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Case Report

Bronchopulmonary malinosculation: a case report

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ABSTRACT

Pulmonary malinosculation, which include a broad spectrum of disorders that involve abnormalities in one or more of the three main components of the lung, namely, the airways and lung parenchyma, arteries and veins. A case of bronchopulmonary malinosculation was presented due to its rarity.

Keywords: Bronchopulmonary malinosclulation, Sequestration, Embolisation

INTRODUCTION

The concept of sequestration was first described by Pryce in 1946.[1] However, this terminology could not define all variants of sequestration and led to the concept of a sequestration spectrum described in 1974. This concept was reviewed in 1984 and the importance of the venous drainage was discussed, leading to the wheel theory by Clements, who coined the term pulmonary malinosculation which include a broad spectrum of disorders that involve abnormalities in one or more of the three main components of the lung, namely, the airways and lung parenchyma, arteries and veins.[2] Malformations can involve the lung parenchyma, airways, pulmonary or systemic arteries, pulmonary or systemic veins, fistulas with the gastrointestinal tract or defects in the diaphragm and various combinations of these. In 2007, Lee proposed the haphazard theory to substantiate this concept and described seven types of pulmonary malinosculation, based on whether the abnormal communication involved the bronchi, arteries of the lungs or veins of the lungs in isolation or in different combinations.[3] We presented a case of isolated systemic arterial supply to the normal lung with normal bronchial tree, normal venous drainage and absent pulmonary artery branches, suggestive of a type B or isolated arterial malinosculation because of its rarity. They could also be classified as a Pryce type 1 anomaly.

CASE REPORT

15 year old, 10th standard student, admitted to us with complaints of fever for 5 days 2 weeks back, cough for 2 weeks and coughing out of blood since 1 day. Fever was low grade, intermittent, not associated with chills/rigor or sore throat, rhinitis, myalgia. Fever subsided in 5 days. Cough was productive associated with scanty mucoid expectoration, which was non purulent or foul smelling. No postural or diurnal variation noticed for cough. Also had complaints of coughing out of blood mixed with sputum, associated with occasional passage of clots since day 1. No massive episodes, epistaxis, malaena or other bleeding manifestations. He did not give history of breathlessness, chest pain, loss of appetite or loss of weight. He gave the history of atopic symptoms. No history of hoarseness of voice, dysphagia, small joint pain, stiffness, rash, photosensitivity, symptoms s/o raynauds or GERD symptoms.

Patient gave history of atopic symptoms from childhood and episodic breathlessness for the past 1 year with history of nebulisations and injections two times last year with good symptomatic relief. There is no history of any inhaler use or hospital admissions. The patient did not give history of antitubercular treatment in the past. There was no history of recurrent sinusitis or ear infections or history of coronary artery disease, diabetes mellitus or hypertension. Sleep and appetite were normal. No alteration in bowel or bladder habits. He was not a smoker or alcoholic and doesn't give history of passive smoke or firewood smoke exposure.

On examination he was conscious, oriented, moderately built and nourished. He had a BMI 18.91 kg/m^2. There was no pallor, icterus, cyanosis, clubbing, lymphadenopathy or pedal edema. Pulse rate was 63 per min, BP was 110/60 mm Hg, afbrile. Respiratory rate was 17 per min, SpO_2 was 95%. His respiratory system examination was normal. His blood routine examination showed Hb=11.5, TC=9090 with 52% neutrophils and ESR=5. His LFT and RFT were normal. Sputum AFB smear examination was negative and sensitivity showed normal pharyngeal flora. His viral markers were non-reactive. Chest X-ray showed a retrocardiacopacity (Figure 1).

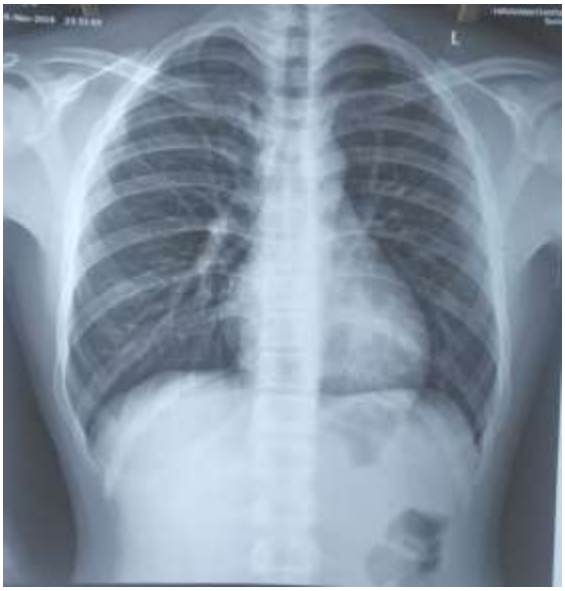

Figure 1: Chest X-ray showing retro cardiac shadow.

HRCT thorax showed large area of ground glass attenuation in the anteromedial segment of left lower lobe-consistent with alveolar hemorrhage. Aberrant vascular mass in the left hilum suggestive of dilated vessels with a draining channel from aorta. Left pulmonary vein appeared dilated, seemed to be entering into the vascular nidus but no direct channel to nidus can be seen (Figure 2 and 3).

Digistal substraction angiography (DSA) showed a large arterial feeder (12.8 mm), supplying left lower lobe and venous drainage was into the pulmonary veins (Figure 4 and 5). No A-V fistula was noted. In view of these findings a diagnosis of isolated arterial supply to normal lung was made. Using progreat microcatheter over microwire, multiple coils were deployed and check angiogram showed no filling of the collateral with no aortic supply to left lower lobe (Figure 6). Post procedure patient was stable and no further episodes of hemoptysis. Patient remains asymptomatic on follow up.

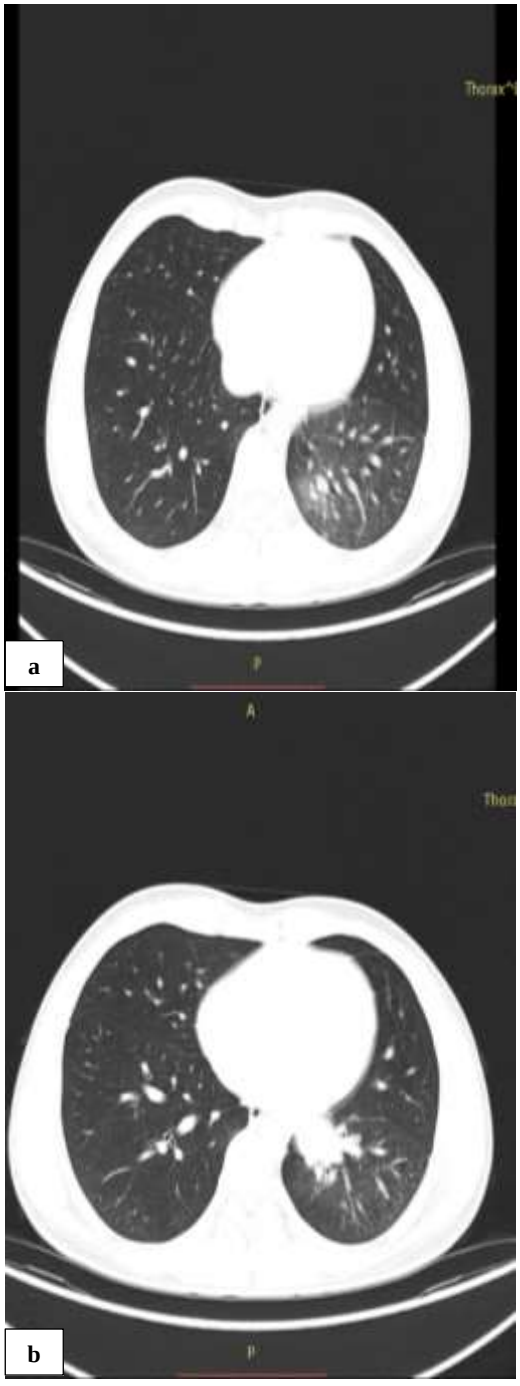

Figure 2 (a and b): Ground glass attenuation in the antero medial segment of left lower lobe-consistent with alveolar hemorrhage and vascular mass.

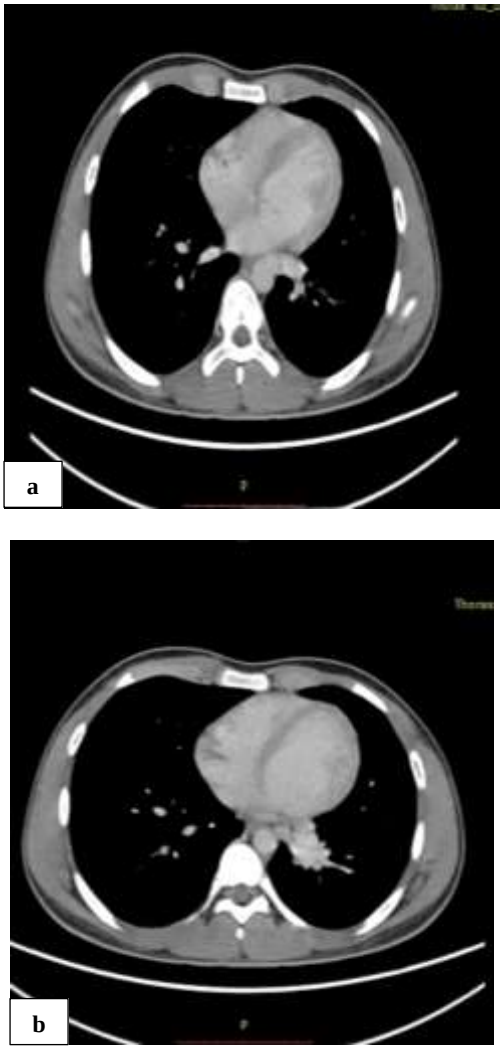

Figure 3: Aberrant vascular mass in the left hilum dilated vessels with a draining channel from aorta; left pulmonary vein appears dilated, seems to be entering into the vascular nidus but no direct channel to nidus can be seen.

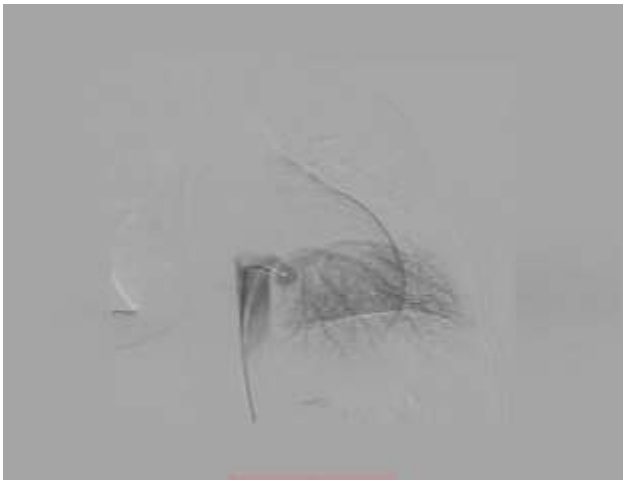

Figure 4: DSA showing arterial feeder (12.8 mm), supplying left lower lobe.

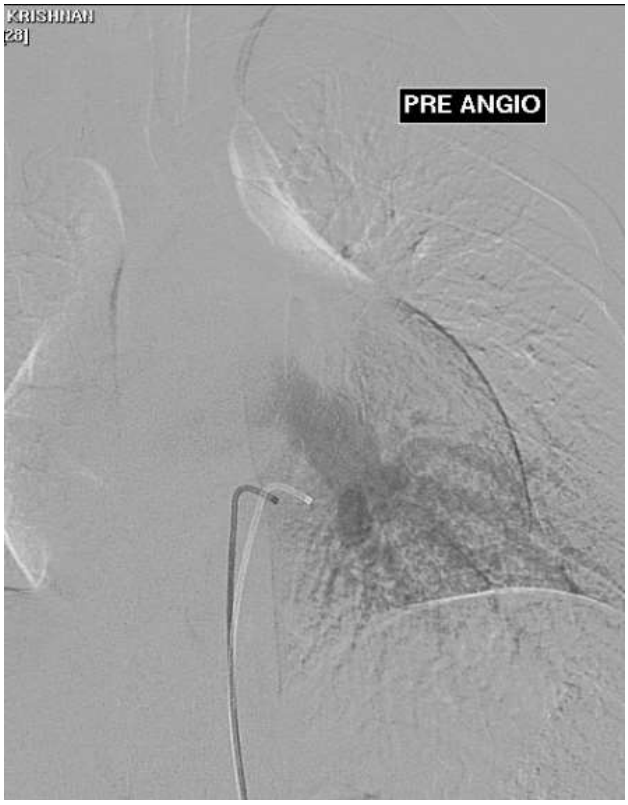

Figure 5: DSA showing venous drainage was into the pulmonary veins.

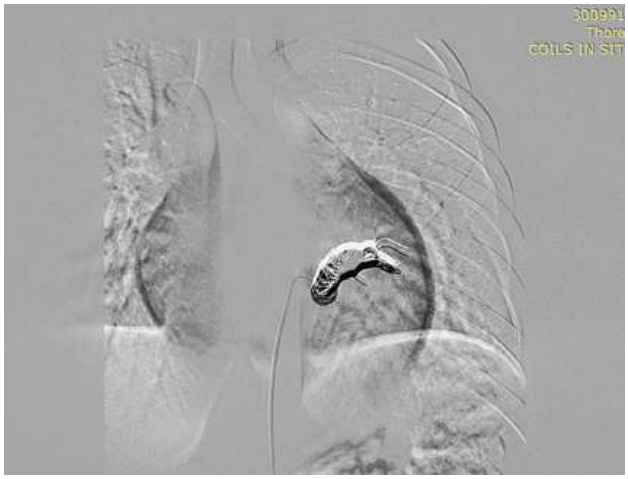

Figure 6: Check angiogram after coil embolization showing no filling of the collateral.

DISCUSSION

Congenital bronchopulmonary vascular malformations (BPVMs) include a broad spectrum of disorders that involve abnormalities in the form of disruptions of normal communication and/or presence of abnormal communication between one or more of the three main systems of the lung, namely, the airways, arteries and veins.[2] These were classified in a systemic manner as bronchopulmonary malinosclulation. Seven types of

bronchopulmonary vascular malinosculation are described. Type A, B and C represent isolated bronchial, arterial and venous malinosculations, respectively. Various combinations of type A, B and C result in bronchoarterial (D), bronchovenous (E), arteriovenous (F) and bronchoarteriovenous (G) malinosculations.[4]

Anomalous systemic arterial supply to normal lung is an anatomical variant where a portion of the lung (usually a basal segment) is supplied by a systemic vessel without a distinct pulmonary sequestration. It was traditionally called a Pryce type 1 sequestration. This entity was also known as pseudosequestration as the lung shows normal communication with tracheobronchial tree. It was described under type B bronchopulmonary malinosculation.

Developmentally, it is thought to arise as a result of the failure of regression of the primitive aortic branches to the developing lung bud. It is more commonly described on the left side where the left lower lobe is supplied by an artery arising from descending thoracic aorta and rarely from the left subclavian and internal mammary arteries. When the right lower lobe is involved, it is usually supplied by an artery arising from celiac trunk or abdominal aorta.

Patients with isolated systemic supply to normal lung (ISSNL) can be asymptomatic or present with hemoptysis and exertional dyspnea due to left side volume overload.

Treatment strategies include surgery, often lobectomy and segmentectomy, when aberrant systemic artery is the sole supply. Occlusion of aberrant vessel by surgical ligation or endovascular treatment (embolization), when the involved segment has a dual to multiple blood supply are also done.

CONCLUSION

A case of bronchopulmonary malinosclulation in a 15 yrs old boy who presented with hemoptysis, managed with coil embolisation is presented due its rarity.

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