IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)
e-ISSN: 2279-0853, p-ISSN: 2279-0861.Volume 21, Issue 1 Ser.13 (January. 2022), PP 01-05
www.iosrjournals.org

Lemierre's syndrome with right IJV thrombosis extending to right subclavian, axillary and brachial vein and managed by thrombolysis- A case report of a missed and forgotten entity of life threatening condition.

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Abstract
Lemierre's syndrome is a septic thrombophlebitis of the internal jugular vein (IJV) commonly caused by anaerobic oropharyngeal flora that is usually followed by fulminant sepsis and rapid death in the pre-antibiotic era. Since the introduction of antibiotics, the morbidity and mortality associated with this syndrome have been dramatically decreased. However, delay in diagnosis and antibiotic treatment resulting in poor clinical outcome is not uncommon as physicians are less familiar with this infection and initial manifestations are often non-specific. We describe a case of a patient with Lemierre's syndrome with thrombosis of right internal jugular vein extending to subclavian, axillary and brachial veins and managed with broad spectrum antibiotics and thrombolysis with streptokinase.

Date of Submission: 07-01-2022 Date of Acceptance: 21-01-2022

I. Review

In 1936, Andre Lemierre published a case series of 20 patients with a syndrome characterized by a history of recent oropharyngeal infection, clinical or radiological evidence of internal jugular (IJ) venous thrombosis, and anaerobic septicemia caused primarily by Bacillus funduliformis (now known as Fusobacterium necrophorum) [1]. Lemierre classified this syndrome as "anaerobic postanginal sepsis" because of the onset of sepsis occurring shortly after the patients had experienced a sore throat. It was not until the 1980s that anaerobic postanginal sepsis was routinely referred to as Lemierre's syndrome [2]. Andre' Lemierre stated that septicemia and distant septic emboli could originate from the nasopharynx, mouth, jaws, otitis media, and urinary passages or during mastoiditis, purulent endometritis, and appendicitis [3-5]. With the introduction of antibiotics in the 1940s and their widespread use for streptococcal pharyngitis, the incidence of Lemierre's syndrome fell dramatically. In fact, some authors in the 1980s and 1990s referred to it as a "forgotten disease" [6-7].

II. Case Report

A 52 yr old male was admitted in emergency department with history high grade fever, swelling of right side of face, neck and right upper limb for last 7 days. Patient gave history of sore throat and pain neck prior to swelling. The sore throat was severe enough to affect his intake of food, as he was unable to swallow solids, but liquids were unaffected. Later patient developed severe swelling and heaviness of right upper limb associated with mild pain. On admission, there was gross edema extending from right side of neck, shoulder till right hand. Oral examination revealed injected pharynx with indurated and swollen right tonsil and tonsillar pillars (Figure 1). His temperature was 39.9∘ C, respiratory rate was 18 breaths/min, blood pressure was 140/90 mmHg, and heart rate was 118 bpm.

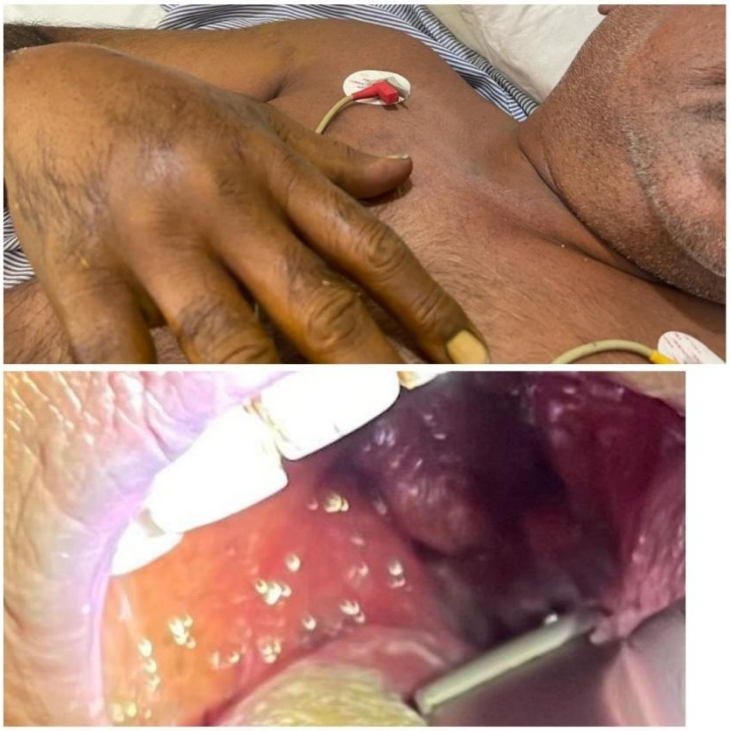

Figure 1- Patient presented with swelling of right side neck and upper limb with injected pharynx and edematous right tonsil.

The patient's blood results showed raised C-reactive protein (CRP) of 108 mg/L, haemoglobin of 140 g/L, the white cell count was 17900/cmm with raised absolute neutrophil count at 8.50 × 109 /L. Platelet count was also normal at 212 × 109 /L. The chest X-ray showed prominent vascular appearances to the hilar contours, but no obvious paratracheal hilar lymphadenopathy. There was also a slight increase in perihilar bronchovascular markings. Doppler ultrasound of the neck and right upper limb showed dilated right internal jugular vein(IJV), subclavian vein (SCV), axillary vein (AV) and brachial vein (BV) and filled with hypoechoiec material showing the thrombosis, marked soft tissue subcutaneous edema, reactive lymphadenopathy in neck(Figure 2 and Figure 3). At this point a diagnosis of Lemierre's syndrome was made. Blood cultures were taken on admission and were returned negative five days later as patient had already received antibiotics outside.

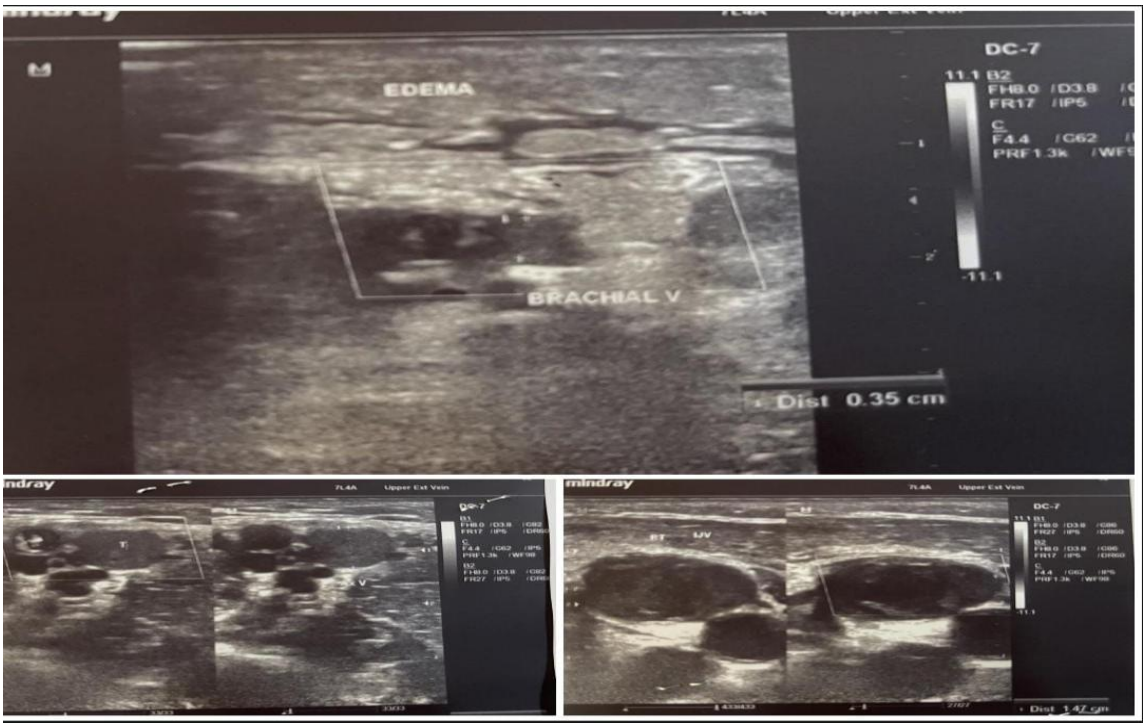

Figure 2- Doppler USG suggestive of thrombosed right IJV, SCV, AV, BV with no flow on color doppler

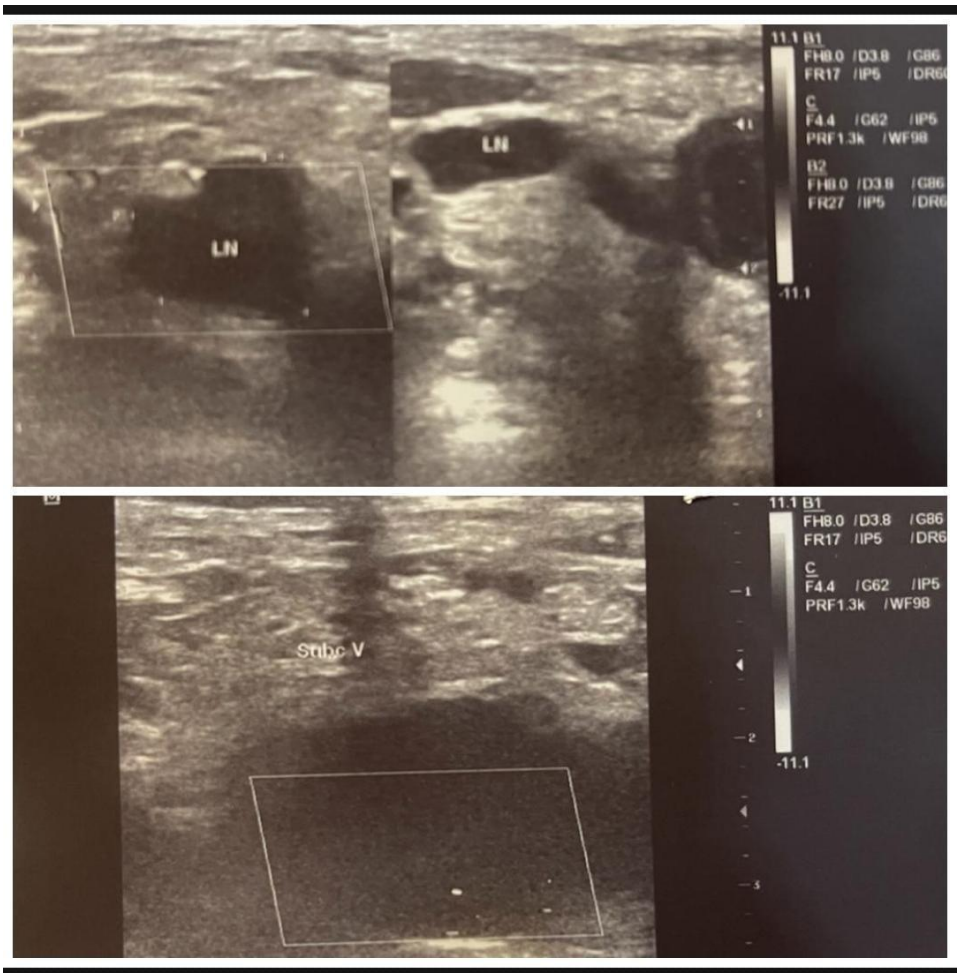

Figure 3- Color Doppler revealed no flow in right subclavian vein and lymphadenopathy on USG neck

The patient was started on Piperacillin/tazobactam (4.5gm, IV, tds) and clindamycin (600mg, IV, bd) was continued for 5 days. Decision for thrombolysis with streptokinase was taken in consultation with intensivists owing to extension of thrombosis in deep veins of right upper limb. Patient's swelling subsided substantially after thrombolysis with streptokinase and partial recannalisation and color flow was seen in right IJV, SCV and axillary veins on color doppler. He was discharged home with complete 2 weeks course of antibiotics and anticoagulation. At a clinic visit 1 month later, the patient was doing well with only a complaint of mild swelling in right shoulder region. The patient attended a follow-up outpatient appointment eight weeks later. His inflammatory markers had fallen to within the normal range, no fever, sore throat, neck an upper limb swelling subsided substantially.

III. Discussion

Lemierre's Syndrome is a largely forgotten disease in the era of antibiotics, with an incidence of 1 in 1 million per year. In 1936, Andre' Lemierre described a syndrome with an illustrative focus on post-anginal septicemia due to Fuso-bacterium necrophorum and complicated with internal jugular vein (IJV) thrombophlebitis and distant septic emboli [1,2]. Before the development of antibiotics, this syndrome was common and had a fatal course within 7-15 days [3].The incidence of Lemierre's syndrome decreased during the antibiotic era.2,3It has gained legendary status with few cases reported and as a "forgotten" and delayed diagnostic disease resulting in various complications and mortality.

A review of the typical presentation of Lemierre's syndrome highlights some of the classical signs and symptoms described in this case presentation. The illness typically begins with a fever reaching 39–41◦ C, the first sign of septicaemia, which may or may not be accompanied by rigors [5]. The septicaemia is most commonly preceded by a sore throat which usually occurs 4-5 days before all other symptoms, but in some cases has been up to 12 days before [4]. The presentation of the sore throat varies, with many showing a normal appearance of the oropharynx [5]. However, in some cases, a severe exudative tonsillitis accompanied by peritonsillar abscess has been documented and may be severe enough to cause dysphagia [6]. Neck pain and stiffness are commonly described, and bilateral or unilateral cervical lymphadenopathy may be present, commonly in the anterior triangle. Patients may also exhibit an induration of the internal jugular vein. Lemierre himself described a triad of pleuritic chest pain, dyspnoea, and haemoptysis and the presence of localised crackles and a pleural rub on auscultation [7]. Chest radiographs frequently show multiple nodular infiltrates throughout both lungs, although it is not unusual for radiographs to be normal as reported here. A metastatic infection found in Lemierre's syndrome can also manifest as septic arthritis, osteomyelitis, meningitis, pericarditis, and hepatic abscesses [8].

Diagnosis of Lemierre's syndrome is established on the presence of thrombus in IJV and positive blood culture. Computed tomography of neck with contrast is the diagnostic modality of choice to demonstrate the thrombus. Doppler ultrasonography is an alternative since it is less invasive and can be done at bedside though it is less sensitive particularly in the area deep to clavicle and mandible and can miss newly formed thrombus with low echogenicity.[10]

Prolonged antibiotic therapy constitutes the mainstay of treatment of Lemierre's syndrome in the modern era. Since no controlled clinical trials exist to identify an optimal antibiotic regimen, decisions must be based on known in vitro sensitivities together with anecdotal clinical evidence

The use of anticoagulation is controversial, and no controlled studies exist. Case series have reported 21%−30% of patients are treated with anticoagulation [7]. A few authors have advocated for the use of anticoagulants in all cases of Lemierre's syndrome [11]. Others have recommended anticoagulation only if thrombosis extends into the cerebral sinuses or if there has been no improvement in symptoms with antibiotic therapy. In this case, however, owing to extensive thrombosis and clinical presentation thrombolysis was done which has yet not been reported in these cases.

Surgical treatment of Lemierre's syndrome may involve drainage of abscesses in the neck, most commonly peritonsillar or lateral pharyngeal abscesses

IV. 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 IJ venous thrombosis and anaerobic bacteremia caused primarily by F. necrophorum. Blood cultures, chest radiographs, and contrast enhanced CT scanning should be definitive enough to provide a diagnosis. In this individual, the diagnosis was consistent with this syndrome, but confirmation and subspeciation of the infective aetiological agent were elusive due to prior antimicrobial therapy. . Diagnosis is usually clinical with supporting radiological evidence and growth of a typical causative organism, usually from blood. It is often mistakenly diagnosed as primary bacteremia from an infected organ or tonsillitis. Treatment consists of prolonged antibiotics and management of its complications.

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