Bronchoscopic and angiographic findings in Dieulafoy's disease of the bronchus

Ben Hope-Gill, Bandipalyan V Prathibha

There have been several case series describing Dieulafoy's disease of the gastrointestinal tract, the majority presenting with massive haematemesis. The defining feature is an aberrant arterial vessel protruding into the lumen of a viscus, usually the stomach (Farrell and Bennett, 1992). The spectrum of disease has been extended to include the bronchial tree (Sweerts et al, 1995; Van der Werf et al, 1999). This article reports the bronchoscopic and angiographic images of a case of Dieulafoy's disease of the bronchus which have not previously been published. It is also the first reported case that has been successfully treated by embolotherapy.

CASE REPORT

A 49-year-old man presented with a 2-week history of increasing haemoptysis. This had been approximately 150 ml on the day of admission to hospital. There were no other associated symptoms. He had a 30-pack year smoking history and had been treated 14 years earlier for pulmonary tuberculosis. There had been four previous hospital admissions over the past 10 years with moderate haemoptyses, each of which settled spontaneously.

On this admission, clinical examination revealed no abnormal findings. The chest radiograph demonstrated several small, tissue-dense opacities confined to the upper lobes bilaterally, more marked on the right than the left. Initial haemoglobin estimation was 13.0 g/dl, coagulation screen was normal and arterial blood gas analysis revealed slight hypoxia; pH 7.43, PaO_2 = 10.39 kPa, $PaCO_2$ = 4.56 kPa. Active pulmonary tuberculosis was excluded; serum aspergillus precipitins were negative, as were antineutrophil cytoplasmic antibodies, antinuclear factor antibodies and antiglomerular basement membrane antibodies.

The possibility of recurrent pulmonary emboli was considered in the initial differential diagnosis, and therefore a radioisotope-labelled ventilation and perfusion lung scan was performed. This revealed unmatched segmental perfusion defects in the posterior segments of the right and left upper lobes and in the right middle lobe. The patient continued with daily haemoptysis of increasing severity such that his haemoglobin dropped to 11.1 g/dl.

Bronchoscopic examination showed no visible endobronchial abnormality in the right lower and middle lobes. In the posterior segment of the right upper lobe at the medial wall, a non-pulsatile lesion was seen arising from the mucosa and extending into the posterior segmental lumen (*Figure 1*). Owing to the possibility of tumour, this was washed with 0.9% wt/vol saline, which precipitated a moderate haemoptysis such that the procedure was abandoned before exploration of the left bronchial tree was possible. The haemoptysis resolved spontaneously with no fall in blood pressure or haemoglobin. Bronchial arteriography showed a right bronchial artery arising from the third right intercostal artery supplying a hypervascular area in the right mid-zone with visible cross-communication pulmonary artery branches (*Figure 2*). The bronchial vessel was selectively entered with a 4F catheter and embolized with 500–710 mm polyvinyl alcohol. This was stopped when the pulmonary artery communication was lost. There has been no further haemoptysis 12 months later. Subsequent high resolution computed tomography of the chest revealed normal lung architecture throughout both lung fields. Specifically, there was no cavity formation or bronchiectasis.

DISCUSSION

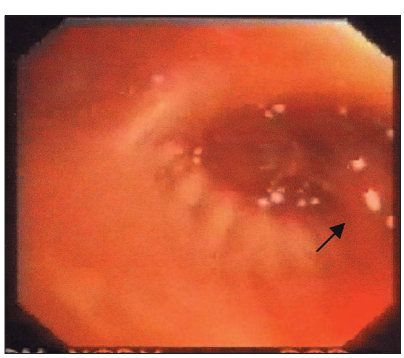
This case follows three previously reported cases of Dieulafoy's disease of the bronchus (Sweerts et al, 1995; Van der Werf et al, 1999). Although histological confirmation was not obtained, the angiographic and bronchoscopic findings support the diagnosis of Dieulafoy's disease in the absence of alternative lung pathology.

As with previous cases, this patient presented with massive haemoptysis in middle age. Fibreoptic bronchoscopy also preceded vascular imaging, yet in this case, and that reported by Van der Werf et al (1999), this precipitated large haemoptysis. Angiography is therefore the most appropriate initial investigation in these patients.

This is the first case that has been successfully treated with embolotherapy, the pulmonary arterial abnormality being embolized. This suggests that a pulmonary rather than a bronchial arterial anomaly is the most probable abnormality and may explain why in previous cases bronchial arterial embolization was unsuccessful.

The assumed aetiology in Dieulafoy's disease of the stomach is peptic ulceration. In the two cases reported by Sweerts et al (1995), no obvious cause existed and the lesions were therefore assumed to be congenital. Interestingly, in the case reported here, and in that of Van der Werf et al (1999), there was a previous history of pulmonary tuberculosis. Furthermore, the anomaly in this case occurred in the right upper lobe, a site typically associated with tuberculosis.

Figure 1. Bronchoscopic image of an aberrant pulmonary artery arising from the posterior segment of the right upper lobe (black arrow).



Dr Ben Hope-Gill is Respiratory Research Fellow in the Respiratory Unit, Morriston Hospital, Swansea SA6 6NL and **Dr Bandipalyan V Prathibha** is Consultant Physician in the Department of Respiratory Medicine, Neville Hall Hospital, Gwent Healthcare NHS Trust, Abergavenny

Correspondence to: Dr B Hope-Gill

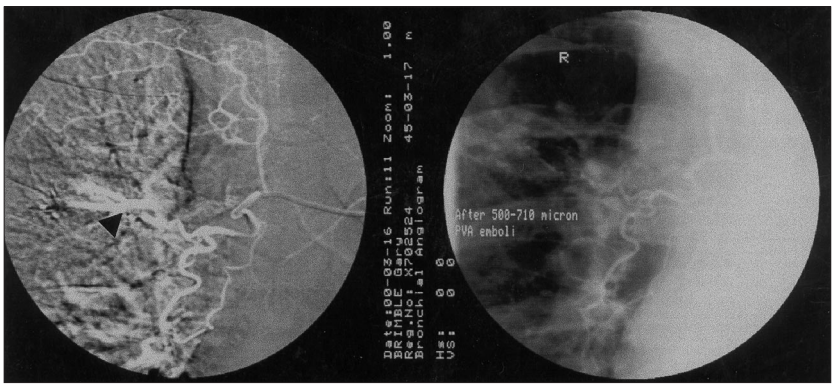


Figure 2. Bronchial angiogram demonstrating the abnormal pulmonary cross-communicating vessel (black arrowhead) and subsequent appearance after embolization.

It has long been recognized that chronic inflammation is associated with bronchial hypervascularity and bronchopulmonary anastomoses. However, it is rare in the absence of co-existent bronchiectasis, cavity formation or active tuberculosis (Ishihara et al, 1974). This suggests that Dieulafoy's disease of the bronchus may be acquired, particularly in patients who have had pulmonary tuberculosis. It should therefore be considered as a cause of haemoptysis in such patients who have no evidence of structural lung damage. HM

Farrell DJ, Bennett MK (1992) Dieulafoy's vascular malformation as a cause of large intestinal bleeding. *J Clin Pathol* **45:** 363–6

Ishihara T, Inoue H, Kobayashi K et al (1974) Selective bronchial arteriography and haemoptysis in non-malignant lung disease. *Chest* **66:** 633–8

Sweerts M, Nicholson AG, Goldstraw P, Corrin B (1995) Dieulafoy's disease of the bronchus. *Thorax* **50:** 697–8

Van der Werf TS, Timmer A, Zijlstra JG (1999) Fatal haemorrhage from Dieulafoy's disease of the bronchus. *Thorax* **54:** 184–5

Images in Medicine

A builder in a 'staple' condition

A Misra, CT Huins, RA Miller, HRH Patel

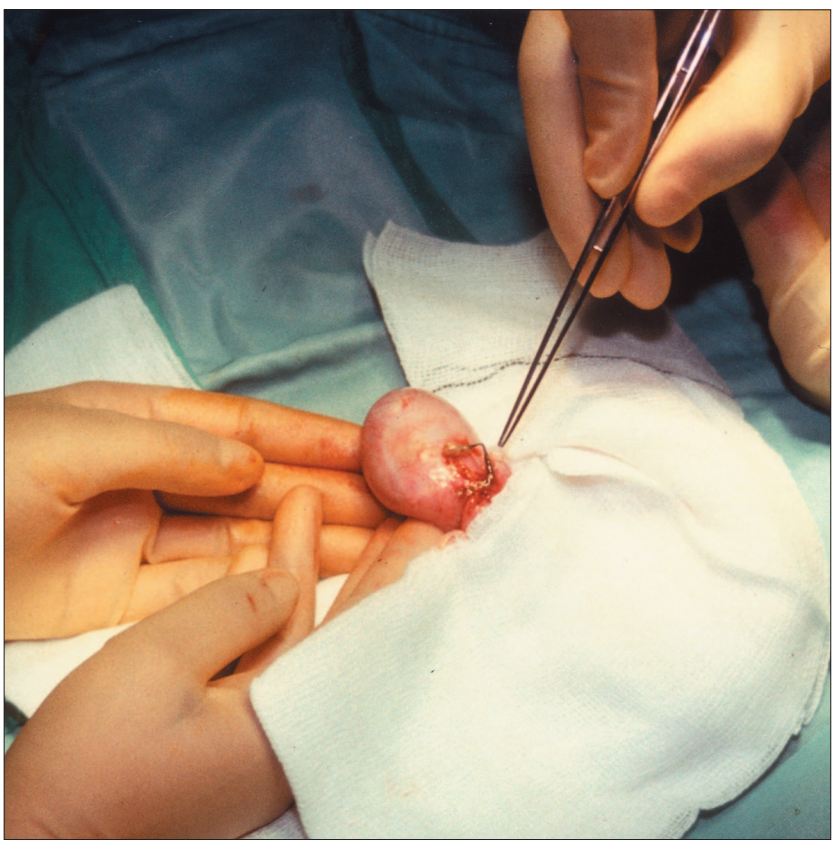


Figure 1. An industrial staple (pointed to) with the right testicle sitting below.

CASE REPORT

A 21-year-old man had an ultrasound 3 years earlier for testicular discomfort which revealed a small right-sided hydrocele and no other abnormalities. No treatment was given at this time.

Recently, on examination, a hard mass was noted under the scrotal skin, but not attached to the testicle. The patient was worried about cancer and requested a scrotal exploration. Surprisingly, an industrial staple was found under the tunica vaginalis of the right testicle (*Figure 1*).

After prompting, the patient remembered having used a staple gun at the time of presentation 3 years earlier, but had no recollection of the injury. HM

Dr A Misra is Preregistration House Officer, **Dr CT Huins** is Senior House Officer, **Mr RA Miller** is Consultant Urologist and **Mr HRH Patel** is Honorary Clinical Lecturer and Specialist Registrar in Urology, The Whittington Hospital, London N19 5NF

Correspondence to: Mr HRH Patel