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Case Report

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Facultative Anaerobes as a Lemierre Syndrome Cause: A Pseudomonas Aeruginosa Infection, Managed with Aggressive Preemptive Therapy

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Introduction

Lemierre's Syndrome (LS) is a rare but lethal disease, which was first described in its core spectrum, by Andre Lemierre. The characteristic LS symptoms include acute infection of the head and neck region, internal jugular vein thrombosis and isolation of the bacterial agent from blood or other infected tissues.

During the first decades of the antibiotic era, both LS incidence and mortality decreased significantly and respectively the rate of suspicion amongst physicians decreased. However, the low rate of suspicion, combined with the fact that the progress on molecular medicine allowed quicker, unequivocal LS diagnosis, resulted on increased rates of LS cases around the globe [1]. The decreased use of antibiotics against tonsillitis, the bacterial resistance phenomenon and the systematic underreport of relevant cases in previous decades, may have contributed on the higher levels of LS prevalence.

Case Report

A forty-nine year old male was referred to our department for buccal swelling, trismus and fever. Physical examination revealed tender cervicofacial swelling on the left, from the level of the zygomatic arch to the respective submandibular area. Intraorally, there was obliteration of the vestibule and unilateral soft and hard palate edema. Trismus, difficulty in swallowing and caries were also observed. Laboratory evaluation revealed leukocytosis (WBC-White blood cells: 19,960/dL) and chest X-ray was normal.

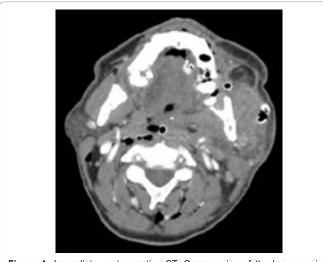


Figure 1: Immediate post-operative CT. Compression of the large cervical vessels.

Under LS suspicion, immediate intra- and extra-oral incision and drainage of the involved anatomical spaces were performed. Incision of the lateral pharyngeal space at the level of the soft palate was effective and a tracheostomy was performed to secure the airway. Post-operatively, a computed tomography (CT) was performed, which revealed occupation of the spaces, obstruction of the oropharynx and -expected- compression of the cervical carotid sheath (Figure 1).

The culture sample was taken during the operation and the results occurred after 5 days. The chemotherapy started before the laboratory results. The patient was placed on metronidazole and tazobactam. However, the pyogenic sample cultures from the lateral pharyngeal space demonstrated infection by *Pseudomonas aeruginosa* and the treatment was modified to ticarcillin-clavulanic acid. The patient's clinical status improved over the next days. CT scan on post-operative day #3 revealed improved internal jugular venous flows. The laboratory tests during day #7 implied an ideal response to the treatment (WBC: 11,560/dL) and the chest X-ray was normal.

During post-operative day # 9, the tracheostomy was closed. However, on a repeat CT scan, internal jugular vein thrombosis was present and the patient was immediately started on enoxaparin 50 mg twice daily followed by acenocoumarin (Sintrom). After anticoagulation treatment the INR (International Normalized Ratio) was 3.5.

Finally he was discharged on day 16, on per os chemotherapy. The discharge day, WBC levels (5,890/dL) and chest X-ray were normal. Follow-up CT scan and Doppler scan, after seventeen days, confirmed patency of the internal jugular vein (Figure 2) and the anticoagulation treatment stopped.

The case report's final version was reviewed and approved by the "Hippokrateion" General Hospital Scientific Board (President: Alexandropoulos Nikolaos MD).

Discussion

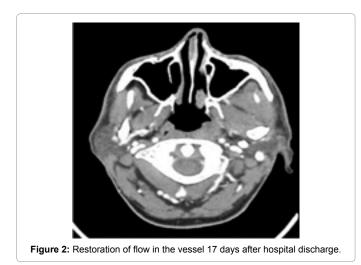
Lemierre syndrome is a rare form of metastatic septic thrombophlebitis, characterized in its fullblown form by superinfection with *Fusobacterium necrophorum*, internal jugular vein thrombosis and presence of distal septic emboli [2]. Tonsils and peritonsillar tissue are

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usually the primary sources of infection. Pharyngitis, parotitis, otitis media, mastoiditis and dental infections have been described as causes of the syndrome.

Fusobacterium Necrophorum is the most common bacterial agent isolated from cases of LS [3]. However, other pathogens such as *Porphyromonas assaccharolytica* were have been isolated [4]. In our case the pyogenic sample cultures demonstrated *P. aeruginosa* infection while the patient was immunoefficient. To our knowledge this is the first case report that correlates LS symptomatology with *P. aeruginosa* infection.

A pre-laboratory clinical LS diagnosis could be established based on clinical examination, CT- Doppler- and MRI (Magnetic Resonance Imaging)-scan [5]. Therapy cannot delay until the final confirmation and under suspicion of LS, the physicians should administer therapy immediately including minimization of risk factors (e.g. removal of intravenous catheter), administration of Intravene-antibiotics, aggressive treatment of the primary source of infection, and possibly anticoagulation. The recommended duration of therapy varies from 2 to 6 weeks. Surgical treatment is necessary in cases with recurrent septic emboli despite antibiotic therapy, and anticoagulation is indicated in cases of thrombus expansion. In our case, it seems that anticoagulation was beneficial, since patency of the vessel was restored in a short period of time while the in time chemotherapeutic treatment and the aggressive operative management prevented the expression of the full spectrum of LS symptoms. Reported mortality of similar cases nowadays varies between 0%-18%.

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