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Case Report

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Lemiere's Syndrome Caused by Klebsiella Oxytoca; A Case Report

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ABSTRACT

Lemiere's syndrome is a rare but life-threatening condition which is most often preceded by an oropharyngeal infection. The most common organism involved is fusobacterium necrophorum, which is a part of normal flora of oropharynx. This case report describes a rare case of Klebsiella oxytoca associated Lemiere's syndrome in a patient with poorly controlled Diabetes Mellitus. To our knowledge there has been only one single reported case of Lemiere's syndrome caused by Klebsiella Oxytoca and this should be considered among the possible organism causing Lemiere's syndrome. Though rare Lemiere's syndrome carries a high mortality rate. Therefore clinicians should be familiar with signs and symptoms of the disease as well as the preemptive examinations, procedures and treatments.

Key Words: *klebsiella oxytoca, diabetes mellitus, thromboplebitis, Lemiere's syndrome, case report*

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INTRODUCTION

Lemiere's syndrome also known as human necrobacillosis is a rare and almost forgotten yet potentially life threatening condition. It is commonly caused by an oropharyngeal infection when it extends into the carotid space resulting in internal jugular vein thrombophlebitis and subsequent metastatic infections. Although initially described in 1900 by Courmont and Cade [1], it was the French microbiologist Andre Lemiere [2] who best characterised this disease in 1936. Pathognomonic findings include swelling and tenderness at the angle of jaw and along the line of sternocledomastoid muscle, along with signs of sepsis (spiking fever, chills), and evidence of IJV thrombosis [3]. Recent diagnostic criteria includeshistory of recent oropharyngeal infection, clinical or radiographic evidence of thrombophlebitis of the internal jugular vein (IJV), isolation of an anaerobic pathogen, and metastasis to distant areas [4, 5]. This infection is commonly caused by Fusobacterium necropherum and F. nucleotum. However, with the increase in variety of comorbidities, use of imunnosuppressiveagents and prevalence of drug resistance bacteria, Lemiere's syndrome has been reported with atypical pathogens and presentations. In the antibiotic era, the incidence and mortality of this disease had drastically reduced. Nowadays, Lemiere's syndrome is not a well known illness because of its rarity. We report a rare case of Lemiere's syndrome caused by Klebsiella oxytoca in a patient with poorly controlled DM.

CASE PRESENTATION

A 40 year old female presented to emergency room with complaint of swelling and pain on left lateral aspect of neck and fever since 5 days. On further probing, Patient gives history of sore throat prior to the development of present complaints which has subsided. She denied history of trauma, night sweats, weight loss, intravenous drug use, recent dental infection or procedure. The patient is a known case of Diabetes Mellitus and is on medication since one year.

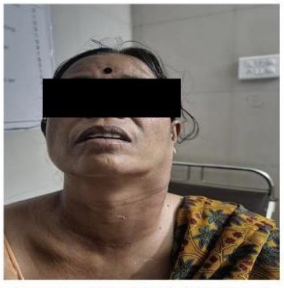

Figure 1: Picture of the patient at the time of presentation with swelling over left angle of mandible and along the line of left Sternocleidomastoid muscle.

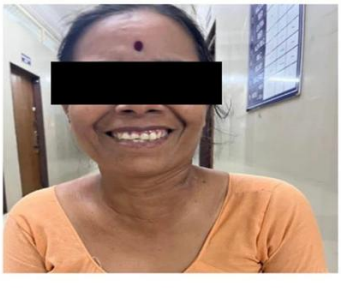

Figure 2: Picture of the patient on the day of discharge. Swelling has completely resolved.

Physical examination at the time of admission revealed temperature of 38.6, blood pressure of 126/90 mmhg, heart rate of 104/min, respiratory rate of 18/min and spo2 of 99% in room air. The patient was conscious, oriented to time, place and person. Swelling and redness were observed on left angle of jaw extending down along the line of sternocledomastoid muscle. It was tender on touch and skin over swelling was inflammmed.

Laboratory investigations showed white blood cell count of 17,500 cells/mm3(75%-neutrophils, 20% lymphocytes and 4% monocytes), haemoglobin level of 11.8gm/dl and platelet count of 3,72,00 cells/mm3. The patient was hyperglycaemic with a glucose level of 424. The patient had uncontrolled diabetes with glycated haemoglobin level of 11gm%.

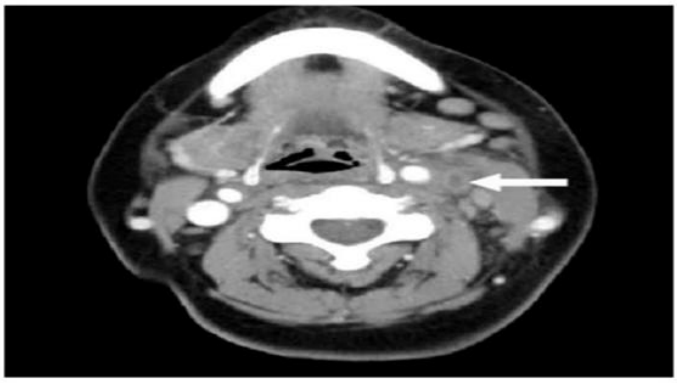

Figure 3 : Axial neck contrast CT demonstrating filling defect in left Internal Jugular Vein

Usg doppler was performed and revealed a collection of size approx. 4x3cm2 in the inter muscular plane underlying sternocleidomastoid muscle and the left internal jugular vein coursing close to the lesion showing increased wall thickness with increased surrounding fat stranding suggesting IJV thrombosis. CT scan was done which revealed a filling defect in left internal jugular vein.

The patient underwent incision and drainage and the pus drained was sent for pus culture sensitivity. Intravenous ceftazidime was started at a dose of 1 gm every 12 hrs. Klebsiella oxytoca was identified from the pus culture. The organism was susceptible to a wide range of antibiotics and resistant only to Ampicillin. Initial antibiotic was then replaced with IJV meropenam. With daily dressing of the wound and antibiotic treatment, the patient began to recover and by 10 th day post surgery patient completely recovered and was discharged with continuation of oral antibiotics.

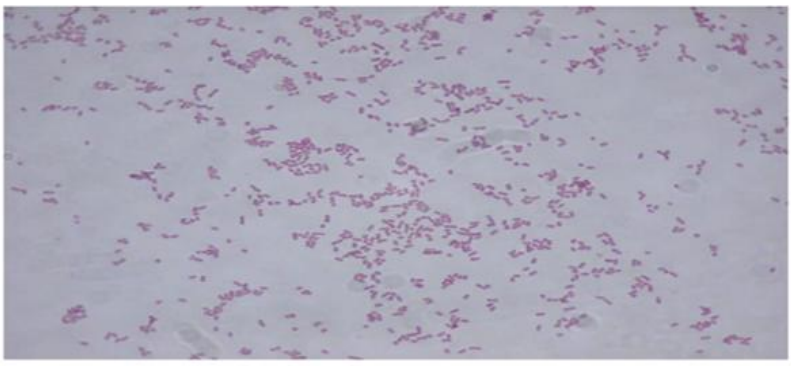

Figure 4: gram stain showing Klebsiella oxytoca

DISCUSSION

Lemiere's syndrome while quite common in pre-antibiotic era, it is regarded as very rare nowadays. In the 90's, the incidence of this syndrome was so low that it was regarded as an almost forgotten condition [6, 7]. Over the past 25 years, Lemiere's syndrome has only been reported in approximately 150 cases, partly due to antibiotic therapy preventing disease progression and general unawareness of the syndrome In the 21st century, the incidence of Lemiere's syndrome seems to be higher[6,8]. The reason for this phenomenon might just be higher awareness and better availability of sensitive diagnostic nodalities, such as computed tomography and magnetic resonance [8]. Nevertheless, it is still considered as very rare with an estimated incidence of 1 case per million [6, 8 & 9]. The term 'forgotten disease has been used to describe the rarity of Lemiere's syndrome in antibiotic era. It typically occurs in adolescents and young adult.

Lemiere's syndrome is primarily caused by Fusobacterium. Among the 13 species of fusobacterium, F. necropherum is the most common followed by f. nucleotide. In other case reports, Bacteroides species, Streptoccus, Enterococcus, and proteus mirabilis have been identified as pathogens.

In the present case, K. Oxytoca was the causative pathogen. Lemiere's disease caused by this bacterium is extremely rare. In a meta-analysis conducted by Dartment of Otorhinolaryngology, State University of New York, out of 427 patients with Lemiere's syndrome, only a single case of Lemiere's syndrome caused by Klebsiella oxytoca has been reported. It has been observed that Klebsiella associated Lemiere's syndrome usually occurs in patients with poorly controlled diabetes [10].

Similarly, our patient also had poor blood glucose control with a glycated haemoglobin level of 11% and a serum glucose level of 424mmol/L at the time of admission which further supports the study.

Patients with type 2 diabetes mellitus are vunerable to infection because of the decreased activity of neutrophils [11]. The underlying mechanism of the increased susceptibility of the patients with type 2 Diabtes Mellitus to Klebsiella species, especially k1/k2 isolates, is resistant to phagocytosis [12].

Beta-lactamase resistant beta-pa tam antibiotics are recommended for typical Lemiere's syndrome. Beta lactamase produced by F. necrophorum may lead to therapy failure. But once culture sensitivity report is available and the causative organism is identified, treatment should be changed from empirical antibiotics to targeted antibiotics based on result.

The role of anticoagulation for IJV thrombosis is controversial. As Lemiere's syndrome has a low incidence rate, there is lack of controlled investigations for anticoagulation treatment. A meta-analysis of 427 cases of Lemiere's syndrome on the effect of anticoagulation on vessel recanalization failed to show a statistically significant benefit for either outcome. Aggressive antibiotic therapy combined with surgical intervention when indicated appears to be the mainstay of treatment in Lemiere's syndrome, with a low mortality rate overall with or without anticoagulation. In our case, no anticoagulant was administered because the patient responded well to antibiotics.

CONCLUSION

In Lemiere's syndrome, once thrombosis of IJV has occurred, pain and swelling or in duration at the ipsilateral angle of mandible extending along sternocleidomastoid muscle can occur along with high fever or trismus. Confirmation of IJV thrombosis is best made by a CT scan with contrast showing luminal filling defect, distension, and enhancement of the IJV with swelling of the soft tissues. Once septic thrombophlebitis has occurred, complications include metastatic disease to various organs, intracranial involvement or septic shock.

Lemiere's syndrome is a rare disease. Therefore it is difficult for doctors with no experience to suspect it. In addition, since original oropharyngeal infection could have resolved by the time of patient presentation like in our case and there might be few, if any, on physical examination. Control of infection at the primary site with appropriate empirical antibiotics and surgical interventions remains the critical strategy to prevent metastasis, achieve clinical responses and reduce mortality.

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