CASE REPORT

Coexisting pulmonary haemorrhage and venous thrombosis: a tricky but novel case

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SUMMARY

We present a case of Lemierre's disease complicated by pulmonary artery pseudoaneurysm. A previously well woman aged 43 years presented with a history of a sore throat and worsening dyspnoea. She was diagnosed with a severe cavitating bilateral pneumonia complicated by internal jugular vein thrombosis consistent with a diagnosis of Lemierre's disease. She had an episode of massive haemoptysis which was confirmed to be due to a pulmonary artery pseudoaneurysm which required radiologically guided embolisation. She then required surgical decortication for bilateral empyemata. Blood cultures revealed growth of *Slackia exigua*, an organism yet to be linked with both of these complications concurrently. We believe this to be a novel case of this bacterium causing thrombosis and haemorrhage.

BACKGROUND

This is a rare but important condition with a high mortality if left untreated. This is the first case to link *Slackia exigua* to Lemierre's disease and highlight the difficulties of managing patients with haemorrhagic and thrombogenic complications of this disorder.

CASE PRESENTATION

A woman aged 43 years with no significant medical history presented with a 7-day history of worsening dysphagia, non-productive cough, hoarseness, fever, rigors, general malaise and anorexia. She was a never smoker and was on no regular medication but had completed a recent course of clarithromycin. At presentation, her cough had become productive of foul-smelling brown sputum.

On examination, the patient was tachypnoeic and tachycardic without haemodynamic compromise. She was feverish at 38.5°C and oxygen saturations were 94% on air. Chest auscultation revealed reduced air entry with crackles at the left lung base. Her tonsils were erythematous and there was right-sided cervical lymphadenopathy.

INVESTIGATIONS

Laboratory investigations show white cell count $29.2 \times 10^9/L$, C reactive protein 354 mg/L, albumin 16 g/dL and sodium 127 mmol/L. Chest radiograph demonstrated multiple cavitating nodules and bilateral pleural effusions.

DIFFERENTIAL DIAGNOSIS

This is likely to be community-acquired pneumonia. However, the presence of multiple cavitating lesions is not typical and one should also consider

atypical pneumonia, mycobacterial disease (although the history is short), immunodeficiency, vasculitis and septic emboli. Appropriate specimens for culture, including throat swabs, sputum and blood cultures are vital at this stage.

TREATMENT

The patient received intravenous fluids and broad-spectrum antibiotics. A right-sided chest drain was inserted draining straw coloured exudative fluid.

A CT scan of the neck and chest revealed bilateral gas locules in the tonsillar beds (figure 1), a right-sided internal jugular vein (IJV) thrombus (figure 2), extensive bilateral cavitating pulmonary abscesses (in keeping with septic emboli) and bilateral pleural effusions (figure 3). Low molecular weight heparin was started.

The patient had persistent high fevers. Serial blood cultures were negative, echocardiogram was normal, respiratory viral and bacterial blood and urine screens were negative, autoimmune and blood-borne virus screens were negative. A further chest ultrasound revealed a left-sided loculated effusion suspicious for empyema. However, before a drain could be inserted, the patient had an episode of ~100 mL haemoptysis. Therapeutic heparin was stopped. A repeat CT scan demonstrated bilateral

Figure 1 CT of the neck demonstrating gas within the right tonsillar bed (arrow).

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Figure 2 CT of the neck, which demonstrates a thrombus within the right internal jugular vein (arrow).

Figure 3 Cavitating lesions in both lungs with bilateral pleural effusions.

gas-containing empyemata, cavitating lung lesions and a 10 mm aneurysmal dilation (figure 4), which CT angiogram confirmed as a pseudoaneurysm arising from the right upper lobe segmental pulmonary artery (figure 5). Pseudoaneurysms are considered at high risk of bleeding, and therefore, embolisation was felt to be appropriate, particularly in the context of ongoing thromboembolic disease. The patient was referred for radiological embolisation.

The right femoral vein was punctured and a 5 Fr pigtail catheter was advanced to the pulmonary artery origin. Superselective cannulation of the feeding vessel was performed with a microcatheter, and the vessel was successfully embolised with two detachable coils (figure 6). The minor trauma from the microcatheter was sufficient to cause extravasation of blood into the extracellular space, indicating the fragility of the pseudoaneurysm's false wall.

Figure 4 (A) Cavitating lesion in the right upper lobe with a pseudoaneurysm within it (arrow). (B) Pseudoaneurysm from a segmental branch of the right pulmonary artery (arrow).

An intercostal chest drain was inserted into the left-sided empyema and frank pus was drained. However, the patient still continued to spike high fevers; so on day 19, she underwent a left-sided pleural decortication, with marsupialisation of two large lung abscesses. An organising peel was observed over 70% of the lung conferring grade III empyema.¹ This advanced stage is usually not amenable to treatment with antibiotics and fibrinolytics² and the fibrous covering necessitates direct removal, with excision of the thickened pleura along with the superimposed fibrous capsule. This was performed through the minimally invasive VATS approach to minimise postoperative complications.³

OUTCOME AND FOLLOW-UP

The patient was transferred to level 2 care postoperatively initially requiring cardiovascular support. She made good progress with a decrease in inflammatory markers.

Blood cultures were subsequently reported as positive for *Slackia* sp and 16S rDNA sequencing revealed *S. exigua*.

Follow-up CT scan at 6 months revealed complete resolution of the pulmonary artery pseudoaneurysm and cavitating nodules, with only small areas of scarring and atelectasis.

The diagnosis is Lemierre's disease.

DISCUSSION

Lemierre's disease was first reported in 1890,^{4 5} and the syndrome was characterised by Andre Lemierre in 1936 as recent oropharyngeal infection with consequent thrombophlebitis of surrounding veins resulting in metastatic septic emboli.⁴⁻⁷ It is a rare phenomenon, estimated at around 1–3 cases per million per year and primarily affects children and young adults.^{8 9}

Patients present with a history of recent oropharyngeal infection fever >39°C, rigors and often respiratory distress¹⁰ in the absence of haemodynamic instability.¹¹ Alternative primary infections have included odontogenic infection, sinusitis, mastoiditis and parotitis.^{7 12-15}

Septic emboli most commonly manifest as necrotic, cavitating lesions in the lungs (97% of cases).^{10 16 17} The second most common focus for septic emboli are the large joints, including shoulders, hips and knees.^{15 16} There have also been reports of emboli resulting in splenic abscess, hepatic abscess, renal abscess, haemolytic uraemic syndrome and pericarditis.^{11 15 16} Presumed retrograde extension^{18 19} of the infection from the IVJ has also rarely resulted in cerebral infarcts,^{20 21} meningitis,^{20 22} encephalopathy²³ and cavernous sinus thrombosis.⁵

Other less common complications include pleural effusion, lung abscess and necrotising mediastinitis^{9 15} and disseminated intravascular coagulation. There are no previous reports of septic emboli causing bronchopulmonary haemorrhage as seen here. Mortality is thought to be between 6% and 22%.²³

The majority of the cases are attributed to *Fusobacterium necrophorum*, a Gram-negative, anaerobic bacillus.⁷ We believe

Figure 5 Selective angiogram of a segmental branch of the right upper lobe pulmonary artery demonstrating the pseudoaneurysm (arrow).

Figure 6 Selective angiogram of the right upper lobe segmental pulmonary artery following the successful embolisation with detachable coils (arrow).

this is the first recorded case of Lemierre's disease caused by *Slackia* species.

Slackia exigua is a Gram-positive, obligate anaerobe, known to be associated with oral infections.²⁴ There are difficulties in identification of *S. exigua* as they grow very slowly on conventional culture media²⁵ are easily missed using standard laboratory protocols.²⁶ *Slackia exigua* has recently been molecularly characterised by 16S rRNA sequencing.²⁷ *Slackia exigua* was identified as causing a bacteraemia in an immunocompetent patient, secondary to periodontal infection.²⁷ Mass spectrometry has been used to detect *S. exigua* as the cause of lung abscess.²⁵

We believe that this is the first case to link *S. exigua* with Lemierre's disease and pulmonary artery aneurysm.

Learning points

- ▶ Lemierre's disease should be considered in younger patients with a persisting high fever not responding to antibiotics, antecedent upper respiratory tract infection, respiratory distress, neck pain and/or septic emboli.
- ▶ A surgical approach for empyema is considered necessary if there is no improvement within 48–72 hours.⁵
- ▶ Most patients will be anticoagulated, but the role of anticoagulation in these patients is under debate²⁸ and the potential complication of pulmonary haemorrhage from septic pseudoaneurysms should be considered.

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