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].

<table>
<thead>
<tr>
<th>Authors</th>
<th>Journal / Year</th>
<th>Tissue Involvement</th>
<th>Age (year)/ Gender</th>
<th>Comorbidity</th>
<th>Symptom onset to presentation</th>
<th>Offending microbials</th>
<th>Antibiotics</th>
<th>Initial visual acuity</th>
<th>Final visual acuity</th>
<th>Sinus involvement</th>
</tr>
</thead>
<tbody>
<tr>
<td>Allegrini et al</td>
<td>J Med Case Reports / 2017</td>
<td>Bilateral CST, right orbital abscess</td>
<td>46 / F</td>
<td>dyslipidemia</td>
<td>6 hours</td>
<td>Streptococcus, peptostreptococcus</td>
<td>Piperacillin/tazobactam</td>
<td>20/20, 20/20</td>
<td>N/A</td>
<td>Right maxillary</td>
</tr>
<tr>
<td>Kiddee et al</td>
<td>J Med Assoc Thai Vol 93 (9), 2010</td>
<td>BI CST, bl SOV Masticator/parapharyngeal space, R upper molar</td>
<td>49 / M</td>
<td>alcoholism</td>
<td>5 days</td>
<td>Pseudomonas aeruginosa</td>
<td>Ceftriazone, clindamycin</td>
<td>CF, 20/200</td>
<td>20/25, 20/25</td>
<td>None</td>
</tr>
</tbody>
</table>

Chandler Class V Odontogenic orbital cellulitis reported since 1992 (past 25 years)
**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 periodontal pain. External examination revealed severe periorbital swelling, purulent periocular 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 funduscopic view on the left. Computer tomography of maxillary face on admission showed intraconal abscess of left orbit and preseptal abscess on the 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.

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

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<tr>
<th>Authors</th>
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<th>Tissue Involvement</th>
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<th>Sinus involvement</th>
</tr>
</thead>
<tbody>
<tr>
<td>Ogundiya et al</td>
<td>J Oral Maxillofac Surgery 1989</td>
<td>Left Buccal cellulitis, pterygomaxillary space, inferior orbital fissure, posterolateral subperiosteal abscess, Left CST</td>
<td>35 / F</td>
<td>Exposure to TB alcoholism</td>
<td>11 days after third molar extraction</td>
<td>B hemolytic strep</td>
<td>IV Penicillin, clindamycin, Cefotaxime, chloram phenicol</td>
<td>OS: 20/100</td>
<td>OS: 20/20</td>
<td>None</td>
</tr>
<tr>
<td>Bullock &amp; Fleishman</td>
<td>J Maxillofac Surg, 1989</td>
<td>Maxillary/ethmoid/sphenoid abscess, R. orb abscess meningitis</td>
<td>21 / M</td>
<td>Nephrotic syndrome</td>
<td>1 month</td>
<td>streptococcus</td>
<td>Tobramycin, nafcillin, clindamycin, intrathecal gentamicin</td>
<td>N/A</td>
<td>LP</td>
<td>&quot;Right maxillary, sphenoid, and ethmoid&quot;</td>
</tr>
<tr>
<td>Bullock &amp; Fleishman</td>
<td>J Maxillofac Surg, 1985</td>
<td>Maxillary abscess, left orbital abscess, large subural empyema of supraorbital dura,</td>
<td>12 / M</td>
<td>none</td>
<td>2 days post extraction</td>
<td>streptococcus</td>
<td>Cephalothin, gentamicin, clindamycin, methicillin, chloram phenicol</td>
<td>NA *</td>
<td>N/A *</td>
<td>Right pansinusitis</td>
</tr>
</tbody>
</table>

* Patient expired from cardiopulmonary arrest

**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 ophthalamic 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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References


