Case Report

Actinomyces of the Hard Palate Presenting as Acute Sinusitis

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Abstract Actinomyces-related osteomyelitis of the face and jaw is an uncommon complication following invasive dental procedures or trauma, but can have an idiopathic etiology. This case involved a 71-year-old woman who presented to her primary physician for acute sinusitis over a two-week period. CT scan revealed sinusitis with associated bony destruction of the hard palate. She was subsequently referred over concern for invasive fungal sinusitis. She underwent biopsy and debridement yielding a diagnosis of Actinomyces-related osteomyelitis of the hard palate. Debridement and intense antimicrobial therapy for several months eventually resolved her infection. Though her outcome was favorable, patients with this disease can present after this invasive and aggressive bacteria has already caused irreversible damage.

Keywords Actinomyces; osteomyelitis; cervicofacial; hard palate; rhinosinusitis

1. Introduction

Actinomyces israelii is an endogenous organism that exclusively inhabits the oral and oropharyngeal cavity of humans. It is an anaerobic or microaerophilic, non-acid-fast, gram-positive organism. Infections of the head and neck by this organism, and to a lesser extent Actinomycosis naeslundii and Actinomycosis bovis, are relatively rare. When they do occur, they can aggressively invade the surrounding tissues, enter bone, and if untreated cause irreversible destruction of the central nervous system. It is paramount that the clinician maintains a high index of suspicion for infections from this atypical organism when encountering atypical presentations of acute or chronic rhinosinusitis or a necrotic oral lesion.

2. Case presentation

The patient was a 71-year-old African American female who presented to her primary care clinic with a complaint of acute rhinosinusitis of a two-week duration. Her presentation included mucopurulent yellow and green, foul smelling drainage from the left naris and low grade fever of 99.4°F. She complained of intermittent high-grade fevers and diaphoresis at home of unknown etiology. She denied at that time epistaxis, pain, facial swelling, nasal congestion, or any cranial nerve deficits. Her previous medical history was significant for chronic right hip, right leg and lumbar pain, hyperlipidemia, an unspecified endocarditis and hypertension. She denied a history of diabetes mellitus or immunosuppression but had been intermittently treated with steroid injections for her lumbar pain. Her vital signs at that visit were otherwise stable. Physical exam revealed a well-developed, well-nourished elderly female with normal facial symmetry and mucopurulent drainage from the left naris. Pertinent negatives included no lymphadenopathy, ear effusions, or obvious polyps or masses in her oropharynx. Upon dental inspection, caries, foci of calculi, and gingivitis were readily visible although there were no records of overt findings documented in her medical history.

Her primary physician ordered a CT of her sinuses and referred her to the otolaryngology clinic for further evaluation. CT results showed an apparent bony devascularization and necrosis with surrounding lucency involving the posteromedial hard palate on the left. Also there was an associated opacification of the adjacent left nasal cavity and possible erosion of the inferior and middle turbinates and lateral nasal wall, suspicious for an infectious process with neoplasm not excluded (Figures 1 and 2).

She presented to her first otolaryngology clinic appointment two weeks later with the same symptoms. At that time, she was afebrile and denied any additional symptoms; her white blood cell count was 6,200 cells/mm3. Flexible nasal endoscopy showed a significant purulence on the floor of the left nasal cavity extending posteriorly along the inferior and middle turbinates with friable tissue throughout (Figure 3); samples were collected and sent for cultures via traditional plating methods.

The following day she underwent a nasal endoscopy and hard palate biopsies with debridement in the operating room. Frozen sections of nasal polypoid changes revealed inflammatory polyps; no tumors or malignancies were present. A
left paramedian hard palate irregularity was biopsied multiple times and verified to be the same location as the lesions in the left nasal cavity (Figure 4).

The patient presented for follow-up two weeks later. The biopsy results revealed fragments of devitalized bone with intraosseous inflammation consistent with active chronic osteomyelitis and overgrown colonies of *Actinomyces*-like organisms, however there was no mention of sulfur granules present. Additionally, prior nasal culture returned a light growth of coagulase-negative *Staphylococcus*, likely related to a superimposed infection of the normally present nasal flora. Given pathology results and her clinical presentation, she was taken back to the operating room for further debridement of the hard palate. She was also admitted for infectious disease consultation and initiation of IV antibiotics. Infectious disease recommended 3 million units of penicillin G IV per day divided into 6 doses for 2 weeks, and then 500 milligrams of amoxicillin by mouth every 6 hours for 6 months. Given the need for extended IV antibiotics, the PICC team was consulted and placed a left upper extremity central venous catheter for at-home administration of antibiotics. She was discharged home on hospital day three with a follow-up appointment in the otolaryngology clinic in six weeks to assess the efficacy of therapy.
At her six-week follow-up appointment, the patient was completely asymptomatic. Nasal endoscopy at that visit revealed a small area of scab surrounded by white mucous on the nasal floor where the primary lesion had been, and was scheduled for a continued close follow-up.

At a follow-up appointment nearly six months later, she remained asymptomatic from her hard palate lesion. As of the publication date of this report, she has not needed any further intervention for this particular disease.

3. Discussion

The etiology of acute rhinosinusitis is most often viral in origin in adults, though differentiation from bacterial causes can be challenging [11]. Symptoms such as sudden onset of fever and accompanying purulent malodorous drainage as in this patient should prompt the investigating physician to consider frank infection of a bacterial or fungal nature. There are only a few other cases of osteomyelitis of the hard palate secondary to *Actinomyces* reported in the literature, and this is the only case presenting as acute sinusitis [4, 9]. Risk factors for *Actinomyces*-related infections include a history of immunosuppression, uncontrolled diabetes mellitus, long-term steroid use, malnutrition and poor dental hygiene [3]. This patient’s major risk factor was her persistent neglect of appropriate oral hygiene which likely caused a violation of mucosal integrity near the original lesion, allowing the organism to thrive and proliferate unopposed [1]. There have, however, been case reports in the pediatric population of disease with no known clear inciting event or etiology [8].

*Actinomyces* is a unique gram-positive filamentous branching bacteria that resides exclusively in the oropharynx of humans. It is known for gaining access to tissues via mucous membrane breakdown or trauma to the oral cavity via dental cleaning or surgery. Cervicofacial *Actinomyces* is a chronic infection characterized by inoculation of soft tissue, abscess formation, and local tissue destruction. Sinus fistula tracts will sometimes form with the exterior skin surface, indicating advanced invasion. This organism has no regard for fascial planes and aggressively invades deep tissue, bone, and if untreated, eventually neural tissue. The neck and face are the most common locations, comprising 50–60% of all documented infections [7].

Distinguishing *Actinomyces* from fungal infection is achieved with histologic exam as traditional fungi exhibit unfragmented hyphae and a traditional branching pattern whereas bacterial branching will easily fragment and appears irregular. This bacteria is quite fastidious and requires a special growth medium such as brain-heart infusion broth, however both require an average two-week incubation period [6]. One of the distinguishing characteristics of this organism is its production of sulfur-colored granules in vivo. They are composed of an internal mass of mycelial fragments surrounded by neutrophils and are visible to the naked eye. Caution is warranted, however, as granule formation is not exclusive to *Actinomyces* sp. (also included are nocardiosis, chromomycosis, eumycetoma and botryomycosis infections), and their absence does not rule-out an *Actinomyces* infection [5].

Unlike the majority of pathogenic micro-organisms today, *Actinomyces* is surprisingly susceptible to penicillin G, albeit given intravenously at high doses for an extended period of time. The specific duration of therapy is guided by a close clinical follow-up and the patient’s clinical response to treatment, and can extend to 12 months [10]. Additionally, with advanced disease (defined as bony invasion) as seen in this patient, surgical debridement to remove fibrotic, purulent, and necrotic tissue is necessary for definitive treatment [2]. For this extent of disease the prognosis is generally favorable with surgery and long-term antibiotic therapy. Long-term surveillance is mandatory as patients that were affected in the past now have a higher susceptibility for reinfection given the degradative bony erosion that has occurred.

4. Conclusion

We experienced a rare sinonasal soft tissue infection with *Actinomyces* with resultant osteomyelitis of the left posterior hard palate. *Actinomyces* is a benign commensal inhabitant of the mouth and oropharynx and can be problematic in patients who are immunosuppressed, with uncontrolled diabetic patients, or after oral trauma or surgery. The result of an untreated infection can be disastrous as the organism unchecked will aggressively invade to deeper structures including bone and brain. Patients can present with symptoms of a typical rhinosinusitis, so the primary physician should maintain a high index of suspicion to accurately and efficiently diagnose and treat this uncommon disease.

References


