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Case Report

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Lemierre's Syndrome in an Immunocompetent, Middle-Aged Female: A Case Report

Lal, Shankar[1]; Bazyler, Caleb[2]; Counts, John[2]; Youssef, Dima[1*]

[1]East Tennessee State University, Department of Internal Medicine, Division of Infectious Diseases, TN, USA
[2]East Tennessee State University, Quillen College of Medicine, Johnson City, TN, USA

[*]**Correspondence:** Dima Youssef, East Tennessee State University, Department of Internal Medicine, Division of Infectious Diseases, Quillen College of Medicine, Johnson City, TN, USA.

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Abstract

Lemierre syndrome is a rare complication of oropharyngeal infection, especially acute pharyngo-tonsillitis, associated with septic, and thrombophlebitis of the internal jugular vein (IJV). Although the incidence of this syndrome has fallen dramatically since the widespread use of antibiotic therapy to treat streptococcal pharyngitis, it should still be suspected in otherwise healthy young patients. It is most commonly caused by Fusobacterium necrophorum. It is life threatening condition therefore, a timely diagnosis, close clinical/imaging monitoring and multidisciplinary management are necessary.

Keywords: *Lemierre, fusobacterium, life threatening, sepsis, timely diagnosis.*

Introduction

Lemierre syndrome (LS) is a rare life-threatening condition presenting as septic thrombophlebitis of the internal jugular vein (IJV) secondary to an oropharyngeal infection often leading to septic embolisms to distant sites. Anaerobic gram-negative bacillus *Fusobacterium nucleatum* and *Fusobacterium necrophorum* are commonly isolated organisms in patients with LS [1]. The syndrome originates from an upper respiratory infection that spreads to the IJV, causing septic thrombophlebitis, which results in multiorgan metastasis, and more frequently pulmonary complications. Septic emboli and end-organ effects can result in long-term morbidity. Therefore, a timely diagnosis, close clinical/imaging monitoring and multidisciplinary management are necessary [2].

Case presentation

This is a 45-year-old female who presented with complaints of sore throat for the last 2 weeks with nausea and vomiting over the last 3-4 days. She suspected flu and tried gargling salt water, but it did not work. She noticed worsening of her sore throat associated with nausea and vomiting. She denied fever, but felt chills. She stated she went to urgent care earlier today and tested negative for strep, flu A and B, and Covid. On physical examination, right cervical lymph nodes were enlarged and right tonsil erythema was noticed. Her workup here showed a white blood cell count of 16,100 (normal, 3.5-10.5 K/ul), platelets 119,000 (normal 150-450 K/ul), and creatinine 2.19 (normal,0.7-1.2 mg/dl). A CT scan of the abdomen and pelvis was performed and revealed no abnormalities. There was a small nodule in the right lower lobe. Chest x-ray showed mild central pulmonary vascular congestion without airspace consolidation. She was admitted for acute kidney injury and was started on IV fluids.

During admission she began having intermittent fever spikes and tachycardia, and was subsequently started empirically on Vancomycin and Zosyn. A CT scan of the neck soft tissues showed thrombosis of the right IJV (figure 1) and common facial vein with near complete occlusion (figure 2). There was also presence of thrombophlebitis of the IJV and mildly heterogeneous enhancement of the right palatine tonsil suggestive of tonsillitis. A CT chest angiogram showed no pulmonary embolism, but there was presence of bilateral lung nodules suggestive of multifocal pneumonia and possible septic emboli (figure 3 and 4). Her blood culture grew gram negative rods identified as *Fusobacterium necrophorym*. A multidisciplinary team was involved and collectively started meropenem and heparin. She significantly improved during the hospital course and was discharged on Zoysn and Eliquis and advised to follow up with chest CT for pulmonary emboli after 4 weeks.

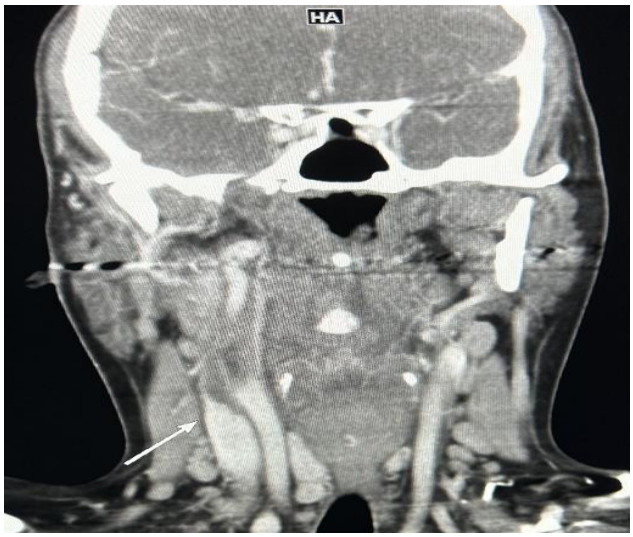

Figure 1: Thrombus is seen within the right internal jugular vein with near complete occlusion. IJ: internal jugular.

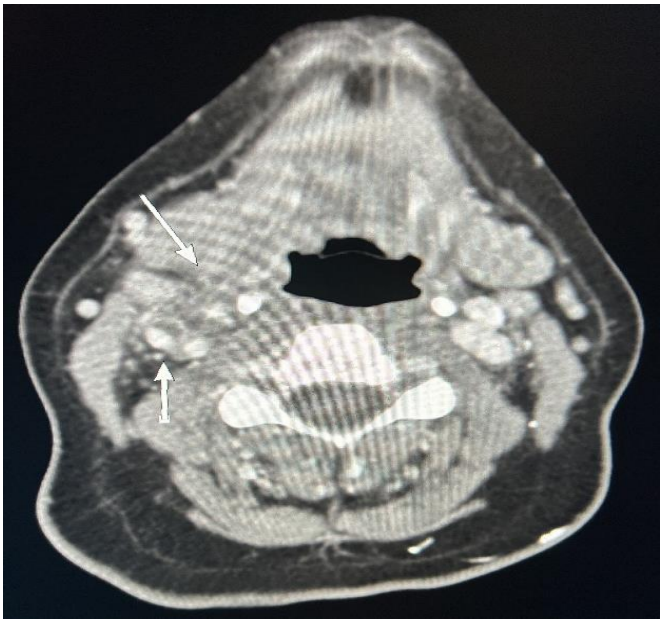

Figure 2: Thrombosis of the right internal jugular and common facial vein with near complete occlusion of the internal jugular vein and occlusion of the common facial vein. IJ: internal jugular.

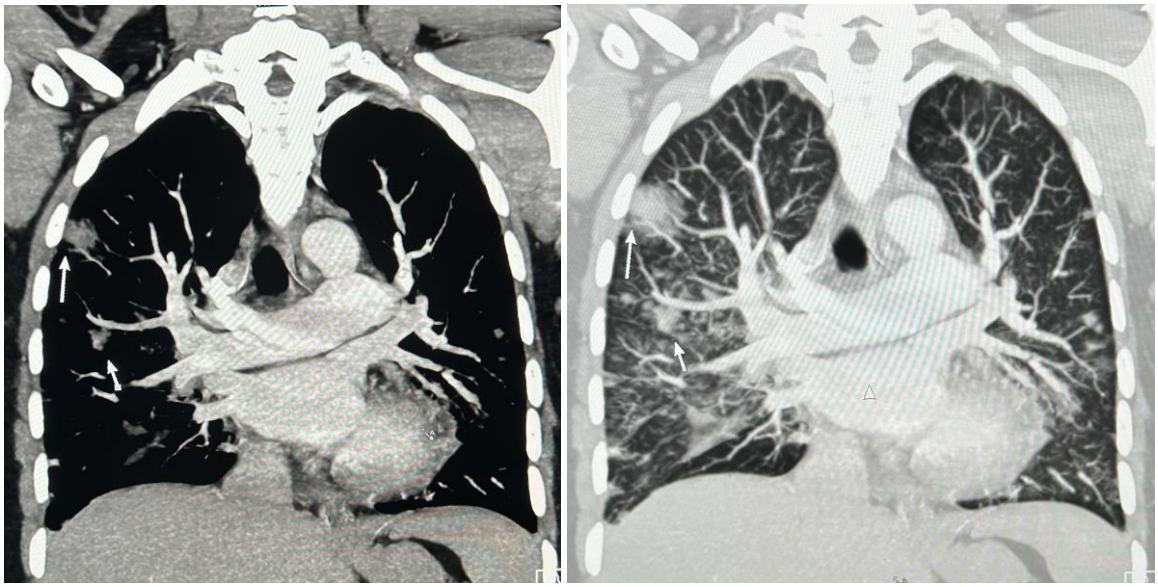

Figure 3: Multiple nodular opacities within visible portions of the lung apices consistent with septic emboli.

Discussion

Classical LS is diagnosed in the presence of anaerobic primary infection of the oropharynx, septicaemia with at least 1 positive blood culture, metastatic septic embolism to distant sites and thrombophlebitis of the IJV [3].

Fusobacterium, commonly found as a commensal organism in the oral cavity, gut and female genital tract, possess a myriad of virulence factors responsible for the development of necrotic abscess, septic thrombophlebitis, and thrombosis. For example, bacterial haemolysins lyse the erythrocytes, reducing oxygen transport to the site of infection and creating a favourable anaerobic environment for its growth. Similarly, hemagglutinin activity facilitates platelet aggregation leading to local site thrombosis, heat mediated endotoxin mediates an inflammatory response and stimulates production of tumor necrosis factor alpha, and lipopolysaccharides in the bacteria cell wall have strong endotoxin properties, which collectively contribute to the bacteria's pathogenicity [3-4]. A breach in the mucosal surface due to various infectious, inflammatory or neoplastic causes creates a pathway for bacterial invasion which subsequently, via direct invasion to local site of infection or lymphatic or haematogenous spread, leads to the suppurative thrombophlebitis or distant metastasis of septic emboli [3,5].

In our case, the suspicion of LS did not arise until the pathogen was identified in blood culture. The CT-scan revealed signs of septic emboli in both lungs, and if this examination had been performed before the results of the blood culture were available, LS would have likely been suspected sooner. Regardless, the final diagnosis would have been made when the blood culture was available because only then were all 3 diagnostic criteria of the disease met.

Although apical cavitary lesions, as shown in the CT-scan of our patient, could have other causes, such as tuberculosis, nontuberculosis mycobacteria (NTM), or fungal infections [6], we ascribe these radiological findings to the infection caused by *Fusobacterium necrophrorum*. First, these infections are most often associated with certain risk factors, such as immunosuppression [6,7] and exposure in an endemic setting. Our patient neither showed signs of immunosuppression nor had any history of likely previous exposure to these pathogens. Secondly, since the patient quickly responded to the initiated treatment, we did not find indication to investigate for other, less likely causes.

The principles of treatment include appropriate antibiotic therapy as well as consideration regarding the need for surgical intervention and anticoagulation. The regimen should consist of an antibiotic regimen that is resistant to beta-lactamase, since *F. necrophorum* beta-lactamase production and treatment failure with penicillin has been reported [1,8]. We do not favor use of empiric ampicillin-sulbactam because resistance rates are higher than for piperacillin or carbepenams [09,10]. It is uncertain whether anticoagulation may reduce propagation of thrombus or septic embolic events originating from IJV thrombosis; data are limited to case reports [11,12]. It is reasonable to discontinue anticoagulation once the patient has improved clinically and imaging suggests that thrombus extension has ceased.

Thus, it is likely that the diagnostic criteria, as they are established today, are not sensitive enough to include all patients with LS and need to be revised in order to effectively encompass all patients with LS. Finally, it is highly possible that due to the rarity of the disease, the physician has never experienced similar cases and therefore cannot narrow down the diagnosis quickly enough.

Conclusion

LS is a rare disease, and often the suspicion of the disease arises only when the blood cultures become available after several days. LS should be particularly suspected in young, otherwise healthy patients with a history of prolonged oropharyngeal infection and fever, especially if there are signs of metastatic infection, which most commonly occurs in the lungs. We hope physicians take the importance of chest X-ray, ultrasound, and CT scan results into consideration, as they can bring tremendous diagnostic value to initiating appropriate antibiotic therapy promptly. Furthermore, because of the rarity of the condition and lack of evidence-based treatment recommendations, management approaches for each patient should be individualized with involvement of multidisciplinary teams.

Conflict of interest statement – The authors declare no conflicts of interest.

All authors have contributed significantly to the writing and editing of this manuscript. It has been seen and approved by all authors. This manuscript has not been previously purchased and it is not being considered for publication elsewhere.

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