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Clinical case

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Empyema secondary to a septic embolus in a patient with septic cavernous sinus thrombosis: a case report and review of the literature

Empiema secundario a embolismo séptico en un paciente con trombosis del seno cavernoso: reporte de un caso y revisión de la literatura

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ABSTRACT. Septic cavernous sinus thrombosis is an unusual and fatal pathology that can develop as a result of infectious or non-infectious conditions. There are very few reports of septic pulmonary emboli secondary to septic cavernous sinus thrombosis. We describe a case of a 31-year-old female patient who was immunocompromised due to methylprednisolone intake, had chronic pansinusitis and developed fever, headache, facial pain, eyelid swelling and proptosis with ophthalmoplegia in the left eye, loss of visual acuity. Computed tomography revealed thrombosis of the cavernous sinus, blood, sinus and pleural cultures isolated *Streptococcus pneumoniae*. The computed axial tomography scan revealed a right multiloculated empyema and a cavitated nodule corresponding to an infectious process, highlighting the presence of multiple nodules scattered between both lungs, suggestive of septic pulmonary embolism. A multidisciplinary strategy was implemented and after discharge, the patient is under surveillance with adequate recovery. This peculiar case shows the importance of a multidisciplinary approach to the management of this rare entity. We have noted all kinds of scenarios and the basis for this is a timely diagnosis and avoidance of possible complications to prevent serious and permanent consequences.

Keywords: empyema, septic cavernous sinus thrombosis, septic embolism.

RESUMEN. La trombosis séptica del seno cavernoso es una patología inusual y mortal que puede desarrollarse como resultado de afecciones infecciosas o no infecciosas. Hay muy pocos informes de émbolos pulmonares sépticos secundarios a una trombosis séptica del seno cavernoso. Describimos el caso de una mujer de 31 años inmunocomprometida por la ingesta de metilprednisolona, contaba con pansinusitis crónica y desarrolló fiebre, cefalea, dolor facial, hinchazón de párpados y proptosis con oftalmoplejía en ojo izquierdo, pérdida de la agudeza visual. La tomografía axial computada reveló trombosis del seno cavernoso, en los cultivos de sangre, senos nasales y pleural se aisló *Streptococcus pneumoniae*. La tomografía axial computada reveló un empiema multiloculado derecho y un nódulo cavitado que corresponde a un proceso infeccioso, destacando la presencia de múltiples nódulos dispersos entre ambos pulmones, sugestivos de embolia pulmonar séptica. Se implementó una estrategia multidisciplinaria y luego del alta, la paciente se encuentra en vigilancia con adecuada recuperación. Este peculiar caso muestra la importancia del abordaje multidisciplinario para el manejo de esta rara entidad. Hemos notado todo tipo de escenarios y la base de ello es un diagnóstico oportuno y evitar posibles complicaciones para prevenir consecuencias graves y permanentes.

Palabras clave: empiema, trombosis del seno cavernoso, embolismo séptico.

Abbreviations:

CAT = computed axial tomography.
SCST = septic cavernous sinus thrombosis.

INTRODUCTION

Cavernous sinus septic thrombosis (SCST) is a rare and serious thrombophlebitic process arising from sinus infections and less common otogenic, odontogenic, and

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pharyngeal sources.[1] Fever, headache, periorbital edema, and ophthalmoplegia are some of the most common symptoms of SCST, and early identification is critical for a favorable outcome.[2]

The cornerstone is broad-spectrum parenteral antibiotic therapy, although surgery is reserved for intracranial processes and control of the underlying focus of infection.[3]

In this article we report a case of SCST complicated by pansinusitis and multiple pulmonary septic emboli in a 31-year-old woman leading to loculated empyema.

CASE PRESENTATION

A 31-year-old woman with a history of poorly controlled Graves-Basedow disease, schizophrenia, and chronic sinusitis. It is important to mention that the patient reports having been treated with methylprednisolone 500 mg three times a day for five days due to her Graves-Basedow treatment. Four days before admission to the emergency room, the patient began experiencing high fever, left-sided headache, facial pain, eyelid swelling, and proptosis with ophthalmoplegia in the left eye. She reports that the reason for admission was loss of visual acuity *(Figure 1)*.

Physical examination revealed vital signs within normal limits, and the patient exhibited no signs of pallor or jaundice. She was oriented to time, place, and person and spoke comprehensively. Ocular examination revealed marked swelling, redness, and ptosis of her right eyelid, accompanied by hemorrhagic conjunctival hyperemia and exophthalmos. She also reported blurred vision. She presented with limited extraocular movement due to pain on abduction and adduction, intraocular pressure of 13 mmHg, lagophthalmos of 5 mm, eyelid erythema, and telangiectasias. There were no signs of meningeal irritation or sepsis/septic shock. A review of the presenting symptoms leads to a series of differential diagnoses,

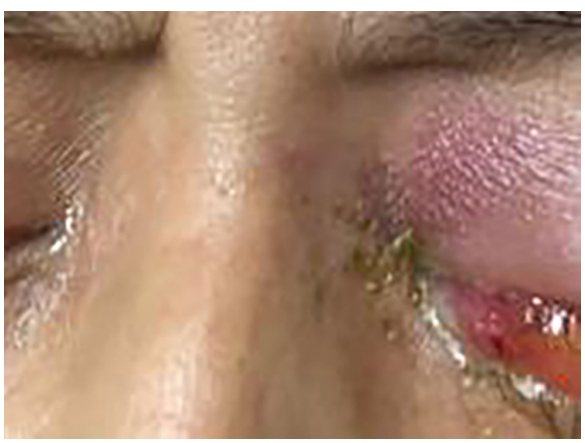

Figure 1: Case presentation photo.

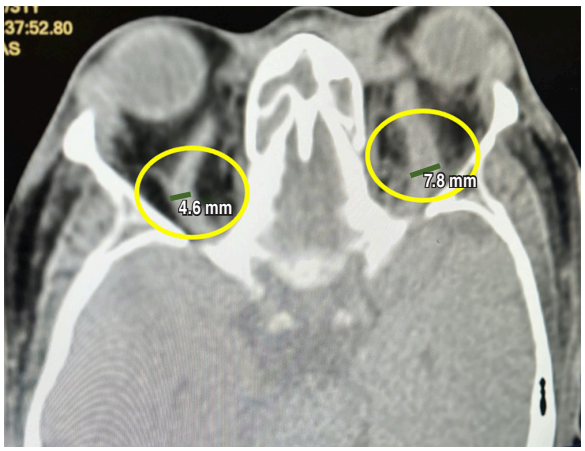

Figure 2: Computed tomography scan revealing bilateral superior ophthalmic vein thrombosis (yellow circles).

including, but not limited to, SCST, acute angle-closure glaucoma, subdural hematoma, subarachnoid hematoma, epidural and/or orbital infections. The most likely diagnosis was SCST due to the patient's history of chronic sinusitis and the immunodeficiency state secondary to the methylprednisolone overdose. It was also the most worrisome because of its higher probability of mortality and complications or disastrous outcomes.

A blood test showed a leukocytosis of 21.3 × 10^3/uL, with a slight shift to the left (92.5% neutrophils), elevated C-reactive protein of 27.8 mg/L, and D-dimer of 3.15 µg/mL. The platelet count and prothrombin time were within normal limits.

Orbital computed tomography (CT) showed chemosis, extraocular muscle thickening, ocular proptosis predominantly in the left eye, cellulitis and soft tissue edema. The right ophthalmic vein was enlarged to 4.6 mm and the left to 7.2 mm. The brain parenchyma was normal. A very marked pansinusitis was observed *(Figure 2)*.

Chest CT showed bilateral pulmonary nodules, multiloculated empyema was observed in the right hemithorax and a cavitated nodule in the left hemithorax, suggesting an infectious process *(Figure 3)*.

An MRI was ordered because it is the gold standard for diagnostic testing, but it was not performed for socioeconomic reasons. Two sets of blood cultures were performed and were positive for *Streptococcus pneumoniae*. Broad-spectrum antimicrobial treatment with ceftazidime and linezolid was initiated, along with low-molecular-weight heparin (LMWH), and was subsequently changed to enoxaparin. Transthoracic echocardiography showed no valvular involvement.

Due to marked pansinusitis, the patient was evaluated by the otorhinolaryngology service, endoscopic drainage of the ethmoid sinus was performed and cultures were taken, resulting in a positive result for *Streptococcus pneumoniae*.

The patient was also assessed by the thoracic surgery service for evacuation of multiloculated empyema, video-assisted thoracoscopic surgery transfusion (VATS), yielding 300 cm^3 of the locules and 2 mm visceral pleura. She maintained stable vital signs and did not develop complications, after 20 days in hospital she no longer had fever and the swelling in both eyes went down, she completed the antibiotic treatment, was discharged home and continues her follow-up as an outpatient *(Figure 4)*.

DISCUSSION

SCST is a rare, life-threatening thrombophlebitic process arising from sinus infections and less common otogenic, odontogenic, and pharyngeal sources. It can also result from severe injury or surgery, especially in the presence of thrombophilic disease.[3] In the pre-antibiotic era, it had a fatal prognosis, with a reported mortality rate of up to 80-100%; however, following the introduction of antibiotics and advances in diagnostic imaging, the incidence and morbidity and mortality rate have decreased to 30%.[3] Around 50% of patients have neurological sequelae, hence the importance of the disease and the need for early recognition, diagnosis and treatment.[4]

A variety of infectious organisms are capable of causing SCST, the vast majority of which are bacterial. *Staphylococcus aureus* is the most common pathogen (66%), followed by *Streptococcus species* (20%), oral anaerobic flora (10%), and gram-negative bacteria (5%). Fungal etiologies (*Aspergillus* and mucormycosis) of SCST are less common and usually arise in immunocompromised patients. Because SCST is so rare, statistics on its occurrence are scarce, so the incidence of SCST can be estimated at two to 13 cases per million per year, with a higher incidence in children.[1]

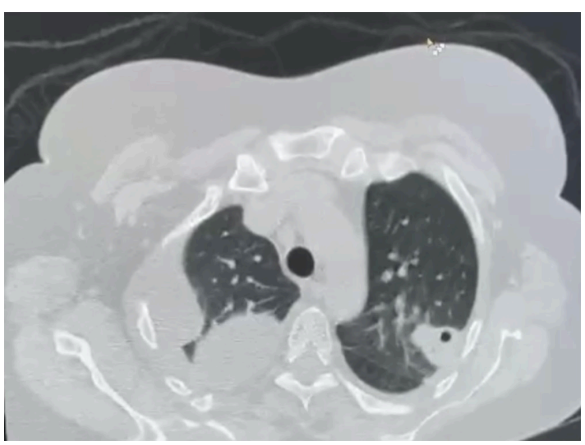

Figure 3: Chest scan demonstrating cavitary lung lesions and multiloculated empyema.

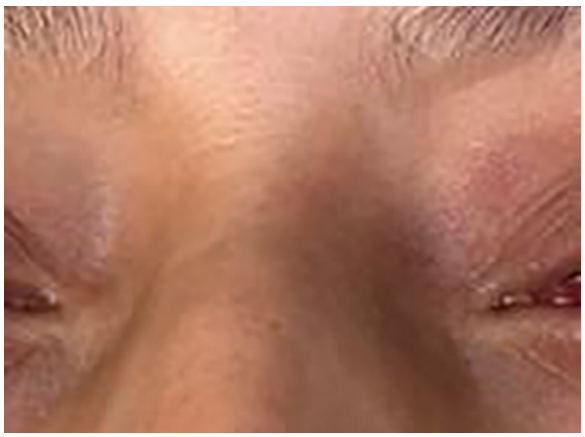

Figure 4: High photo.

Dural venous sinuses, blood can flow in either direction, allowing infectious agents to reach different parts of the brain and cause complications such as meningitis, dural empyema, or brain abscess. Most unilateral cavernous sinus infections eventually become bilateral due to communication between them via the intercavernous sinuses. Infection can also spread through the jugular vein to the pulmonary vasculature, resulting in septic emboli or abscesses, pneumonia, or empyema.[5] Most present with prominent acute features of sepsis, including tachycardia, vomiting, hypotension, confusion, and coma. Headache is present in 52-90% of cases.[3]

The ocular manifestations of SCST are the most universal features of this disease and result from obstruction of venous drainage from the orbit and dysfunction of the central nervous system traversing the cavernous sinuses, resulting in periorbital edema, ptosis, proptosis, chemosis, ophthalmoplegia, and vision loss.[3]

Chemosis, eyelid erythema, periorbital edema, and proptosis are most commonly seen (80-100% of cases), and symptoms present unilaterally at first, followed by bilateral progression within 24-48 hours of symptom onset.[3] Funduscopic examination reveals papilledema and/or retinal vein dilation in approximately two-thirds of patients; however, cloudy media may make visualization difficult in some cases with concurrent intraocular involvement.[3]

Thrombophlebitis associated with SCST may extend to the internal jugular vein, causing sore throat, neck mass/tenderness, anterior cervical lymphadenopathy, toothache or ear pain, dyspnea, hemoptysis, pleuritic chest pain, or trismus (Lemierre's syndrome).[3] Although ocular changes are the most common presenting symptoms and constitute a hallmark of this disease, other manifestations are common and will require appropriate treatment.

SCST can be complicated by intracranial processes such as meningitis, encephalitis, brain abscess, and subdural

empyema, as infection in the cavernous sinuses can spread through the valveless dural sinuses or the cerebral and emissary veins. In addition, infection can spread through the jugular vein to the pulmonary vasculature, causing lung abscesses, pneumonia, or empyema.[5]

Diagnosis is made on a clinical basis with appropriate radiologic imaging. Magnetic resonance imaging (MRI) is the most sensitive imaging modality (~95%) and has higher image resolution; however, contrast-enhanced CT may be preferable as it is easier to obtain, more cost-effective, and better able to identify the integrity of bony structures and/or the source of the underlying infection.[3]

There are no specific diagnostic laboratory tests for SCST; however, they may be useful in the evaluation of a patient with suspected SCST. The complete blood count is abnormal in most patients, and 90% demonstrate marked polymorphonuclear leukocytosis. Elevated levels of C-reactive protein, erythrocyte sedimentation rate, and D-dimer are also frequently observed.[3]

Due to the rarity of the disease, there are no randomized controlled trials (RCTs) to guide treatment.

Patient management, stabilization, acute resuscitation (including parenteral fluids and oxygen supplementation), and treatment of the underlying infection remain important first steps in all patients with suspected SCST.[5]

Parenteral antibiotics are the cornerstone of treatment and should be started immediately in all patients with SCST.

The initial empiric antibiotic regimen should include a third-generation cephalosporin, nafcillin, and metronidazole; however, if there is concern for methicillin-resistant *Staphylococcus aureus* (MRSA) or resistant strains of *Streptococcus pneumoniae*, then a regimen consisting of ceftriaxone, vancomycin, and metronidazole is preferred.[3]

Drainage of the cavernous sinus is rarely performed. Surgery, often performed endoscopically, is reserved for treatment of the primary site of infection; surgical procedures include sphenoidectomy, ethmoidectomy, maxillary antrostomy, mastoidectomy, craniotomy (for subdural empyema), orbital decompression, or ventricular shunt placement.[3]

The presence of a cavernous sinus thrombosis in our patient was an unexpected finding; this is the first time in the literature that a multiloculated empyema secondary to SCST has been reported in an immunocompromised patient. Morbidity and long-term sequelae remain high (50-75%) in patients who receive successful treatment for SCST, and the most common residual deficits observed in survivors include partial or complete vision loss (7-22%), cranial nerve palsy (predominantly III or VI), pituitary dysfunction, seizures, hemiparesis, facial disfigurement, and cortical vein thrombosis, with complete recovery achieved in less than 50% of cases.[3]

CONCLUSIONS

Cavernous sinus thrombosis remains a rare disease. Its rapid progression, high mortality rates, and potential for significant neurological disability in recovered patients require prompt diagnosis and treatment. This is the first case reported in the literature of multiloculated empyema secondary to SCST. We hope this case report can provide clinicians with insights into understanding SCST and, subsequently, lead to appropriate treatment of this disorder.

Conflict of interests: the authors declare no conflict of interests.

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