A Rare Case of Orbital Cellulitis with Progressive Calvarial Osteomyelitis

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Abstract
Orbital cellulitis is a potentially sight and life-threatening complication of acute sinusitis, and the association with osteomyelitis is rare in the era of antibiotic-use. A 13-year-old girl presented with coryzal symptoms and severe headache, with a CT head being consistent with a diagnosis of pansinusitis and orbital cellulitis with abscess formation. She proceeded to have surgical drainage through a combined endoscopic and external approach to intraorbital abscess drainage with frontal trephine. She was also diagnosed with progressive calvarial osteomyelitis involving the right frontal bone, treated with a prolonged course of intravenous antibiotics. Our case highlights the importance of a high index of suspicion for complications of sinusitis. Multimodal imaging is essential to establish the extent of infection, and a multi-disciplinary approach is integral to manage this rare complication.

Keywords
sinusitis, cellulitis, osteomyelitis, infection, surgery

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Background
Orbital cellulitis is a potentially sight and life-threatening disease, and often develops as a complication of acute sinusitis. It is seen more frequently in children compared with adults,1 and most commonly presents with swelling of the eye, limited eye movements, and loss in visual acuity and color vision.2 While it can often be managed conservatively through the use of broad-spectrum antibiotics, the development of a subperiosteal or orbital abscess is likely to require surgical intervention.3 In cases secondary to sinusitis, the frontal bone can be affected due to a direct extension of a frontal infection.4 Here we report our case of orbital cellulitis with progressive calvarial osteomyelitis, to increase awareness of this rare development in the pediatric population.

Case Presentation
A 13-year-old girl presented to the Emergency Department with a 1-week history of coryzal symptoms and severe headaches. In the week leading up to her presentation the headache intensity had continued to increase up to the point where she could barely move. The presence of bilateral periorbital swelling, particularly on the right side, and reduced range of motion indicated an emergency ophthalmology review within 24 hours. The patient had a past medical history of recurrent headaches and dizziness which first presented 5 months ago, however an MRI brain taken during this period showed no pathological changes and at the time she was discharged. The provisional diagnosis at the stage of initial presentation included preseptal or orbital cellulitis most
likely secondary to pansinusitis, with a differential diagnosis of subperiosteal or orbital abscess.

She was admitted and started on intravenous antibiotics on admission: ceftriaxone, vancomycin, and metronidazole. The initial ophthalmology review indicated preserved optic function; however, this was associated with a significant soft tissue swelling along with reduced color vision. Both were seen in conjunction with diplopia and painful eye movements, which warranted close monitoring of her vision and further radiological investigation.

As shown in Figure 1 below, a CT scan of the head and sinuses indicated a 20 × 18 × 3 mm right subperiosteal orbital collection insinuating around the superior orbital margin, extending posteriorly into the orbit for approximately 14 mm. Furthermore, a right frontal intracranial, extradural collection, with a depth of approximately 5 mm was noted.

These findings were consistent with a diagnosis of pansinusitis and orbital cellulitis with abscess formation. Due to the presence of the abscess without apparent involvement of the cavernous sinus, a diagnosis of orbital abscess was made (Stage 4 as per Chandler’s Classification).

The results of a brain and sinus MRI in the subsequent week showed a reduced volume of the previously discovered anterior epidural collection. There was enhanced tissue overlying the convex region of the frontal bone, suggesting progressive calvarial osteomyelitis involving the right frontal bone. A neurosurgical review was advised, and this continued to be monitored with repeat imaging, seen below in Figure 2.

**Treatment**

Following the combined input of the infectious diseases, ophthalmology, rhinology, and neurosurgical teams, it was concluded that in view of the radiological findings and the severity of signs on examination, surgical intervention would be recommended.

The surgical intervention comprised of a combined endoscopic and external approach to intraorbital abscess drainage, with a frontal trephine to access the superolateral extent of the abscess. The anterior epidural collection was managed conservatively with IV antibiotics. Due to the unusual superolateral extension of the abscess, it would be inaccessible by endoscopic approach alone. The surgery occurred the day after her initial presentation, with IV fluids given overnight and ondansetron prescribed PRN to help prevent nausea and vomiting.

Post-operatively, a 7-week course of IV antibiotics was organized by the Outpatient Parenteral Antibiotic Therapy (OPAT) team in view of the calvarial osteomyelitis.

**Outcome and Follow-Up**

She was regularly reviewed via telephone consultations and outpatient reviews by the pediatric ENT, ophthalmology, neurosurgical, and infectious diseases team, with a total follow up period of 6 months.

During this period, she underwent serial MRI scans to monitor the anterior epidural collection, and the calvarial osteomyelitic changes on the initial MRI scan.
A scan taken 8 weeks after her initial presentation showed reduced dural thickening and subperiosteal enhancement of the right frontal bone compared to prior imaging, with no features of progression. Her final MRI taken 2 months later showed only subtle abnormality remaining, with Figure 3 below demonstrating the resolution of the osteomyelitis over time. Her most recent outpatient review revealed a complete

Figure 2. Post-contrast fat-suppressed sequences through the orbits, anterior cranial fossa and frontal bone at the time of diagnosis (A–C) and 1 month following treatment (D–F). The extensive right periorbital and lateral extraconal inflammation seen at diagnosis (A—arrowheads) demonstrates interval regression (B—arrowheads). Similarly, the extradural subfrontal collection (C—short arrows) reduces significantly to be replaced by enhancing phlegmon on the subsequent study (D—short arrows). At diagnosis, there is right frontal subperiosteal and dural enhancement (E—arrowheads) but no marrow enhancement. On the subsequent study, there is persistent dural and subperiosteal enhancement focal thinning of the bone (F—arrowheads) and enhancement of the marrow compatible with osteomyelitis (thin arrows).

Figure 3. Series of axial post-contrast fat-suppressed MR sequences from September (A), October (B), and December (C) demonstrating progressive reduction in intra-osseous, dural, and periosteal soft tissue enhancement (arrowheads) in keeping with gradual resolution of osteomyelitis.
resolution of symptoms and she was discharged from care.

Discussion

The most common predisposing factor for orbital cellulitis development is sinus disease, especially in the pediatric population.6

Due to the widespread use of broad-spectrum antibiotics, osteomyelitis is decreasing in occurrence. While it remains a problem in lower socio-economic areas,7 our case represents how it must still be considered in all countries worldwide. Infection spreads through the diploic veins of Breschet communicating with the marrow cavity of the frontal bone, allowing spread to the frontal bones of the skull.8 However, osteomyelitis still remains an incredibly rare condition, only affecting 0.0002% of the population.9 The annual incidence is usually much higher in males and in adults older than 50 years,10 making our case particularly rare. In cases of acute presentation, there may not be any noticeable changes detected radiographically, taking up to 10 to 12 days for bone loss to appear.11

When the frontal sinus is involved, there is a higher prevalence of intracranial complications compared to infections of other sinuses.12 This further highlights the need for a high index of suspicion of complications from orbital cellulitis in cases similar to the one reported here.

While orbital cellulitis can often be diagnosed from the physical examination and presenting symptoms alone, multimodal imaging with CT and MRI is essential in establishing the extent and nature of infection, helping guide the multidisciplinary management. The use of CT can confirm the extent of the infection and provide evidence to guide surgical intervention. This however must be balanced against the risk of radiation exposure in the pediatric population. CT scanning is often used as the first modality due to its rapid acquisition times, availability, and delineation of osseous structures; however, the high sensitivity for detecting inflammatory changes in bone marrow and soft tissues has made MRI the modality of choice for detecting osteomyelitis. In this case, it is unclear whether the osteomyelitis developed later in the disease course as early osteomyelitis may not be apparent on CT.13

Conclusion

This case represents a unique clinical presentation rarely seen in the pediatric population, demonstrating how we must maintain a high index of suspicion for complications of sinusitis, such as orbital cellulitis. In cases of advanced orbital cellulitis, multimodal imaging can be a useful tool to diagnose rare intracranial and bony complications, such as osteomyelitis. In order to appropriately manage complicated cases such as this, a multi-disciplinary team approach is integral.

Author Contributions

TK contributed to the conception of the work, acquisition and interpretation of the data, and drafting the manuscript. SG contributed to the design of the work, acquisition of the data, and revising the manuscript. EB contributed to the design of the work, interpretation of the data, and revising the manuscript. PT contributed to the design of the work, acquisition of the data, and revising the manuscript. All authors gave final approval of the version to be published and agree to be held accountable for all aspects of the work.

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Ethical Approval and Informed Consent

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