Radiology Case Reports ■■ (2017) ■■–■■

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: http://Elsevier.com/locate/radcr

Case Report

Craniofacial *Actinomyces* osteomyelitis evolving from sinusitis

Joseph Y. Shen BSc[a], *Neal D. Futran MD, DMD*[b], *Maya G. Sardesai MD, MEd*[b,*]

[a] *Department of Dermatology, Stanford University, Stanford, CA, USA*
[b] *Department of Otolaryngology—Head & Neck Surgery, University of Washington, Seattle, WA, USA*

ARTICLE INFO

Article history:
Received 5 October 2017
Received in revised form 20 October 2017
Accepted 24 October 2017
Available online

Keywords:
Actinomyces
Osteomyelitis
Cervicofacial actinomycosis

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

Financial Disclosures: None.
Conflicts of Interest: None.
Poster presented at Triological Society Combined Sections Meeting, January 22-24, 2015, San Diego, CA, USA.
* *Corresponding author.*
E-mail address: sardesai@uw.edu (M.G. Sardesai).
https://doi.org/10.1016/j.radcr.2017.10.018

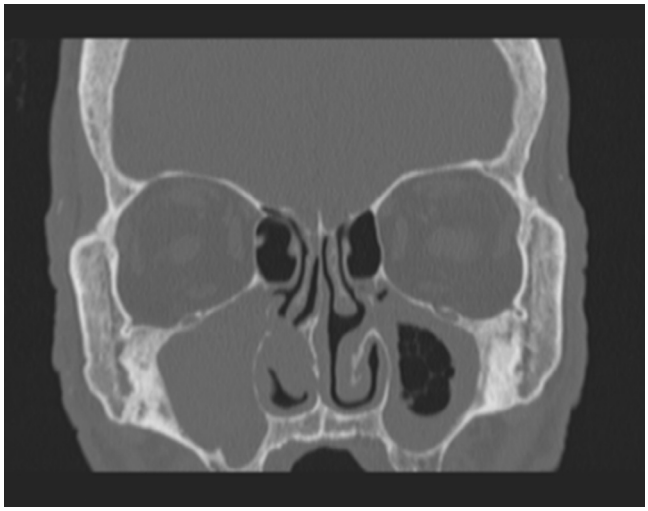

Fig. 1 – Computed tomography of coronal sinus in bone windows demonstrating mucosal thickening and frothy secretions in the left maxillary sinus with complete opacification of the right maxillary sinus consistent with acute on chronic rhinosinusitis during initial presentation with sinusitis symptoms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

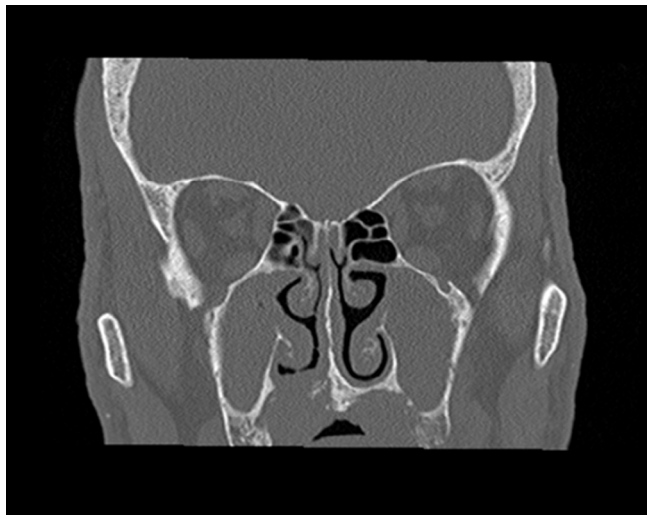

Fig. 2 – Computed tomography of coronal sinus in bone windows demonstrating interval progression with opacification of the maxillary sinus and erosion of the right hard palate 1 year after initial presentation.

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

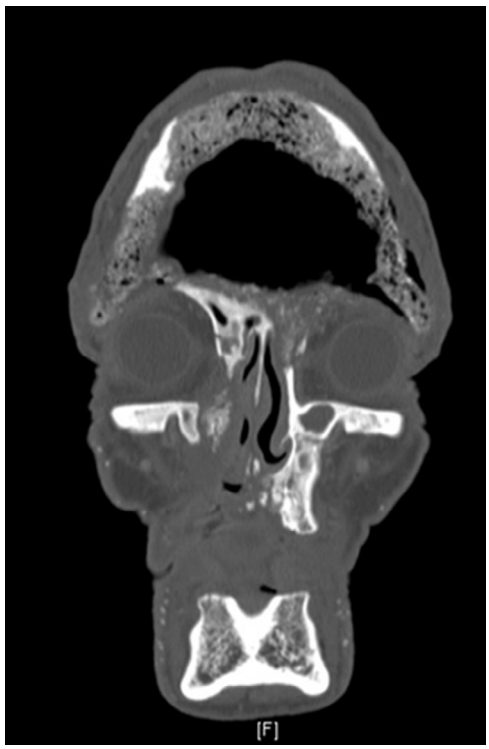

Fig. 3 – Computed tomography of coronal sinus in bone windows demonstrating disease progression with bony erosion extended through the midfacial skeleton and frontal bones bilaterally with intracranial involvement.