at a time when a firm diagnosis would be most helpful. There is a suggestion that measurement of blood gases and lung volumes may help, with the finding of hyperventilation but no hyperinflation suggesting a functional cause. However, it is recommended that, despite suspicions that an individual's acute wheeze or stridor is factitious, they should initially be given full asthma treatment. When evidence has built up, and reassuring patterns of rapid recovery are established, then one can reduce the emergency treatment next time and show that it is not necessary to achieve equally rapid recovery. Once confident of the diagnosis, a referral to a skilled speech therapist can be very helpful, along with psychiatric referral if appropriate.

It is also possible to get variable laryngeal function following *partial* damage to recurrent laryngeal nerves. We recently saw a patient

who presented initially with variable inspiratory stridor due to bilateral cord adduction. The real diagnosis (a subaortic arch lung tumour) only became clear when there was progression to left sided permanent cord paralysis which prompted a computed tomographic scan of the thorax. It appears that early in the process of recurrent laryngeal nerve compression or infiltration there can be differential effects on the fibres, and hence on adductor and abductor function. The bilateral effects are difficult to explain initially but may be due to stimulation of sensory fibres in the nerve with reflex laryngeal narrowing.

These three case reports indicate how important it is for chest physicians to know about the larynx; unfortunately it is sometimes regarded as a bit "too far north" to be of interest and more in the territory of the ENT surgeon.

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Dieulafoy's disease of the bronchus

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Abstract

Dieulafoy's vascular malformation has not been described outside the gastrointestinal tract. Two cases are reported in which this vascular abnormality arose in right lower lobe bronchi, both of which presented with massive haemoptysis.

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Keywords: Dieulafoy's disease, bronchus.

Dieulafoy's disease is an abnormality of the gastric or, less frequently, the intestinal arteries, characterised by wide mucosal branches that are susceptible to ulceration and bleeding.¹⁻³ This abnormality has not been reported in the respiratory tract, but we have recently encountered two patients who each had such a vascular anomaly in a bronchus and presented in middle age with massive haemoptysis.

Case reports

CASE 1

A 35 year old woman, a non-smoker, was admitted with massive haemoptysis. In the previous 14 years she had had numerous similar but less severe episodes, usually preceded by an upper respiratory tract infection. Repeated examination of the nose and throat and

multiple bronchoscopies had not revealed the source of the bleeding. Pulmonary arteriography, chest radiography, computed tomographic scans, and a ventilation-perfusion scan had all been normal. However, bronchoscopy had shown clot confined to the right lower lobe bronchus on four occasions, suggesting that a local abnormality was responsible for the haemorrhage. It had also been noted that she had unusually long index fingers and first metacarpal bones.

On this admission bronchoscopic examination revealed a pulsating bleeding point in the right lower lobe bronchus. Diathermy and selective bronchial embolisation failed to control the haemorrhage and a right lower lobectomy was performed. Postoperative recovery was uneventful and four months later the patient has had no further haemoptysis.

CASE 2

A 59 year old retired male publican, who smoked 40 cigarettes and drank 10 units of alcohol a day, was admitted to another hospital as an emergency with haemoptysis (or haematemesis) and melaena. There was no history of any previous such haemorrhage. Endoscopic examination showed no gastrointestinal abnormalities and he was transferred to the Royal Brompton Hospital for control of the haemoptysis. There was no evidence of a coagulopathy and bronchoscopic examination revealed that the right lower lobe bronchus was filled with blood. A right lower and middle lobectomy was performed. No follow up data are available.

HISTOLOGY

The histological picture was similar in the two cases. In both, a large artery showing focal thinning of the media extended between the

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Figure 1 Case 1: a wide thin walled artery (arrows) communicates directly with the bronchial lumen. Stain: elastin van Gieson, magnification $\times 20$.

Figure 2 Case 2: a wide thin walled artery (arrow) is seen close to the bronchial mucosa and internal to the glands and cartilage. Stain: elastin van Gieson, magnification $\times 40$.

bronchial cartilages into the submucosa, and in case 1 through the mucosa to open into the bronchus (figs 1 and 2).

Discussion

The vascular abnormality we describe appears to be identical to that known as Dieulafoy's disease of the gastrointestinal tract.¹⁻³ Whether it is congenital or acquired has been the subject of debate, probably because peptic ulceration and haemorrhage are so common in the stomach. Its recognition in the colon,¹³ and now in the bronchus, weakens the case for it resulting from peptic ulceration and, in the absence of any obvious cause, it seems reasonable to assume that the lesion is congenital. If so, it may presumably bleed at any time, yet our second patient was 59 years old before this happened. The gastric lesions also tend to present late. This late presentation has been attributed to the general tissue atrophy of advancing age being a contributory factor.¹ That our first patient displayed a further congenital anomaly which affected her hands is possibly relevant to the supposed congenital nature of her bronchial lesion.

Such abnormal bronchial arteries are obviously a potential source of haemorrhage and need to be considered in patients presenting with unexplained haemoptysis, especially if the bleeding recurs and is confined to one lobe. Selective bronchial arteriography should be considered if results of all other investigations are normal. Embolisation of the abnormal vessels is probably the initial treatment of choice, although it is not always successful as our first case shows. Lobectomy appears to have cured our first patient, suggesting that the vascular anomaly was an isolated one.

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