Case Report

A Case of Septic Thrombophlebitis of Internal Jugular Vein - Lemierre's Syndrome

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ABSTRACT

Lemierre's syndrome refers to septic thrombophlebitis of the internal jugular vein (IJV). The condition typically begins with oropharyngeal infection and frequently involves inflammation of the vein wall, infected thrombus within the lumen, surrounding soft tissue inflammation, persistent bacteremia, and septic emboli.

Introduction :

Lemierre's syndrome was first described by a French bacteriologist, Andre-Alfred Lemierre, in 1936, when he examined 20 patients presenting with oropharyngeal infection followed by anaerobic bacteremia, thrombophlebitis of internal jugular vein and secondary septic emboli most commonly to the lungs.[1] Fairly common in the pre-antibiotic era, Lemierre's syndrome is now a rare entity with a mortality rate ranging from 4-18%, and an incidence between 0.6-2.3 per million people.[2] Indeed, it had been labelled as a 'forgotten disease'. But there has been an increasing trend of incidence or reporting of this condition over the past 10 years.[3]

Case Report :

A 55 year old male was brought to the emergency department with complaints of fever and altered sensorium since one day. The fever was of high grade, and associated with chills and rigors. Patient had a history of right-sided tooth ache for fifteen days prior to presentation. Patient developed swelling over the right cheek which gradually extended to the right eye and periorbital tissue over two days prior to presentation. The swelling was associated with increased watery discharge from the right eye. Patient had a history of dental caries in the past for which he was taking treatment from a local doctor. He had no comorbidities. He was a chronic bidi smoker and a tobacco chewer. No h/o past surgeries or hospital admissions was present.

On general examination, the patient was found to be febrile with a core body temperature of 38.7ºC. He was semi-conscious and disoriented. His pulse rate was 122 beats/min, and he was tachypneic. His blood pressure was 80/50 mm Hg. On local examination of his face, there was erythematous swelling around his right eye and over his right cheek. On oral examination, purulent discharge was seen coming through the periodontal area of his right upper premolar. On auscultation of his chest, there were basal crepitations bilaterally with rhonchi in both the lung fields. Air entry was slightly reduced in right infra-axillary area. His pupils were of normal size and were reactive to light. He was moving all four limbs and his plantar response was equivocal. Cardiac auscultation and per abdominal examination were within normal limits. The patient was immediately admitted to the Intensive Care Unit (ICU) for further evaluation and stabilization.

A complete blood count showed a total WBC count of 21,000/uL, with 83% neutrophils. Liver and renal parameters were within normal limits. Serum electrolytes were normal. NT-Pro BNP was significantly raised (25,000 pg/mL). Pus swab culture showed colonies of Pseudomonas aeruginosa and Methicillin sensitive Staphylococcus aureus. CT head (plain) showed no significant abnormality.

Local USG of the face and neck region revealed evidence of a hyperechoic thrombus in the right Internal Jugular Vein with mild to moderate luminal

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Received on 21st December 2020 Accepted on 27th December 2020

compromise. Right sided orbital, maxillofacial, pre-auricular and post-auricular regions showed diffuse subcutaneous edema and increased vascularity, suggestive of infective etiology/cellulitis. Acontrast CT study of the face and neck region revealed a relatively well-defined, peripherally enhancing hypodense collection of approximate size 2.5 X 1 X 3.1 cm (TR-AP-CC) involving right medial pterygoid muscle, suggestive of abscess formation. There was also evidence of a partial eccentric filling defect in the right Internal Jugular Vein (IJV) ***(Fig. 1),*** extending from the lower endplate of C2 vertebra to lower endplate of C6 vertebral level, suggestive of right IJV partial thrombosis.

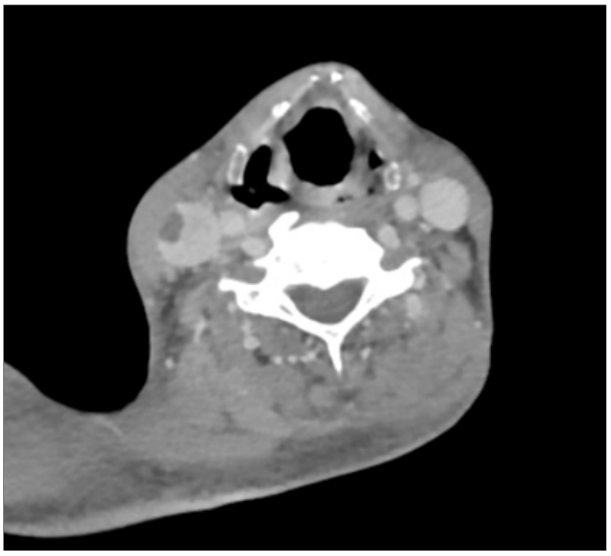

Fig. 1 : **CT neck revealing thrombus in the right Internal Jugular Vein (Black arrow)**

Chest CT study revealed multiple small (10-15mm) non-enhancing nodular areas in the lung parenchyma in the right upper lobe with surrounding consolidatory changes, suggestive of septic emboli with abscess formation. There was also evidence of gross right-sided and moderate left-sided pleural effusion with underlying segmental atelectasis of right upper and lower lobes ***(Fig. 2).***

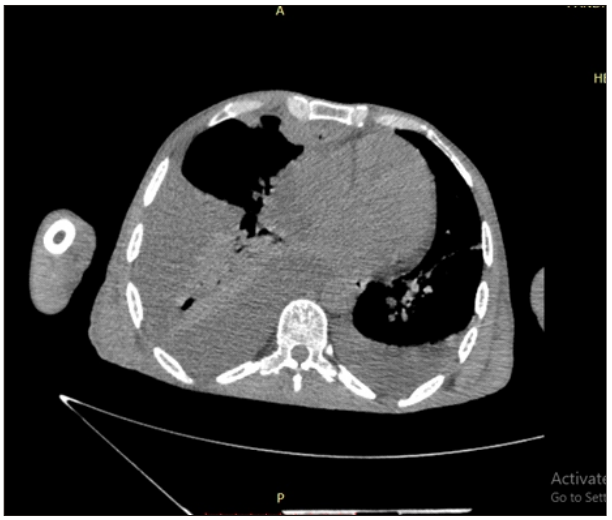

Fig. 2 : **High resolution CT of the chest revealed bilateral pleural effusion with multiple nodules in the right lung upper lobe**

Final Diagnosis :

Septic thrombophlebitis of right IJV (Lemierre's syndrome).

Discussion :

The diagnosis of Lemierre's syndrome is based on the following criteria - 1. History of recent oropharyngeal / dental infection, 2. Evidence of Internal Jugular Vein thrombosis on imaging study (Doppler imaging / CECT neck veins), 3. Evidence of septic embolization to distant sites (mainly, lungs), as seen on HRCT thorax in our case, or isolation of Fusobacterium necrophorum, an anaerobic bacteria living as a commensal in oropharyngeal mucosa, on blood cultures.[4,5] Other pathogens that could be involved in causing *Lemierre's syndrome include Streptococcus, Staphylococcus aureus, Pseudomonas aeruginosa and Enterococcus.F. Necrophorum* is a gram positive obligate anaerobic bacteria that is rather difficult to culture, and has a longer incubation period for growth than other bacteria.6Thus, a clinical diagnosis of Lemierre's syndrome is still valid if the bacteria go undetected or the causative organism turns out to be any other pathogen.

The palatine tonsils and peritonsillar tissue are the primary source of infection in the majority of cases, although pharyngitis, parotitis, otitis media,

sinusitis, odontogenic infection and mastoiditis have been described as causes of the syndrome.[7,8] The infection spreads to the IJV via several anatomic routes, including propagation of thrombophlebitis from local oropharyngeal veins to the IJV, drainage of septic lymph leading to perivenous inflammation and ensuing IJV thrombophlebitis, and direct extension of infection through the neck tissue. The septic thrombus formed in the Internal Jugular Vein then usually embolize to the pulmonary capillaries.[9] As a consequence, the most frequently involved site of septic metastases are the lungs, followed by the joints (knee, hip, sternoclavicular joint, shoulder, and elbow).[10] Other sites involved in septic metastasis and abscess formation are the muscles and soft tissues, liver, spleen, kidneys, and central nervous system.[9]

Ultrasonography with Doppler imaging is an easily available modality for imaging and identifying thrombosis in the neck veins, although it is less sensitive in detecting early thrombophlebitis and visualizing deeper tissues of neck and that beneath mandible and clavicle.[11] CT has the ability to show distended veins with enhancement of the walls, intramural filling defects, and swelling of adjacent soft tissues.[12]

The main modes of treatment include antibiotics and, if necessary, surgical drainage of the source of infection. Penicillin in combination with metronidazole or clindamycin monotherapy is usually preferred. Many reports describe prolonged intravenous therapy, i.e., 6 weeks or more.[13]

Phan and So suggested the use of anticoagulation especially in patients with poor clinical response despite antibiotics therapy and also patients with predisposing thrombophilia.[14] Schubert et al. and Rebelo et al. in their studies recommended anticoagulation in all cases of septic thrombosis of the ENT region, confronted with the serious clinical consequences of cranial thrombosis.[15,16] Overall, patients who show poor clinical response to antibiotics in the first 48 hours[14], those with intracranial thrombosis[17], and those with predisposing thrombophilia[18,19], are more likely to benefit from anticoagulation. The decision to start anticoagulants ultimately depends on the treating physician. Surgical treatment of Lemierre's syndrome may involve drainage of abscesses in the neck, most commonly peritonsillar or lateral pharyngeal abscesses.

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. Diagnosis is often confirmed by the identification of IJ vein thrombophlebitis by an imaging study and growth of anaerobic bacteria on blood culture. Prolonged antibiotic therapy is the cornerstone of treatment, occasionally combined with anticoagulation.

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