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Lemierre's syndrome: A case report of an infant with Acute Suppurative Otitis Media and Poly-microbial infection with Leuconostic Citreum and Acinatobacter Baumannii

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ABSTRACT

Lemierre's syndrome is quite a rare condition characterized by septic thrombophlebitis of the internal jugular vein as a complication of head and neck infection, and septic embolization to other organs. The most common etiological agent is Fusobacterium necrophorum. It is very rarely reported in infants or secondary to Acute Suppurative Otitis Media (ASOM) or secondary to bacteria other than Fusobactrium Necrophorum. Here, we report rare findings of a rare disease, in the case of a 6month old female child who developed Lemierre's syndrome as a complication of ASOM and her blood culture grew Acinatobacter baumannii and Leuconostic citreum. By the time of this study, very few cases of invasive infection in humans due to Leuconostic citreum have been reported.

Keywords: Lemierre's syndrome, Internal jugular vein thrombosis, Infants, Acute suppurative otitis media, Acinatobacter baumannii, Leuconostic citreum.

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INTRODUCTION

Lemierre's syndrome a very rare disease with about one case reported per million, per year [1]. It was first described by Andre Lemierre in 1936, when he described internal jugular vein thrombophlebitis in 20 patients with tonsillar and pharyngeal infections [2]. It is characterized by septic thrombophlebitis of the internal jugular vein due to septic emboli originating from head and neck infection in advanced stages, it can lead to septic metastasis to other organs [1,2]. It is mostly reported due to Fusobacterium necrophorum, but can also be caused by other microorganism like streptococcus, staphylococcus, proteus etc. Poly-microbial bacteremia is reported in one-third of patients [2]. During pre-antibiotic era, mortality due to Lemierre's syndrome was as high as 90% [3]. However, the incidence of Lemierre's syndrome decreased drastically after the year 1940, as Penicillin was used extensively for pharyngitis due to Streptococcus [2,4]. Because of its rare occurrence, authors started referring to Lemierre's syndrome as "forgotten disease" [5].

Despite much advancement in medical fields, a mortality rate of 10-25% is reported in Lemierre's syndrome if there is delay in appropriate treatment by 4 days [2,6,7]. This emphasizes the importance of attending physicians" awareness about the syndrome.

Lemierre's syndrome is diagnosed mostly in previously healthy adolescents and young adults [8]. It is reported mostly as a complication of bacterial infection of the palatine tonsils [8,9]. In the current study, we report on the case of a 6month old female child, who developed Lemierre's syndrome as a complication of Acute Suppurative Otitis Media (ASOM). Her blood culture grew Acinatobacter baumannii and Leuconostic citreum. To our best knowledge, this is the first case where Leuconostic citreum is one of the etiological agents causing Lemierre's syndrome, and invasive diseases in humans due to Leuconostic citreum have been reported very rarely all around the world [10].

Case report

A 6 month old female child with no significant past medical history was brought to hospital with complaints of fever for 5 days and swelling in the right side of her face around the ear and jaw.There was no history of recent travel, or exposure to a sick patient. There was no history of drug allergy. She was not given any medication other than antipyretics. The child was a product of non-consanguineous marriage and she was born by normal vaginal delivery. Immunization was normal for her age. On physical examination, the child was febrile with a core temperature of 102.2 ^{0} Fahrenheit (F), blood pressure of -85/60 mm Hg, the pulse rate was 155 beats / minutes, the respiratory rate was 38 breaths per minute and saturation by pulse oximeter was 98%.Anthropometry examinations were as follows: weight 7.5 kg (between median and + 1 standard deviation), length 67 cm (between median and +1 standard deviation) and weight for length was at the median. Hence, the child was well nourished. Local examination showed swelling in the right sided parotid and sub mandibular region. There was no redness, warmth, fluctuance or crepitus around swelling. The right anterior cervical lymph nodes were enlarged and tender on palpation. Throat examination was normal. Central nervous system examination was normal. The child was active, playful and oriented to person, place and time. There were no signs of meningeal irritation or focal neurological deficits. Respiratory system examination showed normal findings. Chest was clear, bilateral air entry was equal with no added sounds. There was no tachypnea or respiratory distress. Per abdominal examination was normal. Abdomen was soft, non-tender, there was no organomegaly. Cardiovascular system examination showed tachycardia (secondary to fever), S1 and S2 were heard and there was no murmur. After sending laboratory blood tests including aerobic and anaerobic blood cultures, the child was started on intravenous injection Amoxicillin–Clavulanate and intravenous (IV) injection Amikacin. A neck ultrasound was planned, where the findings were multiple enlarged necrotic and non-necrotic lymph nodes in the right sub mandibular, upper, middle and lower jugular with largest conglomerate nodal measuring around 24.9 x 14.4 mm. The parotid gland was normal in size with hetero echoic echo-texture.

Complete blood count was as follows: Hemoglobin -

8.5gm/dl, White blood cells -23,700/ mm^3: Neutrophil-52 %, Lymphocyte -43%, Platelets - 2.5 lakhs/mm^3, C-reactive protein -285mg/dl. The liver function test, kidney function

test, serum electrolytes, coagulation profile were normal. On day 3 of admission, slightly bloodstained ear discharge was seen in the right ear. There was no improvement in clinical condition and core temperature spiked to 104 ° F.The ear, nose, throat department assessed the child and made the diagnosis of ASOM. The blood culture reports were not available at that point. In view of deteriorating clinical condition and delay in blood culture reports, antibiotics were upgraded to IV Meropenem, Vancomycin and Metronidazole. In the meantime, a Contrast Enhanced Computed Tomography of the neck (CECT neck) was done, which showed suppurative lymphadenitis involving right cervical, retro-pharyngeal, parotid gland, carotid space and upper para vertebral space. The right distal cervical segment of the internal carotid artery was not visualized with distal optimal opacification, suggestive of right internal jugular vein thrombosis. (Figure 1, Figure 2).

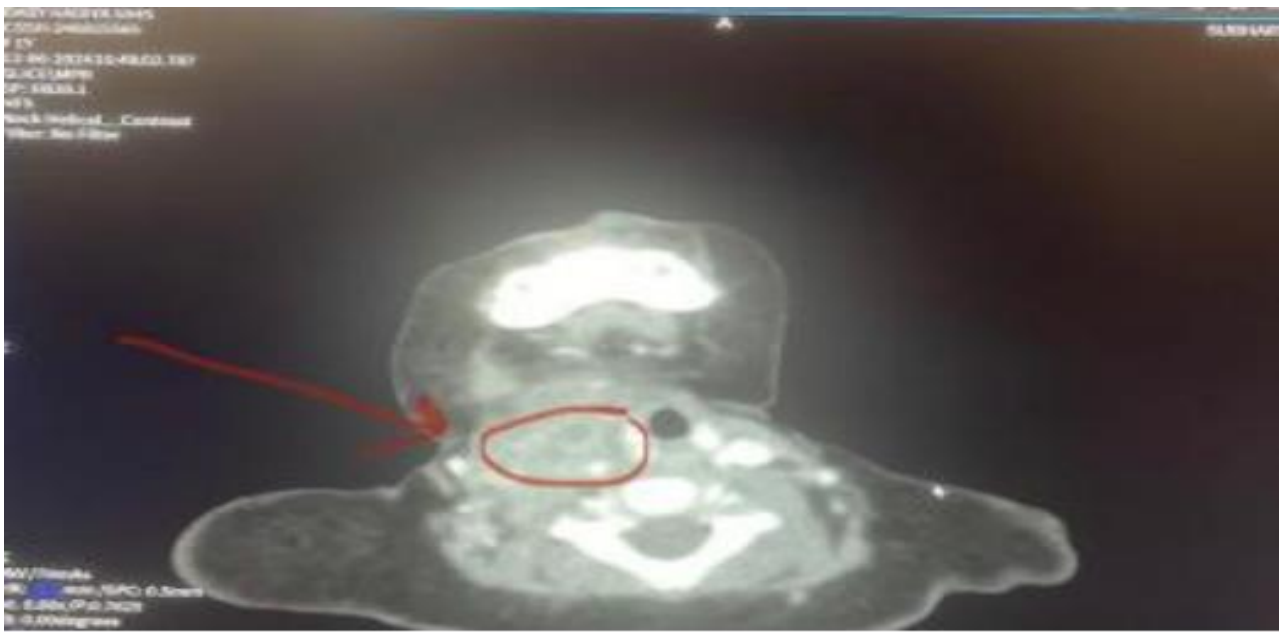
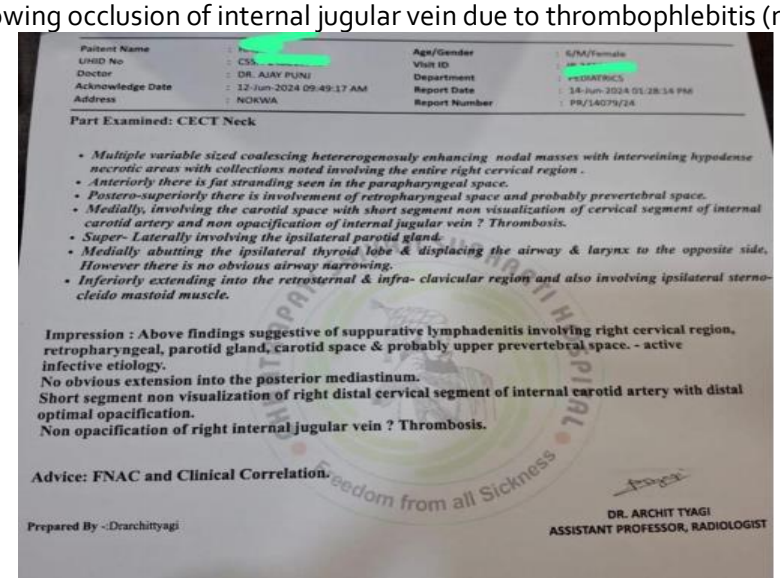


Figure 1. CECT Neck showing occlusion of internal jugular vein due to thrombophlebitis (marked by arrow)



Patient Name : [redacted] Age/Gender : 6/M/Female
UHID No : CSS[redacted] Visit ID : [redacted]
Doctor : DR. AJAY PUNJ Department : PEDIATRICS
Acknowledge Date : 12-Jun-2024 09:49:17 AM Report Date : 14-Jun-2024 01:28:14 PM
Address : NOKWA Report Number : PR/14079/24

Part Examined: CECT Neck

- *Multiple variable sized coalescing hetererogenoxuly enhancing nodal masses with interveining hypodense necrotic areas with collections noted involving the entire right cervical region .*
- *Anteriorly there is fat stranding seen in the parapharyngeal space.*
- *Postero-superiorly there is involvement of retropharyngeal space and probably prevertebral space.*
- *Medially, involving the carotid space with short segment non visualization of cervical segment of internal carotid artery and non opacification of internal jugular vein ? Thrombosis.*
- *Super- Laterally involving the ipsilateral parotid gland.*
- *Medially abutting the ipsilateral thyroid lobe & displacing the airway & larynx to the opposite side. However there is no obvious airway narrowing.*
- *Inferiorly extending into the retrosternal & infra- clavicular region and also involving ipsilateral sterno-cleido mastoid muscle.*

Impression : Above findings suggestive of suppurative lymphadenitis involving right cervical region, retropharyngeal, parotid gland, carotid space & probably upper prevertebral space. - active infective etiology.
No obvious extension into the posterior mediastinum.
Short segment non visualization of right distal cervical segment of internal carotid artery with distal optimal opacification.
Non opacification of right internal jugular vein ? Thrombosis.

Advice: FNAC and Clinical Correlation.

Prepared By -:Drarchittyagi

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ASSISTANT PROFESSOR, RADIOLOGIST

Figure 2. CECT report of the neck

Blood culture and sensitivity reports were available on day 5, and showed growth of Acinetobacter baumannii (carbapenemase screening positive and resistant to all drugs, Meropenem, Imipenem, Gentamicin that it was tested for) and Leuconostic citreum (sensitive to Penicillin). However, as the child had started responding to injectable Meropenem, Vancomycin and Metronidazole with decreasing intensity and frequency of fever, the same antibiotics were continued. In view of the above clinical presentation and laboratory and radiological reports, diagnosis of Lemierre's syndrome secondary to complication of acute suppurative otitis media was made. The child was continued on IV antibiotics for 2 weeks, followed by oral antibiotics for 4 weeks. We did not give any anti coagulation medication. During her whole duration of hospital stay, the child showed no signs of septic emboli metastasis, and was successfully discharged after 6 weeks.

Discussion

The most common etiological agent for Lemierre's syndrome is Fusobacterium necrophorum, an anaerobic bacterium that is normally found in the oral cavity [1]. However, other microorganisms have also been also reported. One recent study observed that out of 96 patients with Lemierre's syndrome, blood culture grew Fusobacterium spp. in 52 %, streptococcus spp. in 18%, staphylococcus aureus in 6.3%, while 23.7% grew other bacteria - mostly gram negative [2]. In our case, there was a poly-microbial infection, as blood culture grew Acinatobacter baumannii and Leuconostoc Citreum. We could not find any case of Lemierre's syndrome associated with these microorganisms. What adds to the rarity of this case is that reports of invasive infection due to Leuconostoc citreum in humans is very rare. It is reported in immunosuppressed patients like patients with malignancy, malnutrition, chronic diseases, after some major surgery or in children with history of pre maturity or low birth weight. Patients receiving Total Parenteral nutrition and with central catheters have also been observed to be at risk of Leuconostoc citreum infection [10,11,12]. However, in our case, the child was born at term gestation with a birth weight of 2.9 kg, had no history of repeated infections or any major surgery, was not suffering from any chronic disease and was well nourished. The child was orally allowed and was never on parenteral nutrition. Hence, in our case, unlike previously reported cases, the child was apparently healthy. The primary source of infection in Lemierre's syndrome are the tonsils and the para tonsillar area in most studies [1,2]. However, other primary sources of infections are also reported less commonly and include the middle ear, mastoid, lungs, sinus, and teeth [8,9]. A study by Karkos et al. revealed that the majority of patients with this syndrome presented in their second decade of life (51%) followed by third decade (20%), and first decade (8%) [8]. It was observed by Baig et al. that Lemierre's syndrome has a stepwise pattern in presentation [13]Step 1 - a triggering primary infection like tonsillitis, otitis media, pharyngitis etc.
Step 2- local microbial invasion via lymphatics from tissues of primary infected site to internal jugular vein.
Step 3- septic metastasis spread from internal jugular vein to other organs.
Our patient presented in step 2 of disease, and further advancement of disease was prevented by timely and appropriate treatment.Septic emboli is most frequent in the lungs. 79-100% of patients develop metastasis to lungs and this can present as infiltrates, pneumonia, lung abscess, empyema etc [2,14] 11-27% of patients have metastasis to joints [6]. Less commonly, metastasis is also reported in the liver, spleen, heart, kidney, brain and soft tissues [4,7,15]

In our case, there were no clinical features suggesting septic emboli metastasis to other organs. Therefore, we did not do radio-imaging of different organs to look for septic emboli metastasis.Despite internal jugular vein thrombophlebitis, optimal anticoagulation regimens for Lemierre's syndrome are not clearly defined. Use of anticoagulation has been reported in 63.7% of pediatric patients with Lemierre's syndrome [16]. However, routine use of anticoagulants is not recommended and until the role of anticoagulation is well defined, antibiotics remain the mainstay of treatment of Lemierre's syndrome [2,6,14]. We did not give any anticoagulation treatment to our patient.

Conclusion

We presented the case of a 6 month old female child admitted with complaints of fever and swelling in the face, around the ears and jaw. Later on, as ear discharge was seen on day 3 of admission ASOM was detected. However, awareness about disease and timely laboratory and radiological tests including ultrasound neck and parotid, CECT neck and positive blood cultures helped in confirming a diagnosis of Lemierre's syndrome. IV antibiotics were started, and when there was no clinical improvement, antibiotics were escalated timely and were continued for sufficient duration. Timely initiation and escalation of antibiotics prevented the advancement of disease to the septic emboli metastasis stage and the child could be discharged successfully.Through this case report we wish to increase awareness about Lemierre's syndrome, a rare disease that can be successfully treated with early diagnosis and prompt interventions, but which can be life threatening otherwise, due to septic emboli metastasis.

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