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CASE REPORT

A case of blindness caused by Lemierre's syndrome

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Key Clinical Message

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.

Abstract

Lemierre's syndrome is a systemic septic embolism resulting from thrombophlebitis of the internal jugular vein. We report a case of blindness caused by Lemierre's syndrome, despite prompt diagnosis and early treatment. A 36-year-old woman was transported to our hospital. She presented with fever, facial swelling, and right visual acuity deterioration. Contrast-enhanced computed tomography revealed a retropharyngeal abscess and right internal jugular vein thrombosis. Blood culture revealed *Fusobacterium necrophorum*, suggesting Lemierre's syndrome. The patient had septic shock and disseminated intravascular coagulation. We diagnosed her visual impairment as orbital-apex syndrome due to spread of inflammation. After 6 weeks of antimicrobial drug treatments, her general condition had improved, and the retropharyngeal abscess had disappeared. On Day 49 of her illness, she was transferred to a rehabilitation hospital, but her visual acuity was not restored. Clinicians should be aware that Lemierre's syndrome can, although rarely, cause blindness.

KEYWORDS

blindness, Fusobacterium, Lemierre syndrome, retropharyngeal abscess

1 | INTRODUCTION

Lemierre's syndrome is a systemic septic embolism caused by thrombophlebitis of the internal jugular vein. Septic embolization is common, most often affecting the lungs, but may also involve bones, joints, or any organ.[1,2] Various diagnostic criteria exist, but there is no standardized definition.[3–5] Lemierre's syndrome is generally believed to be caused by infection with anaerobic bacteria around the oropharynx.[6] The most common pathogen of Lemierre's syndrome is *Fusobacterium necrophorum*, and the causative focus mostly originated from pharyngitis or tonsillitis.[7–9] The possible routes of infection include direct invasion, and lymphatic or hematogenous spread to the

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connective tissue and associated with abscess or distant septic embolic metastasis. Delayed diagnosis can lead to dyspnea and deep neck infections due to acute airway obstruction, which can lead to fatal outcomes.[10] Ophthalmologic complications of Lemierre's syndrome are relatively rare.[11–13] In this study, we report our experience with a patient with Lemierre's syndrome who lost her vision.

2 | CASE REPORT

A 36-year-old, previously healthy woman developed a fever 1 week before the hospital visit. Two days later, she developed difficulty in swallowing, 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.

When she came to our hospital, she was conscious but had a fever of 38.0°C and a tachycardia of 110 bpm. SpO2 was maintained at 95% in room air, but she was tachypneic at 27 bpm. Her face was swollen predominantly on the right side, and it was difficult to examine her oral cavity and pharynx. Cranial nerve abnormalities included right visual acuity loss and right eye midline immobilization. She also had difficulty in opening the right eye and had trismus. Arterial blood gas analysis (room air) showed pH, 7.409; $PaCO_2$, 38.9 Torr; PaO_2, 80.2 Torr; 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 1.8×10^4 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. The contrast-enhanced computed tomography (CT) showing a low-density area in the C2-6 prevertebral muscles (Figure 1) and a poorly enhanced area in the right internal jugular vein (Figure 2).

There are also multiple ground-glass nodules are in both lungs, predominantly on the pleural side (Figure 3).

The magnetic resonance imaging (MRI) showing a thrombus in the right sigmoid sinus (Figure 4).

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, she was at risk of

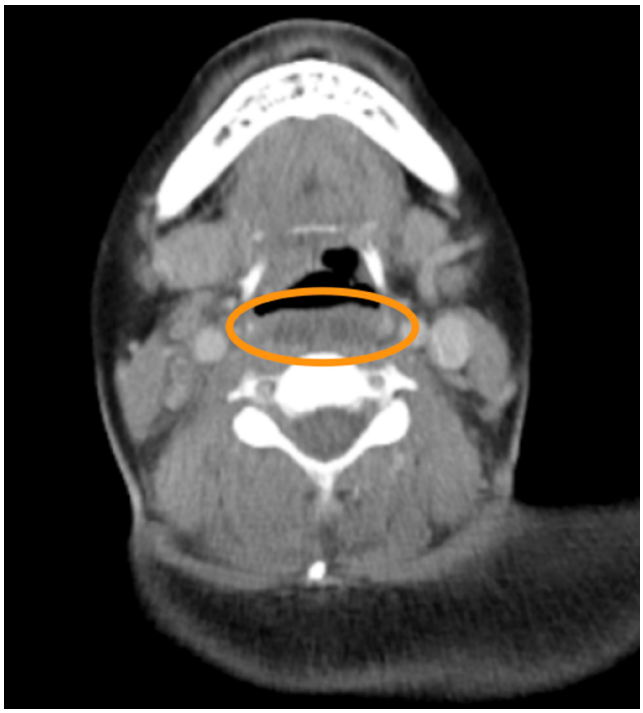

FIGURE 1 The cervical contrast-enhanced CT image showing a low-density area in the C2-6 prevertebral muscles. This is denoted by the orange circle.

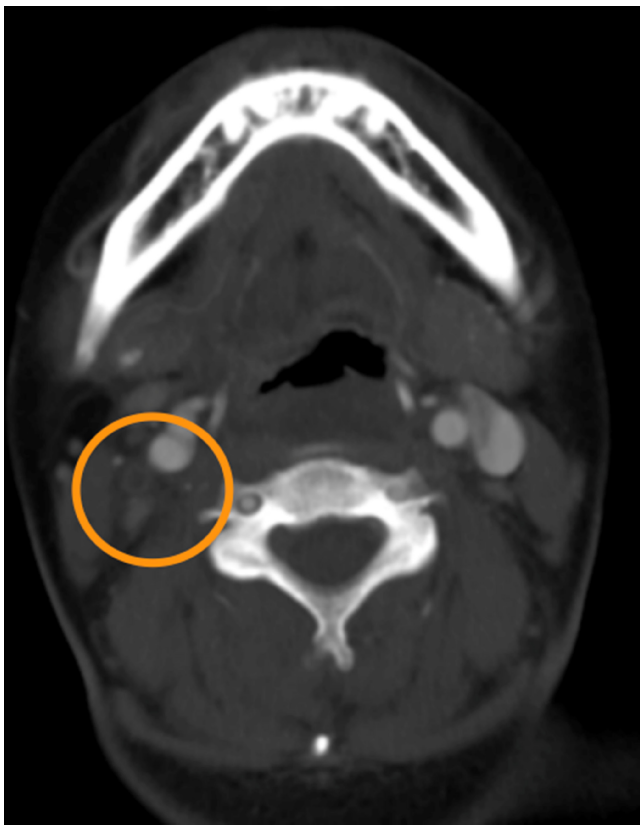

FIGURE 2 A poorly enhanced area in the right internal jugular vein. This is denoted by the orange circle.

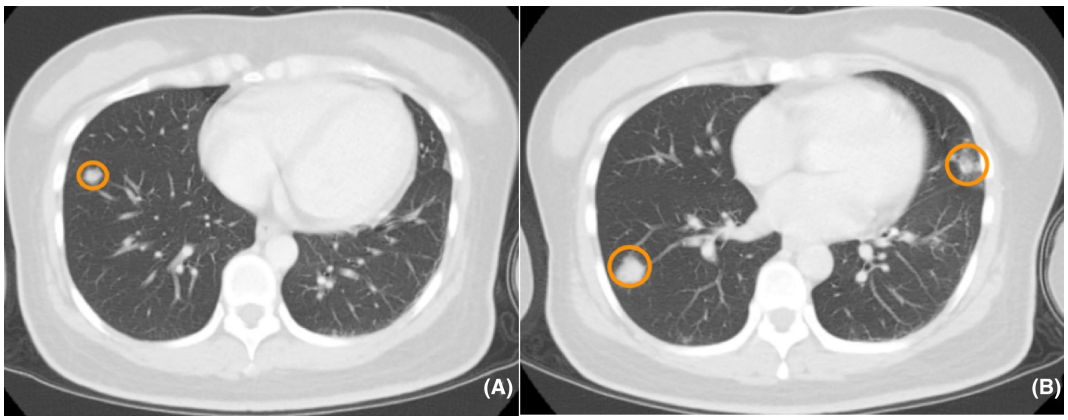

FIGURE 3 (A, B) Multiple ground-glass nodules are in both lungs, predominantly on the pleural side. This is denoted by the orange circle.

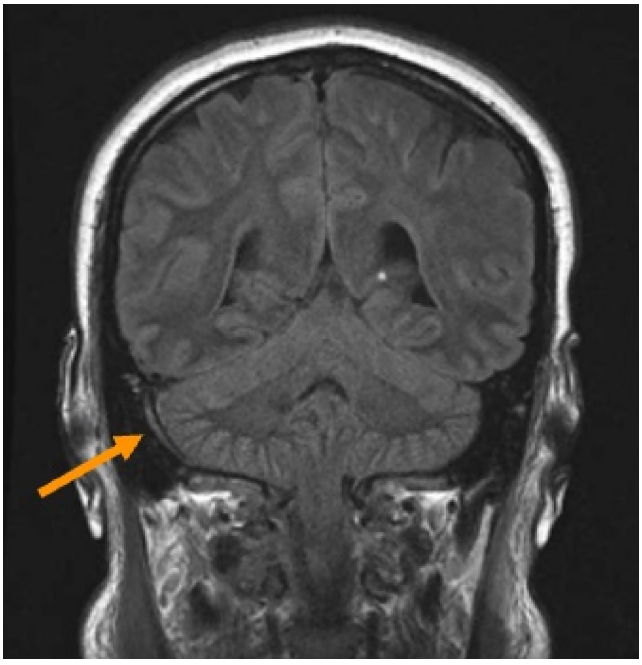

FIGURE 4 MRI showing a thrombus in the right sigmoid sinus. This is denoted by the orange arrow.

airway obstruction due to difficulty in expectorating sputum and saliva caused by trismus. 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. In general, an external approach through a transcervical skin incision is possible for huge abscesses. However, it is also highly invasive; therefore, conservative treatment with antibiotics was administered. Meropenem 1 g IV every 8 h and vancomycin 1 g IV every 12 h 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 h 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.

After de-escalation of the antimicrobial agent, there was no local exacerbation of infection, and WBC and CRP decreased; thus, antimicrobial treatments were de-escalated to amoxicillin 500 mg orally 3 times daily on Day 35 (Figure 5).

Antimicrobial treatments were terminated on Day 42, after confirming that the retropharyngeal 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.

3 | DISCUSSION

To begin with, retropharyngeal abscess usually affects children under 5 years of age because retropharyngeal lymph nodes tend to regress by about 6 years.[14,15] Therefore, it is rare for a retropharyngeal abscess to occur in adults. The most common cause in adults is trauma, such as foreign body ingestion or instrumental procedures. Nontraumatic origin of retropharyngeal abscess is rare in adults and is usually associated with immunosuppression.[14,15]

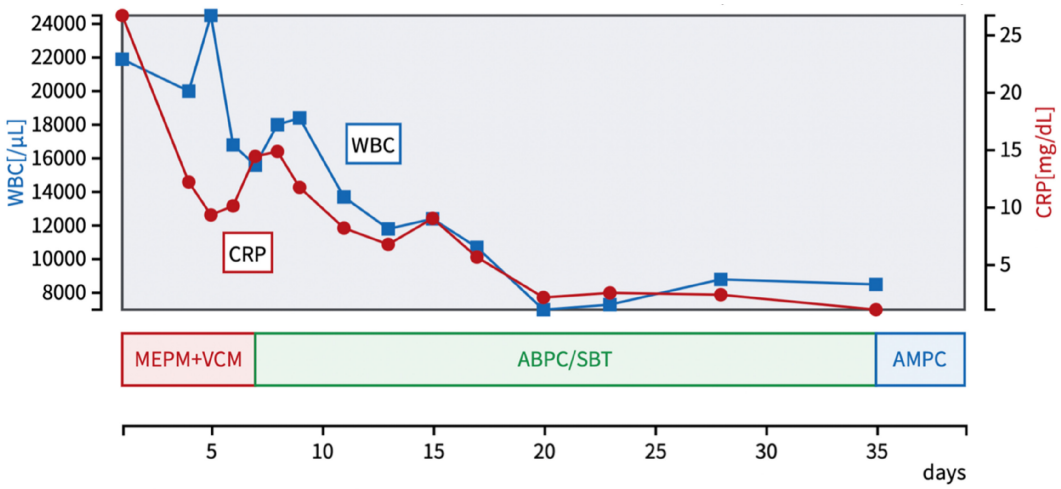

FIGURE 5 This figure shows that the treatments with antimicrobial agents gradually improved the inflammatory parameters.

This case of a retropharyngeal abscess in a woman is very unusual in that there was no history of trauma or immunosuppression.

The most common organ affected by septic embolism in Lemierre's syndrome is the lungs, followed by joints such as hip joints, knees, and shoulders; skin and soft tissue; and the endocardium.[1,2] Ophthalmologic complications of Lemierre's syndrome are rare, and very few cases have been reported.

In a systematic review in 2020, Dasari et al.[11] found only 27 cases of ophthalmologic complications with Lemierre's disease reported between 2009 and 2019.[11] 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 total of 27 cases, only one showed permanent visual impairment.[11]

Ophthalmologic complications of Lemierre syndrome may be caused by inflammation spreading from the cavernous sinus to the nerves and muscles. The cavernous sinus is formed by the meningeal and periosteal layers of the dura mater and is separated from the sphenoid sinuses by a thin bone. It receives blood from various veins such as the superior ophthalmic vein, intracranial vein, and parietal sinus (Figure 6A,B). These extensive connections cause retrograde septic embolism from the internal jugular vein, resulting in cavernous sinus thrombosis.[11–13] In Lemierre's syndrome, the abducens nerve is most likely to be affected because it is in the cavernous sinus and is susceptible to inflammation in the cavernous sinus, whereas the oculomotor, trochlear, trigeminal, and other nerves are in the dura mater of the cavernous sinus.[11–13] In the present case, ocular motility disorder was seen in all directions, suggesting that in addition to the abducens nerve, the oculomotor nerve and the trochlear nerve were also affected. This suggests that the inflammation in the cavernous sinus was very strong.

Furthermore, in addition to her ocular motility disorder, she also had a visual impairment, suggesting that her optic nerve was also affected. In general, inflammation of the cavernous sinus alone does not damage the optic nerve, so when the optic nerve is affected, the patient may have a complication of orbital-apex syndrome due to anatomical proximity.[16]

Orbital-apex syndrome is a complex neurological disorder characterized by vision loss from optic neuropathy and ophthalmoplegia due to the involvement of ocular motor nerves in the orbital apex.[16] It is often caused by various neoplastic, vascular, infectious, or inflammatory conditions.[17]

From the above, it also confirms that the inflammation in the cavernous sinus was so severe that it extended anteriorly to the superior orbital fissure and optic nerve tract, which was thought to have complicated the orbital-apex syndrome (Figure 6C,D).

It is not clear what caused the visual impairment that persisted after recovery from Lemierre's syndrome; in this case, failure to perform local drainage despite strong disease may be one of the causes.

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.

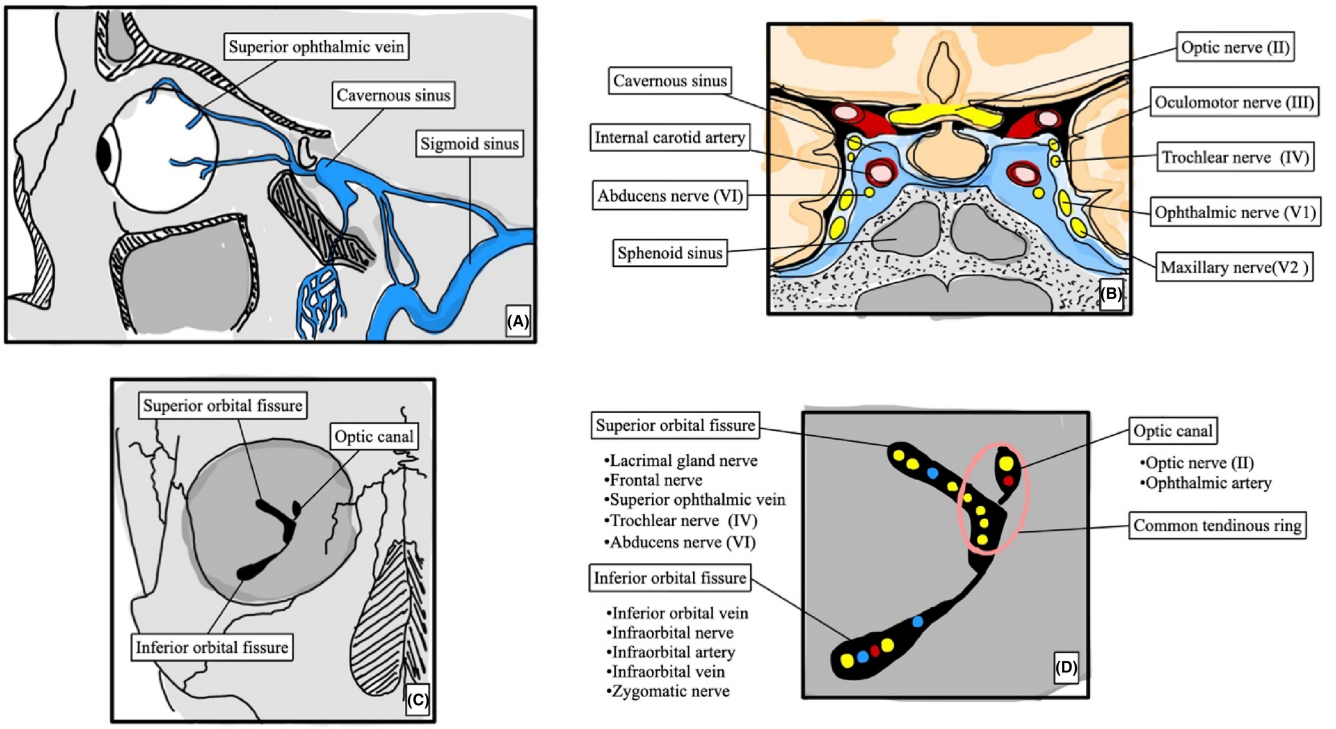

FIGURE 6 (A) Schema of sagittal section around cavernous sinus. (B) Schema of coronal section around cavernous sinus. (C) Schema of the orbital apex. (D) Schemas of the superior orbital fissure, inferior orbital fissure, optic canal, and the arteries, veins, and nerves running within each.

AUTHOR CONTRIBUTIONS

Tensei Suzuki: Data curation; writing – original draft. **Mitsuaki Kojima:** Supervision; writing – review and editing. **Raira Nakamoto:** Writing – review and editing. **Keiichi Kuriyama:** Writing – review and editing. **Shu Tanizawa:** Writing – review and editing. **Yuzuru Mochida:** Writing – review and editing. **Yuri Asakura:** Writing – review and editing. **Ayaka Shibano:** Writing – review and editing. **Tomohisa Shoko:** Funding acquisition; supervision; writing – review and editing.

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Authors declare no Conflict of Interests for this article.

DATA AVAILABILITY STATEMENT

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CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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