# A case of blindness caused by Lemierre's syndrome

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## Introduction

Lemierre's syndrome is a systemic septic embolism caused by thrombophlebitis of the internal jugular vein and presenting with various distantly infected abscesses. Various diagnostic criteria exist, but there is no standardized definition. Lemierre's syndrome is generally believed to be caused by infection with anaerobic bacteria around the oropharynx. Delayed diagnosis can lead to dyspnea and deep neck infections due to acute airway constriction, which can lead to fatal outcomes. Ophthalmologic complications of Lemierre's syndrome are relatively rare. In this study, we report our experience with a patient with Lemierre's syndrome who lost her sight.

## Case

*Patient:* A 36-year-old woman.

*Chief complaints:* Fever, posterior cervical pain, facial swelling, right visual acuity loss

*Medical history:* No major diseases, including immune disorders.

*Family history:* Nothing in particular.

*Lifestyle history:* Nonsmoker and occasional drinker. The patient was not on any oral medications.

*History of present illness:* The patient developed a fever 1 week before the hospital visit. Two days later, she developed difficulty in swallowing and trismus and sore throat. By the following day, she had facial swelling and posterior cervical pain. She kept monitoring her condition at home. However, the symptoms did not improve, and she began having difficulty in moving, which prompted an ambulance request 1 week later and the patient's being transported to our hospital.

*Physical findings at the hospital visit:* Height, 155 cm; body weight, 55 kg; clear consciousness; body temperature, 38.0°C; heart rate, 110 bpm (regular); blood pressure, 98/76 mmHg; $SpO_2$, 95% (room air); respiratory rate, 27 breaths per minute; no chills and shivering; no night sweats; right-dominant facial swelling; examination of the oral cavity and pharynx proved difficult due to trismus; swelling and tenderness noted in the right cervical area; no stiff neck; no chest pain; clear pulmonary sounds; no clear abnormalities in the cardiac sound; flat and soft abdomen; no spontaneous pain and tenderness; and no limb swelling.

*Abnormal neurological findings (cranial nerves):* II, right visual acuity loss; III/IV/VI, right eye midline immobilization, no oculomotor disorder in the left eye; V, no left/right differences in facial sensation; VII, trismus/difficulty in opening the right eye; VIII, no left/right differences in hearing; IX/X, difficulty in swallowing; XI, no weakness or left/right differences in the sternocleidomastoid and trapezius muscles; XII, the tongue could not extend beyond dentition because of trismus.

*Laboratory findings at the hospital visit:* Arterial blood gas analysis (room air) showed pH, 7.409; $PaCO_2$, 38.9 Torr; $PaO_2$, 80.2Torr; and bicarbonate, 24.1 mmol/L. She had no respiratory failure. Blood work revealed a

white blood cell count of 21,900/μL, platelet count of $1.8\times 10^4$ μL, C-reactive protein level of 26.73 mg/dL, quantitative fibrinogen level of 680 mg/dL, Fibrin degradation product level of 4.8 μg/dL, and D-dimer level of 1.8 μg/dL, which indicated an elevated inflammatory response, thrombocytopenia, and high D-dimer level. The acute stage score of disseminated intravascular coagulation (DIC) was 4 points. Therefore, she was diagnosed with DIC.

*Images at the hospital visit:* The contrast-enhanced computed tomography (CT) showing a low-density area in the C2-6 prevertebral muscles, and a poorly enhanced area in the right internal jugular vein. (Fig.1.2) There are also Multiple ground-glass nodules are in both lungs, predominantly on the pleural side.(Fig. 3)

The patient was diagnosed with Lemierre's syndrome, based on the images indicating retropharyngeal abscess, thrombophlebitis, and septic pulmonary embolism.

After admission, the patient was managed in the intensive care unit.

At the time of admission, trismus was observed, and CT showed airway stenosis. Securing the airway was necessary. Therefore, the patient underwent emergency tracheotomy in the primary care room, after undergoing a platelet transfusion.

Local drainage of the retropharyngeal abscess was attempted, but it was impossible to visualize and treat the locality of the pharynx because of trismus; therefore, conservative treatment with antibiotics was administered. Meropenem 1 g IV every 8 hours and vancomycin 1 g IV every 12 hours were the empiric antimicrobial agents. Anticoagulant therapy was not started on admission because the patient was in a state of DIC at the time of admission. The patient was diagnosed with orbital-apex syndrome due to the spread of inflammation from the retropharyngeal abscess.

Treatments with antimicrobial agents gradually improved the inflammatory parameters, facial swelling, and posterior cervical pain. Blood culture sampled upon admission revealed *Fusobacterium necrophorum* . Therefore, the antimicrobial treatments were de-escalated to ampicillin/sulbactam 3 g IV every 6 hours on Day 6 after admission, based on the susceptibility results. Anticoagulant therapy was also started with systemic administration of heparin. Heparin was switched to a direct-acting oral anticoagulant on Day 11, as the patient had no exacerbation in her general condition.

On Day 35, antimicrobial treatments were de-escalated to amoxicillin 500 mg orally 3 times daily. Antimicrobial treatments were terminated on Day 42, after confirming that the abscess had disappeared completely on the follow-up CT scan.

However, even after the abscess disappeared, the patient's reduced visual acuity and oculomotor disorder did not improve. The patient was discharged with these symptoms remaining.

**Discussion**

In our patient, the period from the preceding infection to the appearance of Lemierre's syndrome symptoms was short; thus, we understand that an extremely intense condition manifested in our patient.

*Fusobacterium necrophorum* , a gram-negative anaerobic bacteria, is the causative agent of Lemierre's syndrome in most cases. The same bacterium was identified in our patient. The most common organ affected by septic embolism is the lungs, followed by joints such as hip joints, knees, and shoulders; skin and soft tissue; and the endocardium.[1,2]In a systematic review in 2020, Dasari et al.[3] found only 27 cases of ophthalmologic complications with Lemierre's disease reported between 2009 and 2019.[4] Among these 27 cases, the most common ophthalmologic complications were cranial nerve III/IV/VI palsy and oculomotor disorder due to external ophthalmoplegia. Abducens nerve palsy was noted in 12 (44.44%) cases. Cavernous sinus thrombosis was observed in 19 (70.37%) cases. The next most common symptoms were blepharoptosis (9 cases, 33.33%) and visual impairment (8 cases, 29.63%). Of the three patients with visual impairment, the condition of two patients recovered to some extent, but that of the remaining one patient did not.[3]

Ophthalmologic complications of Lemierre syndrome may be caused by inflammation spreading from the

cavernous sinus to the nerves and muscles. The cavernous sinus receives blood from various veins such as the superior orbital vein, intracranial vein, and parietal sinus. These extensive connections cause retrograde septic embolism from the internal jugular vein, resulting in cavernous sinus thrombosis.[3,4]

## Conclusion

We describe a patient who lost her vision because of Lemierre's syndrome. Ophthalmologic complications of Lemierre's syndrome are rare, and very few cases have been reported. Clinicians need to recognize that it is a serious condition that can lead to blindness in some cases.

## Conflict of interest

Authors declare no Conflict of Interests for this article.

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## Ethics Statement

**Approval of the research protocol: N/A**

**Informed consent:** The patient provided informed consent for treatment and for the report to be published.

**Registry and the registration no. of the study/trial** : N/A.

**Animal studies:** N/A.

**Conflict of interest:** Authors declare no Conflict of Interests for this article.

**Data Sharing and Data Accessibility** : Not applicable.

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## Figure Legends

Fig. 1 The cervical contrast-enhanced CT image showing a low-density area in the C2-6 prevertebral muscles. This is denoted by the orange circle.

CT, computed tomography

Fig. 2 A poorly enhanced area in the right internal jugular vein. This is denoted by the orange circle.

Fig.3 Multiple ground-glass nodules are in both lungs, predominantly on the pleural side. This is denoted by the orange circle.

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