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Concomitant cavernous sinus thrombosis and central serous chorioretinopathy in a patient with total ophthalmoplegia and monocular blindness: a case report

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Introduction: Cavernous sinus thrombosis (CST) is an unusual condition that can result in high mortality and morbidity rate if not treated immediately.

Case presentation: An Indonesian male, 47 years old, presented with total right ocular ophthalmoplegia followed by blindness, headache, ptosis, periorbital swelling, and hypoesthesia over the left V1 region. MRI of the brain showed suitable cavernous thickening until the right orbital apex, which in contrast showed enhancement suggestive of right Tolosa–Hunt syndrome. The patient was treated with a high dose of steroids, but the patient's complaints did not improve. The patient underwent digital subtraction angiography and found CST. The optical coherence tomography was found to be central serous chorioretinopathy. He was treated with an antibiotic, anticoagulant, and extraction of the right maxillary molar was performed to remove the source of the infection. After 3 weeks, visual acuity and optical coherence tomography examination were improved.

Discussion: A comprehensive examination, such as digital subtraction angiography, is essential to confirm CST diagnosis for the patient to obtain the right therapy. This report highlighted the value of the prompt diagnosis of CST through neuroimaging and the importance of proper therapy in patient management.

Conclusions: Early diagnosis, comprehensive examination, and proper treatment of CST will increase good prognosis.

Keywords: cavernous sinus thrombosis, central serous chorioretinopathy, total ophthalmoplegia

Introduction

Cavernous sinus thrombosis (CST) is so rare that data on the incidence are difficult to estimate. CST comprises about 1–4% of cerebral veins and CST epidemiology of about two to four per million people per year, and patient gender predominance in CST is uncertain^[1,2]. CST is a major life-threatening complication of orofacial infections^[1,3]. However, central serous chorioretinopathy (CSC) is characterized by serous detachment from the neurosensory retina and detachment of the retinal pigment epithelium (RPE) at the posterior pole, which usually affects young

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HIGHLIGHTS

- Cavernous sinus thrombosis (CST) is a very rare case.
- Early diagnosis and comprehensive examination play an essential role in CST management.
- Cerebral angiography is helpful in diagnosing CST.

and middle-aged adults^[4]. The pathogenesis of idiopathic CSC remains unclear. Recent studies involve multifactorial etiology and complex pathogenesis. It has been described in patients with high levels of endogenous or exogenous steroids, types A personality, and stress^[5]. This study aimed to report on an Indonesian male with CST and CSC. We report based on SCARE (Surgical CAse REport) 2020 guidelines^[6].

Case report

A 47-year-old Indonesian male presented with double vision, especially when he stared at an object. The right eyelid had been drooping for a few months, and within 2 months, it had been getting worse and had now completely closed. His eye movements were restricted in all directions, and his right face felt tingling. The patient complained of a headache for 2 months. The headache felt like a knot, especially on the right side of the head, with a Numerical Rating Scale of 5-6.

Physical examination showed right eye ptosis, swelling, total ophthalmoplegia in the right eye, and hypoesthesia over the left

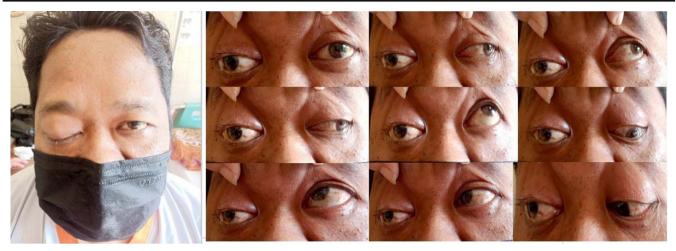


Figure 1. Physical examination shows right eye ptosis, swelling, and total ophthalmoplegia in the right eye.

V1 region (Fig. 1). Laboratory investigations showed increased C-reactive protein of 5.2 mg/dl, increased D-dimer of 1100 ng/ml, increased erythrocyte sedimentation rate of 89 mm/h, and increased fibrinogen of 864.6 mg/dl. Other laboratory results were within the normal limit – international normalized ratio (INR): 1.03, partial prothrombin time (PPT): 11.7 s, activated partial thromboplastin time (APTT): 30.2 s, blood urea nitrogen (BUN): 13 mg/dl, creatinine (Cr): 0.6 mg/dl, blood sugar (BS): 127 mg/dl, sodium (Na): 131 mEq/l, potassium (K): 3.7 mEq/l, chloride (CL): 97 mEq/l, serum glutamic-oxaloacetic transaminase (SGOT):

17 U/l, serum glutamic-pyruvic transaminase (SGPT): 10 U/l, hemoglobin: 12.6 g/dl, hematocrit: 38.6, leukocytes: $9.39 \times 10^{3}/\mu$ l, and thrombocyte: $409 \times 10^{3}/\mu$ l.

MRI of the brain showed suitable cavernous thickening until the right orbital apex (on the right cranial nerves III, IV, and VI course), which in contrast showed enhancement suggestive of right Tolosa–Hunt syndrome. With magnetic resonance angiography of his brain, there were no pictures of stenosis, aneurysm, or vascular malformation (Fig. 2). The patient was treated with a high-dose steroid IV and steroid topical in the right eye, but the

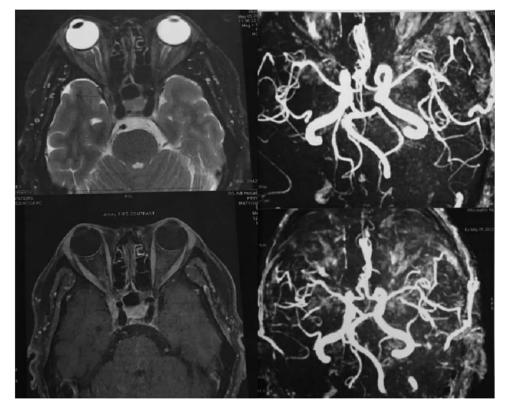


Figure 2. MRI of the brain presents suitable cavernous thickening until the right orbital apex (on the right cranial nerves III, IV, and VI course), which shows enhancement suggestive of Tolosa–Hunt syndrome. There is no picture of stenosis, aneurysm, or vascular malformation in the brain.

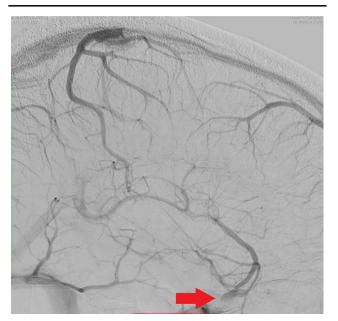


Figure 3. Digital subtraction angiography reveals a partial thrombus in the right cavernous sinus.

complaints did not improve. After treatment, he complained of blurred vision and blindness in the right eye, as well as continuous correct eye-watering. The visual acuity was 1/60 in the right eye.

The patient received a digital subtraction angiography examination. Cerebral angiography revealed a partial thrombus in the right cavernous sinus (Fig. 3). Optical coherence tomography showed subretinal fluid with pigment epithelial detachment in the right eye with a central retinal thickness reaching $625 \,\mu\text{m}$ (Fig. 4). An oral and maxillofacial surgeon first saw the patient. His panoramic photo showed opacity at the tips of the right second and third maxillary molars (Fig. 5). The extraction of the right maxillary molars (Fig. 5). The extraction of the right maxillary molars was carried out immediately. The patient was treated with the antibiotic metronidazole (500 mg) two times per day and cefixime (200 mg) two times per day. An anticoagulant was added in the form of rivaroxaban 10 mg/day. The patient improved in 3 weeks, his visual acuity became 5/60 in the right eye, and the optical coherence tomography examination got better with the central retinal thickness reaching $436 \,\mu\text{m}$.

Discussion

The cavernous sinuses (one on each side of the sella turcica, above and lateral to the sphenoid sinus, anterior superior orbital fissure, and posterior part of the petrous temporal lobe) are cavernous trabecular spaces created by the dura mater and filled with venous blood. This blood drains the superior and inferior ophthalmic veins and the superficial cortical veins anteriorly and then drains into the basilar plexus posteriorly via the superior and inferior petrosal sinuses^[1,7]</sup>. The structures that pass through the walls of the cavernous sinus include the divisions of the internal carotid, abducens, oculomotor, trochlear, and trigeminal nerves^[8]. Clinical manifestations of this patient were total ophthalmoplegia and ptosis. The anatomic course of these nerves explains cranial nerve palsy as they pass through the cavernous sinus, which can result in compression of the third, fourth, and sixth cranial nerves^[9]. Headaches in patients can be caused by secondary headaches and neurological deficits^[10]. Cerebral angiography revealed a partial thrombus in the right cavernous sinus. The evaluation of blood examination in cerebral venous thrombosis may reveal elevations in the C-reactive protein, erythrocyte sedimentation rate, and D-dimer^[1,11].

An MRI of the brain showed suitable cavernous thickening until the right orbital apex (on the right cranial nerves III, IV, and VI course), which in contrast showed enhancement suggestive of right Tolosa–Hunt syndrome. The patient was treated with highdose steroid IV and steroid topical in the right eye. Tolosa–Hunt syndrome responded well to corticosteroid treatment. Faster improvement of symptoms was seen in younger patients, and retro-orbital pain has been reported to settle within 72 h of steroid treatment^[12].

Von Graefe first described CSC in 1866 and named it 'relapsing central luetic retinitis'^[13,14]. Various names have since been used to describe this idiopathic detachment of the neurosensory retina^[15]. The incidence ratio of males to females is 6:1. Research's population was 5–6 per 100,000 people^[5]. CSC is an idiopathic, serous detachment of the neurosensory retina located in the macula^[16] that may be accompanied by RPE detachment^[17]. The pathogenesis is believed to involve hyperchoroidal perfusion and RPE barrier dysfunction^[17,18]. Previous reports have correlated the onset of CSC with various steroid use modes, including systemic and local applications^[19]. In this patient, a case of central serous chorioretinopathy was suspected due to the use of steroids both intravenously and topically in previous treatment or because of the inflammation

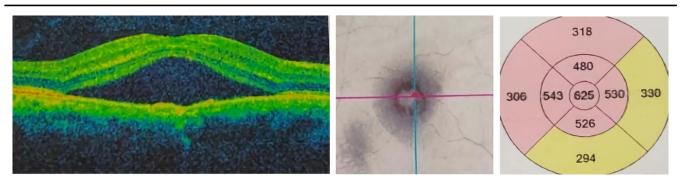


Figure 4. Optical coherence tomography before treatment presents subretinal fluid (A) with pigment epithelial detachment in the right eye with a central retinal thickness reaching 625 µm (B).



Figure 5. Panoramic photo shows opacity at the tips of the right second and third maxillary molars.

process. When the cause is treated, the serous retinal detachments can involve spontaneous resolution with the recovery of visual function^[20,21].

Digital subtraction angiography was performed because the patient's complaint did not improve with steroid therapy. Cerebral angiography revealed a partial thrombus in the right cavernous sinus. The treatment of CST complicating acute bacterial sinusitis relies on antibiotic therapy directed against *Staphylococcus aureus*, Gram-positive organisms, and anaerobes. However, its duration remains controversial and depends on the microbiologic findings and associated intracranial complications. Surgical drainage of the infected sinuses is usually performed through an endoscopic approach. Anticoagulation therapy could statistically lower mortality and morbidity rates if initiated within 7 days of the patient's admission to the hospital. This therapy should be given for at least 3 months^[22].

Conclusions

Patients with risk factors and ocular symptoms suspected of CST should get comprehensive diagnostic imaging studies with cerebral angiography. The clinician must immediately find the cause so the patient can receive the proper treatment.

Ethical approval

This case report does not require any ethical approval.

Patient consent

Written informed consent was obtained from the patient to publish this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author contribution

All authors contributed to data analysis, drafting and revising the paper, giving final approval of the version to be published, and agreeing to be accountable for all aspects of the work.

Conflicts of interest disclosure

The authors declare that they have no conflicts of interest.

Research registration unique identifying number (UIN)

- 1. Name of the registry: not applicable.
- 2. Unique identifying number or registration ID: not applicable.
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