ECRONICON OPEN ACCESS

EC CLINICAL AND MEDICAL CASE REPORTS

Case Report

Thrombophlebitis of the Internal Jugular Vein: What if it is Lemierre's Syndrome?

A Zerhouni, S Benhamza*, M Lazrak, A Bensaid, Y Miloudi and N El herrar

20 August Hospital, Ibn Rochd UHC of Casablanca, Morocco

***Corresponding Author:** S Benhamza, 20 August Hospital, Ibn Rochd UHC of Casablanca, Morocco.

Received: March 12, 2020; **Published:** April 14, 2020

Abstract

Lemierre's syndrome (LS) is a rare condition characterized by suppurative thrombophlebitis of the internal jugular vein, preceded by a primary oropharyngeal infection with septic embolization and subsequent bacteremia. This condition is commonly associated with Fusobacterium. Due to its rarity, we report a clinical case of Lemierre's syndrome revealed by a pharyngeal abscess associated with *Streptococcus* spp.

Keywords: *Lemierre's Syndrome; Septic Thrombophlebitis, Streptococcus Spp*

Introduction

Post-angina anaerobic germ septicemia or necro-bacillosis was first described by Andre Lemierre in 1936 at the medical school of Middlesex hospital in London as follows: the appearance and repetition, several days after the onset of sore throat (and para-pharyngeal abscesses), febrile peaks, with initial pain or even more pulmonary infarctions and arthritic manifestations, which constitute a syndrome so characteristic that error is impossible [1]. This association will be subsequently reported under the name of "Lemierre syndrome" which is currently defined as a septic thrombophlebitis of the internal jugular vein or one of its collaterals caused by a focal sepsis, localized in most cases in the oropharynx, leading to metastatic infections. The advent of antibiotics has drastically reduced the incidence and mortality of this condition. We report here a case of Lemierre syndrome in a 54 years old patient and review the literature on this rare condition.

Case Presentation

54 years old patient treated for type II diabetes on Insulin, initially admitted for the management of a right cervical swelling associated with solid dysphagia evolving for a week in a febrile context. On admission we found a conscious patient, hemodynamically and respiratory stable, with a soft right inflammatory cervical swelling in addition to a trismus. The temperature had reached 39.5°C. After conditioning and stabilization, a paraclinical assessment was requested. A Ct-scan with contrast had revealed a right tonsil abscess, associated with multiple sub-mandibular abscessed lymphadenitis narrowing the pharyngeal lumen associated with a thrombosis of the right internal jugular vein. The laboratory analysis revealed an important inflammatory syndrome. An extension assessment was carried out, including a chest X-ray, an abdominal ultrasound as well as a transthoracic echocardiography, showing no metastatic locations.

After the treatment with a surgical drainage of the abscess, intravenous antibiotics and oral anticoagulation therapy at the intensive-care unit. Microbiological identification of intraoperative samples found a *Streptococcus* spp. The adaptation of antibiotic therapy followed the patient gradually recovered.

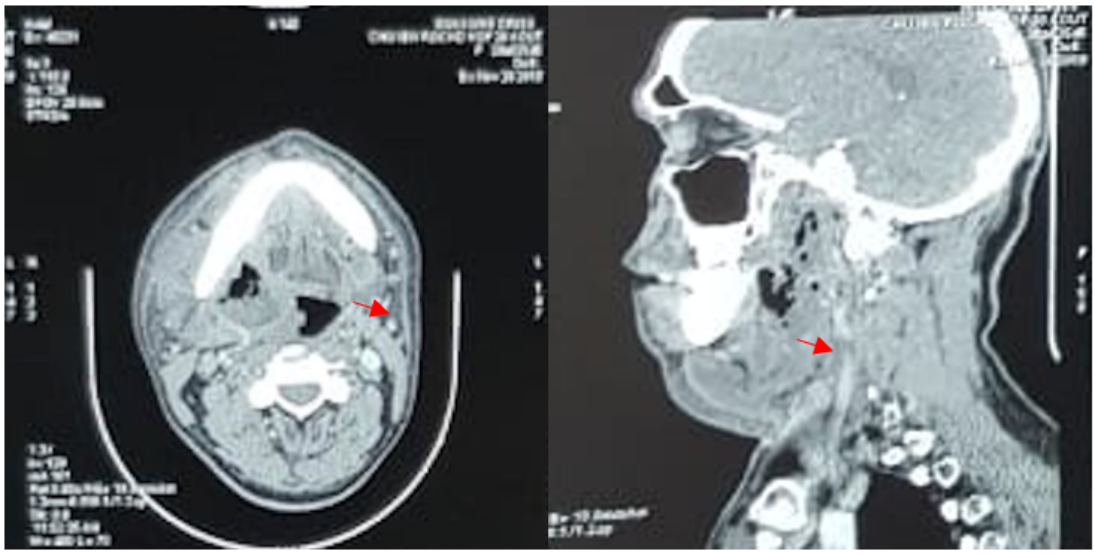

Figure 1: *Axial (left) and sagittal (right) CT scan with contrast of the neck revealing a thrombus (arrows) in the right internal jugular vein.*

Discussion

Lemierre's syndrome is a condition characterized by three key elements:

- Primary oropharyngeal infection
- Suppurative thrombophlebitis of the internal jugular vein
- Metastatic septic emboli.

Epidemiology

Lemierre's syndrome mainly affects young adults with 89% of patients between 10 and 35 years old [2]. The reported cases fell drastically with the advent of antibiotics from the 1940's so much so that it earned the pseudonym of the 'forgotten disease'. A resurgence has also been noted in recent years [3,4] due to a reflection of two main changes in medical practice: first, restriction in the use of antibiotics to treat upper respiratory tract infections, and second, tightening of the criteria for tonsillectomy.

Pathogenesis

The sequence of events leading from tonsillitis to full-blown Lemierre's syndrome or post-anginal septicemia are complex. A key step in developing of post-anginal septicemia is spread of organism from the tonsil to the internal jugular vein [2]. This dissemination may be hematogenous via the tonsil vein, lymphatically or by abscesses which were found in the proximity of the tonsil and that these pus collections spread deeper into the loose connective tissue of the pharynx and attach themselves to the walls of the veins, producing purulent periphlebitis and endophlebitis [5-7]. Once thrombophlebitis develops, metastatic embolization occurs. The most common embolization

sites are the lungs, affecting 85% - 92% of patients, leading to cough, pleuritic chest pain and/or hemoptysis [8]. Joints, liver, spleen, muscles, kidney, brain, bones, heart and meninges can also be involved.

Diagnosis

LS usually occurs a few days to 3 weeks after an ENT infection which is the essential diagnostic element to collect during the anamnesis. Symptoms are linked to septic locations: fever, sweating, chills, tachycardia, dysphagia, trismus, stiff neck, neck, pleural, abdominal or joint pain.

The laboratory analysis finds an important inflammatory syndrome, a hyperleukocytosis a leukopenia, a thrombocytopenia in addition to other signs evidence of damage to other organs. Microbiological identification remains the key examination for the diagnosis of LS, especially in atypical forms. The main pathogen implicated is *Fusobacterium necrophorum* (FN), anaerobic germ, Gram negative bacillus, commensal of the oropharynx, digestive and female genital tract. Its pathogenic nature results in a "necrobacillosis" of which SL is a form. Other pathogens have been reported either in combination with FN, in fact a polymicrobial flora is involved in 10 to 30% of cases, or exclusively. The bacterial ecology involved in this syndrome includes several species of *Fusobacterium, Streptococcus* (A, B, C, *oralis*) *Staphylococcus* (*aureus, epidermis*), *Enterococcus, Proteus mirabilis* [9,10]. Ultrasound Doppler of the IJ vein or CT neck with contrast can be used to detect thrombophlebitis of the IJ vein. Ultrasound is less sensitive, as recently formed thrombi have little echogenicity, and it only evaluates tissue above the clavicle; however, it is rapid, non- invasive and relatively inexpensive [8].

Management: Includes three important aspects:

Antibiotics: Despite the lack of randomized trials, must cover anaerobic germs (including FN), *Streptococcus* and *Staphylococcus* [10]. First line dual antibiotic therapy combines beta lactam with beta lactamase inhibitor (piperacillin-tazobactam) or third generation cephalosporin with metronidazole [5,6]. As a second intention, it is possible to use a monotherapy with imipenem or moxifloxacin [9]. Most sources recommend between 2 - 6 weeks of antibiotics in total.

Surgery: Drainage of infected cervico-facial tissue and abscessed collections may be necessary. Surgical ligation or excision of the internal jugular vein is exceptional, reserved for situations of unfavorable evolution: persistence of septic embolisms under optimal antibiotic treatment, extensive septic thrombosis, or even uncontrolled severe sepsis.

Anticoagulation: The risk of anticoagulant therapy would be to promote the spread of septic thrombosis, usually progressing favorably on antibiotics alone, weighed against a possible retrograde septic extension in the absence of anticoagulation. The indications for anticoagulation will therefore be reserved for high-risk situations linked to thrombosis: retrograde septic extension from the internal jugular vein, sigmoid sinus thrombosis, thrombophilia, lack of response to antibiotic treatment, or cerebral infarction [11].

Conclusion

Lemierre's syndrome is a rare but potentially fatal condition occurring primarily in young, otherwise healthy individuals. It's characterized by sepsis often evolving after a sore throat or tonsillitis and then complicated by various septic emboli and thrombosis of the internal jugular vein. The syndrome is often associated with an infection with Fusobacterium necrophorum, but other pathogens can be identified. Management is based on antibiotic treatment; anticoagulation is reserved for high-risk situations linked to thrombosis, and surgical treatment in special cases. To improve the prognosis, the early diagnosis and treatment of Lemierre's syndrome is essential.

Bibliography

1. Lemierre A. "On certain septicæmias due to anaerobic organisms". *The Lancet* 227.5874 (1936): 701-703.
2. Riordan T. "Human infection with *Fusobacterium necrophorum* (Necrobacillosis), with a focus on Lemierre's syndrome". *Clinical Microbiology Reviews* 20.4 (2007): 622-626.
3. Hagelskjaer Kristensen L and Prag J. "Lemierre's syndrome and other disseminated Fusobacterium necrophorum infections in Denmark: a prospective epidemiological and clinical survey". *European Journal of Clinical Microbiology and Infectious Diseases* 27.9 (2008): 779-789.
4. Brazier JS. "Human infections with *Fusobacterium necrophorum*". *Anaerobe* 12.4 (2006): 165-172.
5. "Lemierre's syndrome: current perspectives on diagnosis and management". IDR (2020).
6. Kuppalli K., *et al*. "Lemierre's syndrome due to *Fusobacterium necrophorum*". *Lancet Infectious Diseases* 12.10 (2012): 808-815.
7. Habert P., *et al*. "Pleuro-pneumopathie révélant une thrombophlébite septique de la veine jugulaire : pensez au syndrome de Lemierre". *Revue des Maladies Respiratoires* 33.1 (2016): 72-77.
8. Vijay V and Fattah Z. "Lesson of the month 1: Lemierre's syndrome: a reminder of the 'forgotten disease'". *Clinical Medicine* 18.1 (2018): 100-102.
9. Osowicki J., *et al*. "The long shadow of Lemierre's syndrome". *Journal of Infection* 74.1 (2017): S47-S53.
10. Ravi Jariwala., *et al*. "Methicillin-resistant *Staphylococcus aureus* as a Cause of Lemierre's Syndrome". *The Pediatric Infectious Disease Journal* 36.4 (2017): 429-431.
11. Mesrar H., *et al*. "[Lemierre's syndrome: Diagnosis, exploration, treatment]". *Revue de Médecine Interne* 39.5 (2018): 339-345.

Volume 3 Issue 5 May 2020