus (arrow in figure 2C). The fistula was clipped (figure 2D) and dysphagia remarkably improved. The bronchoalveolar lavage culture turned out to be positive for *Mycobacterium tuberculosis* and sensitive to all first-line antitubercular therapies. He was screened for HIV and other infectious diseases which were negative.

TREATMENT

He was kept nil per oral and was started on total parental nutrition. Based on history and CT findings suggestive of tuberculosis and PPD(purified protein derivative) test of 13 mm, he was started on empirical antitubercular therapy with four first-line drugs. After 20 days, he developed pain in the abdomen and vomiting, and liver function tests showed raised ALT(alanine aminotransferase) 595 and AST(aspartate aminotransferase) 739. INH(isoniazid), rifampicin and pyrazinamide were stopped as the patient developed drug-induced hepatotoxicity. Second-line drugs for tuberculosis, amikacin and moxifloxacin, were started. After 10 days, his symptoms subsided and liver function tests became normal. He was rechallenged with INH and rifampicin, and this time he tolerated them well without any evidence of hepatotoxicity. Amikacin was stopped and discharged with INH(isoniazid) 300, ethambutol 800, moxifloxacin 400 and pyridoxine 40. He is being followed up in the clinic.

OUTCOME AND FOLLOW-UP

This young man improved clinically and was discharged to be followed up in the clinic.

We are planning to perform repeat endoscopy after completing 6 months of antitubercular therapy.

DISCUSSION

The anomalous connection between the oesophagus and the respiratory tract, most commonly the trachea or bronchus, is known as esophagorespiratory fistula (ERF). In adults, the usual common cause of this ERF is the invasion of malignancy or trauma due to intubation.¹ Infections like Candida, cytomegalovirus, tuberculosis and Varicella zoster, especially in immunocompromised cases like HIV, can cause ERF.²⁻⁴ Congenital fistulas are rare in adults as they usually present during the neonatal period. Many cases reported in literature suggest that broncho-oesophageal fistula due to tuberculosis is usually associated with extensive pulmonary or disseminated tuberculosis. In broncho-oesophageal fistula due to tuberculosis, the mediastinal lymph nodes are involved first with inflammation in and around these enlarged lymph nodes, which then spread to neighbouring structures or organs particularly the oesophagus, trachea near its bifurcation and the bronchus resulting in perioesophagitis and peritracheitis. If untreated, necrosis and caseation can occur in the lymph nodes with local abscess formation and subsequently rupture into the oesophagus, trachea or mainstem bronchi resulting in a fistula. Our patient's CT scan of the chest showed multiple enlarged, matted lymph nodes with central caseation close to carina and the left main bronchus, which could have resulted in the broncho-oesophageal fistula. It is rare to find mediastinal tuberculous lymphadenopathy directly resulting in broncho-oesophageal fistula without extensive lung parenchyma involvement as in our case.⁵⁻⁷

This fistulous tract can lead to repeated episodes of aspiration pneumonia, which can be life threatening. Therefore, it is very important to detect and treat broncho-oesophageal fistula. Barium oesophagography can demonstrate fistula tract. However, endoscopy is the investigation of choice as it allows direct visualisation of the broncho-oesophageal fistula.¹⁸

CT can be helpful in detecting tracheo-oesophageal fistula and associated mediastinal lymphadenopathy, although smaller size fistulas can be missed.^{9 10}

CT scan of the chest in our case showed multiple mediastinal lymph nodes with central caseation (figure 1A,B) and clear fistula communicating the left main bronchus to the oesophagus (figure 1C,D) along with tree-in-bud opacities in the basal segments of the left upper lobe. Based on these findings, the radiologist concluded that the fistula is due to tuberculosis. Our diagnosis was confirmed with bronchoalveolar lavage culture, which was positive for tuberculosis by PCR and sensitive to all first-line antitubercular drugs.

The treatment of tracheo-oesophageal fistula depends on the size and the aetiology. The larger fistulas may require surgical repair, whereas smaller ones can be treated by endoscopy-guided clipping of the fistula. Some cases might require Ryle's tube feeding or PEG(percutaneous endoscopic gastrostomy) tube feeding until the fistula is healed. In case of advanced malignancy, palliative treatment in the form of insertion of an endoscopy-guided oesophageal stent can alleviate symptoms and improve the survival rate.¹¹⁻¹⁴

However, our case of ERF was treated effectively by upper gastrointestinal endoscopy-guided clipping of the fistula combined with antitubercular chemotherapy without the need for surgical intervention.¹⁵

Learning points

- ▶ Tuberculosis can rarely present with extrapulmonary manifestation like broncho-oesophageal fistula.
- ▶ High index of suspicion is needed to detect and treat broncho-oesophageal fistula of tuberculous ethology.
- ▶ Extrapulmonary tuberculosis like tuberculous broncho-oesophageal fistula can very rarely affect young and healthy individuals.
- ▶ Endoscopy-guided clippings can result in instant alleviation of symptoms like dysphagia and choking.
- ▶ Antitubercular therapy can cause permanent cure for broncho-oesophageal fistula secondary to tuberculosis.

Acknowledgements we authors acknowledge Dr Ibrahim Shalabi for selecting CT images

Contributors AS has contributed in planning, conduct, reporting, conception and design, acquisition of data or analysis and interpretation of data. EHA has contributed in planning, conduct, reporting, conception and design. ABA has contributed in conception and design. NABG has contributed in conception and design.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

© BMJ Publishing Group Ltd (unless otherwise stated in the text of the article) 2017. All rights reserved. No commercial use is permitted unless otherwise expressly granted.

REFERENCES

- 1 Diddee R, Shaw IH. Acquired tracheo-oesophageal fistula in adults. *Continuing Education in Anaesthesia, Critical Care & Pain* 2006;6:105–8.
- 2 Dieterich DT, Wilcox CM. Diagnosis and treatment of esophageal diseases associated with HIV infection. Practice Parameters Committee of the American College of Gastroenterology. *Am J Gastroenterol* 1996;91:2265–9.
- 3 Hirano I, Richter JE. Practice Parameters Committee of the American College of Gastroenterology. ACG practice guidelines: esophageal reflux testing. *Am J Gastroenterol* 2007;102:668–85.
- 4 Couraud L, Ballester MJ, Delaisement C. Acquired tracheoesophageal fistula and its management. *Semin Thorac Cardiovasc Surg* 1996;8:392–9.
- 5 Sasaki M, Mochizuki H, Takahashi H. A bronchoesophageal fistula that developed shortly after the initiation of antituberculous chemotherapy. *Intern Med* 2013;52:795–9.
- 6 Cooperman R, Paragi PR, Shah S. Adult congenital bronchoesophageal fistula. *Thorac Cardiovasc Surg* 2007;55:334–6.
- 7 Narayanan S, PV S, Majeed K A A, et al. Tuberculosis presenting as bronchoesophageal fistula. *IDCases* 2017;8:19–21.
- 8 Ahn JY1, Jung HY, Choi JY, et al. Benign bronchoesophageal fistula in adults. Endoscopic closure as primary treatment. *Gut Liver* 2010;4:508–13.
- 9 Im JG, Kim JH, Han MC, et al. Computed tomography of esophagomediastinal fistula in tuberculous mediastinal lymphadenitis. *J Comput Assist Tomogr* 1990;14:89–92.
- 10 Williford ME, Thompson WM, Hamilton JD, et al. Esophageal tuberculosis: findings on barium swallow and computed tomography. *Gastrointest Radiol* 1983;8:119–22.
- 11 Marchese R, Poidomani G, Paglino G, et al. Fully covered self-expandable metal stent in tracheobronchial disorders: clinical experience. *Respiration* 2015;89:49–56.
- 12 Mizobuchi S, Kuge K, Maeda H, et al. Endoscopic clip application for closure of an esophagomediastinal-tracheal fistula after surgery for esophageal cancer. *Gastrointest Endosc* 2003;57:962–5.
- 13 Manca S, Fois AG, Santoru L, et al. Unusual clinical presentation of thoracic tuberculosis: the need for a better knowledge of illness. *Am J Case Rep* 2015;16:240–4.
- 14 Aggarwal D, Mohapatra PR, Malhotra B. Acquired bronchoesophageal fistula. *Lung India* 2009;26:24.
- 15 Joshi JM, Gothi D. Post infectious obliterative bronchiolitis. *Lung India* 2006;23:75.

Copyright 2017 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow