CASE STUDY

Lemierre Syndrome: Thrombosis of the Cavernous Sinus and Internal Carotid Artery Occlusion Secondary to Acute Sphenoid Sinusitis

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Abstract Lemierre syndrome typically consists of a septic thrombophlebitis of the internal jugular vein caused by Fusobacterium necrophorum. We present an unusual variant of Lemierre syndrome with cavernous sinus thrombosis and occlusion of the ipsilateral internal carotid artery secondary to sphenoid sinusitis caused by Streptococcus viridans.

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Introduction

Lemierre syndrome (LS) was first described in 1936. It consists of a septic thrombophlebitis of the internal jugular vein or one of its affluentes caused by an oropharyngeal infection produced by Fusobacterium necrophorum. Previously, it had a mortality rate of 90%, but this has currently been reduced to 5% through the use of antibiotic therapy. The literature also describes variants produced by non-opharyngeal infections and/or caused by other germs.

PALABRAS CLAVE
Síndrome de Lemierre; Sinusitis esfenoidal; Trombosis seno cavernoso; Oclusión carotidea

Síndrome de Lemierre: trombosis de seno cavernoso y oclusión de arteria carótida interna secundarios a sinusitis esfenoidal aguda

Resumen El síndrome de Lemierre consiste típicamente en una tromboflebitis séptica de la vena jugular interna causada por Fusobacterium necrophorum. Presentamos una variante excepcional del síndrome de Lemierre con trombosis del seno cavernoso y oclusión de la carótida interna ipsilateral secundaria a una sinusitis esfenoidal causada por Estreptococo Viridans.

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We present a rare case of LS variant with cavernous sinus thrombosis and occlusion of the ipsilateral internal carotid artery, secondary to an acute infectious process of the sphenoïd sinus by *Streptococcus viridans*.

**Case Report**

The patient was a 21-year-old male with no medical history of interest who presented upper respiratory tract catarrh of 1-week evolution, as well as right ocular discomfort with mild holocranial headache of 3 days evolution. His family doctor prescribed 875/125 mg of oral amoxicillin-clavulanate every 8h. Two days later, he attended the Emergency Unit due to an increase in ocular pain and onset of diplopia.

The patient was afebrile and presented no pharyngeal symptoms. An exploration detected anisocoria and paresis of the right sixth cranial nerve, with the rest of the examination being normal.

Laboratory tests found leukocytosis (22,000) and neutrophilia (88.3%), with the remainder of the parameters being normal. A cranial computed tomography (CT) scan without contrast was performed, which showed occupation of the rights maxillary and sphenoïd sinuses, with presence of some pneumocephalus “bubbles” in the ipsilateral temporal fossa adjacent to the cavernous sinus, which appeared hyperdense (Fig. 1a and b).

The patient was admitted and treated with ceftriaxone (2 g/24 h, intravenously [iv]) vancomycin (1 g/12 h, iv) and metronidazole (500 mg/6 h, iv). The next day, a cranial magnetic resonance imaging (MRI) scan with intravenous contrast was performed, which confirmed thrombosis in the right cavernous sinus and decreased calibre of the homolateral internal carotid artery (Fig. 1c).

Using an endoscopic approach, we proceeded to the surgical drainage of sinus structures by right ethmoidectomy and sphenoidotomy. The culture of the mucopurulent material obtained from the sphenoïd sinus was positive for *S. viridans*, sensitive to vancomycin.

At first, the patient did not undergo anticoagulation treatment while awaiting surgery, but 2 days later we prescribed 80 mg/12 h of subcutaneous enoxaparin.

Two weeks after admission, we obtained a CT-angiography which showed occlusion of the right internal carotid at its origin which had been asymptomatic (Fig. 2).

We conducted a new biopsy of the septal mucosa and cavum, which obtained negative results for *S. viridans* and positive results for *F. necrophorum*. The patient was prescribed a treatment consisting in 5 mg/kg twice/week of amphotericin B and 300 mg/24 h of oral acetylsalicylic acid for 6 weeks and discharged.

The final diagnosis was LS caused by sphenoïd sinusitis with thrombosis of the right cavernous sinus and occlusion of the ipsilateral internal carotid artery.

**Discussion**

Lemierre syndrome is defined as a case of septic thrombosis of the jugular vein with septic emboli caused by an oropharyngeal infection produced by *F. necrophorum*. Other pathogens, such as *Streptococcus*, *Peptostreptococcus*, *Bacteroides, Enterococcus* and *Eikenella* have subsequently been accepted as possible causative agents.

The diagnosis is established through the clinical symptoms, radiological findings and microbiological cultures. In our case, the symptoms included involvement of the right sixth cranial nerve, anisocoria, ocular pain and headache. All were suggestive of right cavernous sinus involvement, which was confirmed by radiographic observation of thrombosis secondary to acute sphenoïd sinusitis. The latter usually appears with nonspecific symptoms, the most common being frontal headache. The culture was positive for *S. viridans*.

Thrombosis/occlusion of the internal carotid artery is a very rare complication. Karkos et al. found 2 cases of thrombosis of the internal carotid artery. Toutou et al. published a case of LS with mastoiditis and pansinusitis, which presented right cavernous sinus thrombosis and stenosis of both carotid arteries. Recently, another case of left carotid thrombosis without jugular vein involvement has been published. In our case, the clinical course of the occlusion was asymptomatic and it was an incidental finding on a radiographic control.
The absence of thrombosis of the internal jugular vein does not exclude the diagnosis of LS, as this can occur in a wide variety of cranial and/or cervical veins.\(^6\)

Our patient presented an atypical case of LS, as we found an uncommon origin (sphenoid sinusitis)\(^4\) and causative germ (Streptococcus),\(^3\) and no septic embolism.\(^9\) This was the second case reported in the literature with occlusion of the internal carotid artery in the absence of jugular thrombosis.

The treatment of this condition is aimed at eradicating the focus of infection through surgical treatment and specific antibiotic therapy. Treatment with penicillin, clindamycin or chloramphenicol for 2–6 weeks can be prescribed empirically.\(^4\) The indication for anticoagulation and its duration have not been established.\(^1,5\)

In conclusion, despite being a rare condition, it is vital to be familiar with it in order to obtain an early diagnosis and good therapeutic response.

Conflict of Interest

The authors have no conflict of interest to declare.

References