Actinomycosis: Report of a Case with a Focus on its Uncommon Etiology of Chronic Sinusitis

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ABSTRACT

The palate is considered as a site of a variety of pathologies ranging from developmental disturbances, infections to malignancies. The most severe clinical appearance among these pathologies is a palatal perforation. A 42-year-old male patient reported to our institute with a complaint of regurgitation of fluids through his nose since last 4-5 months. His past medical history was significant as the patient revealed that he was diagnosed with lepromatous leprosy 5 years back. Computed tomography scan showed bony destruction on the left side of the palate and irregular thickening of bilateral maxillary sinuses suggestive of chronic sinusitis. As the microbial culture and other investigations were not significant to reach a final diagnosis, histopathological evaluation was carried out which was suggestive of actinomycosis histopathological examination revealed bacterial colonies composed of basophilic branched filaments surrounded peripherally by eosinophilic clubs with polymorphonuclear leukocytes. After surgical debridement of the lesion, the patient was treated with a regimen of oral ampicillin 500 mg 4 times a day. It was planned to continue the treatment for 6 months. Follow-up revealed a significant decrease in the size of the necrotic lesion. The patient advised palatal obturator for the closure of the defect.

Keywords: Actinomycosis, Lepromatous leprosy, Palate, Palatal perforation, Ray fungus, Sinusitis

INTRODUCTION

“Whatever are the benefits of fortune, they yet require a palate fit to relish and taste them.”

- Michel de Montaigne.

The word “ palate” is derived from Latin word “ palatum” or “ palat” which refers to the “ roof of the mouth.” The palate is a one complex structure of the oral cavity which forms tissue intervening between oral and nasal cavity.¹ Understanding the variety of pathologies of palate ranging from developmental disturbances, infections to malignancies requires an appreciation of its embryology, development of the different tissues native to the palate and their complexity.² The most severe clinical appearance among these pathologies is a palatal perforation.

There are various potential causes of palatal perforation. Failure of the palatal shelves to close during the 6th week of prenatal period results in cleft palate. Maternal alcohol consumption and cigarette smoking, folic acid deficiency, teratogenic drugs, certain viruses, corticosteroid use, and anticonvulsant therapy are some of the environmental factors known to cause cleft palate. Various infectious and granulomatous diseases, such as leprosy, tertiary syphilis, tuberculosis, rhinoscleroderma, naso-oral blastomycosis, leishmaniasis, actinomycosis,
histoplasmosis, coccidiomycosis, and diphtheria, are reported to perforate the palate. Palatal perforation is also seen in autoimmune diseases such as lupus erythematosus, sarcoidosis, Crohn’s disease, and Wegener granulomatosis. Minor salivary gland malignancy adenoid cystic carcinoma is reported to cause palatal perforation. Tumors can extend from maxillary sinus or nasal cavity and perforate the palate. Palatal perforation due to cocaine abuse is a well-known situation. Other drugs (heroin and narcotics) can be responsible for palatal perforation. Herein, we report an unusual case of palatal perforation due to bacterial infection—actinomycosis.

**CASE REPORT**

A 42-year-old male patient reported to our institute with a complaint of regurgitation of fluids through his nose since last 4-5 months. The patient was apparently alright 1 year back when he noticed a persistent small ulcer in the midline of the palate (Figure 1). After a couple of months, ulcer got deeper with associated dull pain and foul odored pus discharge. He complained of nasal congestion at that time and was prescribed medications by a private practitioner for the same 3 months back. He denied fever, chills, loss of appetite, weight loss, and other constitutional symptoms. He was diagnosed with lepromatous leprosy 5 years back, and lesions were present on forearm and palm of both hands. After taking antileprotic medications for 6 months, the lesions had resolved, and patient discontinued the medications. On general examination of body parts, no active lesions of leprosy were seen when the patient reported to us. The patient had a habit of tobacco chewing since last 15 years 3-4 times a day. Family history was inconclusive.

Intraorally, a tear shaped necrotic bony lesion of the midline palatal mucosa, exposing underlying bone with perforation was seen measuring approximately 2 cm × 1 cm in dimensions, grayish-yellow, extending anterioposteriorly from posterior part of palatal rugae to the first molar region. On inspection, no pus discharge was seen. It was tender on palpation (Figure 1). Paper blow test to check palatal perforation and nasal regurgitation showed positive results.

The occlusal radiograph revealed radiolucent area on the midline of palate more on the left side suggestive of palatal perforation. Computed tomography revealed destructive bony lesion with single communication between oral and nasal cavity. Bilateral irregular thickening of ethmoidal and maxillary sinuses which was suggestive of chronic sinusitis was noted (Figure 2). Blood investigations revealed no abnormality. A swab from the lesional area for microbial culture revealed no significant bacterial or fungal growth.

Histopathological examination revealed a granulomatous inflammatory lesion with large collections of polymorphonuclear leukocytes was seen. Bacterial colonies composed of basophilic branched filaments surrounded peripherally by eosinophilic clubs with surrounding polymorphonuclear leukocytic infiltration.
demonstrated bright magenta colored branched filaments which were PAS positive and confirmed the diagnosis actinomycosis of the hard palate. After surgical debridement of the lesion, the patient was treated with a regimen of oral ampicillin 500 mg 4 times a day. It was planned to continue the treatment for 6 months. Follow-up revealed a significant decrease in the size of the necrotic lesion. The patient was asked to follow stringent oral hygiene instructions. Palatal obturator was fabricated with heat cured acrylic plate with ball end clasps between canine and first premolar and C clasp on the 3rd molars of both the sides with palatal post dam (Figure 5).

**DISCUSSION**

Oral microbiome is a complex ecosystem where several species of microorganism have been identified. It harbors a diverse, abundant, and complex microbial community on varied surfaces of the oral cavity including both hard and soft oral tissues as biofilms. The normal commensals of healthy mouth include *Streptococcus, Actinomyces, Veillonella, Fusobacterium, Prevotella, Neisseria, Lactobacterium*, and *Capnocytophaga*. Most of these microorganisms exist in our oral cavity in a symbiotic capacity, maintaining relationships with the host that are based on mutual benefits (Los Alamos National Library, 2009). Not only do they not cause harm, but also the commensal populations may keep pathogenic species in check by not allowing them to adhere to mucosal surfaces. The bacteria do not become successful pathogens, causing infection and disease, until they breach the barrier of commensals (Jenkinson and Lamont, 2005).

Disturbing the homeostasis of the oral cavity can stir pathogen activity and lead to oral diseases. *Actinomyces israelii* is one of these endogenous organism that exclusively inhabits the oral and oropharyngeal cavity of humans. It is an anaerobic or microaerophilic, non-acid-fast, saprophytic, and gram positive organism. In humans, *Actinomyces* are often normally found in the oral cavity, the gastrointestinal tract, and the female genital tract. Disruption of normal mucosal barrier is necessary for *Actinomyces* species to penetrate subcutaneous tissue and cause an infection known as “Actinomycosis.”

Bollinger first reported the yellow granules in jaw masses of cattle in 1877. When this disease affects the jaws, it leads to a hard board like lesion that has a lumpy hence the earliest descriptions of actinomycosis as the “lumpy jaw” disease. In 1878, Wolf and Israel described the first human case. In 1879, Hartz first observed the microscopic appearance of granules of *Actinomyces* infection. In 1938, Cope classified actinomycosis infection into three distinct clinical forms as cervicofacial (50%), pulmothoracic (30%), and abdominopelvi (20%). *Actinomyces israelii* accounts for 52% of the infections, whereas *Actinomyces viscosus, Actinomyces odontolyticus, Actinomyces rachnia propionica*, and *Actinomyces myeri* contribute to 40%, 5%, 2%, 2%, and 1%, respectively.

Actinomycosis is a subacute to chronic bacterial infection characterized by contiguous spread, suppurative, and granulomatous inflammation, and formation of multiple abscesses and sinus tracts that may discharge “sulfur granules.” Granule formation is not exclusive to *Actinomyces* sp., and their absence does not rule-out an *Actinomyces* infection. It follows a pattern of remissions and exacerbations. When left untreated it involves the bone in 15% of the cases, with a gradual cortical erosions, which give way to localized lytic bone destruction. Invasion of the cranium or the blood stream may occur if the cervicofacial actinomycosis is left untreated.

Because of its rarity, there is a chance of missing its diagnosis and proper treatment leading to substantial morbidity and mortality.
The pathogenesis of actinomycosis is not completely clear. To become invasive, organism requires mucosal breakage to gain access to the submucosal tissue. Hematogenous spread is rare and lymphatic spread is rare due to the large size of the organism. Actinomyces being anaerobic it resides as a commensal in periodontal pockets and gingival crevices, in carious teeth, dental plaques, tonsillar crypts, or in the periodontium. Open wound, like dental extraction and trauma, facilitates the penetration.

In addition to Actinomyces microorganisms, almost all actinomycotic lesions contain so-called companion bacteria. The most important of these bacteria is Actinobacillus actinomyetemcomitans, followed by Peptostreptococcus, Prevotella, Fusobacterium, Bacteroides, Staphylococcus, and Streptococcus species, and Enterobacteriaceae, depending on the location of actinomycotic lesions. These companion bacteria appear to magnify the low pathogenic potential of actinomycetes. It has been described as polymicrobial nature of Actinomycosis. According to this concept, there are 5-10 companion bacteria which act as a co-pathogen that enhance the relatively low invasiveness of Actinomyces. Specifically, they may be responsible for the early manifestations of the infection and for treatment failures.

Once the infection is established, the host mounts an intense inflammatory response and fibrosis may develop subsequently. Infection typically spreads contiguously, frequently ignoring tissue planes and invading surrounding tissues or organs. Ultimately, the infection produces draining sinus tracts.

Laboratory studies generally present with anemia, mild leukocytosis, increased erythrocyte sedimentation rate (ESR), and C-reactive protein (CRP) levels. Actinomyces are very difficult to grow in culture with <30% diagnostic yield, which limits the usefulness of microbiological identification in such infections. Similarly, other laboratory findings such as anemia, leukocytosis, and an increase in ESR are non-specific and are not supportive to establishing the diagnosis. 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.

Unlike the majority of pathogenic microorganisms 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. In addition, 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. For this extent of disease, the prognosis is generally favorable with surgery and long-term antibiotic therapy.

In the differential diagnosis of this case, the leprotic ulcer was first thought due to patient’s history of lepromatous leprosy and the most commonly affected site in the oral cavity is hard palate. The second differential diagnosis was given as fungal infections such as mucormycosis. Due to tobacco habit history and typical location; malignancies, such as squamous cell carcinoma and mucoepidermoid carcinoma, were considered. Histopathology revealed the final diagnosis.

In our case, we could not demonstrate sulfur granules probably because the infection was just transitory between the acute and chronic forms and the mineralization might not have taken place. When the histological section proved it to be an actinomycotic lesion, literature, and clinical situation was reviewed, and a positive history chronic sinusitis was considered as the conclusive etiological factor.

CONCLUSION

In conclusion, although cervicofacial actinomycosis is known to be the most common type, localized intraoral lesions on palate occurs rarely. An initial clinical examination and past medical history of lepromatous leprosy first led us to the differential diagnosis of a leprotic ulcer on the palate. As the microbial culture and other investigations were not significant to reach a final diagnosis, histopathological evaluation was carried out which was suggestive of actinomycosis. The result of an untreated infection can be disastrous as the organism unchecked will aggressively invade to deeper structures. Patients can present with symptoms of a typical sinusitis, so the primary physician should maintain a high index of suspicion to accurately and efficiently diagnose and treat this uncommon disease. It is important to consider chronic sinusitis as a potential etiological factor in this case.

REFERENCES

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