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# Blindness caused by septic superior ophthalmic vein thrombosis in a Lemierre Syndrome variant

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ABSTRACT

A 65-year-old man presented with right facial cellulitis and right blindness. Enhanced CT and MRI showed right facial cellulitis involved with pterigopalatine fossa. Additionally, orbital cellulitis, superior ophthalmic vein thrombosis, and pulmonary multiple nodules were observed. $^{18}$F-FDG PET/CT supported these findings. He was diagnosed with septic superior ophthalmic vein thrombosis accompanied with Lemierre Syndrome variant and was treated mainly by the administration of intravenous antibiotics. His symptoms and image findings improved after a few days of treatment, but the right visual loss has not recovered. Since septic superior ophthalmic vein thrombosis and Lemierre Syndrome both have life-threatening potential, early diagnosis and appropriate treatment are important and may contribute to reduce the incidence of severe complications. Septic superior ophthalmic vein thrombosis accompanied with Lemierre Syndrome is exceeding rare, and this case is the first report of blindness in Lemierre Syndrome. A literature review and discussion of septic superior ophthalmic vein thrombosis and Lemierre Syndrome are included.



## 1. Introduction

Septic thrombosis of the superior ophthalmic vein (SOV) is a rare disease which is often caused by infections of the mid-face. It may or may not involve cavernous sinus thrombosis [1].

Lemierre syndrome (LS) is defined as septic thrombosis of the internal jugular vein (IJV), secondary to an acute otolaryngologic or upper airway infection, resulting in metastasis septic complications [2]. Although this syndrome is now called the "forgotten syndrome", there has been an increase in reports recently [3].

This paper describes the case of a patient with septic SOV thrombosis with blindness caused by facial and orbital cellulitis originating from a mandibular decayed tooth. This is an exceeding rare case of an LS variant that exhibited some unusual characteristics, including blindness and SOV thrombosis.

## 2. Case report

A 65-year-old man was referred to the hospital because of right-sided facial cellulitis and blindness. He had a past history of diabetes and hypertension. He initially consulted a dentist for toothache in his right mandibular second molar and right-sided facial swelling. The dentist diagnosed a decayed tooth accompanied by facial cellulitis, and oral antibiotics were given; however, his symptoms worsened. He consulted an otolaryngologist at a general hospital 9 days after onset. Enhanced CT scan showed right-sided facial cellulitis involving the pterigopalatine fossa, and pulmonary multiple nodules were observed. A blood test indicated strong inflammation: C-reactive protein (CRP) was 29.8 mg/dl and the white blood cell (WBC) count was 19,600 μl with 80.5% neutrophils. The otolaryngologist suspected generalized sepsis and treated the patient with intravenous administration of meropenem (MEPM, 1.5 g/day). Heparin (10,000 U/day) intramuscular injection and oral administration of betamethasone sodium phosphate (4 mg/day) were also given. Lung nodules were suspected of being septic emboli, although multiple distant metastasis of a malignant tumor was not ruled out. Although his systemic state was stable and blood tests improved after 2 days of treatment, he had strong bilateral eye pain and eyelid edema. His right eye was blind without a light sense. He was therefore referred to our hospital.

Physical findings on admission were almost normal. $SpO_2$ was 98%, and body temperature was 37.2 °C. CRP: 12.6 mg/dl, WBC: 20,800/μl, Hb: 14.3 g/dl, platelets: $34.2 \times 10^4$/μl, GOT: 50 IU/l, GPT: 87 IU/l, and blood cultures: negative. He was unable to open his eyes due to severe edema of his bilateral eyelid and lower conjunctiva, and he also had trismus. He had no stiff neck, neck pain, or abdominal pain. His nose showed no abnormal findings.

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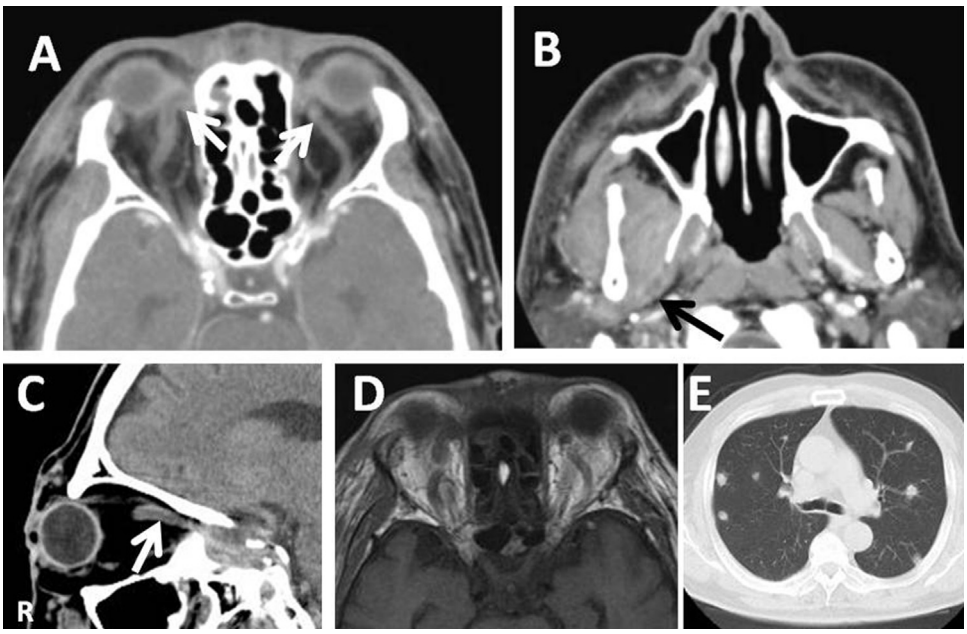


**Fig. 1.** (A) Axial enhanced CT image; arrows indicate bilateral superior ophthalmic vein dilation (the diameter of the right was 5.4 mm, and the left was 3.3 mm) with central hypointensity lesion on the right. No involvement of the cavernous sinus. (B) Hyperplasia of the right pterygoid muscle, percutaneous swelling of bilateral facial tissue, and right maxillary vein thrombosis (arrow) are observed. (C) Sagittal enhanced CT image; arrow indicates right-sided superior ophthalmic vein thrombosis. (D) T1-weighted MRI; bilateral orbital tissue swelling and dilated bilateral superior ophthalmic veins with no involvement of cavernous sinus. (E) Multiple pulmonary nodular lesions.

The CT scan showed dilation of the bilateral SOV with a right central hypointense lesion. Pulmonary multiple nodules were still present without changes (Fig. 1). There were no findings on the bilateral nasal or paranasal sinuses. MRI showed swelling of the bilateral orbital contents and dilation of both SOVs, but it was clear of intraorbital abscess and intracranial pathological changes (Fig. 1). There was no involvement of the cavernous sinus and IJV.

Ophthalmological examination disclosed right-sided visual loss without severe limitation of eye movement, and fundoscopic examination indicated no clear features of central retinal artery occlusion. Examinations of the left side were within normal ranges. Bilateral conjunctival vessels showed enlargement and tortuosity, more prominent on the right side. Fluorescein angiography disclosed marked prolongation of the arm-to-retina and retinal circulation time (Fig. 2).

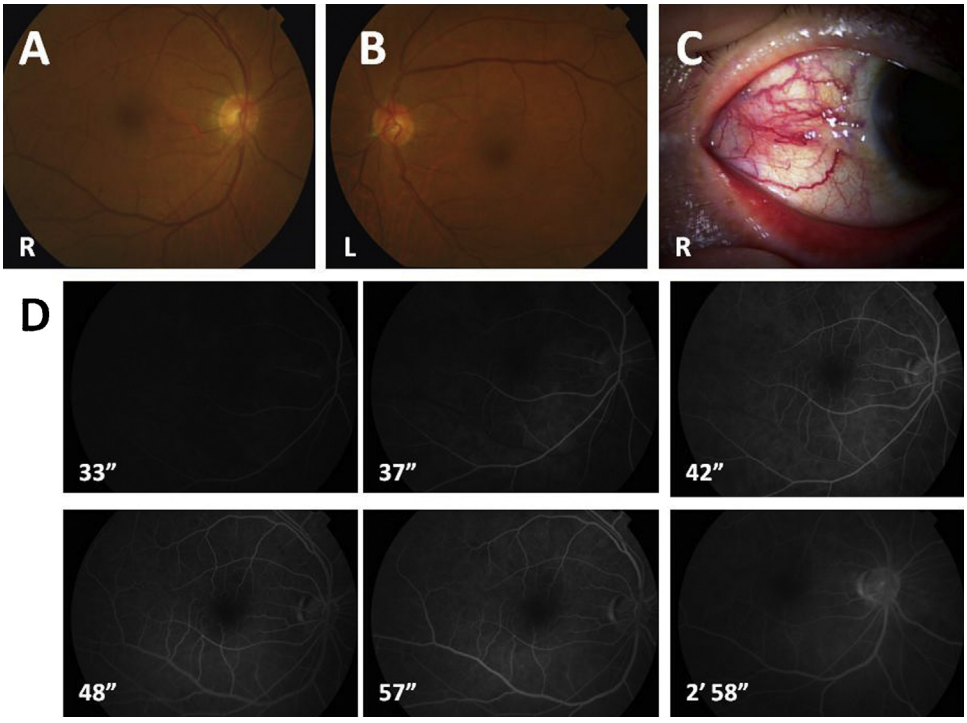


**Fig. 2.** (A, B) Image of both ocular fundus; no specific feature suggesting central retinal artery occlusion or venous stasis retinopathy. (C) Image of right eyeball; conjunctival vessels showed enlargement and tortuosity. (D) Fluorescein angiography performed on the 8th day of admission; markedly prolonged arm-to-retina circulation time (approximately 33 s; normal average 10–15 s) and retinal circulation time (approximately 24 s; normal average within 11 s) were confirmed.