

Intraoral Actinomycosis-A Systematic Review of Case Reports

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ABSTRACT

Introduction: Cervicofacial actinomycosis is a more frequently occurring disease compared to its abdominal or thoracic variants. Actinomyces persists as normal commensals of the oral cavity and gastrointestinal tract that can become pathogenic. Several factors like trauma, chronic hyperglycemia or other systemic diseases promote structural and functional disturbances in oral tissues. They are associated with delays in wound healing or cause changes in the immune mechanisms of the oral mucosa thus opening the portal of entry for these microbes.

Materials and Methods: This review incorporates Oral dysbiosis, clinical features, and differential diagnosis of intra-oral actinomycosis based on a few case reports. Case reports of intra-oral actinomycosis affecting the soft tissues published between 2000 and April 2020 were searched. Only applicable 16 cases with oral actinomycosis occurring in the soft tissues were included in the final analysis. Actinomycosis infections are hard to diagnose, since the symptoms remain unknown to dentists because of their varied presentations.

Conclusion: Dysbiosis due to various etiological factors can directly affect the oral mucosa through altered immune functions. The correlation of clinical presentation with other details is paramount to avoid morbidity due to extensive tissue involvement.

Keywords: Actinomyces species, Oral actinomycosis, Tongue, Palate, Gingiva

INTRODUCTION

Actinomycosis is a chronic type of an infectious disease, first described in 1896 by Kruse. The most recognised are cervicofacial clinical forms incidence ranging from 40 to 60% compared to thoraco-pulmonary and abdomino-pelvic. The causative obligate anaerobe actinomyces species reside on mucosa surfaces in great numbers as one of the oral cavity's normal commensals. When they gain, access to deeper tissues it can infiltrate the underlying structures. Actinomyces species commonly pathogenic to humans are *A. odontolyticus*, *A. israelii*, and *A. gerencseriae*. Among them, *A. israelii* and *gerencseriae* cause 70% of the infections in the cervicofacial region.¹⁻³ Intraoral actinomycotic infections can be present in the bone and soft tissue. Debilitating diseases, trauma to the oral cavity or poor oral hygiene can be a few causes of the pathogenesis. Infection can spread into the surrounding tissues with the sinus opening into the outer surface. It usually presents with a slow-growing indurated lesion; as the lesion progresses, it can be suppurative and mimic other granulomatous lesions with multiple abscesses, fistulas, and sinuses. The immunological reactions bring macrophages to the target area; multinucleated giant cells result from this macrophage fusion. Diagnosis of intraoral actinomycosis is challenging for dentists, as it is a fastidious anaerobe and presents as different diseases, including malignancy exhibiting varied symptoms. The disease can closely resemble other granulomatous diseases, suppurative infections by

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other organisms, and neoplasms. Usually the diagnosis is done by histopathological identification of sulphur granules from the exudate or by culture.^{4,5} It is essential to identify the actinomycotic infections inside the oral cavity as the cervicofacial actinomycosis is most common. This article focuses on the review of oral actinomycotic case reports in identifying and comparing the clinical manifestations, prevalence and their differential diagnosis.

MATERIALS AND METHODS

Case reports of oral actinomycosis in soft tissues of the oral cavity published between 2000 and April 2020 were searched. It was done by carrying out a structured search

through the Scopus database search and manually by google scholar. The keywords used were Cervicofacial, intra-oral, oral actinomycosis for the search. Only those applied to the soft tissues of the oral cavity were chosen.

RESULTS

Through a survey of the Scopus database search, 153 case reports were identified, only applicable 16 cases with oral actinomycosis occurring in the soft tissues were included. Cases of actinomycotic osteomyelitis were excluded.

DISCUSSION

The oral microbiota consists of many normal microbes residing in healthy tissue, and the actinomyces is also one of them. Most patients have a history of local trauma resulting in mucosal breakdown; others usually have poor oral hygiene, trauma, extraction or systemic diseases. In the reviewed case reports among tongue actinomycosis, only two reports had a history of trauma, one with poor oral hygiene; the rest of

the five cases did not have any history of trauma (Table 1). In gingiva and palate cases, four had systemic diseases, two had undergone extraction and scaling, and another two cases with poor hygiene and periodontitis (Table 2). Incidences of actinomycotic cases are greater in the fifth decade of life and are more common among men in a ratio of 3:1-4:1. Among our case reports, there were six male and ten female patients (Table 1&2). Actinomycotic infections can affect both females and males equally, as our study shows more female patients in soft tissue actinomycosis. The age group gender predilection in previous reports may be based on environmental factors. Some literature reviews show that the incidence of actinomycosis is greater among men in rural areas. Our review's age group shows patients 50 years and above among most tongue actinomycosis cases. A variation in the age group of those with actinomycosis in the palate was observed at third decade and above, when compared with those in the gingiva where the patients belonged to an older age group. Actinomyces can present with varied signs and symptoms depending on their anatomic location. In our review of case reports, two of the cases show a history of trauma along with poor oral hygiene or periodontitis. Actinomyces usually exist with other microorganisms making isolation by culture methods complex since they are associated with other microbes like streptococci or other Gram-negative bacilli. The presentation of abscess formation with or without multiple drainage sinuses is one characteristic feature that helps diagnose actinomycosis.⁶⁻⁸

Most of the time, during diagnosis, the actinomycotic infection is chronic. It can present in different ways atypical with subacute clinical manifestations. In some cases, the extensive

Table 1: Some of the reported cases of tongue actinomycosis from 2000-2023

Author/Country	Age/sex	Clinical findings
Habibi et al. ⁹ Iran	54 F	Elevated, nontender mass on the right border of the tongue. History of Tongue bite.
D'Amore et al. ¹⁰ San Paolo	52M	Swelling on the anterior part of the tongue. History of benign squamous papilloma 5years before in the same location
Kurtaran et al. ¹¹ Turkey	54F	A solid, painful deep mass in the left side of with abscess. History of dental trauma
Won J et al. ¹² New Zealand	72 M	Fluctuant, tender mass on the right side of the tongue. No history of trauma to the affected area.
Enoz M et al. ¹³ Turkey	39 F	Lesion in the anterior portion of the tongue. Oral hygiene status is not good.
Karla et al. ¹⁴ Brazil	44F	Purplish vesiculobullous lesion on the tongue. History of trauma on the tongue.
Al-Rawee et al. ¹⁵ Iraq	65 F	Mass inside the tongue. No history of trauma.
Sadeghi et al. ¹⁶ Canada	66M	Generalized enlargement of the tongue . No history of trauma.
Ahmed et a. ¹⁷ UK	60F	A firm and tender mass ofthe tongue. No history of trauma.

Table 2: Some of the reported cases ofpalate and gingival actinomycosis from 2000-2023

Author / Country	Age/ Gender	Clinical findings
De et al. ¹⁸ India	32M	Ulceration on the palate with slough History of scaling
Andrade et al. ¹⁹ Brazil	46F	Ulcerative lesion on the hard palate with slough History of diabetes mellitus
Garg et al. ²⁰ USA	49M	Exposed necrotic palatal bone History of extraction/Renal disease
Yadegarynia et al. ²¹ Iran	48F	Indurated mass on the hard palate Poor dental condition
Sakallioğlu et al. ²² Turkey	60F	Ulcerative, desquamative gingiva History of periodontitis
Bruno et al. ²³ Brazil	57 F	A necrotizing lesion in the gingiva History of Lymphoma
Acar et al. ²⁴ Turkey	63M	Erythematous desquamative gingiva Presence of systemic disease



soft tissue infiltrative changes in the underlying tissue from the surface are characteristic. Actinomycotic osteomyelitis of the bone thus can be differentiated with extending sinuses opening through the bone. Our review consists of actinomycosis cases involving the soft tissues only, but few of these case reports show the secondary involvement of the bone. Samuel and Martin classified actinomycotic infections with three different clinical manifestations one with acute swellings that are painful with short duration of one month or less, chronic infections present more than three months and lastly actinomycotic infections that are confirmed after microbiological tests.⁹ Any non-specific infections inside the oral cavity are important as they can prove fatal if not diagnosed. Intraoral Actinomycosis occurs as an asymptomatic slow growing firm mass as the reports shows, but in a longer duration the patient may complaint of pain and abscess formation. The indurated mass can give a clue to actinomycosis as one of the clinical differential diagnosis.^{7,8}

Only 10% of the actinomycotic lesions are diagnosed in the first visit. If undetected, patient might have an extensive destruction of tissue may need a surgery. Though histopathology is proven best for the detection of actinomycotic organism these species can go unnoticed at times. The diagnosis in histopathology is done by identifying the morphology of the microorganism with H and E, PAS or grams stain. FNAC sometimes can be a useful tool. Actinomycosis exists in association with other microbes, they have a typical colony forming growth in the form of human molar on agar and grows as clumps in reddish forming colonies. There is an improvement in polymerase chain reaction and other molecular tests for faster results. The penicillin as drug of choice is effective if allergic than other drugs like clindamycin is preferred along with metronidazole.^{1,7,8}

Actinomycosis of tongue

The common site of occurrence in the soft tissue in the oral cavity is buccal mucosa, other sites include palate, tongue, masseter muscle, salivary glands, maxillary and mandibular bone involvement. Actinomycosis occurring on the tongue was first described by Von Hacker in the year 1885. Intraoral actinomycosis is challenging for dentists as it is a rare disease, especially tongue actinomycosis. The lesions of the soft tissue can progress to involve the bone, causing bony destructions. Clinically it can be manifested as a fibrotic reaction with woody consistency. The common site is anterior two-thirds, our review shows all the cases on the anterior two thirds or the lateral border most of them presenting as a mass.⁽⁹⁻¹³⁾ It can cause speaking difficulties because of the restriction of tongue movements with or without pain. The differential diagnosis for tongue swelling includes hemangioma, lymphangioma, amyloidosis, granulomatous diseases and neoplasms like granular cell tumours, neuroma, neurilemoma, lymphangioma or metastatic tumours. The overall rate of tongue actinomycosis is few, making up only 3% of all reported intraoral site occurrences. Actinomycosis in the tongue is uncommon because of its keratinised mucosal lining, rich vascularity, mobility and self-cleansing properties by the saliva. Most of the lesions manifest

as a swelling in the tongue; some show abscess formation. Multiple abscess formations, swelling, abnormal hardening of the soft tissue. It produces sizeable fibrotic swelling with central necrosis. They are generally non-specific, resulting in misdiagnosing the lesion.¹⁴⁻¹⁷

Actinomycosis of palate and gingiva:

The Actinomycotic cases of gingiva and palate are sporadic. They usually present with an environment conducive to actinomyces' growth. Just as in any other soft tissue region here, the polymicrobial growth leads to an anaerobic atmosphere, thus creating a pathway for the further progress of the actinomyces species. In diabetic patients, due to high glucose in the wound area, there is high bacterial growth, so healing is slow. Our review shows a high chance of contracting palate and gingival actinomycotic infection if the patient has systemic or immunocompromised conditions.⁽¹⁴⁻¹⁶⁾ Studies show that actinomyces from periodontitis or plaque can get absorbed to cause pulmonary actinomycosis. Actinomycosis can present as an acute or chronic lesion, sometimes as desquamation of the gingiva or necrotising ulceration, which can be infiltrative with underlying tissue destruction, often showing the yellow purulent slough. The other lesions on the palate, like necrotising sialometaplasia, mucormycosis, salivary gland lesions or HIV, are possible. It is common for the underlying bone to get involved since the palate has minimal soft tissue. Most gingival actinomycosis is due to periodontal infections, which have disrupted the integrity of the gingiva and supporting tissues leading to infection by the actinomyces species to cause the infection.¹⁸⁻²⁴

Oral mucosa and pathogens

The normal commensals of oral flora consist of a wide habitat of microorganisms. The intact healthy oral mucosa acts as a fort to guard against the invasion by these organisms to deeper tissues. Certain factors can break the barrier like trauma or any systemic diseases, autoimmune disorders, dental caries and periodontitis. In the presence of disease, there can be an increased colonization of these normal microflora. In normal circumstances the breach in the oral epithelium with eventual replacement with the new cells, the microbes are removed. So there are fewer chances of penetration by some of these opportunistic microorganisms.²⁵ But if the condition exists for a long time the immune system may not be able to fight thus affecting the oral mucosa. The salivary flow rate and food consumption habits of the patient can influence the bioenvironment thus the growth of the microbes, increasing the colonizing properties depending on the availability of oxygen, pH of saliva and nutrients. So the nature of the actinomyces species to reside and colonise in the oral mucosa along with other microbes becomes easier. Diagnosis of this disease is challenging because of difficulties in the culture of the organisms since only predominant bacteria can be cultivated and can miss the main pathogen in the process. So dysbiosis to various etiological factors can directly affect the maturation of oral mucosa through altered immune functions.^{26,27}



CONCLUSION

Actinomycosis of the oral cavity presenting as a non-specific infection can be alarming to dentists. If the infection is not diagnosed at the initial stage, it can cause extensive tissue damage. Trauma and other factors that affect oral environment are the possibilities for entry of actinomycetes. This may exist in the oral cavity with periodontal pathologies or extraction incidences and also in cases of immune suppressions. It is challenging for a dentist to catch the culprit at the right time, thus avoiding further surgery.

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