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Recurrence of tracheo-oesophageal fistula 32 years after primary repair

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Recurrence of congenital tracheo-oesophageal fistula after repair is not unusual, occurring in about 10% of cases.[1-3] Most observers agree that symptoms probably related directly to the size of the fistula. Large fistulae produce more symptoms and are diagnosed earlier, while small fistulae may cause little trouble and are difficult to detect.[4] Recurrent fistulae have been diagnosed from seven days to nine years after repair, but more than half are apparent within two months.[3] The purpose of this paper is to describe a case of recurrent tracheo-oesophageal fistula presenting 32 years after the primary repair.

Case report

In 1952 a two day old baby girl was found to have oesophageal atresia with tracheo-oesophageal fistula. Right thoracotomy was performed and the fistula seen to communicate between the trachea and the distal oesophageal segment. After division of the fistula the tracheal side of the fistula was closed with wire sutures and an end to end oesophageal anastomosis was performed with one layer of interrupted silk sutures. After operation she developed sputum retention, from which she recovered quickly with physiotherapy. She continued to make good progress with no symptoms of dysphagia or respiratory tract infection.

In 1985, she attended the clinic with an 18 month history of a dry cough, which disturbed her sleep; and she said that she sometimes felt as though her throat was closing up during swallowing. There was no history suggestive of gastro-oesophageal reflux, dysphagia, respiratory tract infection, or chest trauma. Clinically she was a fit young woman but examination of the respiratory system revealed a few crepitations over the left lower zone. Radiography of the chest showed no abnormality, but bronchoscopy showed a 3 mm elliptical defect in the posterior wall of the trachea about 4 cm above the carina. Oesophagoscopy showed a small defect in the anterior wall of the oesophagus at 25 cm. Barium studies showed the presence of a tracheo-oesophageal fistula, with barium entering the left main bronchus (fig 1).

A right thoracotomy was carried out. Before the pleural cavity was opened an intercostal muscle pedicle graft was prepared. After dissection a short fistulous tract 2 mm in length and 3 mm in diameter was identified between the trachea and the oesophagus below the level of the previous anastomosis and about 4 cm above the carina (fig 2). The fistula was divided flush with the oesophagus, the tracheal defect was closed in two layers with interrupted Ethibond sutures, and the oesophageal defect was closed in the same way. The intercostal muscle pedicle graft was interposed and secured between the trachea and oesophagus under no tension. After operation the patient made a good recovery and a barium meal performed at 10 days showed no evidence to suggest recurrence of the fistula. When seen in the clinic six months later she was well.

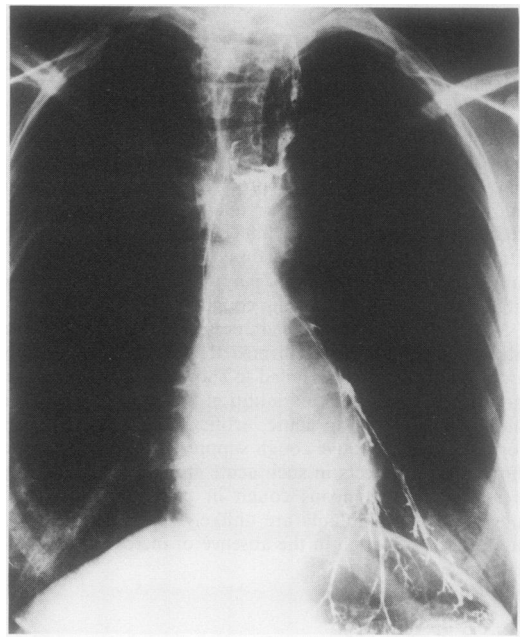

Fig 1 *Barium swallow showing a high tracheo-oesophageal fistula with contrast filling the left bronchial tree.*

Discussion

Recurrent tracheo-oesophageal fistula may be subtle in its presentation or even totally asymptomatic; this explains why some fistulae are not discovered for years, if at all.[5,6]

Recurrent tracheo-oesophageal fistula should be suspected in any patient who presents with the following symptoms at any time after the primary repair: repeated episodes of pneumonia, cough after meals, and abdominal distension.[2] It should also be suspected if the patient presents with a dry cough when lying down, as in this case.

Recurrence usually starts as an abscess between the trachea and the oesophagus at the anastomotic site, which eventually erodes into both organs leading to the formation

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Fig 2 *Comparative photograph showing the recurrent fistula between the oesophagus to the left and the trachea to the right.*

of the fistula. Cowley[7] suggested that the abscess formation is primarily due to the capillary action of the suture material, such as silk, which acts as a wick to draw contaminated material along the interstices of the suture material and leads to the abscess formation.

In this case, the time lapse between the primary repair and the recurrence 32 years later is difficult to explain. Negus in 1929,[8] described a proximal fold of oesophageal mucosa that initially overlapped the opening of the fistula during swallowing. This mucosal fold subsequently forms a less effective barrier leading to the appearance of the symptoms. Demong[9] postulated that the rostral direction of the fistulous tract from the oesophagus to the trachea may cause it to close during swallowing. Another possibility is that the fistula was closed with a thin, weak, membranous covering that ruptured after an episode of coughing, as was postulated by Jackson[10] to explain the late presentation of tracheo-oesophageal fistulae in adults.[10] In this case the recurrent fistula may initially have had an extremely small fistulous diameter, which in the face of the rapid passage of a food bolus and the short negative pressure of inspiration led eventually to a large bore fistula.[11]

It is important to emphasise that a recurrent fistula should be considered in the differential diagnosis in any patient who has had a previous repair.

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