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CASE REPORT

When it's more than just a sore throat: Lemierre's syndrome

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Lemierre's syndrome is a rare condition typically affecting healthy young adults, however its diagnosis is often delayed, which may be fatal. In this report, we present a typical case of this condition: a 25 year old gentleman presenting with fever, cough and sore throat who was found to have superficial thrombophlebitis of the right external jugular vein with blood cultures positive for Fusobacterium necrophorum. Multifocal lung consolidations were identified on CT thorax. He was started on appropriate antibiotic and anticoagulant therapy and made a full recovery. Through this report, we aim to increase awareness of this condition, particularly as a possible cause of sepsis in an otherwise healthy patient.

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CASE REPORT

A previously healthy 25 year old gentleman presented to the Emergency Department with a 1 week history of fever (more than 39°C), sore throat and productive cough which persisted despite a course of antibiotics (azithromycin). He also reported occasional haemoptysis.

On examination he was found to have low oxygen saturations and an ENT review revealed a hard lump on the right side of the larynx and non-specific pharyngeal erythema.

Blood investigations revealed an elevated white cell count with left shifted neutrophils and a high c-reactive protein. Blood cultures and a viral screen were also taken. Routine admission chest X-ray (CXR) showed patchy airspace shadowing in the left middle lung zone. He was thus started on empirical antibiotic therapy (tazobactam-piperacillin and clarithromycin).

In view of the CXR findings, he underwent a CT thorax which revealed multifocal areas of consolidation throughout both lungs and small bilateral pleural effusions (**Figure 1**). No intrathoracic lymphadenopathy was seen and mild splenomegaly was reported in the upper abdomen.

The intrapulmonary findings were believed to be secondary to septic emboli, and he thus underwent a transthoracic echo which was negative for infective endocarditis. In view of the neck mass on examination as well as to look for any other areas of infection, he also underwent an ultrasound of the neck, which revealed superficial thrombophlebitis of the right external jugular vein with extension of thrombosis into the right internal jugular vein (**Figure 2**). No intracranial abnormalities were demonstrated on a CT scan of the brain.

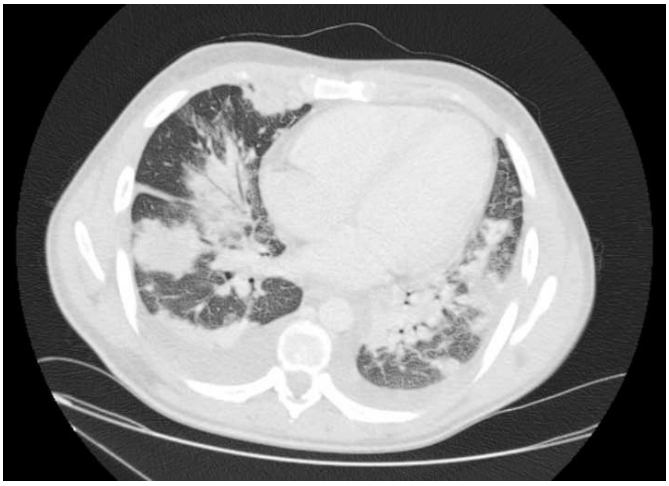

Figure 1 *A CT scan of the thorax showing bilateral multifocal consolidations and bilateral pleural effusions*

Viral screen taken on admission for Epstein-barr virus (EBV), cytomegalovirus (CMV), hepatitis and human immunodeficiency virus (HIV), serum mycoplasma antibodies and serum and urinary investigations for Legionella and Strep. pneumoniae were negative. Blood cultures however grew Fusobacterium necrophorum (sensitive to penicillin, metronidazole, clindamycin, meropenem and imipenem).

Therefore, a diagnosis of Lemierre's syndrome was suspected and antibiotics were modified according to sensitivities. There was a good clinical response to meropenem and clindamycin. He was also started on anticoagulation – initially on intravenous heparin, which was then switched to a direct oral anti-coagulant (rivaroxaban).

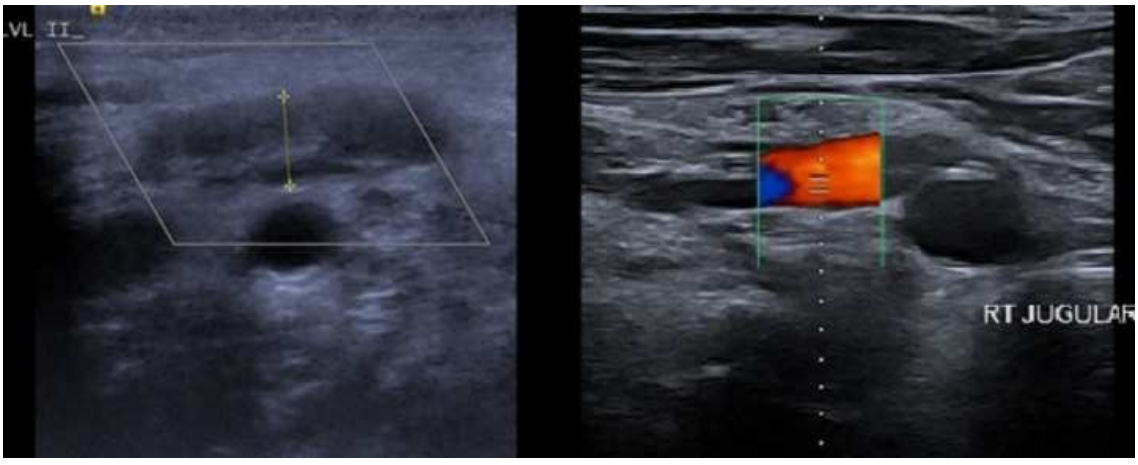

Figure 2 *Ultrasound scan of the neck showing thrombosis of the right external jugular vein (EJV), with slight extension into the right internal jugular vein (left image), with the right image showing patency of the right jugular vein 3 months after treatment.*

Figure 3 *Comparison of Chest X-rays, prior to treatment and two months after treatment*

OUTCOME AND FOLLOW-UP

The patient improved remarkably on the upgraded antibiotics and anticoagulation and was thus discharged home on oral antibiotics (clindamycin for a total course of 21 days) and rivaroxaban for a total of 3 months.

He was followed up with repeat ultrasound scans of his neck (**Figure 2**) and CXRs (**Figure 3**) over the next few months, which showed complete resolution of both the jugular vein thrombosis and the areas of consolidation within the lungs.

DISCUSSION

Lemierre's syndrome is a rare condition typically affecting healthy young adults. It is characterised by pharyngitis followed by thrombophlebitis of the internal jugular vein (IJV) with possible spread to the lungs, liver or brain.[1] The diagnosis is often delayed, hence it is associated with significant morbidity and mortality if not diagnosed and treated promptly.[2]

It is a rare condition often referred to as the 'forgotten disease' due to the low reported incidence after the pre-antibiotic era.[3] However, it is thought that it is not as rare as previously believed, possibly due to increasing antibiotic resistance or decreased antibiotic prescription for upper respiratory tract infections (URTIs)[4], which are more commonly due to a viral aetiology.

Early accurate diagnosis requires a high degree of clinical suspicion, particularly since it presents with nonspecific symptoms similar to benign URTIs.[2] It is however essential, as this condition may be life threatening if untreated, and effective treatment options are available if a timely diagnosis is carried out.

This case highlights the fact that diagnosis is often delayed; it was only suspected after internal jugular vein (IJV) thrombosis, a recognized typical complication of the condition, was noted on imaging, and further confirmed by identification of the most common causative organism (Fusobacterium necrophorum) on blood cultures.

Treatment involves antibiotics; penicillins given for 2 to 6 weeks, together with metronidazole or clindamycin due to macrolide- and increasing penicillin-resistance.[5] As happened in our case, most patients are treated empirically with antibiotics for a bacterial infection or pneumonia suspected on blood results or chest imaging, prior to diagnosis.

SUMMARY BOX

- The physician should consider Lemierre's syndrome as a possible cause of sepsis particularly in previously healthy patients. Early diagnosis and treatment is paramount as the condition can be life threatening.
- The following should prompt the possibility of Lemierre's syndrome: (i) pharyngitis which persists for more than 5 days, particularly if treated with macrolides (most common causative organism tends to be macrolide-resistant), (ii) pharyngitis associated with lateral neck pain or swelling, (iii) pharyngitis followed by sepsis or multiple pulmonary lesions.

The role of anticoagulation is still controversial. Some authors report use only in patients who fail to improve despite antibiotics, and in those with thrombophilia, cerebral venous sinus thrombosis or arterial thrombosis. Other authors have reported use in all patients with this condition.[6] Surgery is often reserved for drainage of neck abscesses. IJV ligation or excision may rarely be required in cases of persistent septic embolization despite medical treatment.[7]

CONCLUSION

In this report we present a typical case of Lemierre's syndrome, in which accurate diagnosis was delayed despite the classical presentation. We thus aim to increase awareness of this condition, particularly as a possible cause of sepsis in an otherwise healthy patient.

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