

A Rare Case of Odontogenic Orbital Cellulitis Leading to Septic Cavernous Sinus Thrombosis and Review of Literature

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Abstract

Septic cavernous sinus thrombosis (CST) is a rare condition that usually extends from the facial venous plexus or sphenoid sinus. Untreated or undertreated odontogenic disease can, although rarely, cause orbital cellulitis, orbital abscess, subperiosteal abscess, and even septic cavernous sinus thrombosis. We report a case of an untreated dental abscess that progressed to an ipsilateral orbital abscess/cellulitis, ipsilateral septic CST, bilateral superior ophthalmic vein thrombosis, meningitis, and ipsilateral blindness. We also reviewed the literature of this disease entity.

Keywords: *Odontogenic Abscess; Orbital Cellulitis; Septic Cavernous Sinus Thrombosis; Superior Ophthalmic Vein Thrombosis*

Introduction

Septic cavernous sinus thrombosis (CST) is an uncommon condition that was first described in 1778. With the advent of broad spectrum antibiotics, orbital complications of odontogenic origin are rarely seen. Septic CSTs of odontogenic origin are very rarely reported as a complication of odontogenic orbital cellulitis. Pubmed review of the English literature over the past 25 years only found three cases of septic cavernous sinus thrombosis of odontogenic origin with a detailed ophthalmic examination (Table 1) [1-3].

Authors	Journal / Year	Tissue Involvement	Age (year) / Gender	Comorbidity	Symptom onset to presentation	Offending microbials	Antibiotics	Initial visual acuity	Final visual acuity	Sinus involvement
Chandler Class V Odontogenic orbital cellulitis reported since 1992 (past 25 years)										
Allegrini et al	J Med Case Reports / 2017	Bilateral CST, right orbital abscess	46 / F	dyslipidemia	6 hours	Streptococcus, peptostreptococcus	Piperacillin/tazobactam	20/20, 20/20	N/A	Right maxillary
Kiddee et al	J Med Assoc Thai Vol 93 (9), 2010	BI CST, bl SOV Masticator/parapharyngeal space, R upper molar	49 / M	alcoholism	5 days	Pseudomonas aeruginosa	Ceftriazone, clindamycin	CF, 20/200	20/25, 20/25	None
Embong et al	Malaysian J Med Sci 2007	R Upper alveolar abscess, R parapharyngeal abscess, bilateral maxillary / ethmoidal sinusitis, L orbital cellulitis, L superior ophthalmic vein, bilateral CST	43 / M	none	3 days	Mixed growth	Vancomycin, Cefazidime, metronidazole	OS: LP	OS: LP	Bilateral maxillary and posterior ethmoidal sinusitis

Authors	Journal / Year	Tissue Involvement	Age (year) / Gender	Comorbidity	Symptom onset to presentation	Offending microbials	Antibiotics	Initial visual acuity	Final visual acuity	Sinus involvement
Chandler Class V Odontogenic orbital cellulitis reported prior to 1992										
Ogundiya et al	J oral maxillofac surgery 1989	Left Buccal cellulitis, pterygomaxillary space, inferior orbital fissure, posterolateral subperiosteal abscess, Left CST	35 / F	Exposure to TB alcoholism	11 days after third molar extraction	B hemolytic strep	IV Penicillin, clindamycin, Cefotaxidine, chloramphenicol	OS: 20/100	OS: 20/20	None
Bullock & Fleishman	J Maxillofac surg, 1985	Maxillary/ethmoid/sphenoid abscess, R. orb abscess meningitis	21 / M	Nephrotic syndrome	1 month	streptococcus	Tobaramycin, nafcillin, clindamycin, intrathecal gentamicin	N/A	LP	"Right maxillary, sphenoid, and ethmoid"
Bullock & Fleishman	J Maxillofac surg, 1985	Maxillary abscess, left orbital abscess, large subdural empyema of supraorbital dura,	12 / M	none	2days post extraction	streptococcus	Cephalothin, gentamicin, clindamycin, methicillin, chloramphenicol	NA *	N/A *	Right pansinusitis

* Patient expired from cardiopulmonary arrest

Table 1: Literature case summary of Chandler Class V orbital cellulitis with detailed ophthalmic exam

Case Presentation

A 68 year-old Caucasian male with no past medical or ocular history presented with 3 weeks of progressive periorbital pain and decreased vision of the left eye, preceded by 6 weeks of left peridontal pain. External examination revealed severe periorbital swelling, purulent periorcular drainage, marked hemorrhagic chemosis, and proptosis of the left eye (Figure 1). Visual acuity was 20/30 on the right eye and hand motion on the left eye with left relative afferent pupillary defect (RAPD). Extraocular movements were full on the right and fully restricted on the left. Intraocular pressures were 21mmHg on the right eye and 9mmHg on the left eye. Slit lamp examination was normal on the right and showed severe corneal edema with no fundusoscopic view on the left. Computer tomography of maxillary face on admission showed intraconal abscess of left orbit and preseptal abscess of left eye. Patient was started on vancomycin and ampicillin/sulbactam on admission by the infectious disease team. Patient underwent incision and drainage of left orbital abscess on day 1. Abscess culture grew methicillin sensitive *Staphylococcus aureus* (MSSA) and *Staphylococcus capitis* (sensitive to cefazoline, imipenem and oxacillin). As a result, on day 2, antibiotic regimen was changed to vancomycin, ceftriaxone and metronidazole for better gram negative coverage.

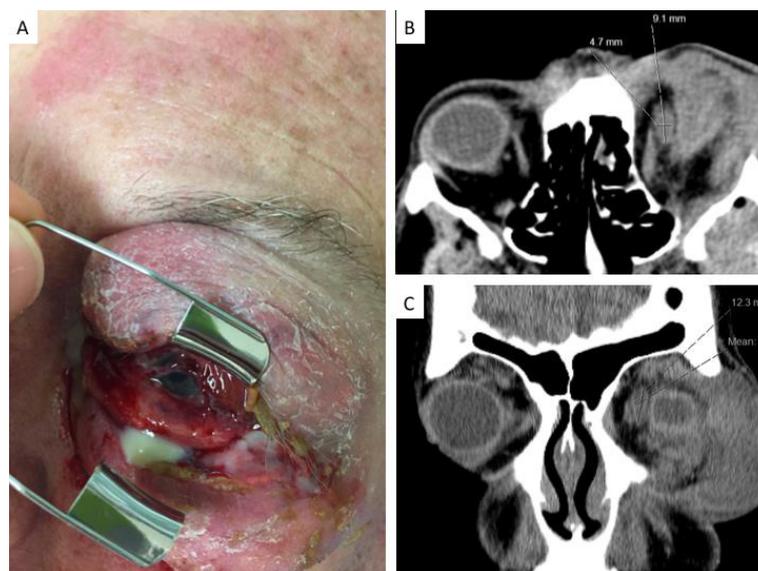


Figure 1: Clinical presentation and contrast computer tomography (CT) of maxillary/facial scans on day-1
 (A) Marked proptosis, hemorrhagic chemosis and purulent drainage noted from lower fornix, left eye
 (B) Post contrast axial CT image shows intraconal orbital abscess lateral to medial rectus muscle (12.3 x 9.1 x 4.7mm)
 (C) Post contrast coronal CT image shows the same intraconal orbital abscess as in Figure 1(B)

His visual acuity of the left eye improved to 20/200 on day 2 but quickly declined to no light perception (NLP) on day 3. On day 3, the visual acuity of the right eye also declined to 20/40 from 20/20 along with new onset chemosis. CT showed prominent conical distortions of both globes and bilateral superior ophthalmic veins thrombosis (Figure 2). Infectious intracranial extension was noted with the bilateral cavernous sinus thrombosis (Figure 2). Also noted was inflammation of the left pterygoid muscles and proximal left retromaxillary region. These findings, along with meningitis, were confirmed on subsequent Magnetic Resonance Imaging (MRI) of the brain and orbits. Repeat orbitotomy and Caldwell-Luc procedure of the left maxillary sinus were undertaken with subsequent extraction of all left upper molars.

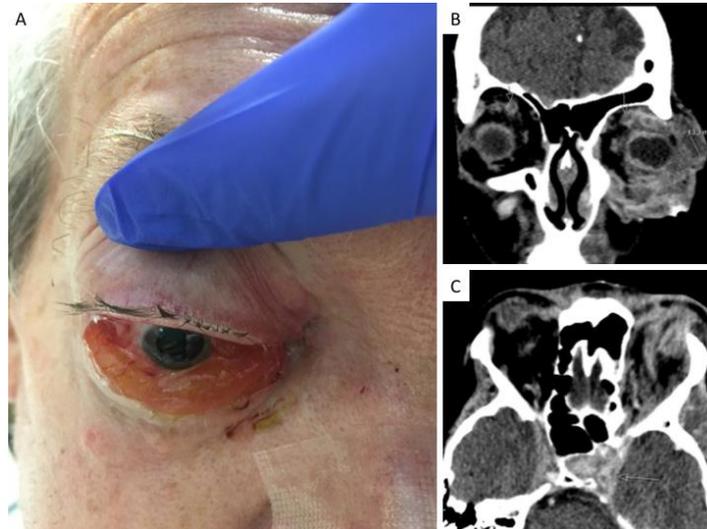


Figure 2: Clinical presentation and contrast computer tomography (CT) of maxillary/facial scans on day-1
 (A) Progression of chemosis of the right eye on day 3
 (B) Coronal contrast CT image shows periorbital infection involving the left orbit with a 1.3 cm abscess involving the soft tissues overlying the lateral orbit. Also noted is thrombosis of bilateral superior ophthalmic veins (arrows)
 (C) Axial CT image shows thrombosis of the left cavernous sinus (arrow)

The abscess culture from the second orbitotomy MSSA and *Staphylococcus capitis* (sensitive to cefazoline, imipenem and oxacillin), blood culture on admission grew Gram positive and Gram negative bacilli on day 5. As a result, antibiotic regimen was changed to vancomycin and cefepime for broader gram negative coverage. The speciation of blood culture was confirmed to be *Streptococcus anginosus* and *Rosamonas gilardii* (no sensitivity study) on day 9, and the antibiotic regimen was then changed to vancomycin and meropenem for *Rosamonas* coverage per infectious disease service as it is mostly sensitive to imipenem and resistant to third generation cephalosporin [4].

The cavernous sinus thrombosis was managed medically with intravenous antibiotics as above and anticoagulation with apixaban. The chemosis of the right eye resolved with 5 days of pulse intravenous methylprednisolone (Figure 3). Vision on the right eye improved to 20/25. Patient was discharged from hospital with 4-week duration of meropenem and fluconazole (for oral candidiasis coverage).

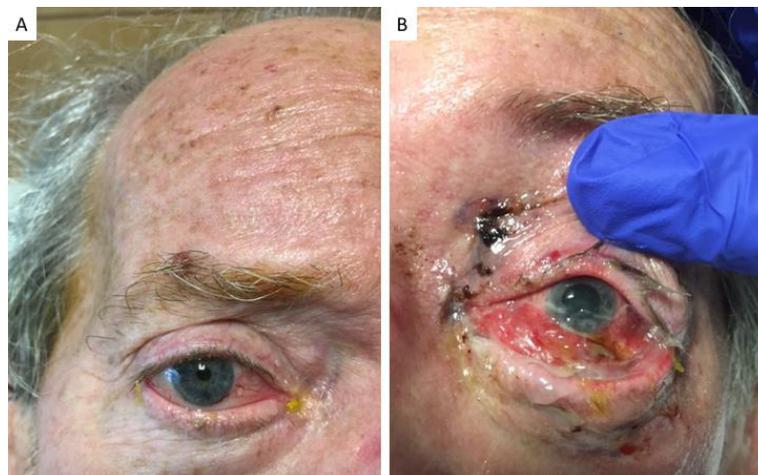


Figure 3: Clinical presentation on day 13
 (A) Complete resolution of chemosis in the right eye
 (B) Improvement of hemorrhagic chemosis and proptosis in the left eye

Discussion

Orbital abscess formation is a serious infectious burden and can arise from a diverse set of niduses. An odontogenic nidus is especially rare, with a prevalence of 1.3% [5]. Youssef et al found that 45.8% of patients had a final vision of light perception or worse [6]. Visual prognosis tends to be poor if the initial loss is severe [6]. The rarity of odontogenic orbital infection, combined with the high visual morbidity, makes diagnosis a challenge and early recognition important. We are going to review the classification of orbital inflammation, and the associated anatomy and path of infectious spread, and microbiology and management of odontogenic orbital abscess.

The Chandler system includes five classes of orbital inflammation and infection based on the anatomic extent: Group 1 for preseptal cellulitis, Group 2 for orbital cellulitis, Group 3 for subperiosteal abscess, Group 4 for diffuse orbital abscess, and Group 5 for cavernous sinus thrombosis [7]. Superior orbital fissure syndrome and orbital apex syndrome can occur when the orbital inflammation spreads along optic canal and ophthalmic vein [7]. Superior orbital fissure syndrome (SOFS) is marked by the involvement of the third, fourth, fifth and sixth cranial nerve and a parasympathetic block. When SOFS progresses to optic neuropathy, it is termed Orbital Apex Syndrome (OAS). Orbital apex syndrome includes signs of superior orbital fissures syndrome and optic neuropathy. The progression from SOFS and OAS to CST is usually abrupt, with worsening orbital signs due to impaired venous drainage from the orbit. When an orbital cellulitis and abscess spreads to the cavernous sinus, the contralateral vision is at risk [8]. Other serious entities of the central nervous system from advanced complication of orbital inflammation and infection include meningitis, cerebritis, epidural and subdural empyema, and brain abscess [7].

Due to the complicated anatomic relation of the orbit and adjacent structures, there are at least three postulated paths for a maxillary molar abscess to spread to the orbit. The first two paths are via adjacent soft tissue spread, while the third is via venous spread of septic thrombosis. First, the abscess penetrates the buccal cortical plate into the paranasal sinuses and then erodes through either the orbital bones or following the natural soft tissue conduit, including infraorbital neurovascular canal. Second, the maxillary molar abscess can spread posteriorly into retromaxillary soft tissues, then into infratemporal and pterygopalatine fossa, which extends into maxillary sinus, and then to the orbit via inferior orbital fissure [6]. Third, odontogenic infection is drained via through the venous system into valveless pterygoid venous plexus which anastomoses with inferior ophthalmic vein, or into the facial vein and the angular vein which drain into the superior ophthalmic vein [1,8,10].

There may be multiple routes of infection spread in our cases. However, the purulent cavity of the affected molars, which was tracked to pterygoid fossa, along with the early extensive maxillary sinusitis, suggests that primary path of spread starts from the abscess of the first upper molar, spreading onto retromaxillary soft tissue, onto pterygopalatine fossa into maxillary sinus, and then into the orbit via inferior orbital fissure.

When cultured, odontogenic orbital abscesses tend to be a polymicrobial mix. Gram-positive aerobes (*S.aureus* and *epidermis*, *Streptococcus*), anaerobes (*Bacteroides*), and a few oral pathogens (*Peptostreptococcus*, *Prevotella*, *Fusobacterium*, hemolytic *Streptococcus*) can be isolated[2,7] *Streptococcus anginosus* is one of the most virulent pathogens and can potentially lead to necrotizing orbital cellulitis [7]. Broad antibiotic coverage is thus recommended at high doses for both aerobes and anaerobes. Progression to septic CST warrants steroid use while being covered with board spectrum antibiotics to decrease orbital inflammation and may aid in preventing vascular collapse secondary to pituitary dysfunction [11]. Anticoagulation is somewhat controversial in the management of CST [11].

Orbital cellulitis of odontogenic origin is a rare entity with a poor prognosis. Inadequate treatment can lead to cavernous sinus thrombosis, meningitis, and blindness. The pillars of treatment remain surgical incision and drainage, antibiotic coverage for both aerobes and anaerobes, and eradication of the source [5]. Prompt surgical explorations and drainage should be performed if there are worsening orbital signs despite broad spectrum antibiotic coverage or if progression to the contralateral orbit occurs. It is important to note that development or resurgence of orbital cellulitis can follow the extraction of an infected tooth [12]. In summary, early recognition remains important in preserving visual function. Considering every source, including the rare odontogenic origin, is an important first step. New or worsening orbital signs following odontogenic infection or dental intervention warrant prompt urgent evaluation and treatment.

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