

# Foregut duplication cyst presenting as neonatal respiratory distress and haemoptysis

D P Burgner, R Carachi, T J Beattie

## Abstract

**The case history is described of an infant, with a thoracic foregut duplication cyst containing a perforated peptic ulcer, who presented with haemoptysis and respiratory distress. This presentation is discussed within the context of thoracic foregut duplications.**

(Thorax 1994;49:287-288)

Duplication cysts of the gastrointestinal tract are rare and often present a diagnostic dilemma. Thoracic foregut duplications tend to present with respiratory symptoms.<sup>1</sup> We report details of a neonate with a foregut duplication containing a perforated peptic ulcer which formed a fistula with the lower lobe of the right lung, who presented with respiratory distress and haemoptysis.

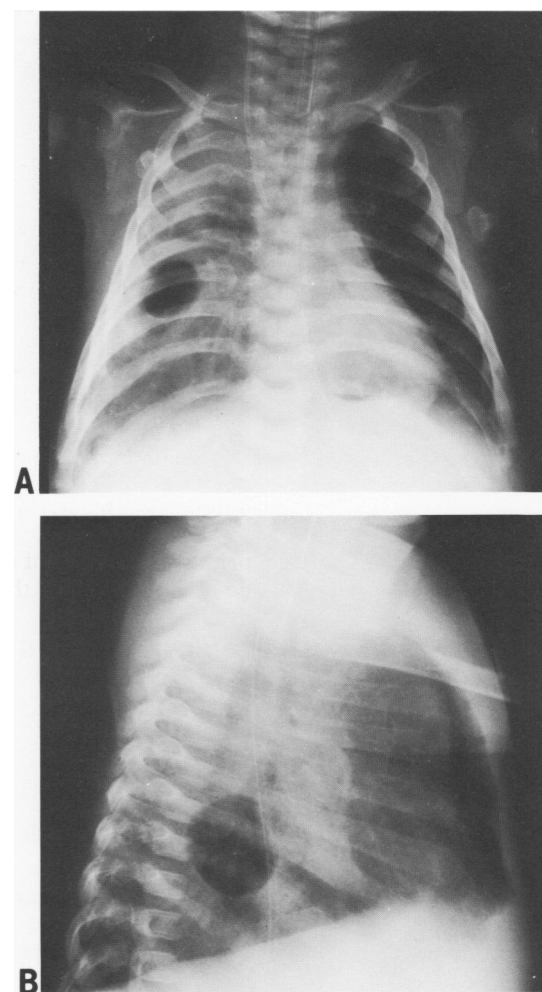
## Case report

A previously well two week old female infant, born by normal delivery at term, presented with a choking episode during a feed. On initial examination she was afebrile with signs of mild respiratory distress. A chest radiograph showed right lower zone opacification. Initial investigations revealed a peripheral blood neutrophilia and sterile blood cultures. Direct immunofluorescence of a nasopharyngeal aspirate was negative for viral pathogens. A presumptive diagnosis of aspiration pneumonia was made and she was started on intravenous Co-amoxiclav. During the following week the infant had several paroxysms of cough associated with cyanosis and vomiting. A chest radiograph showed persisting right lower zone opacification. Perinasal swab culture was negative for *Bordetella pertussis* and sweat electrolyte levels were normal. Erythromycin was added to her treatment.

On the 11th day of her illness she developed acute respiratory failure and required endotracheal intubation and artificial ventilation. A further chest radiograph showed opacification of the right lung with downward displacement of the right hemidiaphragm. In addition, an air-filled cyst was present in the right mid and lower zones (figure). A presumptive diagnosis of a lung abscess was made and the antibiotic treatment was changed to intravenous flucloxacillin, cefotaxime, and metronidazole. Two days after intubation heavily blood stained secretions were noted following endotracheal

suction. The secretions were sterile on culture and stained negative for acid fast bacilli. Recurrent life threatening haemoptysis continued, requiring blood transfusion. Bronchoscopy and upper gastrointestinal endoscopy failed to demonstrate an "H"-type tracheo-oesophageal fistula or other structural abnormalities. However, in view of the persisting haemoptysis an exploratory right thoracotomy was performed on day 36 of the illness.

At operation a large thoracic foregut duplication cyst was found which resembled stomach and communicated through the diaphragm. It contained a perforated peptic ulcer which had formed a fistula with the right lower lobe. There was no evidence of associated vertebral anomalies or extension of the cyst into the neural canal. The cyst was resected, although no resection of pulmonary tissue was necessary as simple closure of the pulmonary defect was possible. Postoperative upper gastrointestinal series failed to show a duodenal diverticulum. The infant made an uneventful postoperative recovery and at two year follow up she is symptom-free and thriving.



Posteroanterior (A) and lateral (B) chest radiographs performed on day 11 showing opacification of the right lung field with downward displacement of the right hemidiaphragm, and the presence of an air-filled cyst in the right lower and mid zones.

Department of  
Medicine  
D P Burgner  
T J Beattie

Department of  
Paediatric Surgery  
R Carachi

Royal Hospital for  
Sick Children,  
Yorkhill, Glasgow  
G3 8SJ

Reprint requests to:  
Mr R Carachi.

Received 19 March 1993  
Returned to authors  
28 May 1993  
Revised version received  
10 June 1993  
Accepted for publication  
15 June 1993

## Discussion

Congenital duplications may arise anywhere in the gastrointestinal tract. Midgut duplications are the most common, but those of foregut derivation (oesophagus, stomach, parts I and II of the duodenum, and rarely the oropharynx<sup>2</sup> account for approximately one third.<sup>3,4</sup>

Bronchopulmonary foregut duplications are duplications involving the pulmonary tree, most commonly the left lower lobe and the right lung. Communication between the gastrointestinal and respiratory tracts occurs most commonly at the distal oesophagus or oesophagocardiac junction,<sup>1</sup> although in this patient the communication originated in the proximal duodenum.

Foregut duplications are associated with the presence of ectopic gastric mucosa<sup>5</sup> and may present with gastrointestinal haemorrhage.<sup>4</sup> However, the presentation with haemoptysis due to a communication between a site of gastrointestinal haemorrhage and the bronchial tree is previously unreported.

Although bronchopulmonary foregut duplications are uncommon, approximately two thirds of patients will present before the age of two with either acute respiratory distress or more insidious respiratory symptoms such as chronic cough and recurrent pneumonia.<sup>1,4</sup>

The diagnosis is often made by radiography,

but ultrasonography and computed tomographic scanning are additional useful diagnostic tools; ultrasonography may define the cystic nature of the duplication,<sup>6</sup> and computed tomographic scanning may elucidate vertebral anomalies and extension of the cyst into the spinal canal, which are recognised associations with foregut duplication cysts.<sup>7,8</sup>

Foregut duplication cysts should therefore be considered as part of the differential diagnosis in patients with chest radiographic appearances of opacification, possibly with a cystic area, who do not respond to conventional treatment.

- 1 Heithoff KB, Sane SM, Williams HJ, Jarvis CJ, Carter J, Kane P, *et al.* Bronchopulmonary foregut malformations. A unifying etiological concept. *AJR* 1976;126:46-55.
- 2 Frenkiel S, Remsen KA. Foregut duplication arising from the pharynx. *J Otolaryngol* 1990;19:279-81.
- 3 Hocking M, Young DG. Duplications of the alimentary tract. *Br J Surg* 1981;68:92-6.
- 4 Ilstrad ST, Tollerid DJ, Weiss RG, Ryan DP, McGowan MA, Martin LW. Duplication of the alimentary tract. Clinical characteristics, preferred treatment, and associated malformations. *Ann Surg* 1988;208:184-9.
- 5 Bissler JJ, Klein RL. Alimentary tract duplications in children: case and literature review. *Clin Pediatr (Phila)* 1988;27:152-7.
- 6 Kangarloo H, Sample WF, Hansen G, Robinson JS, Sarti D. Ultrasonic evaluation of abdominal gastrointestinal tract duplication in children. *Radiology* 1979;131:191-4.
- 7 Mahboubi S, Finkelstein M, Afonami E. Esophageal duplication in children: a report of three cases evaluated by computerised tomography. *Emerg Care* 1985;1:90-3.
- 8 Thurston SE, Lenn NJ. The association of spinal and gastroesophageal anomalies. Case report and review. *Clin Pediatr (Phila)* 1984;23:652-4.