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**Case Report**

# The challenges of a Lemierre's syndrome in COVID-19 times

**Vikram Kemmannu Bhat*, Jino Johns Lalitha, Suhas Suresh Desai, Nidhi Mohan Sreedevi**

Department of Otolaryngology, Head and Neck surgery, Karnataka Institute of Medical Sciences, Hubli, Karnataka, India

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***Correspondence:**
Dr. Vikram Kemmannu Bhat,
E-mail: vikram.ent@gmail.com



## ABSTRACT

Lemierre's syndrome is a type of necrotizing fasciitis of the head and neck that is an uncommon rapidly spreading infection involving the skin, subcutaneous tissue and the fascia leading to life-threatening complications like septicaemia, bone marrow suppression, disseminated intravascular coagulation and multi-organ failure. The management of this disease, especially in COVID-19 pandemic is a challenge as it is associated with high morbidity. A 46-year-old male presented to us with complaints of swelling in the neck with discoloration of skin in the neck and chest. Computed tomography (CT) scan of the neck revealed thrombosis of the right internal jugular vein (IJV) and diffuse abscesses on both sides of the neck. A high index of suspicion is required for the diagnosis of Lemierre's syndrome in the absence of typical clinical features. The rapid, unpredictable dissemination of infection and occult thrombus within the IJV make both diagnosis and management a challenge especially, during COVID-19 times.

**Keywords:** Necrotizing fasciitis, Deep neck infection, Lemierre's syndrome

## INTRODUCTION

Lemierre's syndrome is a condition characterized by clinical and radiological evidence of IJV septic thrombophlebitis and bacteraemia caused primarily by the anaerobic organism *Fusobacterium necrophorum*.[1] It was once called the forgotten disease because of its rarity, but it may not be that uncommon at all.[2] It was brought to broader evidence and was described by Andre Lemierre in 1936.[3]

The disease is notorious for causing abscesses at distant sites like the liver, lung, kidney as a result of septic emboli. The management of this disease, especially in COVID-19 times, is a challenge as it is associated with high morbidity. There is no consensus or proper guidelines in addressing IJV thrombosis and tackling multi-organ failures.

## CASE REPORT

A 46-year-old male presented with complaints of swelling in the anterior aspect of the neck for the past 5 days with skin discoloration on the neck and chest (Figure 1). He also had a history of fever for 5 days with chills and rigors. There was difficulty in breathing and swallowing for 2 and 4 days respectively. He had tooth ache 3 weeks back in the lower jaw. He had no history of diabetes mellitus, hypertension or tuberculosis. On examination, a soft, fluctuant tender swelling was noted under the lower jaw with gangrenous changes of skin below the swelling. The patient underwent a contrast-enhanced computed tomography scan (CECT) of the neck and upper chest after admission. It revealed right IJV thrombosis (2.7 cm long), right submandibular sialolithiasis along with necrotizing features of multiple pockets of deep diffuse abscesses and gas on both the sides of the neck (Figure 2).

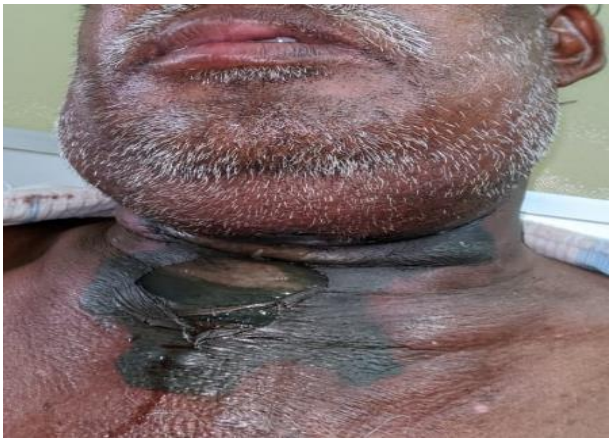

**Figure 1: The pre-operative gangrenous involvement of the skin.**

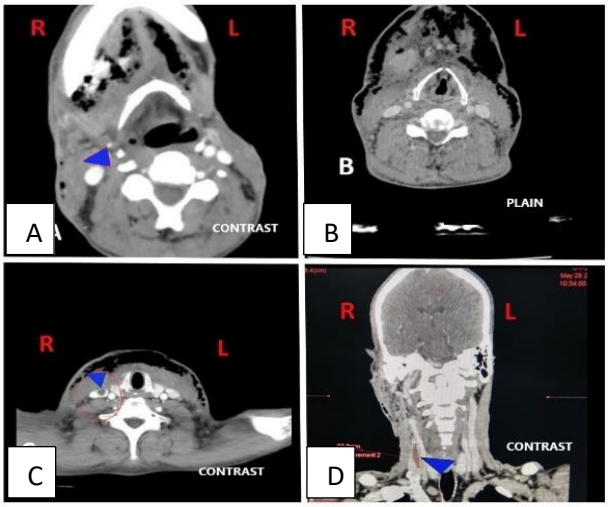


**Figure 2: Pre-operative CT of involvement of neck spaces, (A and B): pus and gas collection in tissue spaces, (C and D): a filling defect in the right IJV.**

The incision and drainage of the abscesses were done under local anaesthesia and the swab was sent for microbial analysis. The swab reported *Fusobacterium necrophorum* and the patient was started on injection metronidazole (500 mg TID) and injection amoxycillin with clavulanic acid (1.2 gm BD). Test for COVID-19 infection (reverse transcriptase polymerase chain reaction: RT-PCR) was done for the first time, which was negative. Emergency wound debridement under general anaesthesia was undertaken in view of expanding cervicofacial necrotizing fasciitis (Figure 3). Due to dysphagia, a nasogastric tube had to be inserted for adequate feeding. CECT neck and chest were repeated after 6 days to monitor the progress of the treatment. It revealed a reduction of the air pockets and no reduction in the size of the right IJV thrombus. Regular dressing was done on the neck and chest and therapeutic nutrition with Curilux® RTFED™ in a dosage of 4 scoops in 200 ml water (Curilux biotechnology, Dharwad, India) was started and continued for 1 month. CT scan of the neck was repeated 12 days after the second one again to evaluate the response to the treatment. However, no new pockets of pus were found and there was a reduction in the size of the thrombus within the right IJV (2 cm).

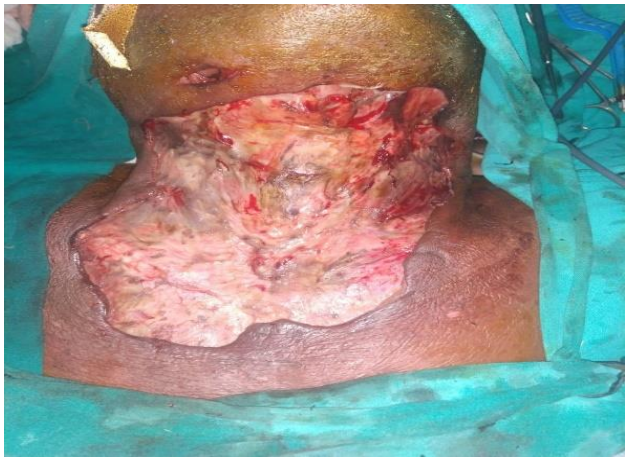

**Figure 3: Operative defect after debridement.**

After 20 days, the patient developed a fever with chills and rigors. He also developed acute kidney injury (blood urea 70 mg/dl and creatinine 2.4 mg/dl). A complete hemogram revealed reduced platelets (90000/mm$^3$) and total white cell count (2600/mm$^3$). There was also hypokalaemia (2.3 meq/l), hyponatremia (120 meq/l). A repeat COVID-19 RT-PCR test was, however, negative again. The next day patient developed spontaneous bleeding from the wound site. The coagulation profile was deranged and the INR was 2.6. This was managed with 4 pints of fresh frozen plasma over 2 days and vitamin K injection for 3 days as advised by the departments of internal medicine and nephrology. An ultrasound abdomen revealed bilateral grade 1 to 2 renal parenchymal disease changes. Even though the renal parameters improved over the next 8 days, the haematological profile worsened further to display a picture of pancytopenia (Hb 6.9 gm/dl, WBC count 1900/mm$^3$, platelets 8000/mm$^3$). The patient developed a cough with expectoration and had bilateral crepitation in the chest. Radiography of the chest revealed bilateral consolidation. However, sputum was negative for acid-fast bacillus. Serum alkaline phosphatase was elevated (370 IU/L) along with hypoalbuminemia. Hence disseminated intravascular coagulation (DIC) and multiple myeloma were suspected. However, plasma D-dimer (3500 ng/ml) and bone marrow biopsy showed reactive plasmacytosis. Also, urine Bence Jones protein was negative and serum electrophoresis of plasma proteins was normal. Hence multiple myeloma was ruled out. The patient, fortunately, responded to the treatment and his general condition gradually began to improve. Nasogastric tube was removed and then another formula for oral feeding (Curilux® Orofeed™ 2 scoops in 200 ml water twice a day) was given for 2 more months. In the meantime, the next bed patient turned COVID-19 positive and hence this test was repeated for our patient. The report was negative once again. As advised by the

physician, 2 pints of whole blood along with 4 pints of random donor platelets were given. Fortunately, the rigorous treatment paid off and the haematological parameters started to improve. Regular wound dressing and appropriate IV antibiotics were continued in consultation with the physician. The patient underwent skin grafting of the wound under local and spinal anaesthesia after his general condition improved. A revision skin grafting had to be undertaken a week later as the first one was not complete. In the meantime, the attendant of the patient turned COVID-19 positive and hence the patient had to be tested yet again. However, luck was on his side once again and he escaped the fourth time.

Nearly 2 months after admission, the patient again had a fever with chills and rigors and hence COVID-19 test had to be done for the fifth time. Yet again, the report was negative. The pus from the wound revealed Proteus vulgaris sensitive to levofloxacin. Hence injection levofloxacin 500mg bd was given for 1 month. After repeating all the blood parameters, (within normal limits) patient was posted for revision skin grafting yet again to make good the skin defects following reinfection. After 3 months of admission and treatment, the patient finally got well and was discharged safely (Figure 4). After 15 days, the patient underwent carotid doppler and no evidence of thrombus in IJV was found in the first follow up.

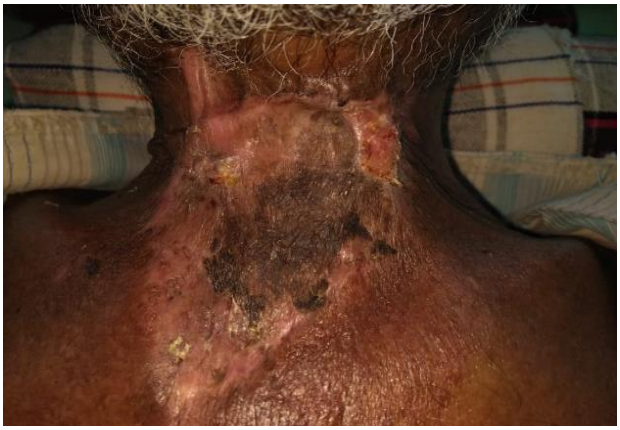

**Figure 4: After healing of the defect with a skin graft.**

## DISCUSSION

Deep neck space infections secondary to oropharyngeal or dental infections are quite common in developing countries. They frequently progress to the stage of abscess and cellulitis. However, rarely they may spread rapidly and progress further to the stage of necrosis and gangrene of soft tissues despite prompt treatment with antibiotics, especially, in a non-immunocompromised patient. The loss of skin and other tissues left a defect that took a long time to heal, thus requiring skin grafts several times. The sudden cascade of events heading towards multiple organ failures was indeed a challenge to recover. Safeguarding the patient from subsequent nosocomial COVID-19 infection was also a herculean task in a COVID-19 hospital as this patient could not be referred elsewhere.

In this patient, there appeared to be no septic emboli in the lungs; however, the kidney injury is suspected of having risen out of this complication. Noh et al however, reported septic pulmonary embolus in their case report.[4] In the times of COVID-19, remaining free of infection was a challenge for a critically ill patient with prolonged stay as the risk of acquiring nosocomial infection was high and also the prognosis in such patients was not favorable. In case the patient turned positive, he would need to be treated in a different isolation ward and operation theatre with no attendants. He would then have to follow the COVID-19 protocol for the treatment and access would be restricted for his follow up. As the pandemic gained momentum in the hospital, even radiological investigations like ultrasound and CT scans became difficult. It was indeed a miracle how our patient managed to be negative in 5 RT-PCR tests and continued to remain uninfected for more than 3 months' hospital stay. As the results of the COVID-19 test would not be known immediately, it was a challenge to plan any intervention like debridement or skin grafting. There were trismus and dysphagia in our patient due to parapharyngeal space involvement and necrosis of digastric and mylohyoid muscles. Hence the patient had to be fed through a nasogastric tube. The therapeutic nutrition could probably have helped the patient to build up the nutrition and regain immunity and hence a dying man was saved!

As this syndrome involved multiple organ systems that, too, in an emergency, various departments were involved in the patient care. Arriving at a consensus that was most appropriate for the patient at that moment was often a challenge. The multi-organ involvement, in this case could be related to DIC and metastatic septic emboli. The thrombus within the IJV was relatively silent in the beginning and was discovered only on a CECT scan. Hence the diagnosis was possible only after the CECT scan.[5] However, Ogunbayo et al have reported a case where there was no IJV thrombosis.[6] Therefore it seems that necrotizing fasciitis is the only essential component for the diagnosis.

Recent studies have examined the role of anticoagulants in the treatment of Lemierre's syndrome, but there is no consensus on the role of anticoagulation and its effect on vessel recanalization.[7,8] There are no data to support the idea that the recanalization of the IJV was an advantage and lowered morbidity and mortality.[9] The anticoagulant therapy was not contemplated in our patient as the thrombus was stable with no signs of progression and also there was no thrombophilia or antiphospholipid antibodies.

## CONCLUSION

Lemierre's syndrome is a challenge in many ways during COVID-19 times. A high index of suspicion is required for diagnosis in the absence of typical clinical features. Management is difficult when it involves multiple organ systems, primarily to protect the patient from getting infected in a COVID-19 hospital and referral elsewhere is not possible.

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