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A rare cause of hemoptysis in childhood: peripheral pulmonary artery pseudoaneurysm to bronchial fistula in a case with tetralogy of Fallot

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Brief Report

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Abstract

We herein report a patient complaining from significant hemoptysis due to secondary fistulisation of the peripheral branch of the left pulmonary artery and the left bronchial tree, which was successfully treated with a transcatheter angiography.

Hemoptysis is a rare but life-threatening symptom in childhood. Infections, particularly infections of the lower respiratory tract, foreign body aspirations, and tracheostomy complications are frequently cited as aetiological factors. Vascular pathologies are rare causes in aetiology.[1–3]

We herein report a patient complaining from significant hemoptysis due to secondary fistulisation of the peripheral pulmonary artery between the peripheral branch of the left pulmonary artery and the left bronchial tree, which was successfully treated with a transcatheter angiography.

Case report

A 6-year-old boy patient was admitted to our emergency department due to increased discharge of bloody sputum. The sputum had been extracted intermittently for the past 2 years and consisted of remnants of blood and mucus. On his medical records, it was discovered that he had a total correction surgery for tetralogy of Fallot performed 4 years ago. His post-operative period was uneventful, and he had used acetyl salicylic acid during this period. When he began to experience hemoptysis, an erythrocyte suspension was transfused due to iron deficiency anaemia. There was no history of chronic respiratory infections or a foreign body aspiration in the patient and had received all immunisations.

Physical examination of the patient, who presented with no genetic disease in his pedigree was conscious, with a withering appearance. Remnants of blood clots were detected in the oropharynx. The heart rate of the patient was 115 beats/min, with a blood pressure of 100/65 mmHg. While the cardiac examination showed 2–3/6º systolic murmur in the left 2.–3. intercostal range, there was no sign of a pathology detected in other organ system examinations, including the respiratory examination. The patient had a leucocyte count of 6470/mm^3, haemoglobin 5.5 gr/dL, and a platelet count of 340,000/mm^3 on complete blood count; liver–kidney function tests, and electrolytes and coagulation tests were normal. A ferritin level of 4.72 ng/mL was detected. A suspicious pericardiac infiltration was detected on chest X-ray.

The patient's antiaggregant dose of acetyl salicylic acid was stopped, an erythrocyte suspension transfusion was conducted, an oral iron supplementation was begun, and the patient's hemoptysis and hematemesis could not be cleared, thus a proton pump inhibitor was started. Nasolaryngoscopic examination indicated no signs of active bleeding. The upper gastrointestinal tract endoscopy was conducted to rule out possible gastrointestinal tract pathologies, and the findings were normal.

A thorax CT imaging to rule out possible vascular pathologies (aortic pulmonary collateral artery, etc.) and to display lung parenchyma in the patient who underwent full correction surgery due to tetralogy of Fallot was conducted. An aneurysmatic dilatation reaching 1.5 cm was observed in the lower lobe segmentary branch of the left pulmonary artery (Figure 1 a, b). No signs of haemorrhage were present on rigid bronchoscopy, and a catheter angiography was planned for the patient, to better visualise the aneurysmatic structure as well as for therapeutic purposes. Angiography showed extravasation of the contrast agent to the left main bronchus and trachea immediately after contrasting the aneurysm with an injection of radio-opaque material into the left pulmonary artery. The proximal part of the aneurysm was sealed with 10- and 12-mm vascular plugs. A control angiography conducted following the procedure indicated no sign of contrast material leakage into the aneurysm or the bronchus (Figure 2 a, b, c).

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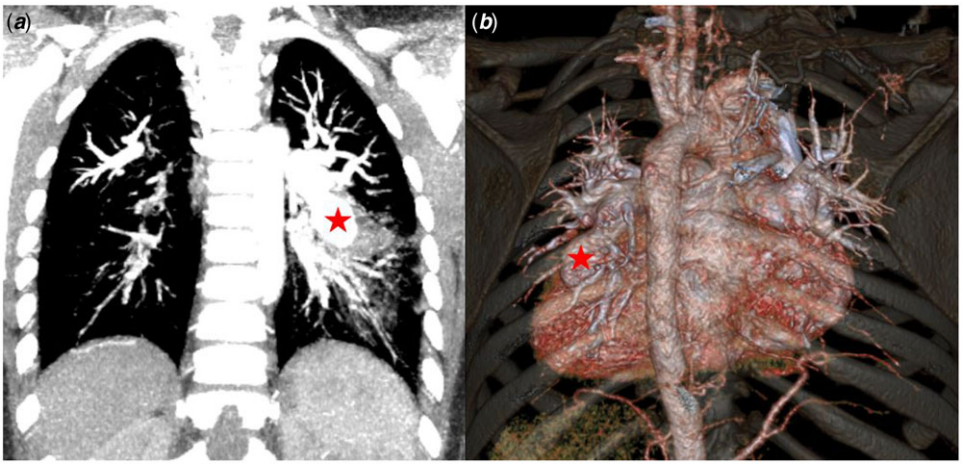

Fig. 1. Aneurysmatic dilatation in left pulmonary artery brunch on computerized tomography (red stars).

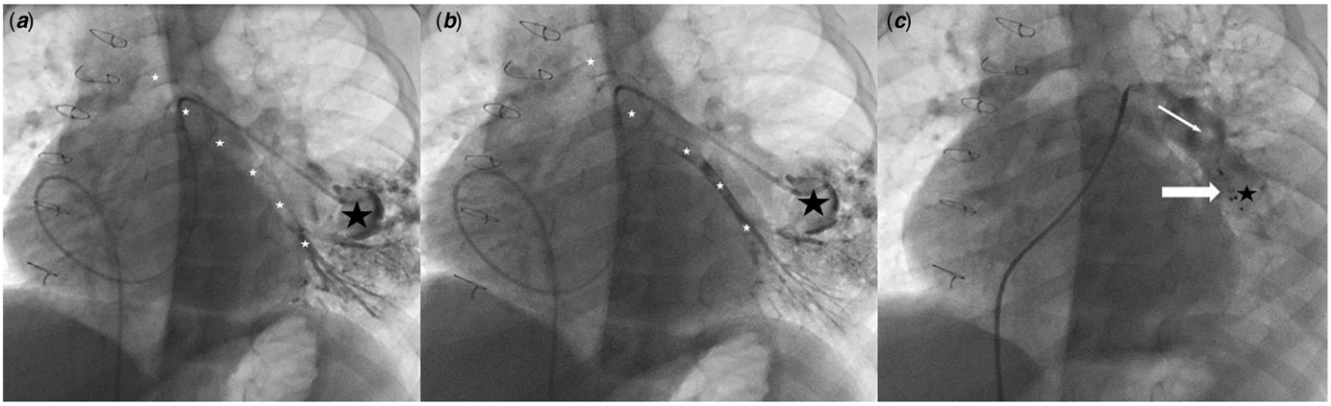

Fig. 2. Aneurysmatic dilatation in left pulmonary artery brunch on angiography (black star), filling with opaque substance and blood via pulmonary-bronchial connection to left bronchus (white stars) and occlusion of pulmonary aneursyms with vascular plug (white arrows).

The patient, who was stable after angiography, was discharged after 2 days. The patient did not present with a recurrence of hemoptysis, after a 3-month follow-up.

Discussion

Hemoptysis is a rare but life-threatening symptom in childhood. Infections, particularly infections of the lower respiratory tract, foreign body aspirations, and tracheostomy complications are frequently cited as aetiological factors. Other rare aetiological causes include chronic lung diseases (cystic fibrosis, bronchiectasis, tuberculosis, and pulmonary hemosiderosis), upper respiratory pathologies (tonsil haemorrhage, nose bleeds, and laryngitis), certain diseases of the gastrointestinal tract, CHD (tetralogy of Fallot especially accompanied by aortopulmonary collateral arteries), neoplasia, bleeding diathesis, and Munchausen's syndrome and also include diseases such as Good Pasteur syndrome and Wegener's granulomatosis, which are diseases of vascular origin and can infiltrate the lungs and kidneys.[1–4]

In the presented case, the anamnesis, physical examination, and basic laboratory tests rule out both the leading aetiological and other rare aetiological causes. Possible upper respiratory pathologies with ear, nose, and throat are ruled out with flexural nasopharyngoscopy, while gastrointestinal tract pathologies are also ruled out by an upper gastrointestinal tract endoscopy.

Pathologies of vascular origin, arteriovenous malformation, hereditary telangiectasia, tracheal hemangioma, bronchial artery or pulmonary artery pseudoaneurysm, direct pulmonary artery trauma, or lung injury are much rarer causes.[2,3]

The patient had a surgical correction procedure for tetralogy of Fallot 4 years prior to presentation. He underwent a contrasted tomography angiography of the thorax to investigate the pulmonary parenchyma in detail and to rule out any possible bronchial artery pseudoaneurysms which might originate from an aortopulmonary collateral artery. An aneurysm located at the distal segment of the left pulmonary artery was detected leading to bronchial erosion and hemoptysis. A pulmonary artery-bronchial fistulisation was confirmed with conventional catheter angiography, and pathology was treated successfully with percutaneous techniques.

These fistulisations can be occluded with different types of devices such as coils and vascular plugs.[2] In this case, we occluded the pulmonary artery-bronchial fistulisation with vascular plugs.

Secondary pulmonary artery injuries due to catheterisation or surgery and associated hemoptysis may develop. In addition, chronic lung infections, mushroom ball causes, etc.,or traffic

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accidents, stabbings, etc., and subsequent cases of hemoptysis due to lung injury have been reported.[5–7] In addition, fine needle biopsy for the bronchial system, etc., and hemoptysis may develop after such invasive procedures.[8] In this case, in addition to the smooth post-operative period of the patient, no invasive interventions were made towards pulmonary arteries or the bronchial system during both the pre-operative and the post-operative period.

In conclusion, it is well known that sometimes hemoptysis may lead to life-threatening complications. The frequent causes are sought when investigating the aetiology of hemoptysis. On the other hand, although seldom, congenital cardiovascular pathologies (e.g., presence of aortopulmonary collateral arteries in tetralogy of Fallot) should be remembered as an aetiology for hemoptysis and ruled out with contrasted CT, angiography, or conventional angiography. When the cause is detected, it should be managed with appropriate measures to prevent bleeding and further consequences.

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Conflicts of interest. None.

Ethical standards. This case report was appropriate by the Hospital Institutional Ethical Committee.

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