CASE REPORT

An unusual epistaxis

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The case of a man who presented complaining of epistaxis is reported. He had coarctation repair 18 years previously. Subsequent investigation revealed an aorto-bronchial fistula resulting from false aneurysm formation distal to the original vessel anastomosis. This was repaired at surgery, the patient suffering a minor stroke, before rehabilitation and good recovery.

A 37 year old right handed man was assisted at home by paramedics, having called 999. He complained of nose-bleed.

On initial paramedic assessment he was noted to be sweaty and hypotensive, with a systolic blood pressure of 90 mm Hg, and tachycardia of 140 beats/min. There was an estimated blood loss of 1–2 units. The alleged epistaxis had stopped spontaneously.

After transfer to the accident and emergency department he remained tachycardic, but his blood pressure had improved to 106/68 mm Hg after crystalloid infusion. The history given was one of an acute, painless, sudden onset haemorrhage which the patient felt was originating in his throat. He did not report blood coming from the nostrils, but did report having to spit out blood. The haemorrhage lasted around one hour and was associated with light headedness. He gave a history of similar, but less severe, episodes in the preceding weeks, amounting to seven episodes of haemorrhage in a six week period. He gave no history of clotting disorders, and denied any local trauma to the nose.

Past medical history included hypertension, diagnosed one year previously, and a repair for coarctation of the aorta 18 years before. His only medication was quinapril 2.5 mg once daily.

Examination revealed tachycardia and hypotension. There was no active haemorrhage at this time. Fluid resuscitation was started, to which he responded. Before an ear, nose, and throat review the patient suffered a witnessed episode of haemorrhage, resulting in approximately 200 ml of blood loss. The bleed was associated by a fall in systolic blood pressure to 70 mm Hg. The patient described the blood as appearing in the back of the throat. There was no evidence of an anterior nasal bleed. The episode was self limiting.

Early fiberoptic nasendoscopy showed old blood in the nasopharynx and some crusted blood in the nasal cavities bilaterally. No obvious bleeding point was seen. The oropharynx, hypopharynx, and larynx showed bloodstaining, but no bleeding point was seen. He was admitted by the ear, nose, and throat team and managed as a suspected posterior nasal bleed. His haemoglobin concentration on admission was 110 g/l. An electrocardiogram showed a sinus tachycardia; chest radiography was not requested.

Haemoglobin was measured the next day and showed a drop to 80 g/l, and as a result he received a blood transfusion of three units. A gastroscopy and bronchoscopy was organised. The gastroscopy showed limited oesophagitis, and a small oesophageal ulcer, neither of which were thought to be the

Figure 1 Computed tomogram showing reactive changes in left lung base (arrows), which may represent a recent haemorrhage into the left lung; an abnormal calibre of the descending thoracic aorta is also shown.

Figure 2 Computed tomogram showing an unusually shaped descending aorta with focal abnormality posteriorly (arrow), which is consistent with a small aneurysm.

source of the haemorrhage. Examination of the bronchial tree found a trace of blood in the left main bronchus and computed tomography of the thorax was organised (see figs 1 and 2).

Computed tomography showed clips consistent with previous aortic surgery. The descending aorta was an unusual shape and showed a fleck of calcification. There was also a focal 1 cm area posteriorly, which was consistent with a small aneurysm. The lung posterior to this was abnormal, and suspicious of a recent bleed into that area. No haematoma was seen.

The patient was transferred to the regional cardiothoracic centre for definitive management. At surgery a false aneurysm from the previous coarctation repair was found to have invaded the left lung, resulting in the massive haemoptysis seen on admission. A Dacron graft was inserted as repair to the false aneurysm and this was done under circulatory arrest.

Postoperative recovery was hindered as the patient suffered a minor stroke affecting his right hand and arm. He was subsequently referred to the neurorehabilitation unit, on which he had a short stay, and on discharge he had recovered good function in the affected limb. Outpatient physiotherapy continues, and he remains otherwise well.

DISCUSSION

We have presented a case of haemoptysis resulting from a false aneurysm of the aorta invading the bronchial tree. The patient had undergone repair of a postductal coarctation of the aorta 18 years previously.

Coarctation of the aorta is a condition in which narrowing of the aorta occurs over a short distance. Two types are recognised: preductal and postductal, depending on their relationship to the ductus arteriosus. Preductal coarctation tends to occur in infants and is usually very severe leading to a cyanotic infant soon after birth, it is therefore readily recognised. In this case the patient had suffered from postductal coarctation.

Postductal coarctation tends to be less obvious and approximately 20% are diagnosed for the first time in adolescents and adults. It is more common in males with a ratio of 2 or 3:1; it also occurs in syndromes such as Turner's or Noonan's. There is an associated bicuspid aortic valve in at least 50% of cases, and the condition can also be associated with conditions such as Berry aneurysms.

Clinically coarctation has important consequences. Firstly there is a progressive elevation in arterial pressure in the proximal vasculature. Second, collateral circulation develops from the subclavian and axillary arteries through the internal mammary, scapular, and intercostal arteries. Thirdly, there may be aneurysmal dilatation of the aorta immediately before or after the stenosis.^{1,2}

Coarctation is usually repaired early in life, although some debate persists as to when is the ideal time.^{3,4} A variety of methods may be employed to correct the abnormality, the commonest include, resection of the narrowed segment and end-to-end anastomosis, or if it is not an option, the placement of a vessel graft.⁵ It is also possible to use percutaneous transluminal aortoplasty, which has shown promising results.⁶

Complications of aortic surgery are well documented, and include wound infection, bleeding, and cerebrovascular accident but also the more dramatic complications such as paralysis due to emboli in the anterior spinal artery.⁷ It is also documented that aneurysmal formation around, or immediately distal to, the repair can occur. Certainly aneurysms are more common if a patch aortoplasty has been employed.^{2,5} Some authorities have suggested that long term follow up of coarctation repair patients should occur, so as to allow for early identification of problems.⁸ Our patient was having surgical follow up.

What are less well documented are the problems that occur if aneurysms are not detected and the complications that may ensue. Certainly, there is literature showing aortobronchial fistulisation, but most is attributed to be secondary to another cause such as infection or chest trauma.^{9,10} This case is uncommon, as it appears to have occurred without a preceding event. It has also occurred some 18 years after the original surgery. It is noteworthy that the patient reports episodes similar to, but not as severe as, the presenting bleed, in

Learning points

- Young adults compensate well for blood volume loss and early indicators such as tachycardia in the face of normal blood pressure should not be ignored.
- The description of where blood is coming from in the upper airways is not always accurate.
- In patients with previous aortic surgery, a bleed reported in the upper airway, or reported haemoptysis, should at least be considered to have an aortic origin in the differential diagnosis.
- The gold standard for investigation in these situations is computed tomography of the thorax as it allows for a rapid diagnosis to be reached and is more widespread and readily available than magnetic resonance imaging. An aortogram, although useful, is limited by its unavailability and the possibility it may not detect a lumen which is full of blood clot.

the weeks before admission. It is obvious with hindsight that these were also episodes of early communication between the false aneurysm and the lung.

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