Lemierre's syndrome: A forgotten entity with a very rare presentation

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ABSTRACT

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CASE REPORT

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Keywords: Forgotten disease, ICA thrombosis, Lemierre's syndrome, Septic thrombophlebitis of internal jugular vein

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INTRODUCTION

Lemierre's syndrome, also known as postanginal septicemia or necrobacillosis was first reported in 1890. It was named after André Lemierre who explained the disease in 1936 [1]. It is usually caused by gram negative anaerobic bacillus *Fusobacterium necrophorum* [2]. However *Peptostreptococcus*, *Staphylococcus*, *Streptococcus* and *Proteus* have also been isolated. The disease progresses in several steps following a primary infection, which is usually pharyngitis (87.1%). Further invasion of deep neck spaces occurs, leading to internal jugular vein (IJV) thrombophlebitis (71.5%), and infective metastasis to distant sites most commonly to lungs (79.8%) [3]. Imaging and other investigations helps in the diagnosis. Surgery is seldom required as prolonged IV antibiotics have been documented as the mainstay of treatment.

CASE REPORT

A 34-year-old housewife presented with a short history of fever, and headache for 20 days; odynophagia, neck pain, right sided facial swelling for 10 days; and hemoptysis for five days. The patient denied any history

of seizures, altered sensorium, photophobia, and arthralgias. The past history was unremarkable.

On examination, the patient was ill looking, tachypneic, and febrile to touch. However, her vitals were normal with pulse rate 104/min, and blood pressure 130/80. General examination showed right sided facial swelling and tenderness along the right sided sternocleidomastoid. Oral examination revealed inflammation of right tonsillar fossa. In the systemic examination, there were fine infraaxillary crepitations in bilateral lungs; and neurological examination showed terminal neck rigidity; while the examination of other systems was unrewarding (Figure 1).

Laboratory data showed total leukocyte count 18,650 (normal range 4,500–11,000 per μ l) with 86% neutrophils. C-reactive protein (CRP) with value 31 mg/L (normal range 0–10 mg/L) and erythrocyte sedimentation rate (ESR) with value 62 mm/hr (normal range 0–20 mm/hr) were significantly raised; anti-nuclear antibody (ANA), anti-phospholipid antibody (APLA), protein C, protein S, Factor 5 Leiden, antithrombin, homocysteine were normal and HIV was non-reactive. Cerebrospinal fluid analysis revealed 40 cells with 98% lymphocytes while adenosine deaminase (ADA), proteins and sugar were insignificant.

Ultrasonography of abdomen and echocardiography were normal. However, electrocardiography showed sinus tachycardia. The chest radiograph showed bilateral patchy shadows with mild bilateral pleural effusion (Figure 2).

Magnetic resonance imaging scan of head and neck showed right peritonsillar abscess, thrombosis of bilateral proximal IJV along with the thrombosis of right sigmoid sinus and left retromandibular vein. It also suggested bilateral ethmoid and sphenoid sinusitis/abscess, with extension of abscess to bilateral masticator space caudally and cranially to dura leading to subdural abscess and leptomeningitis.

A computed tomography angiography chest (Figure 3) and neck (Figure 4) was done which revealed thrombosed left internal carotid artery (ICA), partially thrombosed left IJV, sphenoid sinusitis and cavitation in bilateral lung fields suggestive of septic emboli. Color doppler bilateral neck was done to assess the extension of thrombus in the vessels. Surprisingly, it showed involvement of both IJV (partially occluded) and ICA (80% occluded) of left side.

Blood (anaerobic and aerobic), urine, sputum, and throat cultures were negative as the patient had received antibiotics prior to admission in our hospital.

The patient was given intravenously ceftriaxone (1 g q12 h), vancomycin (1 g q12 h), and clindamycin (600 mg q8 h) for 4 weeks. She also received anticoagulation with LMW heparin (6 mg SC q12 h) and acenocoumarol (2 mg q24h) and INR was monitored subsequently.

Figure 1: (A) Patient on presentation with right sided facial swelling, (B) Patient after four weeks of receiving treatment, and (C) Oral examination showing right peritonsillar abscess.

Figure 2: (A) Chest X-ray at the time of admission, and (B) Chest X-ray at the time of discharge.

Figure 3: Contrast-enhanced computed tomography chest showing septic emboli.

Figure 4: (A) Computed tomography angiography showing partial left internal jugular vein thrombosis. (B,C,D) Computed tomography angiography showing left internal carotid artery thrombosis.

The patient responded to the therapy as evident on radiographic resolution of the thrombosis both in internal jugular vein and internal carotid artery.

DISCUSSION

Lemierre's syndrome, being a rare entity (incidence 1 in a million) [4] has been drawing our attention because of its increasing incidence and dreaded complications; reemergence of this disease is most likely due to the antibiotic resistance and discouragement of the use of antibiotics for sore throat.

This patient had been worked up to rule out the closely mimicking differentials which were disseminated tuberculosis, cavernous sinus thrombosis, autoimmune vasculitis and thrombophilias.

Lemierre's syndrome can have a spectrum of presentation ranging from mild pharyngitis to peritonsillar abscess. However, sinusitis, otitis media, mastoiditis, and odontogenic infections have also been described rarely. Sinusitis/sinus abscess (sphenoid and ethmoid) was probably the primary source of infection in our case (as per the history given by the patient), which involved the pharynx (peritonsillar abscess), lateral pharyngeal space, masticator space and the meninges. Meningitis in this case could be either due to direct spread from the sinus (sphenoid and ethmoid), or indirect spread through septic emboli (via ICA and IJV), or retrograde involvement of the sigmoid sinus (via IJV). There was involvement of the carotid sheath which was evident on CT angiography and color Doppler neck, leading to thrombosis of ICA and IJV. In our case, ICA thrombosis could be either due to its proximity to IJV or due to septic emboli. Though involvement of ICA is rarely reported in literature but in our case, this finding was very much evident on the imaging. Considering the endovascular nature of the disease, metastatic spread most commonly involves the lungs (79–100%) [3]. However, joints, bones, skin, liver, and kidney may also be involved.

A prolonged antibiotic course (2–6 weeks) is needed in order to eradicate the microorganisms within the thrombus. The role of anticoagulation is controversial, however, some authors consider it if there is extension of thrombosis. Surgical intervention is seldom required in the present era.

Untreated cases may have mortality of up to 90% [5]. The prognosis of Lemierre's syndrome is favorable if there is early diagnosis and treatment. Rare presentation of Lemierre's syndrome, ICA thrombosis along with IJV thrombosis, meningitis and sinusitis are being highlighted in our case report.

CONCLUSION

Lemierre's syndrome occurs primarily in young, otherwise healthy individuals and is characterized by a history of recent oropharyngeal infection, clinical or radiological evidence of internal jugular venous thrombosis and anaerobic bacteremia caused primarily by *F. necrophorum*. This is a rare illness in the modern era of antibiotic therapy, though it has been reported

with increasing frequency in the 21st century. Lemierre's syndrome should be suspected in young, healthy patients with prolonged symptoms of pharyngitis followed by symptoms of septicemia or pneumonia, or an atypical lateral neck pain. Diagnosis is often confirmed by the identification of internal jugular vein thrombophlebitis by an imaging study and growth of anaerobic bacteria on blood culture. Prolonged antibiotic therapy is the cornerstone of the treatment, occasionally combined with anticoagulation.

Author Contributions

Sailesh Kumar Bansiwal – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Prabal Rajvanshi – Substantial contributions to conception and design, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Harshita Sharma – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Rajesh Manocha – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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SUGGESTED READING

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- <https://www.ncbi.nlm.nih.gov/gquery/?term=+Lemierre%E2%80%99s+syndrome>
- <https://www.readbyqxmd.com/read/20107834/eponym-the-lemierre-syndrome>
- www.medscape.com/viewarticle/410830

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