CASE REPORTS

Recurrent Hemoptysis Secondary to an Aortobronchial Fistula

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Aortobronchial fistula is a rare but serious cause of hemoptysis. It can develop from an aneurysm of the descending thoracic aorta in the context of infections or it may appear as a sequel of surgical repair of congenital heart defects. Presenting symptoms include mild bronchial hemorrhages and recurrent chest pain, culminating in a normally fatal massive hemorrhage. Diagnosis by imaging is not always conclusive and clinical suspicion based on medical history is essential. Surgical placement of an endovascular stent graft is the treatment of choice. Post-surgical prognosis is good although there is a risk of recurrence in the case of superinfection.

Key words: *Hemoptysis. Aortobronchial fistula. Endovascular stent.*

Hemoptisis recurrente secundaria a una fístula aortobronquial

La fístula aortobronquial es una causa rara pero grave de hemoptisis. Se produce como evolución de un aneurisma de la aorta torácica descendente en procesos infecciosos o tras la reparación quirúrgica de cardiopatías congénitas. Se suele manifestar con episodios de hemorragia bronquial leve y dolor torácico recurrentes, hasta la aparición de una hemoptisis masiva, mortal en la mayoría de los casos. El diagnóstico definitivo por técnicas de imagen no siempre es posible, por lo que es fundamental la sospecha clínica tras una anamnesis adecuada. El tratamiento de elección es quirúrgico, mediante la colocación de una prótesis endovascular. El pronóstico tras la intervención es bueno, aunque con riesgo de recurrencia si se produce una sobreinfección.

Palabras clave: *Hemoptisis. Fístula aortobronquial. Prótesis endovascular.*

Introduction

Aortobronchial fistula (ABF) is a rare cause of hemoptysis that should be watched for in patients who have undergone surgery of the descending thoracic aorta as the consequences of nontreatment are fatal. We present the case of a patient examined for recurrent hemoptysis caused by an ABF secondary to surgical repair of an aortic coarctation performed 15 years earlier.

Case Description

The patient was a 47-year-old man, employed in the construction industry, active smoker of 20 cigarettes per day (30 pack-years exposure), without signs of chronic bronchitis or history of or known contacts with tuberculosis. He had undergone surgery of an aortic coarctation at another hospital 15 years earlier, without apparent postoperative complications.

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The patient was admitted to the pneumology unit of the hospital in July 2002 with mild self-limited hemoptysis. Analyses, chest x-ray, pulmonary function tests, sputum smears, and computed tomography (CT) without intravenous contrast were carried out. The sputum smears were normal. The patient had no further episodes of hemoptysis in hospital and was discharged. A year later the patient came to the emergency department after a hemorrhage of approximately 100 mL brought on by coughing, and was admitted to hospital again. There were no signs of infection or anomalies in the patient's medical history and the chest x-ray was normal. Fiberoptic bronchoscopy was performed and produced only blood-stained samples with no endobronchial material. After 48 hours' observation the patient was discharged and monitored as an outpatient. A few days later the patient had another episode of hemoptysis, in which the amount of blood was moderate. Fiberoptic bronchoscopy performed at this time revealed sheet bleeding of the left upper and lower lobes. A thoracic CT scan with intravenous contrast revealed a pseudoaneurysm in the thoracic aorta beside the old aortic repair. ABF was suspected and a catheter angiogram (Figure 1) and a magnetic resonance angiogram (Figure 2) were performed, confirming the diagnosis.

Subsequent surgery consisted of the transfemoral insertion of an endovascular stent graft. The patient suffered postoperative

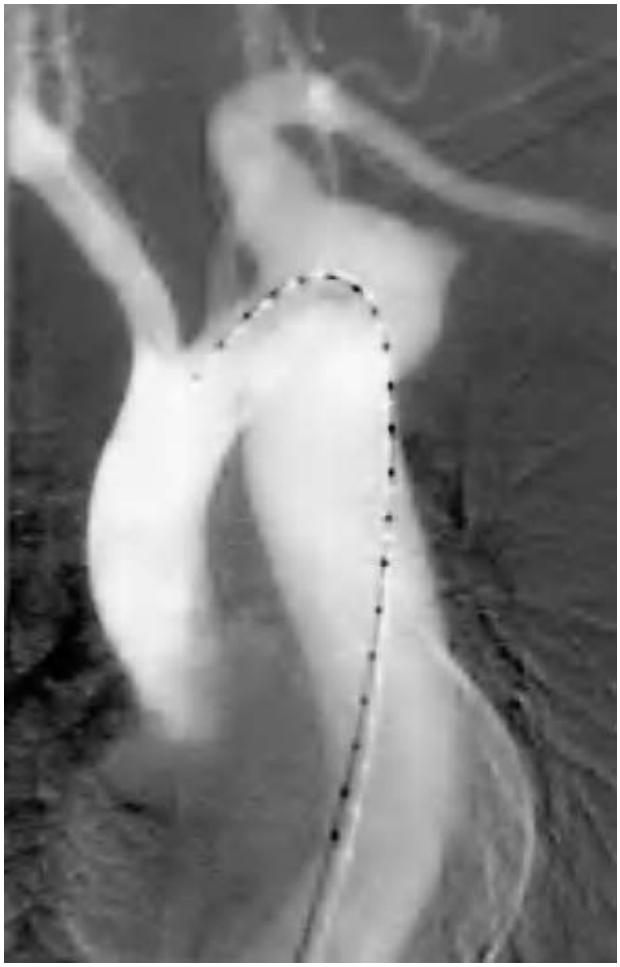

Figure 1. Pseudoaneurysm confirmed by catheter angiogram.

venous thrombosis and anticoagulation with dicoumarin was started. He was moved from intensive care to a pneumology ward where the third, this time massive, episode of hemoptysis (of about 800 mL) caused respiratory failure. He was intubated and readmitted to intensive care. A control catheter angiogram was performed and coils were inserted into the pseudoaneurysm cavity as a safety measure although contrast extravasation had not been observed. The anticoagulation treatment was suspended and, after observation in the ward without incidents, the patient was discharged. He has had no further episodes of hemoptysis and is asymptomatic.

Discussion

ABF is a rare but potentially fatal cause of hemoptysis. Diagnosis on suspicion is essential as diagnosis tests are frequently nonspecific.[1,2] Most ABF develop from an aneurysm or pseudoaneurysm of the descending thoracic aorta. The aneurysm can be caused by tuberculosis, syphilis, mycotic infections, or trauma. In recent years the incidence of ABF secondary to surgical repair of congenital heart defects has increased, particularly among young patients like ours, as has the incidence of atherosclerotic aneurysms among older patients.[3]

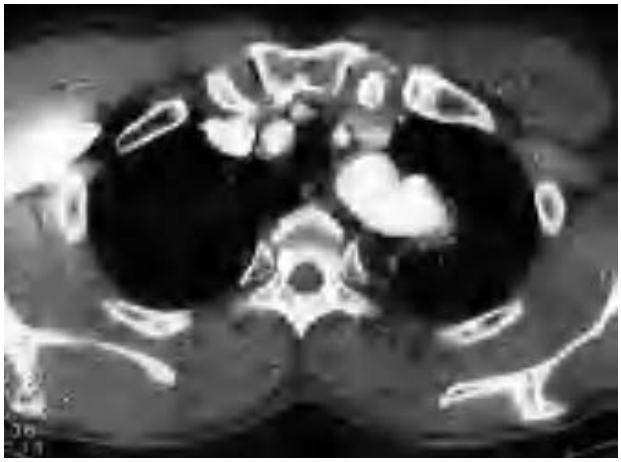

Figure 2. Magnetic resonance angiogram of a thoracic, aortic pseudoaneurysm.

Several factors can cause a fistula. In fistulas secondary to surgical interventions which have involved the insertion of prosthetic material, as in coarctation repair, the aneurysm or pseudoaneurysm forms along the proximal or distal suture lines of the prothesis. Once formed, the continuous and pulsating pressure of blood against a weakened vascular wall damages the aorta and subsequently the adjacent lung parenchyma, and communication is created. Moreover, the foreign material can cause inflammation and become attached to the lung tissue. The pseudoaneurysm can tear and affect the bronchus or the lung, usually the left one.

ABF presents as hemoptysis and can appear between 3 weeks and 25 years after surgery.[4] Episodes tend to be self-limited and recurrent, increasing in severity until a massive hemoptysis occurs. The reason for the interval of days or weeks between episodes is thought to be temporary obstruction of the fistula by clots. Chest pain presents in 45% of patients, but did not in ours.[1]

Diagnosis of ABF is difficult and clinical suspicion is often unconfirmed. Chest x-rays tend to be normal or to show alveolar images of bloody material. High resolution CT and angiography usually identify the aneurysm or pseudoaneurysm but rarely locate the fistula. Fiberoptic bronchoscopy is necessary to identify the point of bleeding and sometimes the fistula itself.[4] In our patient, diagnosis was reached by magnetic resonance angiogram, which is the method of choice according to the literature.[1]

Treatment is always surgical and must be performed immediately, even before diagnosis has been completely confirmed, because of the risk of fatal massive hemoptysis.[5,6] Endoluminal/endovascular treatment is preferred although sometimes local pulmonary resection is also necessary because of parenchymal inflammation.[7,8] In the case we report, an endovascular stent was inserted into the aorta and coils were placed in the pseudoaneurysm cavity. Direct repair with prothesis carries the risk of sepsis and subsequent fistula recurrence and some authors recommend long-term postoperative

antibiotic treatment. Our patient has taken treatment with oral teicoplanin since the intervention.

Postoperative prognosis is good in most cases according to the literature, without recurrences or other complications presenting.[8,9] This has been the case with our patient. However, recent reviews show that while insertion of an endovascular stent is the treatment of choice, serious secondary complications can occur and patients require close monitoring.[10] In any case, mortality is 100% if patients are not diagnosed in time, making clinical suspicion and immediate action critical in all patients with hemoptysis who have a history of surgery on the descending thoracic aorta.

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