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Case report

A rare case of MRSA associated Lemierre's syndrome

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ABSTRACT

Lemierre's syndrome, or postanginal sepsis, is an uncommon but potentially fatal infection of the internal jugular vein. The combination of bacteremia, internal jugular vein thrombophlebitis, and metastatic septic emboli secondary to acute pharyngeal infections is characteristic of Lemierre's syndrome. Isolated pathogens are typically oral anaerobic bacteria, most commonly Fusobacterium necrophorum. While the incidence of Lemierre's syndrome has declined over the years, the proportion of cases caused by uncommonly implicated bacteria have been increasingly cited in the literature (1). In this case report, we introduce a novel presentation of Lemierre's syndrome in a patient who presented to the emergency department with neck swelling and shortness of breath and was found to have infectious myositis and bacteremia with methicillin-resistant *Staphylococcus aureus*. Clinicians should be vigilant of underlying thrombus in patients with neck swelling and infectious myositis as our patient's internal jugular vein thrombus was missed on initial computed tomography read.

Introduction

Lemierre's syndrome is a rare but often fatal infection of the internal jugular vein that is caused most often by *Fusobacterium necrophorum* and other anaerobic bacteria that colonize the oropharyngeal mucosa and gain access to the lateral pharyngeal spaces of the neck. This usually manifests as a septic thrombophlebitis of the internal jugular vein that is often preceded by a primary oropharyngeal infection [1]. There have been a few reported cases of Lemierre's syndrome associated with methicillin-resistant *Staphylococcus aureus* (MRSA) bacteremia, mostly in pediatric populations. In this case, we describe a rare cause of Lemierre's syndrome associated with MRSA bacteremia in a patient with infectious myositis of the sternocleidomastoid muscle.

Case description

A male in his 30 s with a past medical history of intravenous drug use presented to the emergency department with a four-day history of painful left-sided neck swelling and shortness of breath. He also reported neck stiffness and odynophagia. On the day of presentation, he developed fevers and pleuritic chest pain. He admitted to daily use of intravenous fentanyl using his right cephalic vein as an injection site. He denied any history of injections into the neck.

On presentation, the patient's temperature was 39.1 degrees Celsius, the blood pressure 115/70 mm Hg, the heart rate 120 beats per minute, the respiratory rate 32 breaths per minute, and the oxygen saturation 91 % while he was breathing ambient air. The patient was able to speak in full sentences and was in no obvious distress. His physical exam was notable for an obvious area of soft tissue swelling at the insertion of the sternocleidomastoid muscle into the sternoclavicular joint with overlying erythema of the skin. He had pain with active and passive movement of the neck. His lung sounds were coarse with rhonchi at the bilateral bases. He had no murmurs or peripheral stigmata of endocarditis. Focused examination of his skin revealed a raised area of necrotic tissue associated with phlebitis of the right cephalic vein at the site of reported injection.

Labs were remarkable for a WBC of 23.8 K/uL with 7 % bands, a hemoglobin of 9.5 g/dL, and a sodium level of 133 mmol/L. Inflammatory markers were elevated, including ferritin of 1457 ng/mL, ESR of 77 mm/h, and C-reactive protein of 19.8 mg/dL. Urine drug screen was positive for fentanyl, benzodiazepines, and cocaine. Blood cultures returned positive for MRSA for which the patient received intravenous Vancomycin.

Computed tomography soft tissue neck with intravenous contrast (Fig. 1) revealed infectious myositis of the left sternocleidomastoid muscle with surrounding cellulitis as well as a focal area of low attenuation in the inferior left sternocleidomastoid muscle suspicious for early abscess formation. Interventional radiology was consulted and determined that the abscess was not large enough to drain.

Computed tomography chest with intravenous contrast per

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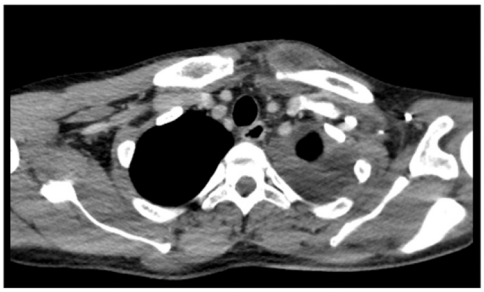

Fig. 1. Infectious Myositis of Left Sternocleidomastoid.

pulmonary embolism protocol (Fig. 2) revealed widespread nodular opacities affecting all lobes, consistent with septic emboli to the lungs. The patient went for transthoracic echocardiogram followed by transesophageal echocardiogram, neither of which demonstrated intracardiac vegetations.

While there were no notable filling deficits of the local vasculature near the site of pyogenic myositis on initial computed tomography read, given the patient's unimproved swelling on serial examination of the neck, radiology was called to determine whether local vascular obstruction was contributing. On re-evaluation of neck imaging, a filling defect in the left internal jugular vein was identified. Subsequent Doppler ultrasound revealed a non-occlusive thrombus of the left internal jugular vein. A diagnosis of Lemierre's syndrome was made based on radiographic evidence of a thrombus in addition to positive culture data. Vascular surgery was consulted for possible surgical intervention. Given that the thrombus was non-occlusive, anticoagulation with heparin was recommended.

The patient received two days of heparin followed by transition to oral Apixaban. He was continued on monotherapy with Vancomycin and was discharged with a peripherally inserted central catheter line to complete a six-week antibiotic course. He was continued on Apixaban for three months with repeat Doppler ultrasound showing resolution of the left internal jugular vein thrombus.

Discussion

Our case is consistent with a classic clinical presentation of Lemierre's Syndrome with an atypical bacterial etiology. Lemierre's syndrome has been described as a septic thrombophlebitis of the internal jugular vein, persistent bacteremia, and septic emboli most often involving the lungs. This entity is most commonly caused by oral anaerobic bacteria, with *Fusobacterium necrophorum* reported in 80 % of cases [2]. The mechanism of thrombus formation in such strains is attributed to lipopolysaccharide in the cell wall, which can activate human platelets in vivo and can lead to subsequent localized coagulation [3]. The production of a heat-stable toxin leukocidin is believed to be responsible for the inflammatory response, which further exacerbates thrombus formation [2]. The result is immunothrombosis; formation of a blood clot of fibrin and infected debris formed by the immune system to seal off bacterial infections, at the expense of thrombotic complications of these infections themselves.

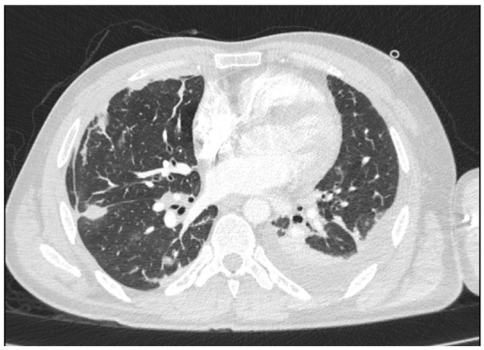

Fig. 2. Septic Emboli to the Lungs.

Other pathogens, such as in our case, are less commonly implicated. *Staphylococcus aureus* associated Lemierre syndrome is of particular concern due to its inherent virulence. *Staphylococcus aureus* has the ability to not only promote thrombosis as is the case in most bacterial infections, but also to degrade thrombosis by activation of fibrinolysis. *Staphylococcus aureus* secretes two coagulases, staphylocoagulase and von Willebrand factor binding protein; both activate prothrombin to generate fibrin [4]. It also possesses thrombolytic enzymes produced by extracellular proteins within the bacteria. In particular, production of staphylokinase (SAK) by *Staphylococcus aureus* serves to promote fibrinolysis by converting plasminogen into plasmin, which acts to cleave fibrin and dissolve clots. In addition to mediating fibrin degradation, the SAK/plasmin complex also activates matrix metalloproteinases, which can facilitate bacterial spreading through tissues by degrading extracellular matrix. The complex also diminishes the activity of host opsonic factors, further contributing to immune-evasion and bacterial spreading [5].

Staphylococcus aureus also has various surface receptors for fibrinogen that facilitate platelet binding and subsequent recruitment to sites of vascular damage. Clumping factor A (ClfA) and the fibronectin binding proteins A and B (FnVPA/B) are two surface receptors expressed by *Staphylococcus aureus* that bind to polymerized fibrin at sites of fibrin clots, and can also mediate platelet binding through bridging with the platelet αIIbβ3 (GPIIb/IIIa) receptor. *Staphylococcus aureus* also expresses von Willebrand factor binding protein, which binds to surface-anchored ClfA. The redundancy of receptors allows very effective binding of *Staphylococcus aureus* to the vascular wall. Furthermore, binding to the vascular wall can not only be direct, but also by binding to platelets that are recruited to sites of vascular damage [5].

While MRSA associated Lemierre's syndrome is rare, it is an important consideration given that standard treatment for Lemierre's syndrome includes mainly anaerobic bacterial coverage. The use of anticoagulants in Lemierre's syndrome remains controversial. It has been proposed that anticoagulation should be reserved for cases where thrombosis fails to resolve following antibiotic treatment, or following cases where thrombosis is extensive [6].

The diagnosis of Lemierre's syndrome can be challenging. In a review of the literature, only 25 % of patients with Lemierre's syndrome had radiographic evidence of thrombophlebitis [2]. A high index of suspicion for Lemierre's syndrome should thus be maintained in patients with swelling or tenderness along the lateral side of the neck. Definitive diagnosis requires specific diagnostic testing; most commonly with computerized tomographic scanning and sonography. This case highlights the importance of recognizing Lemierre's syndrome as a possible entity in patients with MRSA bacteremia, and more importantly, using diagnostic imaging to evaluate for a thrombus of the internal jugular vein in patients with myositis of the sternocleidomastoid muscle.

Ethical approval

All authors have agreed to authorship, read and approved the manuscript, and given consent for publication of the manuscript.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Olivia Mobarakai: Drafted the manuscript and medical care of the patient. Sofia Barrios: Researching the bacterium and discussion section. Ajit Singth: Editing manuscript and medical care of patient. Jarle Stone: Provided feedback on manuscript and medical care of patient.

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Olivia Mobarakai: Drafted the manuscript and medical care of the patient. Sofia Barrios: Researching the bacterium and discussion section. Ajit Singth: Editing manuscript and medical care of patient. Jarle Stone: Provided feedback on manuscript and medical care of patient.

Conflict of Interest

All authors report no potential conflicts of interest.

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