

hospital³. Childhood deprivation and antisocial personality traits may be associated features. Wherever possible, patients should be referred for psychiatric consultation. However, successful therapy for patients with Munchausen syndrome is extremely difficult given their penchant for wandering and taking their own discharge when confronted. As such, early recognition is crucial in order to avoid time and expenditure being

wasted on potentially harmful investigations and treatments in these very willing patients.

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An Unusual Case of Oesophagopleural Fistula

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Summary

In 1985, a 12-year-old patient with pulmonary tuberculosis developed a pneumothorax. It was left untreated because he did not agree to chest tube insertion. After completion of anti-tuberculous therapy the patient failed to come for follow-up. Nine year later he presented with reactivation of tuberculosis and a pyopneumothorax. Chest tube drainage was instituted but rice was seen emerging from the tube and radiological investigations showed an oesophagopleural fistula. The patient refused any surgical procedure and was treated with conventional anti-tuberculous chemotherapy. His symptoms have resolved and the fistula appears to have healed with conservative management.

Key Words: Oesophagopleural fistula, Tuberculosis

Introduction

Acquired oesophagopleural fistulae are fortunately uncommon, but may occur as a complication of empyema or thoracic surgery¹. The finding of food particles in the chest drainage tube, although extremely distressing to both patient and physician, is a key pointer to the diagnosis². Reassuringly, it appears that such fistulae may be satisfactorily managed with non-surgical therapy, as is illustrated by our fascinating case.

Case Report

Our patient was 12-years-old when he first presented to the Chest Clinic in 1985 with a 2 month history of fever, cough and breathlessness. He was found to be smear positive for acid fast bacilli and was treated on a fully supervised regime of streptomycin, isoniazid, rifampicin and pyrazinamide for the first two months. A chest radiograph at the end of this phase showed that he had developed a left sided pneumothorax. Chest tube drainage was suggested but the family refused the procedure and the patient subsequently defaulted treatment. A few months later, he returned to the clinic and this time, successfully completed two months of induction anti-tuberculous treatment and 7 months of consolidation treatment with biweekly streptomycin, rifampicin and isoniazid. Throughout this time the pneumothorax remained untreated and the patient again defaulted follow up after treatment was completed. He was then sputum smear negative for acid fast bacilli as well as culture negative. Nine years later he presented with fever, cough and haemoptysis of a weeks' duration. There was no history of dysphagia or of cough with ingestion of fluids. Clinical examination showed him to be clubbed and cachexic. Sputum proved to be smear positive for acid fast bacilli and the same anti-tuberculous regime was started. Subsequently culture grew *Mycobacterium tuberculosis* which was sensitive to the conventional drugs in the drug regimen. Chest tube drainage was instituted after needle aspiration confirmed the presence of pus in the left hemithorax.

Pus continued to drain from the chest for over a month and it was then noted that particles of rice were coming out of the tube. A barium meal was carried out and this confirmed an oesophagopleural fistula which extended cutaneously due to the presence

of the closed tube thoracostomy. Various surgical options were proposed to the patient but he refused any surgical intervention. The chest tube was left *in situ* for a further 6 weeks with continued drainage of food particles, but the tube had to be removed when the patient took his own discharge from hospital. He was fitted with a "colostomy bag" over the cutaneous fistula exit site and continued to receive fully supervised anti-tuberculous treatment for a total period of 12 months. Serological testing for the human immunodeficiency virus was repeatedly negative.

On follow up several months after completion of therapy, the patient remained symptom free and had repeatedly negative bacteriological tests. The radiological signs of active tuberculosis at the apex of the right lung had improved. There was no longer any discharge from the fistula exit site and the skin wound had healed. Unfortunately, the patient refused to undergo further radiological contrast investigations, saying that he now felt well. Nonetheless, the clear signs of clinical improvement, as well as the absence of symptoms, were consistent with the conclusion that the fistula had healed.

Discussion

In a review of oesophagopleural fistulae, 35 cases which occurred as a complication of thoracic empyema were found in the literature from 1890 to 1960 and about a third of these patients were suffering from pulmonary tuberculosis¹. The authors were of the opinion that fistulae related to empyema were most commonly reported in the pre-antibiotic era.

Nonetheless, there continue to be isolated reports concerning the development of oesophagopleural fistulae. In 2 such reports, the fistulae occurred as a result of tuberculous pyopneumothorax and appear to be similar in nature to ours^{2,3}. In these cases, a number of mechanisms have been proposed to explain the formation of oesophagopleural fistulae¹. Erosion of an empyema through the oesophageal wall is a strong possibility. Alternatively, caseating lymph nodes may have ruptured into the oesophagus and into the extrapleural spaces; this could have happened in our patient since he had an unresolved pneumothorax. Primary oesophageal tuberculosis, although uncommon,

can also lead to fistula formation. While it may be interesting to speculate on the possibilities, it seems that the exact reasons behind our patient's relapse of tuberculosis and fistula formation will remain unknown.

A number of other causes of oesophagopleural fistulae have also been described in the literature. The most commonly reported ones are in post-pneumonectomy patients, occurring as a result of direct surgical trauma or as a recurrence of the underlying malignant or chronic inflammatory process^{4,5}. Perforation of the oesophagus and subsequent fistula formation can occur as a result of foreign bodies⁶, or Barrett's ulcers⁷; more rarely, Boerhaave's Syndrome of spontaneous oesophageal rupture has also been implicated⁸. In the current era, the advent of photodynamic therapy for malignant mesothelioma also appears to have become an iatrogenic cause for the development of oesophagopleural fistulae⁹.

The optimal treatment of oesophagopleural fistula is

not well defined¹, and healing may occur by medical therapy alone^{2,5}, or may require surgical intervention because of failure to resolve after months of conservative management³. Some authors have advocated the use of curative surgical techniques involving muscle flap repair of the oesophagus but the potential morbidity and mortality appears to be quite considerable^{4,6}. Our patient had refused to undergo any surgical procedure and it was fortunate that he had a good outcome.

In conclusion, this case demonstrates that reactivation of tuberculosis and the development of unusual complications such as oesophagopleural fistula may still occur even in the modern antibiotic era.

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