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Challenge dilemma of actinomycosis in the tongue: Review and case report

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ABSTRACT

INTRODUCTION: Rare presentation of tongue actinomycosis makes it challenge, less than 3% of all recorded cases of actinomycosis can be proved in the English literature. In this study we clarify the unexpected presence of actinomycosis wishing that it assist our practitioner colleagues in diagnosing such lesions perfectly.
CASE PRESENTATION: Sixty five years old female was complaining from mass in the tongue submucosally since 1 year last with no history of fever, pain and trauma. It's situated in the anterior third of the tongue, mild discomfort with percussion. Benign neoplasm was the clinical diagnosis. Decision was excisional biopsy. So patient underwent surgery under local anesthesia. Complete excision was done. Specimen sends for histopathological examination. Good healing process. Patient advice to keep on tetracycline capsule 250 mg for 1 month precautionary. Histopathological examination of the biopsy specimen diagnosis was mycetoma, with no malignancy.
DISCUSSION: Clinically actinomycosis can be challenge too when it be silent not manifest as infection more over similarity to other pathological conditions at that time detailed patient history will be invaluable to exclude neoplastic pathology. Surgeons should pay attention to that prolonged antibiotic therapy is usually required is mandatory in some cases, and excellent prognosis confirmed. With better oral hygiene, availability of antibiotics, and advanced surgical techniques, the outcome, and mortality have improved.
CONCLUSION: Varied clinical manifestations of the disease are make achievement of clinical diagnosis of actinomycosis is difficult. Treatment should implement in harmony according to each diseases solely.

1. Introduction

Actinomycosis considered as a rare bacterial infectious disease produced by Actinomyces species. *Actinomyces israelii* or A. gerencseriae is considered the origin of about 70% of infections [1]. The main distinctive feature of chronic infection by genus *Actinomyces* primarily is suppuration and soft tissues cellular invasion [1].

Actinomyces are mutual bacteria concerted in oral, pharyngeal and gastrointestinal tissues forming settlement colonizes. Trauma or previous infection can be pathway for activate infections in the soft or bony tissues by these colonies [2–4]. Cervicofacial region for actinomycotic infections are an important entity to the maxillofacial and otolaryngologist. One of vital symptom in clinical presentation of actinomycosis is unexplained swelling, importance of this symptom that might mirror whether benign and malignant neoplasms, specifically in submucosal area. Rare presentation of tongue actinomycosis makes it challenge, less than 3% of all recorded cases of actinomycosis can be proved in the English literature, for this rarity as well as to promote precise diagnosis; high suspicions is crucial in order to provide appropriate management [5]. Tongue has a unique anatomical and histophysiological features make it resistant to infection. Presence of great mobility and mechanical cleaning of saliva as well presence of keratinized mucosal lining rich with vascular parenchyma make it resistant to bacterial adherence and attack. All this create presence of missed or undiagnosed actinomycosis may end with sever tissue damage. In this study we clarify the unexpected presence of actinomycosis as a submucosal mass in the tongue wishing that it assist our practitioner colleagues in diagnosing such lesions perfectly.

1.1. SCARE compliance

The work has been reported in line with the SCARE criteria [6].

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Fig. 1. Surgical Dissection of the mass.

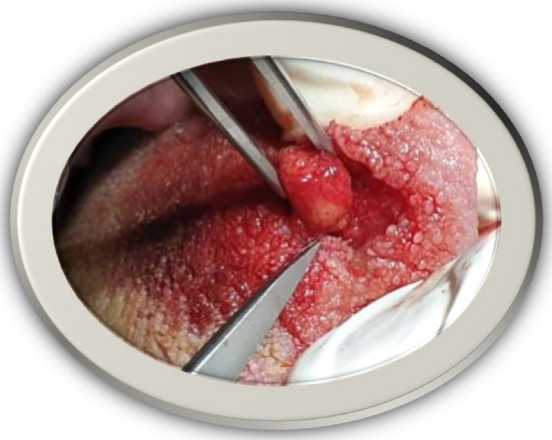
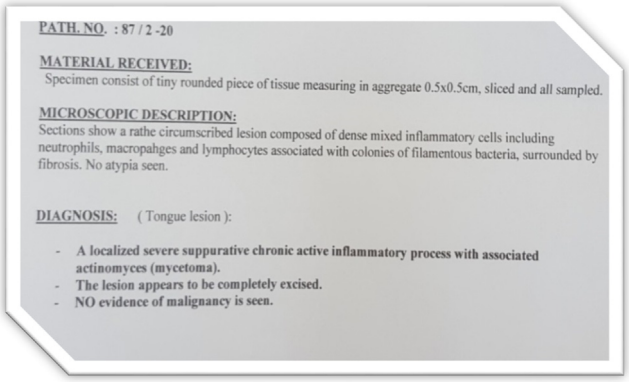


Fig. 2. Complete excision of the mass.

2. Case presentation

Sixty five years old female came to the maxillofacial department in our institution. Mass inside the tongue was the exact descriptive worried wards of the patient. Starting with history taken patient complain from boil like small mass since 1 year last with no history of fever, pain, trauma, sense alteration or disturbance of mobility. Patient is healthy with no any previous medical disease. Intraoral examination of the tongue, inspection show normal mucosal lining with nil abnormality as change color or shape and even size either dorsal or ventral surfaces. With palpation of the anterior third of the tongue we can feel a boil like mass submucosally about 1 cm in diameter slightly hard, mild discomfort with percussion, movable and well circumscribed. No lymph node enlargement shown. Benign neoplasm considered as clinical diagnosis, keeping in mind a differential diagnosis including trauma, infection or malignancy. Decision was excisional biopsy. Full discussion with the patient concerning details of surgery takes place, followed by ethical agreement signed by the patient and their relatives (informed consent). So patient underwent surgery under local anesthesia, infiltration technique (2 cartridges of 1.8 ml lidocaine 2%, adrenaline 1/80.000). Straight line incision over the mass prepared with use of scalpel 15 done followed by dissection to separate the mass precisely (Fig. 1). Complete excision was done (Fig. 2), suturing with 3/0 black silk suture. Specimen sends for histopathological examination. We don't use culture and sensitivity test. Postoperative instruction well explained to the patient relative with follow-up dating. Antibiotics and analgesic scribed as augmentin tab (625 mg) twice time daily, for pain, we advise patient to use paracetamol tab 500 mg on need. Patient was recall after seven days for suture removal and checkup. Good healing process of the dorsal surface of the tongue shown. Patients also seen for the third time after 14 days, re-examine again with the histopathological report. Patient is well no complaining, with normal architecture of the tongue preset. Patient advice to keep on tetracycline capsule 250 mg for 1 month precautionary.



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MATERIAL RECEIVED:
Specimen consist of tiny rounded piece of tissue measuring in aggregate 0.5x0.5cm, sliced and all sampled.

MICROSCOPIC DESCRIPTION:
Sections show a rathe circumscribed lesion composed of dense mixed inflammatory cells including neutrophils, macropahges and lymphocytes associated with colonies of filamentous bacteria, surrounded by fibrosis. No atypia seen.

DIAGNOSIS: (Tongue lesion):

- **A localized severe suppurative chronic active inflammatory process with associated actinomyces (mycetoma).**
- **The lesion appears to be completely excised.**
- **NO evidence of malignancy is seen.**

Fig. 4. Histopathological Report of the mass.

3. Histopathological report notes

Histopathological examination of the biopsy specimen, truly, show a circumscribed lesion composed of dense mixed inflammatory cells including neutrophil, macrophage and lymphocyte associated with collection of filaments bacteria surrounded by fibrosis. Diagnosis was a localized sever suppurative chronic active inflammatory process associated with actinomycosis (mycetoma) completely excised, with no malignancy (Fig. 3a, b, c). Histopathological Report is shown in Fig. 4.

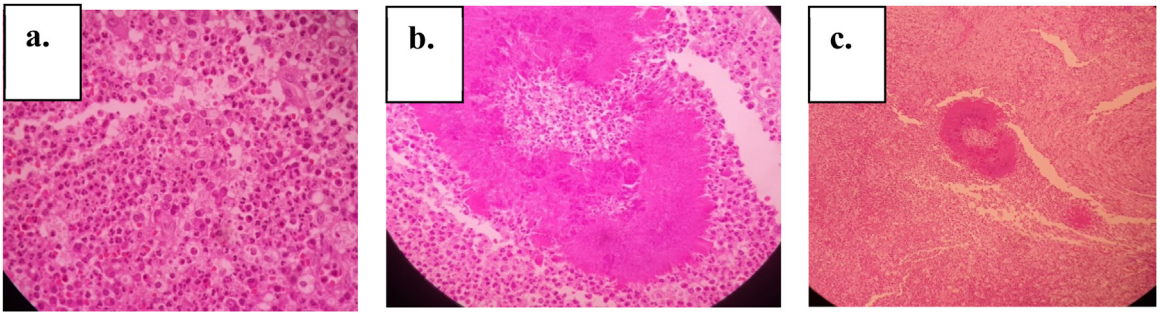

Fig. 3. Microscopical IMG-20200211-. a. WA0012 b. WA0013 c. WA0014.

Table 1
Brief description of different reported cases of tongue actinomycosis.

Author[s] year [ref]	*Patients Age [years]*	*Sex*	*Clinical findings*	*Treatment*
Sodagar and Kohort 1972 [35]	47	Female	Firm palpable mass right lateral border	Excision
Uhler and Dolan 1972 [36]	43	Male	Painful nodular mass right lateral border	Excision Penicillin for 6 months
Kuepper and Harrigan 1979 [5]	35	Male	Round firm tender mass left middle third	Penicillin for 1 month
Brignall and Gilhooly 1989 [14]	55	Male	Non ulcerated, indurated swelling anterolateral and ventral tongue surface	Penicillin for 2 months
Islaska et al. 1991 [37]	64	Female	Painless swelling left dorsal surface	Penicillin for 6 months
Morris et al. 1992 [38]	50	Male	Nodular lesion anterior-dorsal surface	Penicillin
Ficarra et al. 1993 [34]	57	Female	Non ulcerated indurated mass right lateral aspect	Penicillin for 2 weeks
Gerbino, 1996 [6]	44	Female	Nodules	Penicillin for 1 month
Vazquez, 1997 [31]	31	Male	Non painful submucosal Nodule	Excision and Amoxil for 3 weeks
Alamillos-Granados et al. 2000 [39]	74	Female	Painless indurated ulcer involving ant floor of mouth, alveolar process and labial mucosa	Minocycline for 10 weeks [Allergic to Penicillin]
Atespare et al. 2006 [12]	52	Female	Solid painless mass left anterior tongue Excision	Penicillin for 3 weeks
Enoz M 2006 [7]	] 39	Female	Nodular lesion anterior dorsal tongue	Tetracycline for 6 weeks [Allergic to Penicillin]
Habibi et al. 2008 [40]	54	Female	Non ulcerated, non-tender mass right lateral border.	Excision Penicillin for 3 weeks
Kurtaran et al. 2011 [41]	54	Female	Solid painful mass left tongue	Excision + Amoxicillin –Clavulanic acid + Metronidazole for 5 weeks
Escoda, 2013 [42]	49	Male	Two ulcerated lesions Pain asymptomatic	Clindamycin for 1 month
Rocha et al. 2017 [43]	44	Female	Necrotic tissue with purulent right lateral tongue developed after sclerosing agent injection to treat vascular lesion	Local debridement
Jat et al. 2017 [44]	44	Female	Non ulcerated nodular mass dorsal surface	Excision. Doxycycline for 3 weeks [Allergic to Penicillin]
Aneja et al. 2017 [45]	14	Male	nodular mass present on right lateral border	Excision Amoxicillin–Clavulanic acid for 2 weeks
Sadeghi, 2018 [46]	66	male	Macroglossia. Dysphagia and speech impairment	Penicillin for 22 weeks
Bekerecioglu, 2018 [47]	45	Female	uniformly bounded, swollen, painless mass without ulceration a	Excision and penicillin
Siddiq, 2019 [22]	60	Female	gradual onset of painful swelling firm and tender mass	4 week course of oral amoxicillin
Fiorella D'Amore, 2020 [26]	52	Male	solid, fixed, bilobed submucosal nodule	Excision and oral intake of clarithromycin for three week

4. Discussion

Actinomycosis utmost frequently involves the region of head and neck, and it's rarely attack oral cavity. It's one of natural oral microbiota, it occasionally becomes pathogenic. When it became pathogenic it affects essentially soft tissue as well hard tissue too in situations caused by tissue injury and poor oral hygiene. In this case presentation no history of trauma as patient claim also she is healthy, no medically compromised disease that can make her susceptible to infection, no history of trauma add to that she has good oral hygiene. In such circumstances, we think that diagnosis is challenge for this vague silent presentation with no any sign for inflammation or infection just small nidus of bacteria surrounded by fibrous tissues completely hide submucosally. Histopathological examination is highly important as it's the only way present to differentiate simple from worst things. No possibilities for culture and sensitivity test or fine needle aspirate biopsy as its small hidden mass. Even Doppler ultrasound is impractical more over we thought it a small benign lesion from the start but unlikely actinomycosis. In this case report we will review different articles focus on tongue actinomycosis that sharing challenge of actinomycosis dilemma. In Table 1, viewed clinical presentation and treatments available in different articles discuss case reports or case series are listed [5,7].

Actinomycosis defined as a granulomatous anaerobic infectious disease that spreads into adjacent soft tissue without regard for tissue planes or lymphatic drainage; it may also be associated with a draining sinus tract. The term actinomycosis is deceptive some suppose that actinomycosis is a fungal infection, related to the Greek

idiom mykes, inspit of it is not a fungal infection. Ray fungus is a term used to describe the ramified organism found sulfur granule, this term firstly used by Aktino [8]. Richtsmeier in his published article stated that, Humans actinomycosis firstly described in 1857 in medical articles after detection of the granules in human autopsy material by Israel in 1878 [9].

Demographically actinomycosis infections are more common in men (males > females) ratio (3:1). No specific age groups can be attacked by this type of microorganism, young to middle-aged adults (age 20–50 years) slightly forming the utmost age group in reported case reports [10]. Different published article agreed on rarity more over that actinomycosis can give vague different clinical features this is up to its presence in various anatomical sites. Cervical (50%), thoracic (17%) and abdominal (23%) are the most usual sites show clinical manifestations of actinomycosis infiltration specially as fluctuant mass most common is the cervico-facial [11–13]; beside more than 30 species of *Actinomyces* have been described. Sandeep Sharma [14] in his published article estimates that sinus tract and sulfur granules are the distinguished features of chronicity. Highest percentage of actinomycosis isolated clinically in human infections from different sites is *Actinomyces israelii,* mainly considered the predominant species [15]. A. israelii is responsible for about 70% of orocervicofacial infections [16,17]. Ln 1989, Brignall and Gilhooly published a 20 year review of the English literature, which found seven cases and the whole number not more than 15 cases [15]. Articles also confirmed that the unique histophysiologic features of the tongue as keratinized mucosal lining, rich vascularization, excessive mobility and saliva role as mechanical cleansing, for these reason bacteria can not adhere and multiply forming abscess [13,18,19]. T̈rauma playing a catalytic role in initiating a pathway of entry in the hostT̈hese are wards description of Chiang CW [20] in his article insure that actinomycosis it often coincides with dental manipulation or maxillofacial trauma. This approved with Schmidt P, Syed MA [21,22] too. Brad 2019 [10] illustrates that tooth extraction or soft tissue injury, infection includes odontogenic or periodontal, poor oral hygiene, diabetes mellitus, immunosuppression, neoplasm or irradiation, in addition malnutrition can be predisposing factors for actinomycosis in cervicofacial region. Rare presentations of tongue actinomycosis with local trauma as a main cause of infection are confirmed by Siddiq Ahmed [23]. The case presented here claim that no history of trauma and she keep good oral hygiene.

Clinically actinomycosis can be challenge too when it be silent not manifest as infection more over similarity to other pathological conditions at that time detailed patient history will be invaluable to exclude neoplastic pathology. Sezer [24] in his article cited that pain, swelling and induration of the dorsum or lateral tongue are the frequent symptoms complained by the patients [25]. That adds another interesting point to our case presentation. Lesions are diagnosed by a combination of histopathology, microbiological culture and radiological investigations. Time for actinomycosis diagnosis is essential, spread of infection avoidance and early management with antibiotics can prevent unwanted complications [18]. Thomas [26] highlight that diagnosis is based on Gram stain culture, and sensitivity testing. Sulfur granules are aggregates of filamentous forms of the organisms. Its classic presence is one of diagnostic feature of actinomyces. On the contrary, Brad W [10] advocate that cultures can aid in diagnosis but may be complicated by strict anaerobic incubation requirements over an extended period, overgrowth of other associated bacteria, and prior antimicrobial therapy.

Fiorella [27] considers histological examination as the gold standard for the final diagnosis. Diseases as granulomatous lesions (gumma and tuberculosis), infected cyst, pyogenic abscess, and benign and malignant neoplasms should include is differential diagnosis. In this case presentation we really suspect benign neoplasm [5,12,28–30]. To date, the favored drug of choice remains penicillin, sure should combined with excision in small lesions at the early stage of the disease and or drainage with fistula excision if abscess occur [31]. Altay Ateşpare [13] cited that excision can prove the antibiotics efficacy when used as combined therapy in some chronic cases. Wide range of drugs prescribed as antibiotics coverage in the literature even with different doses and safety period of use such as penicillin G, amoxicillin-clavulanic acid therapy. Recurrent actinomycosis infections can be treated by metronidazole (beta-lactamase inhibitors) [32,33]. In case of patient allergies to amoxicillin, macrolides other drugs therapies as cephalosporin can be used [18,34].

Table 2
Brief description of some causes of negative culture result of actinomycosis.

Previous antibiotic therapy
Polymicrobial infection
Failure to maintain an anaerobic environment
Short-term incubation
Technical difficulties in isolation

No agreement in literature on the ideal duration of therapy. Some researches recommend the long duration of treatment can extend from weeks to months in severe cases with recurrence may occur after the cessation of the antibiotic [35]. Table 1 explain the wide range of difference between multiple case reports and series with demographical, clinical manifestation and managements. It's obvious from the table that all authors consider actinomycosis as a vague challenge in diagnosis. Finally it's important to high lighting role of histopathology in diagnosis of such dilemma. Urs AB [49] in his published article stated that in 50% of cases, culture may be negative. Table 2 show causes for such negative result, one of these is possibly attributed to technical difficulties in isolation of actinomyces bacteria in presence of other species. McHugh KE focus on the yellow sulfur granules which is the specific presentation and this should be declared for the microbiologist or pathologist for better result [50,51] seeming like those identified in several cases in the literatures [52,53]. No suppuration or fistula presence in our case makes it considered chronic type also it's a slowly progressing nodule. With this type of pattern of progression, a neoplasm can be suspected.

The diagnosis is done by histopathological examination. No role of other diagnostic aids as culture test here. Histopathological examination may show colonies of filamentous bacteria surrounded by neutrophils [10]. Surgeons should attention to that prolonged antibiotic therapy is usually required is mandatory in some cases and excellent prognosis. Good oral hygiene, proper antibiotics use, and correct surgery as needed, will improve the outcome, and mortality [10,26,48]. In this case, patient treated for one month with complete cure clinically.

5. Conclusions

From all previous details actinomycosis infections is a rare event. Early diagnosis and treatment are pivotal. Severity can be life-threatening situations. Varied clinical manifestations of the disease are make achievement of clinical diagnosis of actinomycosis is difficult. Biopsy is essential for diagnosis because of similarity to other diseases, such as neoplasms, for that actinomycosis can be a challenging problem for the clinician. Treatment should implement in harmony according to each diseases solely.

Conflicts of interest

No Conflict of Interest for All authors in this paper.

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Ethical approval

In the situation of this study is exemption from ethical approval as it's a review article.

"Only two written informed consent was obtained from the patient one as agreement to underwent surgery and other for publication and accompanying images"

Consent

"Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request."

Author contribution

1. Substantial contributions to conception and design, or acquisition of data and interpretation of data. **Dr. Rawaa / Dr. Mostafa**
2. Drafting the article or revising it critically for important intellectual content. **Dr. Rawaa / Dr. Nazar**
3. Final approval of the version to be published. Dr. Rawaa

Registration of research studies

It's case report so I don't register.

Guarantor

Corresponding author is the Guarantor here Dr. Rawaa Y. Khaleel Al-Rawee.

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