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A rare case of painful swelling in the neck in a middle aged uncontrolled diabetic male

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Abstract
Lemierre's syndrome: More than a historic curiosa a prepenicillin era illnees characterized by disseminated abscess and thrombophlebiltis of internal jugular vein after infection of the oropharynx. The predominant pathogen is a gram -ve anaerobic bacillus *Fusobacterium necrophorum*. A prompt clinical diagnosis is of utmost importance for a favourable prognosis in the management of this patient. Here we present a case of lemierre syndrome in a middle aged uncontrolled diabetic male where the unusual and atypical clinical manifestation posed a mounting challenge in the diagnosis of this case. And the peculiarity in this case was that the causative organism was *Klebsiella pneumoniae* which is not known to cause Lemierre's syndrome typically. Till date only few other cases has been reported where the organism was *Klebsiella pneumonae*.

Keywords: *Fusobacterium necrophorum*, painful, swelling, uncontrolled

Introduction
A 44 year gentleman with h/o fever and painful swelling in the neck 5 days prior to admission. There was no associated respiratory distress. However the patient complained of difficulty in neck movement. There was no trismus. Dental examination was done which was unremarkable.

On examination a firm solitary swelling 9x10 cm in size noted over the left side of the neck and face extending to the clavicle. Swelling was tender with marked redness and increased temperature. Swelling was non-pulsatile and firmly fixed to the skin. No active pus points or ulceration seen. Surface was smooth.no pulsation seen or any movement with deglutition or protrusion of tongue. The patient is a known case of type 2 diabetes mellitus for last 3 years and on glimepiride 2mg + metformin 1 gm.

Patient is a known hypertensive for the past 5 years on amlodipine 5 mg. Patient is a known smoker and alcoholic for the past 10 years. After admission the patient was treated with i/v amoxycillin clavulunate 1.2 gm thrice daily and clindamycin 600 mg thrice daily, injection glargine 20 units and regular insulin 15 units before breakfast, lunch & dinner and betadine mouthwash.

Blood investigation reports
Hb 15,4, TC-8370, N-8O, L-13, M-6, E-1, B-0, Platelet-1.5 Lakh, MCV-89.7, Total Bilirubin-0.7, Albumin-3.6, Globulin-3.1, SGOT-26, SGPT-18, Alkaline Phosphatase-74, Urea-51, Creatinine-0.8, Sodium-128, Potassium-4.4, CRP 24, Pro calcitonin-2.2, LDH-143 (225-480), FBS-304, HbA1C-13.5%. Urine for ketones negative, RBS-406.

He was referred to an ENT surgeon and gave a diagnosis of deep neck space infection. Radiological investigations chest X-ray pa view-normal, USG neck-left parotid bulky and increased vascularity. Left sternocleidomastoid appears to be bulky and heterogeneous compared to the right. Few mildly enlarged lymph nodes noted in level 2 & 3. USG carotid doppler-flow through the B/L carotid WNL. Left internal jugular vein shows a echogenic thrombus with air. No colour uptake noted. Heterogeneous ill-defined areas noted in the left side of the neck below the sternocleidomastoid extending to the root of neck. Echo-mildly dilated left ventricle, EF 57%, RWMA noted.

He was thereafter referred to a radiologist for CT guided aspiration of pus and to see the integrity of neck vessels. About 300 ml of pus was aspirated and was sent for aerobic and anaerobic culture which revealed no growth. Meanwhile blood C/S report came in which showed growth of *Klebsiellla pneumonae* which was sensitive to meropenem and clindamycin and the antibiotic regime was changed to a regime of sensitive antibiotic. And low molecular weight heparin 40 mg S/C twice daily and tablet aspirin 75 mg. His latest reports are HB 12.9, TC-9600, N-5, l-29, M-5 E-1, B-0, ESR 16.

Discussion

Lemierre's syndrome is a rare disease originally described by Andre Lemierre in 1936. It is typically caused by the microorganism *Fusobacterium necrophorum*. The most common primary infection being tonsillitis (87.1%), followed by mastoiditis (2.7%), odontogenic infections (1.8%) [1, 2] most cases of Lemierre's syndrome occur in adolescent and young adults with tonsilliopharyngitis or less commonly those with an odontogenic infection, mastoiditis, sinusitis [3]. Oral anaerobes invade the peritonsillar tissue and infection spreads to the adjacent lateral pharyngeal space which contains the internal jugular vein. *F. necrophorum* an anaerobic gram negative rod that commensally inhabits the oral cavity and gi tract and fmelae genital tracts in 82% of cases [3]. From neck it spreads haematogenous to various organs but most commonly to lungs but also to joints, liver, spleen, bone, kidneys, meninges [4].

K. pneumoniae as a causative organism of LS is quite rare with only a few reported cases previously. Curiously all of the previously reported cases the patient was diabetic posing a serious argument whether diabetes predisposes to infection of certain group of microorganism. Many mechanism have been proposed to explain the increased susceptibility to periodontal disease in these patients, such as alteration in immune response, subgingival microbiota aspects, altered collagen metabolism, alteration in oral vascularization, hereditary patterns, altered neutrophil function, reduced phagocyticcapacity and chemotaxis [5]. Inversely poor and persistent glycaemic control has been associated with greater incidence and progression of gingivitis and periodontitis producing a vicious cycle [6-8]. In another paper by lee and kanaglingam the authors had documented that 50% of their patients with diabetes with deep neck absecess had *K. pneumoniae* especially the K1 and K2 isolates [9].

Extensive and prolonged antibiotic therapy is the mainstay of treatment. Suggested antibiotic based on empirical evidence includes metronidazole, clindamycin, penicllin, imipenem, ampicillin sulbctam, ticarcillin-clavulunate [10]. In our case metronidazole was avoided because the patient was an alcoholic. Few clinicians have recommended use of anticoagulants in all cases of Lemierre's syndrome [11, 12].

Conclusion

Lemierre's syndrome maybe caused by other organisms other than *Fusobacterium necrophorum*. Our case advocates the need for search of organisms which do not typically cause Lemierre's syndrome and also highlights the changing patterns of infection with diabetes and the evolving menace of Klebsiella in community acquired infections with diabetes. Primary care physicians should be aware of its existence and its variety and various manifestations. A curious and prompt diagnosis is essential for favourable outcome in the treatment.

Fig 1: Showing swelling over the left side of neck extending to upper chest

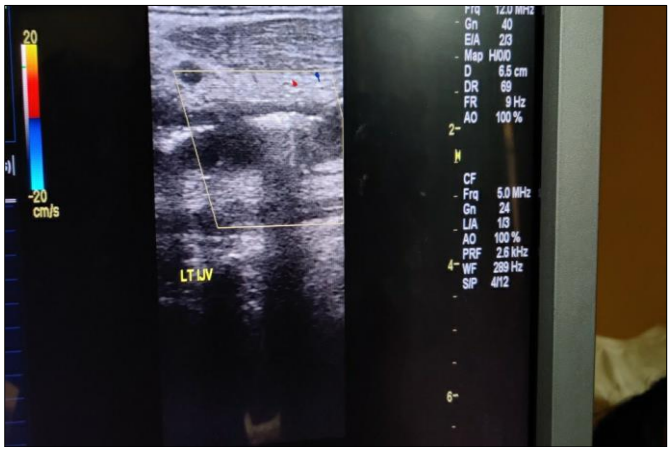

Fig 2: Showing internal jugular vein thrombosis (Left)

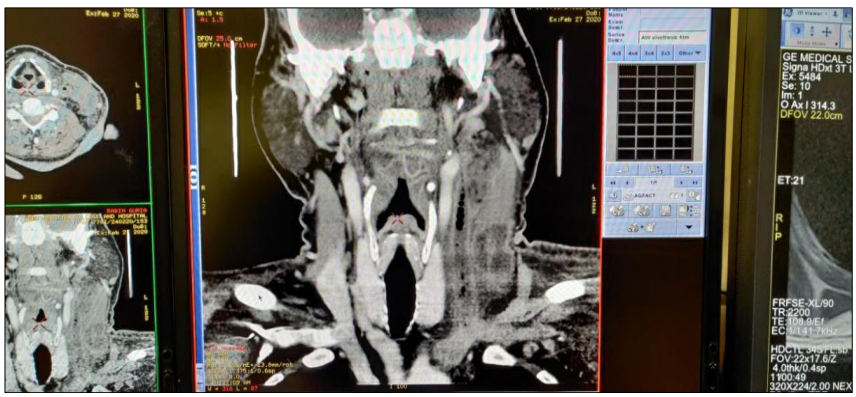

Fig 3: Showing left internal jugular vein thrombosis

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