Images in Vascular Medicine

VASCULAR MEDICINE

Unexpected cause of recurrent haemoptysis

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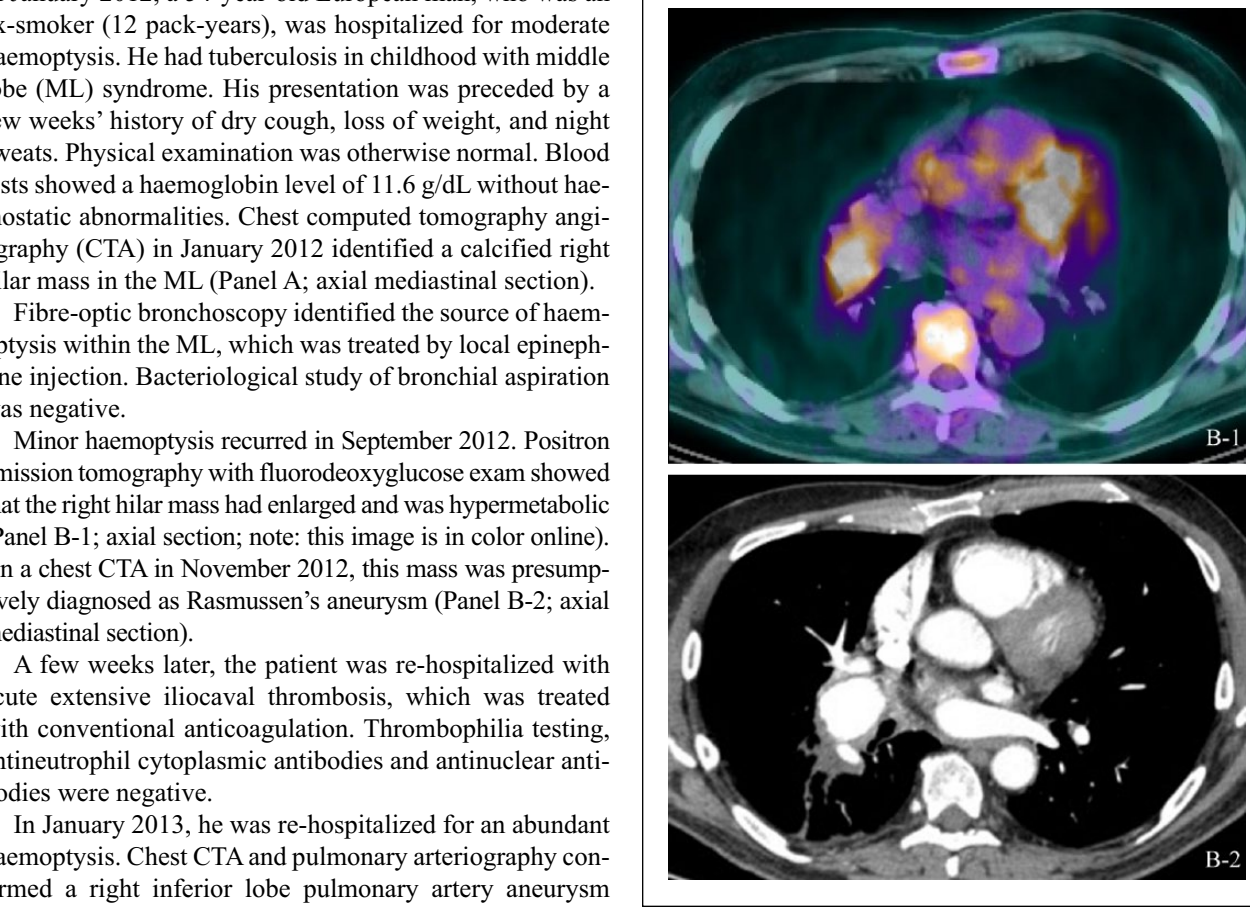
In January 2012, a 54-year-old European man, who was an ex-smoker (12 pack-years), was hospitalized for moderate haemoptysis. He had tuberculosis in childhood with middle lobe (ML) syndrome. His presentation was preceded by a few weeks' history of dry cough, loss of weight, and night sweats. Physical examination was otherwise normal. Blood tests showed a haemoglobin level of 11.6 g/dL without haemostatic abnormalities. Chest computed tomography angiography (CTA) in January 2012 identified a calcified right hilar mass in the ML (Panel A; axial mediastinal section).

Fibre-optic bronchoscopy identified the source of haemoptysis within the ML, which was treated by local epinephrine injection. Bacteriological study of bronchial aspiration was negative.

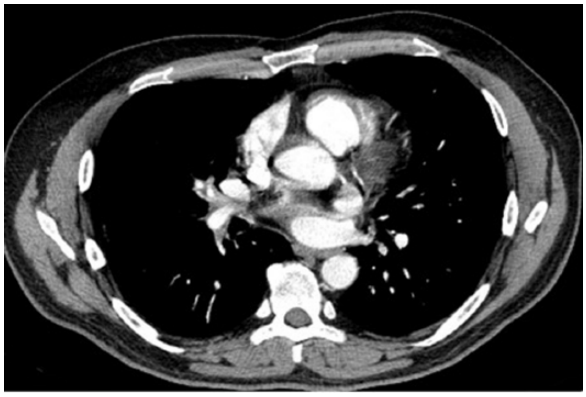
Minor haemoptysis recurred in September 2012. Positron emission tomography with fluorodeoxyglucose exam showed that the right hilar mass had enlarged and was hypermetabolic (Panel B-1; axial section; note: this image is in color online). On a chest CTA in November 2012, this mass was presumptively diagnosed as Rasmussen's aneurysm (Panel B-2; axial mediastinal section).

A few weeks later, the patient was re-hospitalized with acute extensive iliocaval thrombosis, which was treated with conventional anticoagulation. Thrombophilia testing, antineutrophil cytoplasmic antibodies and antinuclear antibodies were negative.

In January 2013, he was re-hospitalized for an abundant haemoptysis. Chest CTA and pulmonary arteriography confirmed a right inferior lobe pulmonary artery aneurysm (PAA) (Panel C; frontal section). Anticoagulation was stopped given the high risk of massive haemoptysis.[1]



Panel B.



Panel A.

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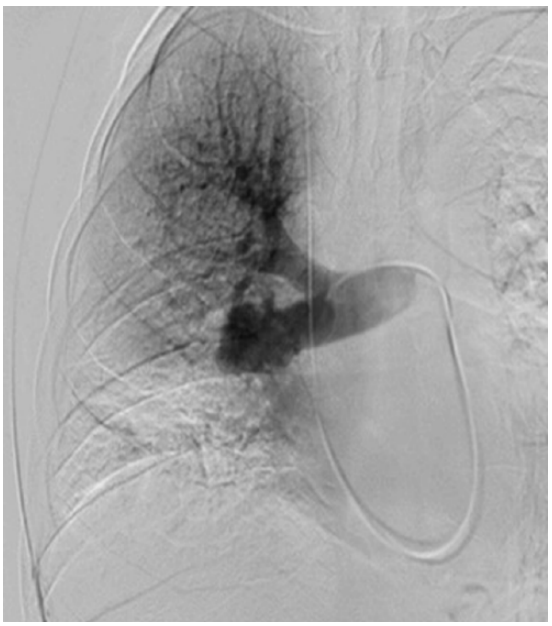

Panel C.

Retrospectively, the patient reported recurrent oral and genital aphtosis from 18 months of age. Although there was no family history of Bechet disease (BD) and pathergy testing was normal, testing for HLA B51 was positive. There was no evidence of anterior uveitis, and magnetic resonance imaging excluded neurological vascular lesions.

A diagnosis of BD variant Hughes–Stovin syndrome was made based on the characteristic combination of PAA and extensive deep venous thrombosis.[2] After six monthly cycles of cyclophosphamide and systemic corticosteroids, chest CTA showed a significant regression of the PAA (June 2013).

Our case illustrates that the diagnosis of incomplete BD or Hughes–Stovin syndrome may be delayed when a European patient (prevalence 3.3/100,000)[2] with personal history of tuberculosis presents with PAA-associated haemoptysis. The composite of rapidly progressive PAA and venous thrombosis should prompt consideration of Hughes–Stovin syndrome as early recognition and immunosuppressive treatment improves the prognosis.[1,3]

Declaration of conflicting interests

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