Lemierre's Syndrome: An Unusual Presentation

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Citation

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Abstract

A 46 year old man with diabetes was admitted for evaluation of high grade fever and oligura following a sore throat. On admission he had acute renal failure. He also had a painful neck swelling the radiograph of which showed the presence of gas inside it. CT scan of the neck showed a gas filled abscess. Doppler study of the neck revealed thrombosis of the left internal jugular vein. Blood culture and culture of the aspirate from the abscess had grown Fusobacterium necrophorum. A diagnosis of Lemierre's syndrome was made and patient was managed by surgical drainage of the abscess, intravenous clindamycin 600mg 12th hourly and metronidazole 500mg 8th hourly, subcutaneous insulin and haemodialysis. He recovered fully in 2 weeks time with the treatment. The case was unique because of the higher age at presentation, presence of acute renal failure necessitating haemodialysis and absence of overt pulmonary complications.

DECLARATION

This work has not been presented anywhere earlier and no grant has been accepted for this work.

CASE REPORT

A 46-year old man was admitted for evaluation of high-grade fever for one week and vomiting and oliguria for 2days. He had sore throat for a few days 2 weeks prior to the admission and had noticed a painful swelling on the left side of the neck 4 days before admission. He also had type 2 diabetes mellitus for the past one year that had been well controlled with glipizide 5mg daily.

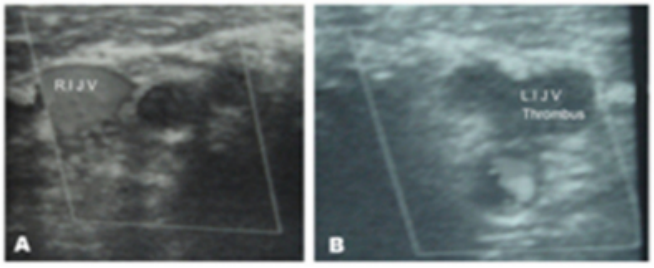
On admission his temperature was 40.5°C, pulse rate 120 beats/min, respiratory rate 33/min and blood pressure 94/70mm of Hg. He was clinically dehydrated. Throat examination revealed mild congestion of the tonsils. A tender swelling was palpable in the left supraclavicular region. Rest of the systemic examination was unremarkable.

The initial laboratory investigations were: haemoglobin concentration 100 g/L, white cell count 14.6 x 10^9/L, neutrophils 12.4 x 10^9/L, lymphocytes 2.2 x 10^9/L, platelet count 190 x 10^9/L, erythrocyte sedimentation rate 110 mm in the first hour, plasma glucose 17 mmol/L, urea 94 mmol/L, creatinine 406 micromol/L and bilirubin level 61 micromol/L. Other liver tests, sodium, potassium, corrected calcium and inorganic phosphates were within the reference range. Urine microscopic examination and the ketostix reaction were unremarkable. Fractional excretion of sodium was 1.2%. Chest radiograph was normal and the serological assays for leptospirosis, dengue virus and hepatotrophic viruses were negative. The electrocardiograph showed only sinus tachycardia. Peripheral blood smear revealed normocytic normochromic anaemia and neutrophilia with toxic granulations.

Ultrasonography of neck swelling showed a focal heterogenous area of 3.5cm diameter with ill-defined borders in the intramuscular plane. Doppler study of the neck revealed absence of blood flow in the left internal jugular vein and a thrombus inside the vein (Fig 1B).

Figure 1

Figure 1: A Doppler study showing normal blood flow pattern in the right internal jugular vein (R.IJV). B. Doppler study on the left showing absence of blood flow and a thrombus inside the vein (L.IJV thrombus).



Lateral view radiograph of the neck showed a gas shadow in the neck (fig 2A: arrow) and CT scan images of the neck (contrast study was not done because of the renal failure)

confirmed the gas filled abscess in the left supraclavicular region (Fig 2B: arrow).

Figure 2

Figure 2a: Radiograph of neck showing the gas shadow (Black arrow).

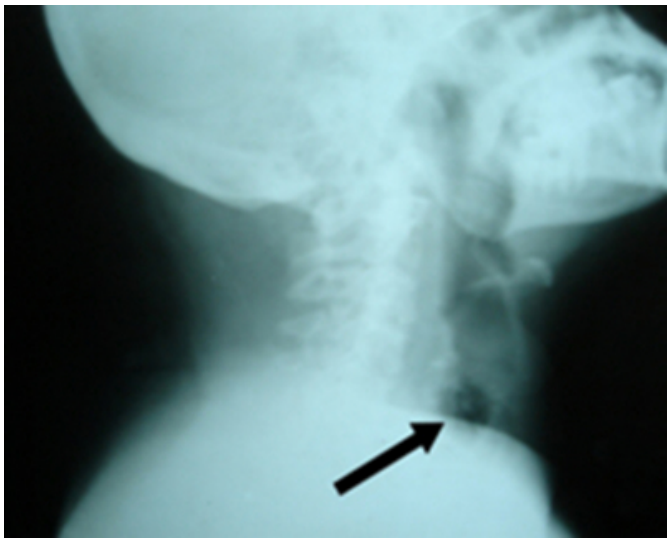
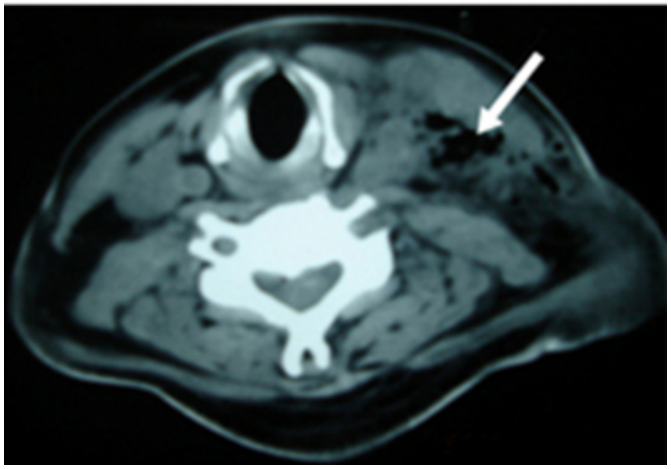


Figure 3

Figure 2b CT scan of neck showing the gas filled abscess (White arrow).



A diagnosis of Lemierre's syndrome was made and the patient was managed by surgical drainage of about 40ml of foul smelling pus from the abscess with a 4 cm long vertical incision in the posterior triangle on the left side of neck. Subcutaneous insulin, intravenous clindamycin 600mg 12th hourly and metronidazole 500mg 8th hourly also were administered. He required 4 alternate day sessions of hemodialysis for renal failure. On the 4th day of the treatment the blood culture and culture of the pus from the neck swelling showed the growth of Fusobacterium necrophorum confirming the clinical diagnosis. After two weeks of treatment the patient recovered fully from acute renal failure and sepsis. A repeated Doppler study of the neck performed 3 months later had shown partial flow through the left internal jugular vein indicating some degree of spontaneous recanalisation of the thrombosed vein.

DISCUSSION

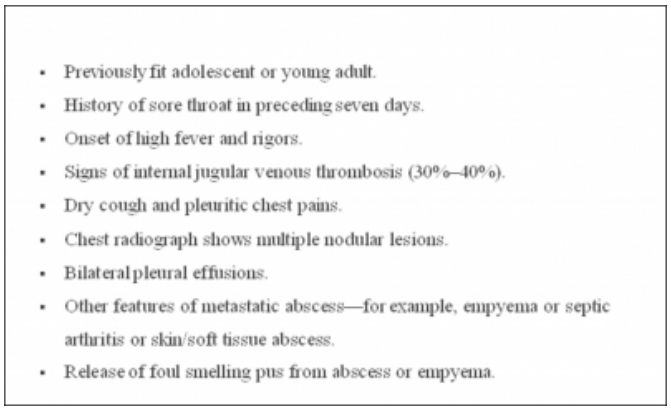
Lemierre's syndrome, also called necrobacillosis/post anginal sepsis, is characterized by acute pharyngeal infection most often with Fusobacterium necrophorum, later resulting in secondary septic thrombophelebitis of internal jugular vein and frequently complicated by metastatic infection[1]. Even though first reported by Courmont and Cade in 1900, Andre Lemierre best characterized the syndrome in 1936. The incidence of this disease seems to be increasing now probably because of lesser use of antibiotics for sore throat.

F necrophorum is an anaerobic gram-negative bacillus that colonizes the oral cavity, female genital tract, and gastrointestinal tract. Lemierre's syndrome has been described as a complication of acute oropharyngeal infection with this organism in adolescents and young adults and classical cases have four findings: 1) primary infection of the oropharynx 2) secondary bacterial infection with at least one positive blood culture 3) thrombosis of the internal jugular vein and 3) at least one distal focus of infection such as pneumonia or arthritis.

Even though F necrophorum is the commonest organism isolated from blood cultures in patients with Lemierre's syndrome, other organisms such as Streptococcus sp, Bacteroides sp, Peptostreptococcus sp and Eikenella corrodens are occasionally grown in blood cultures of patients with this disease[2]. A high degree of clinical suspicion should aid the initial diagnosis, considering the catastrophic complications from septic emboli[1,3,4] and the related high mortality in cases of delayed diagnosis. Features suggestive of Lemierre's syndrome are given in box 1.

Figure 4

Box 1: Suggestive features of Lemierre's syndrome.



Complications described in Lemierre's syndrome include pulmonary lesions (embolism, suppurative pneumonia, abscesses and empyema), clinical jaundice, internal jugular vein thrombosis, septic arthritis, septic shock, skin and soft tissue lesions, renal failure, meningitis, osteomyelitis and clinical DIC in the descending order of frequency[3].

Intravenous antibiotics directed against anaerobes and drainage of abscesses in accessible sites are the ideal treatments recommended. F necophorum has been traditionally susceptible to penicillin, clindamycin and metronidazole, but patients may have a fulminant course even with proper antibiotic therapy[4]. There is no consensus regarding the role of anticoagulation in the management[1,4], even though recent studies show a higher incidence of thrombophilic states in patients with Lemierre's syndrome and venous thrombosis[5]. Excision/ligation of internal jugular vein may be needed if emboli are not controlled with medical management.

For an infection associated with such severe sepsis, acute renal failure requiring dialysis is remarkably uncommon in patients with Lemierre's syndrome[1,3]. Acute tubular necrosis (secondary to hypovolemia and systemic sepsis) was the probable cause for renal failure in this case and the high fractional excretion of sodium and rapid and complete recovery following treatment also suggest the same.

Several features of this case such as higher age at presentation, absence of overt pulmonary complications and development of acute renal failure necessitating hemodialysis were unique.

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