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Case Reports

LEMIERRE'S SYNDROME –A Rare Fusobacterial Complication

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Abstract: Lemierre's syndrome is a rare complication following an acute episode of upper respiratory tract infection. *Dr. André Lemierre*, a French bacteriologist in 1936, who first published 20 cases in lancet out of which only 2 survived. The causative agent is typically Fusobacterium. The number of cases of Lemierre's syndrome subsequently declined with the introduction of antibiotics (1940) and widespread use of antibiotics to treat tonsillitis. With the increase of antibiotic resistance and a greater reluctance to prescribe antibiotics for minor conditions such as tonsillitis and pharyngitis, there are now concerns developing about the re-emergence of the condition. This increasing prevalence along with unfamiliarity of clinicians with the classical features of this syndrome may result in the misdiagnosis or delay in diagnosis of this potentially fatal illness. If left untreated, the mortality rate is over 90%. We report a case of 48-year-old male diagnosed with this condition and successfully treated, any delay in the diagnosis could have been fatal.

Keywords: Lemirre's Syndrome, Fusobacterium, Internal Jugular Vein Thrombosis, Tosillopharyngitis, Fusobaterial Complication

Introduction

The incidence of Lemierre syndrome has been estimated at between 0.6 and 2.3 per million. Fusobacterium necrophorum which is a obligate anaerobe and a gut commensal has been described as the most common etiological agent, with positive cultures in 81.7% of patients (Chirinos *et al*., 2002), occasionally F. nucleatum, F. mortiferum and F. varium can also leads to this complication. Brazier (2006; Huggan and Murdoch, 2008; Kristensen and Prag, 2000; Kushawaha *et al*., 2009). The fusobacterium is considered as the normal commensal of oral cavity and gi tract, the pathogenesis of the syndrome is unknown but some theories believe that Invasion of viral e.g., ebstein barr virus or other bacterial infection in oropharynx can facilitate the invasion of fusobacterium through mucosa, with the formation of a peritonsillar abscess. When the abscess wall ruptures internally, the bacteria can spread through the soft tissue and infects the nearby structures, which can lead to involvement of internal jugular vein (Brazier, 2006; Huggan and Murdoch, 2008; Kushawaha *et al*., 2009). As a consequence of this bacteraemia it can lead to thrombus formation in internal jugular vein and this also acts a source of metastatic septic emboli to distant organs (Lemierre, 1936; Huggan and Murdoch, 2008; Forrester *et al*., 1985). In addition to internal jugular vein it can also involve some branches of external jugular vein (Morris *et al*., 2006). Lemierre's syndrome remains a rare condition, with one retrospective study from Denmark estimated an incidence of around 1 case per 1000000 (Dagan and Powell, 1987).

Case Report

About 48-year-old male presented with fever for 7 days, Cough since last 7 days and neck swelling/pain from last 2 days along with headache and vomiting. Fever was intermittent, cough was productive with blood, neck swelling was acute in onset and progressive. He was treated for fever by general physician with antibiotics and analgesics. On examination there was swelling of right side of neck (Fig. 1) (black arrow), his temperature was 4°C, His chest exam showed bilateral bibasilar crackles. Oral examination demonstrated erythema of the pharyngeal mucosa and tonsils. The neck was tender to palpation but brudinsky and kernig sign were absent. Cervical lymphadenopathy along with dilatation of the internal jugular vein.

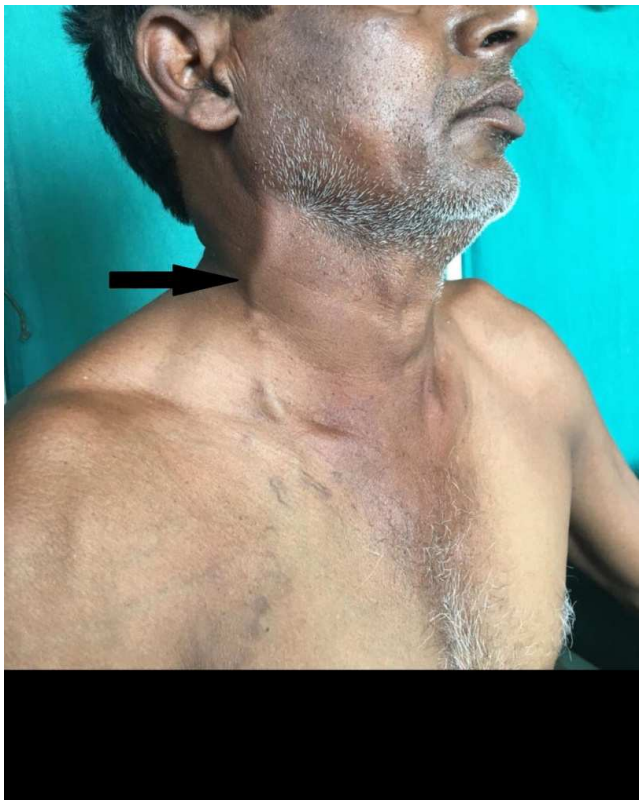

Fig. 1: External ugular vein distension

The lab investigation of patient showed:

- CBC-. Haemoglobin-11.8 gm/dl(13.5-17.5gm/dl) , Total leucocyte count-11500/cumm (4000-11000/cumm), Total RBC count- 3.72 million/cumm (5-6 million/cumm) Alkaline phosphatase – 125U/lt(44-147U/lt), Total protein-6.7(6-8.3), A/G- 1.1(0.8-2.0).Patient was negative for HIV1 AND 2, HBsAG, HCV, VDRL. Urine analysis was normal
- CECT finding- There is dilation of right jugular vein, on post contrast imaging right internal jugular vein reveals filling defect in lower part of neck. Upper part of vein is showing sluggish enhancement of post enhancement contrast (Fig. 2) when a patient with any type of infection of head and neck develop signs of thrombophlebitis of internal jugular vein the level of suspicion for this syndrome become very high. Blood culture were negative, because of antibiotics course. After the cect the patient developed right arm swelling and tenderness which was due to contrast allergy. So, CECT chest was not possible. 2D ECHO was performed which was normal. xRay-Showed unilateral middle lobe consolidation (Fig. 3)

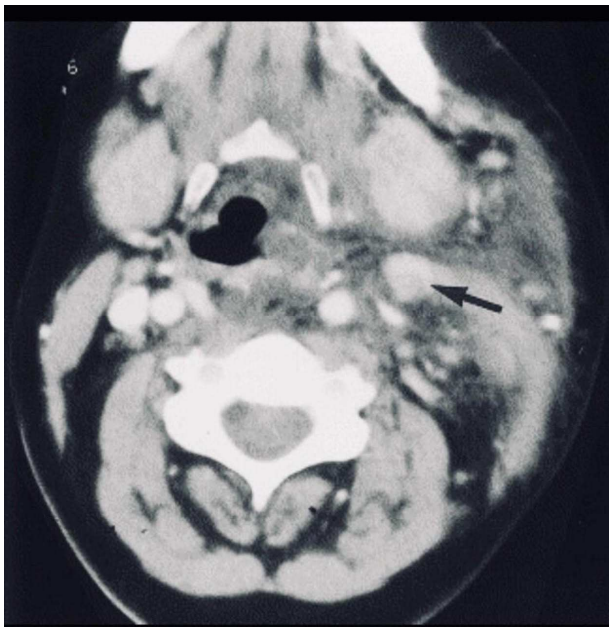

Fig. 2: Right jugular vein dilation on Contrast enhanced computerized tomography

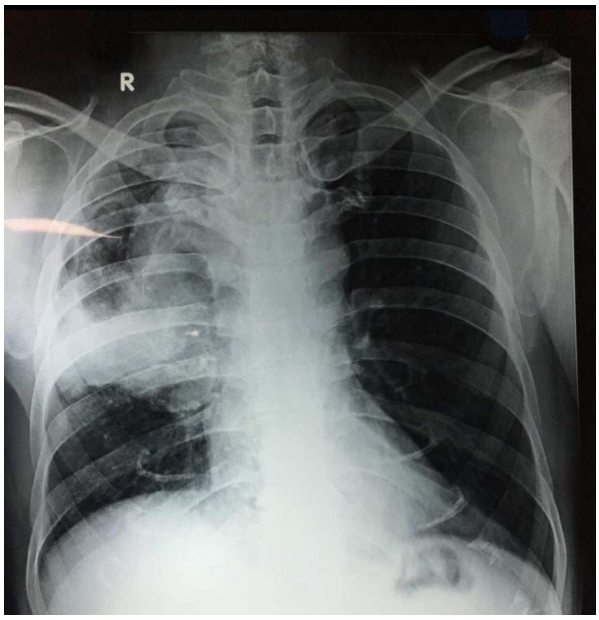

Fig. 3: X-ray showing consolidation in middle and lower part of upper lobe in right lung field

Patient was started with amoxicillin(1g) tds and oral metronidazole (400 mg) tds. He was also started on warfarin(5mg) od and dalteparin at the time of admission. The patient was asymptomatic on discharge, he continued antibiotics till 12 days after discharge and he continued to take warfarin for 90 days. Patient attended follow up after eight weeks, he was asymptomatic and the neck swelling was resolved.

Discussion

Following a more acute presentation and appropriate evidence from computed tomography, we diagnosed Lemierre's syndrome. Detection of a *Fusobacterium* spp. from blood culture will provide the confirmation to diagnosis (Brazier, 2006; Eilbert and Singla, 2013). However, culturing may take upto few days and prior treatment with antibiotics will decrease chance of detection of the bacteria in culture as was in this case (Eilbert and Singla, 2013; Riordan and Wilson, 2004). F. necrophorum and F. nucleatum are discriminated from other species by their abilities to grow in 20% bile, produce indole, display lipase activity and form gas in glucose agar (Hagelskjaer and Prag, 2000; Sinave *et al*., 1989). Furthermore, F. necrophorum have unique property of lactate fermentation to propionate and haemolysis (Sinave *et al*., 1989). It can classically present as fever reaching 39-41 degree Celsius, which may or may not be accompanied by rigors (Riordan and Wilson, 2004). Sore throat usually occurs few days before the septicaemia, it can be with no findings in oropharynx or with exudative tonsillitis (Riordan and Wilson, 2004). Patients may also exhibit an induration of the internal jugular vein, slightly inferior to the sternocleidomastoid muscle's anterior border. The infection can be metastatic leading to osteomyelitis, pulmonary involvement, pericarditis, arthritis, meningitis, hepatic abscess (Sinave *et al*., 1989). Septic arthritis which can occur in 13-27% of cases, typically affecting the hip joint and osteomyelitis in 3% (Riordan and Wilson, 2004). Hepatomegaly due to liver involvement and abdominal pain is common (Alherabi, 2009) Contrast enhanced CT is most specific for the diagnosis (Kushawaha *et al*., 2009). CT scan is always better than ultrasonography (Kushawaha *et al*., 2009).

Antibiotic coverage to include anaerobic organism should be started as soon as diagnosis is suspected and should last from 3 to 6 weeks. We used amoxicillin along with metronidazole in this case however, according to Katrine M Johannesen carbapenem and piperacillin/tazobactam were commonly used (Johannesen *et al*., 2016). Surgical exploration may be indicated.

Use of anticoagulant is controversial, according to Amaro *et al*., routine anticoagulation is not advisable, because of risk of hematogenic dissemination of the infection and should be reserved for the cases in which there is retrograde progression of thrombus in the direction of cavernous sinus.

Conclusion

Through this case report we want to discuss this rare outcome of a common infection, so that the diagnosis should not be delayed and infection like tonsillitis should not be under treated. Primary prevention of this syndrome should be kept in mind while dealing with tonsillitis due to its high mortality. We also wanted to discuss the controversial role of anticoagulants through this case report.

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Author's Contributions

Chandan Kumar: Analysis and interpretation of data, edited the content for critical information. Involved in drafting the manuscript. Final approval for publication.

Abhishek: Contributed to conception and design, revised the content for critical information, wrote the manuscript. Final approval for publication.

Pal Satyajit Singh Athwal: Contributed to conception and design of manuscript as well as collection and analysis of information. Involved in drafting the manuscript. Final approval for publication.

Sunny Khari: Involved in writing the manuscript, edited the content and approved for pulication.

Anil Kumar Kem: Revied the manuscript for critical information, analysis and interpretation of data. Final approval for publication.

Ethics

Written informed consent was obtained from the patient for this case report and the images.

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