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Case Report

Craniofacial *Actinomyces* osteomyelitis evolving from sinusitis

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ABSTRACT

Craniofacial *Actinomyces* osteomyelitis progression is rare, as patients are soon treated. A 56-year-old male smoker presented with sinusitis and was managed medically. This patient failed to follow up and presented 1 year later with erosive bony disease. He was managed medically and surgically; however, his disease evolved to include his midface, skull base, and cranium. He underwent staged debridement and free tissue reconstruction. His disease is controlled but not cured. The literature includes case reports and small series describing limited disease treated successfully with surgical and medical management. Although craniofacial *Actinomyces* osteomyelitis is uncommon, it can become debilitating. This case demonstrates how craniofacial *Actinomyces* osteomyelitis can progress and highlights the benefit of a multidisciplinary approach.

Introduction

Actinomyces species are anaerobic filamentous Gram-positive bacteria that are commensal organisms in the human mouth, digestive, and genital tracts [1,2]. However, these organisms can become pathological, particularly in patients with risk factors such as poor oral hygiene, history of mucosal breach or trauma, male gender, diabetes, immunosuppression, alcoholism, and malnutrition [1,3]. Cervicofacial actinomycosis describes osteomyelitis of the facial skeleton related to actinomycosis and may involve deformity and abscess formation, most frequently affecting the mandible [4]. Treatment often involves debridement and prolonged intravenous antibiotics [1,4]. We describe a particularly challenging case of midface, which progressed to bony destruction extending to the skull base and cranial fossa, and which ultimately required multidisciplinary management.

Case report

A 56-year-old man with history of tobacco abuse and recent dental extraction initially presented with a 4-month history of right facial pressure, right nasal obstruction, and clear nasal drainage. Anterior rhinoscopy and nasal endoscopy demonstrated diffuse mucosal congestion without purulence, and bone windows on computed tomography imaging demonstrated mucosal thickening and frothy secretions in the left maxillary

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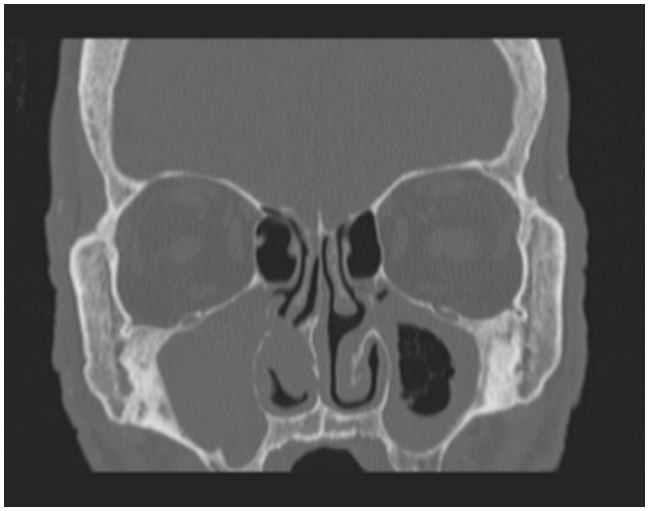

Fig. 1 – Computed tomography of coronal sinus in bone windows demonstrating mucosal thickening and frothy secretions in the left maxillary sinus with complete opacification of the right maxillary sinus consistent with acute on chronic rhinosinusitis during initial presentation with sinusitis symptoms after dental extraction.

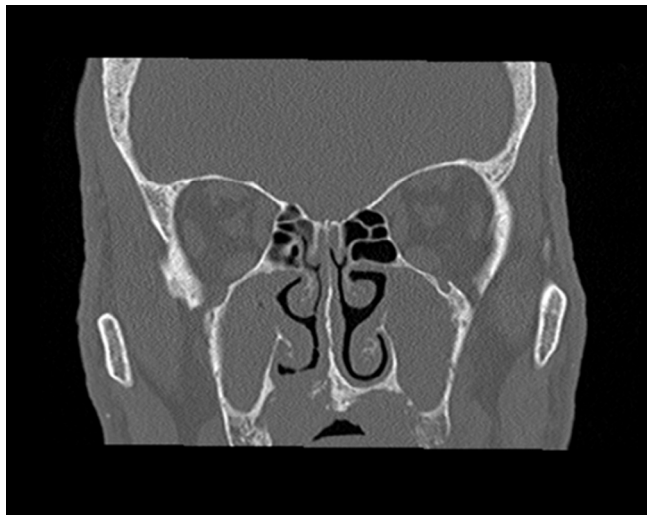

Fig. 2 – Computed tomography of coronal sinus in bone windows demonstrating interval progression with opacification of the maxillary sinus and erosion of the right hard palate 1 year after initial presentation.

sinus with complete opacification of the right maxillary sinus consistent with acute on chronic rhinosinusitis (Fig. 1). The patient was empirically prescribed intravenous (IV) levofloxacin, which soon improved his symptoms, and he was discharged after a few days of admission for observation with plans for follow-up a few weeks after discharge.

Unfortunately, the patient failed to follow up, and after 1 year, he presented to his local emergency department complaining of headache. At this time, computed tomography imaging demonstrated opacification of the maxillary sinus with erosion of the hard palate with disease extending into the upper alveolar ridge (Fig. 2). He was transferred to our tertiary care center, and given the extent of bony destruction, intraoperative biopsy was performed. Invasive *Actinomyces* osteomyelitis was diagnosed. With the guidance of the infectious disease service, IV penicillin was prescribed. He failed to improve despite ongoing antibiotic therapy, and functional endoscopic sinus surgery to open sinus outflow tracts and debride grossly involved tissue was performed. Subsequent cultures identified as coagulase-negative *Staphylococcus*, *Streptococcus viridans*, and *Propionibacterium* species, and anaerobic Gram-negative rods, ultimately speciated as *Klebsiella pneumoniae*, were sensitive to ceftriaxone. Based on sensitivities, the patient was placed on a regimen of IV ceftriaxone, IV vancomycin, and oral metronidazole with a plan to receive IV antibiotic therapy for several months.

The condition of the patient did not improve; in fact, it progressed over the following 7 months such that the bony erosion extended through the midfacial skeleton and frontal bones bilaterally (Fig. 3). During this time, he continued to smoke and found it difficult to attend follow-up appointments regularly because of social issues. It was thus difficult for him to adhere to his IV antibiotic regimen also, and oral antibiotic substitutions were made. The bony destruction ultimately involved his calvarium with epidural abscess and multiple draining fistulae from the paranasal sinuses to the skin along the nasal dorsum and glabella.

Given the extent of disease at this point, and difficulty of control with antibiotics alone, aggressive debridement was planned despite challenging social circumstances. A coronal incision with frontal craniotomy and free flap reconstruction

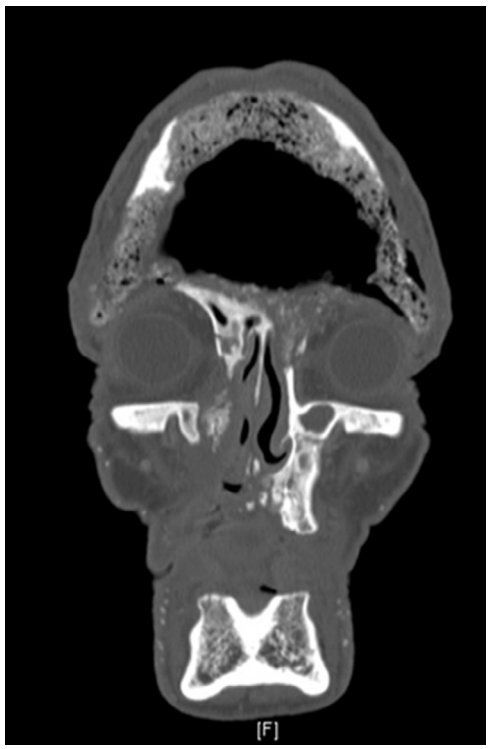

Fig. 3 – Computed tomography of coronal sinus in bone windows demonstrating disease progression with bony erosion extended through the midfacial skeleton and frontal bones bilaterally with intracranial involvement.

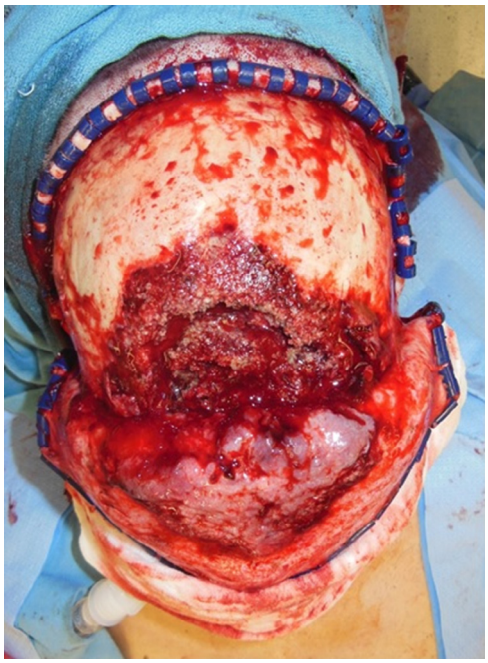

Fig. 4 – Intraoperative photograph with coronal flap reflected inferiorly (toward bottom of image) demonstrating frontal bony destruction and soft tissue involvement of *Actinomyces*.

was performed in a joint procedure between otolaryngology—head and neck surgery and neurosurgery. Extensive necrotic malodorous bone was found and debrided (Fig. 4). The dura was preserved, and Surgicel (Ethicon) and gelfoam were placed for hemostasis, and a latissimus dorsi free soft tissue transfer was performed to seal and support the dura (Fig. 5) but definitive reconstruction was deferred because of the extensive involvement of the craniofacial skeleton and anticipated residual-infected tissue.

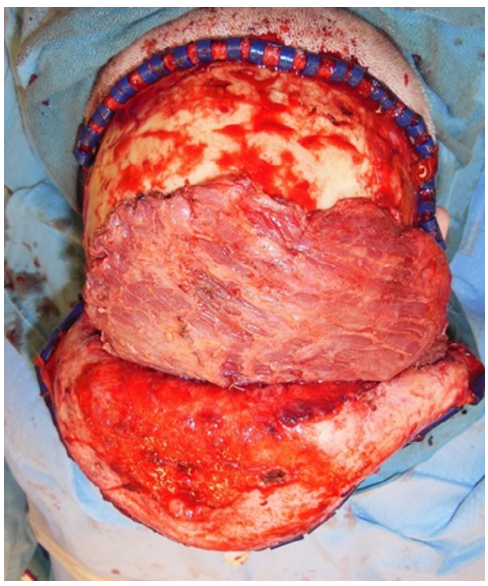

Fig. 5 – Intraoperative photograph after aggressive debridement and placement of latissimus dorsi free flap to fill dural and calvarial defect.

After this major debridement with reconstruction, the patient continued with IV meropenem. He subsequently require a minor debridement 7 months later, but otherwise continued to manage well with maintenance IV antibiotic therapy more than 2 years after his last procedure. He continued to receive routine surveillance in otolaryngology—head and neck surgery clinic with nasal endoscopy, and he is anticipated to require lifelong oral antibiotic therapy with amoxicillin-clavulanate.

Discussion

Cervicofacial actinomycosis is an uncommon condition caused by the pathological behavior of the otherwise commensal organism *Actinomyces* [1,4]. It some cases, cervicofacial actinomycosis can manifest as osteomyelitis and has been well described in the mandible, particularly in immunocompromised patients [5]. Less frequently, cervicofacial actinomycosis has been described in the maxilla, and from here it can extend to the skull base [6,7]. Cervicofacial actinomycosis has also been reported to be associated with epidural abscess, but this is exceedingly uncommon [7–9]. Typical management of cervicofacial actinomycosis consists of conservative debridement and aggressive intravenous antibiotic therapy lasting several months. In most instances, with this regimen, the disease can be well controlled, and even cured [3,5].

We report a case of aggressive *Actinomyces* osteomyelitis originating in the midface after a dental extraction. Unlike typical *Actinomyces* infections, *Actinomyces* osteomyelitis did not initially respond well to culture-directed intravenous antibiotic therapy. The reason for this is not entirely known, although the patient's ongoing smoking and challenging social situation precluding regular follow-up and adherence to IV regimen may have been risk factors for the aggressive behavior of the disease. The patient also appeared to have polymicrobial superinfection on return 1 year after initial presentation, and may ultimately have harbored resistant strains. This was certainly a potential risk, given his challenges with adherence to the prescribed regimen. He eventually required very aggressive debridement to enable adequate control of the disease. Long-term disease control continues to require a combination of IV antibiotic therapy along with multidisciplinary follow-up.

This case highlights the potential of untreated *Actinomyces* to behave aggressively and impact on function, form, and quality of life, particularly in situations where adherence to conventional medical and conservative surgical treatment protocols is compromised. Historically, the mainstay of treatment has been culture-directed IV antibiotic therapy, and surgical management has emphasized treatment of dental caries and apical abscesses, drainage of voluminous abscesses, and marsupialization of chronic sinus tracts [1]. However, removal of recalcitrant fibrotic lesions and/or debridement of necrotic bone have also been described [1]. This case highlights the occasional role of aggressive debridement of nonviable tissue and its ability to enable IV antibiotic therapy to be more effective in management of this potentially devastating disease. In this

case, the combination of modalities has enabled long-term maintenance of disease. We advocate for a multidisciplinary approach with involvement of infectious disease specialists for preoperative, perioperative, and long-term antibiotic therapy planning, ablative and reconstructive head and neck surgeons along with caring for this complex disease.

Conclusion

Although craniofacial *Actinomyces* osteomyelitis is uncommon, it can become debilitating and deforming, particularly if it extends to the skull base or intracranial fossa. This case demonstrates how the disease can progress if left unchecked, and highlights the benefit of a multidisciplinary approach to care of this potentially chronic condition.

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