INTRODUCTION

Actinomycosis species are anaerobic filamentous gram-positive bacteria that are commensal organisms in the human mouth, digestive and genital tracts. However, these organisms can become pathological, particularly in patients with risk factors such as poor oral hygiene, history of mucosal breach or trauma, male sex, diabetes, immunosuppression, alcoholism, and malnutrition. Cervicofacial actinomycosis osteomyelitis describes the situation in which actinomycosis is implicated in osteomyelitis of the facial skeleton with deformity and abscess formation, most frequently affecting the mandible. Treatment often involves debridement and prolonged intravenous antibiotics.

CASE DESCRIPTION

A 56-year-old male with history of tobacco abuse and recent dental extraction initially presented with a four-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 CT imaging demonstrated findings consistent with acute chronic rhinosinusitis (see Figure 1). He was prescribed levofloxacin, which soon improved his symptoms, and he was discharged after a few days of observation with plans for follow up in a few weeks.

Unfortunately, he failed to follow up and ultimately presented to his local emergency department complaining of headache one year later. At this time, CT imaging was performed, and progression to erosive bony disease was identified (see Figure 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, intravenous (IV) penicillin was prescribed. He failed to improve despite ongoing antibiotic therapy and functional endoscopic sinus surgery, and subsequent cultures identified coagulase negative staphylococcus, streptococcus viridans, propionibacterium species, and anaerobic gram negative rods, and ultimately Klebsiella pneumoniae sensitive to ceftriaxone. Based on sensitivities, he was placed on a regimen of ceftriaxone, vancomycin and metronidazole with a plan to receive IV antibiotic therapy for several months.

His condition did not improve, however, and in fact progressed very slowly over the following seven months (see Figure 3). During this time, he continued to smoke and found it difficult to attend follow up appointments regularly because of social issues. The bony destruction ultimately involved his calvarium and involved epidural abscess and draining sinuses on his nasal dorsum and glabella.

At this point, given the extent of disease and difficulty of control with antibiotics, aggressive debridement was planned. A coronal incision with frontal craniotomy and free flap reconstruction was performed in a joint procedure between otolaryngology – head & neck surgery and neurosurgery. Extensive necrotic malodorous bone was found and debridged (see Figure 4). The dura was preserved and gelfoam were placed for hemostasis (Figure 5). Lattisimus dorsi free soft tissue transfer was performed to seal and support the dura (Figure 6). Cranioplasty was performed at a later stage.

Following this major debridement with reconstruction, the patient continued with IV meropenem. He has subsequently required two minor debridements, but has otherwise continued to manage well with maintenance IV antibiotic therapy.

DISCUSSION

Cervicofacial actinomycosis is an uncommon condition that is felt to be a consequence of pathological behavior of the otherwise commensal organism actinomyces. It sometimes can manifest as osteomyelitis, and has been well described in the mandible, particularly in immunocompromised patients. Less frequently, it has been described in the maxilla, and from here it can extend to the skull base. It has also been reported to be associated with epidermal abscess, but this is exceedingly uncommon. Typical management consists for 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.

We reported a case of aggressive actinomyces osteomyelitis originating in the midface, likely following a dental extraction. Unlike typical actinomyces infections, it did not initially respond well to culture-directed intravenous antibiotic therapy. The reason for this is not entirely known, though his ongoing smoking and challenging social situation precluding regular follow up may have been risk factors for aggressive disease behavior. He also appeared to have polymicrobial superinfection on return one year after initial presentation, and may ultimately have bacterial resistant strains. He ultimately required very aggressive debridement in order to enable adequate control of the disease.

This case highlights the potential for untreated actinomyces to have aggressive impact on function, form, and quality of life. In this way, this case demonstrates the potential natural history of the disease. The case also demonstrates the ability for aggressive debridement of nonviable tissue to enable IV antibiotic therapy to be more effective in this potentially devastating disease. Finally, this case emphasizes the benefit of a multidisciplinary approach to care for this complex disease.

CONCLUSIONS

Though cervicofacial actinomyces osteomyelitis 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 condition.

REFERENCES

1. Yoda F, Shimoda K, Tsuchiya C, Kusumoto H, Ichikawa S, Honda K, Fuchita A, Nose N. Efficacy of IV antibiotic therapy to be more effective in this potentially devastating disease. 2014; 710–711