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Case Report

Actinomycosis of tongue: Rare presentation mimicking malignancy with literature review and imaging features

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ABSTRACT

Actinomycosis of the tongue is rare. It may be difficult to differentiate this infection clinically and radiologically from other tongue pathology especially neoplasia. We report a substantial tongue lesion which mimicked malignancy at presentation. The patient was treated successfully with 4 weeks of oral antibiotic therapy. It is also important that clinicians are aware of the radiological differential diagnosis of lingual actinomycosis.

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Case Report

A 60 year-old woman presented to the Maxillofacial Surgery Department with a gradual onset of painful swelling on the dorsum of her tongue. Patient was referred by otolaryngology Department with concern this was a possible malignancy. There was no history of trauma. Past medical history was also noncontributory. Oral examination revealed a firm and tender mass approximately 3 × 4 cm involving left side of the dorsum of tongue and floor of mouth. The

mucosa was intact and tongue movements were unaffected. There was no cervical lymphadenopathy or signs of systemic infection. Nasopharyngoscopic examination did not reveal any synchronous lesion. Haematological examination revealed leucocytosis and raised C-reactive protein (CRP) level. Radiological investigations prior to biopsy included contrast-enhanced Magnetic Resonance imaging (MRI) of the oral cavity and the neck (Figs. 1 and 2) revealed a large heterogenous lesion involving the left side of tongue and floor of mouth. Imaging findings were consistent with a possible neoplasm without significant adenopathy in the

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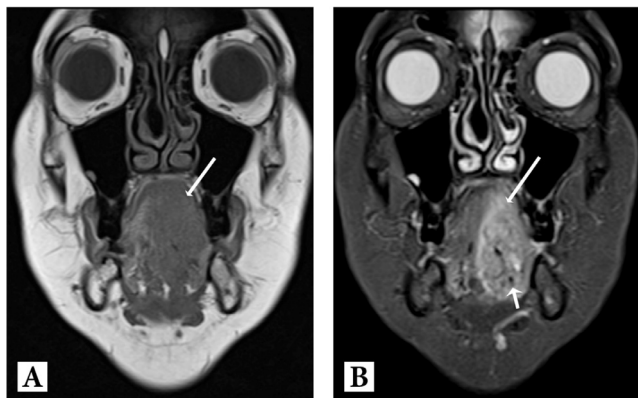


Fig. 1 – T1 (A) and T2 STIR (B) images show a large ill-defined lesion involving the entire tongue (large arrow). It is isointense to muscle on T1-weighted images and heterogenous on T2-weighted images. There are small hypointense areas seen within the lesion (small arrow). The lesion crosses the midline and involved the left genioglossus muscle.

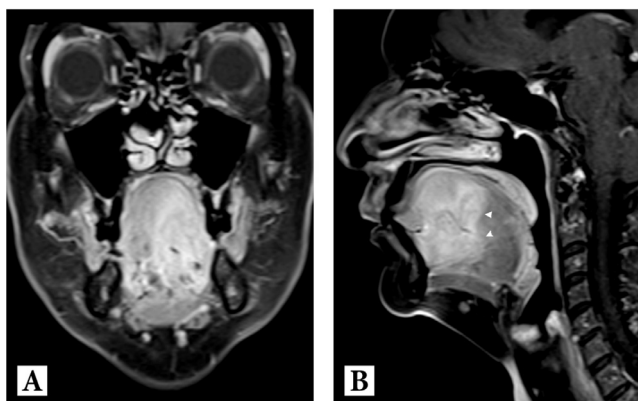


Fig. 2 – Postcontrast fat suppressed coronal (A) and sagittal (B) reveal moderate non-homogenous enhancement. The hypointense areas seen on precontrast imaging did not enhance on postcontrast scans. The involvement of the floor of mouth was better delineated after contrast administration (arrowheads).

neck. Computerized tomography (CT) scan of the chest and abdomen with intravenous contrast did not reveal distant disease. Deep biopsies were obtained and histopathology revealed inflammatory cells with colonies of actinomycosis (Fig. 3). Periodic acid–Schiff–diastase (PAS-D) and immunohistochemistry confirmed the diagnosis of actinomycosis. The patient was treated with a 4 week course of oral amoxicillin and was instructed to use 0.2% chlorhexidine gluconate mouthwash. The lesion improved gradually and eventually resolved completely. The patient has remained disease free both clinically and on follow-up imaging for over 8 months. To the best of our knowledge this is only the second report where MRI has been utilized in the diagnostic work-up.

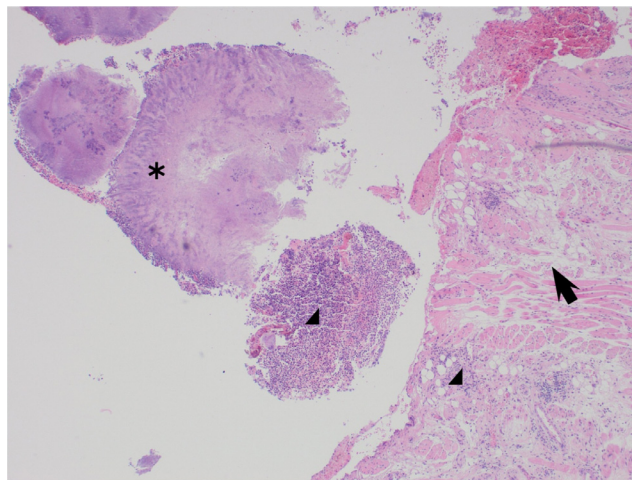


Fig. 3 – Histopathological section (H&E Stain) showing polymorphonuclear leukocytic infiltration (arrow heads), dark staining Actinomyces colonies (asterix) and connective tissues in tongue (arrow).

Discussion

Actinomycosis is a chronic suppurative infection primarily caused by Gram-positive anaerobic bacterium, *Actinomyces israelii*. There are generally 2 forms of actinomycosis infection: acute and chronic. Acute form shows features including a sudden onset of the infection and multiple pus discharging sinuses containing sulphur granules. Our case can be considered as chronic, the other form, since there was a gradual onset of disease and the lesion only had induration and fibrosis but no suppuration.

The most common sites are the cervicofacial region followed by abdomen and lungs [1]. Actinomycosis of the tongue is rare and represents only small percentage of the actinomycosis reported cases in English language literature. A few of those reported cases and their management are described as a brief summary in our report (Table 1). According to these findings, Actinomycosis of the tongue was mostly caused by local trauma but sometimes there was no obvious etiological factor.

The clinical manifestations of Actinomycosis may be confusing and mimic other disease processes. A detailed history is invaluable to determine whether it is an infective or a neoplastic pathology. The commonly reported presentation is pain, swelling and induration of the dorsum or lateral tongue. Direct spread of this infection into the adjacent tissues may cause osteomyelitis of the jaws [17] and facial disfigurement. Cervicofacial actinomycosis can also affect organs such as lung, heart, and brain through blood stream from infected tissue. Although disseminated actinomycosis is uncommon but if occurs it may lead to multiorgan failure and death if left untreated [18].

Lesions are diagnosed by a combination of histopathology and microbiological culture. Radiological investigations

Table 1 – Brief description of few reported cases of tongue actinomycosis.

Author(s) year (ref)	Age of diagnosis (years)	Sex	Clinical findings	Treatment
Sodagar and Kohort 1972 [2]	47	Female	Firm palpable mass right lateral border	Excision
Uhler and Dolan 1972 [3]	43	Male	Painful nodular mass right lateral border	Excision Penicillin for 6 months
Kuepper and Harrigan 1979 [4]	35	Male	Round firm tender mass left middle third	Penicillin for 1 month
Brignall and Gilhooly 1989 [5]	55	Male	Nonulcerated, indurated swelling anterolateral and ventral tongue surface	Penicillin for 2 months
Islaska et al. 1991 [6]	64	Female	Painless swelling left dorsal surface	Penicillin for 6 months
Morris et al. 1992 [7]	50	Male	Nodular lesion anterior-dorsal surface	Penicillin
Ficarra et al. 1993 [8]	57	Female	Nonulcerated indurated mass right lateral aspect	Penicillin for 2 weeks
Alamillos-Granados et al. 2000 [9]	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 [10]	52	Female	Solid painless mass left anterior tongue	Excision Penicillin for 3 weeks
Enoz M 2006 [11]	39	Female	Nodular lesion anterior dorsal tongue	Tetracycline for 6 weeks (Allergic to Penicillin)
Habibi et al. 2008 [12]	54	Female	Nonulcerated, nontender mass right lateral border.	Excision Penicillin for 3 weeks
Kurtaran et al. 2011 [13]	54	Female	Solid painful mass left tongue	Excision Combination therapy of Amoxicillin–Clavulanic acid with Metronidazole for 5 weeks Local debridement
Rocha et al. 2017 [14]	44	Female	Necrotic tissue with purulent right lateral tongue developed after sclerosing agent injection to treat vascular lesion	
Jat et al. 2017 [15]	44	Female	Nonulcerated nodular mass dorsal surface	Excision. Doxycycline for 3 weeks (Allergic to Penicillin)
Aneja et al. 2017 [16]	14	Male	nodular mass present on right lateral border	Excision Amoxicillin–Clavulanic acid for 2 weeks

include MRI, CT and/or Ultrasound ideally performed prior to biopsy. There are no unique radiologic features; therefore, correlation with clinical findings is important. Early diagnosis limits the spread of infection and associated morbidities [19] and avoids inappropriate surgery.

Radiologists should be familiar with the various clinical and radiologic characteristics of Actinomycosis. Kurtaran et al. (2011) reported Actinomycosis of the tongue as high-contrast enhancement mass on T1-weighted MRI image whereas a T2-weighted finding was a hyperintense mass [13]. Most of the described radiological features are not consistent with our MRI findings (Fig. 1) which emphasizes the variation in appearance depending upon amounts of inflammatory granulation and fibrous tissue within the lesion [20].

Actinomycosis can mimic a neoplasm but certain imaging features, especially on MRI, can help differentiate it from malignancy. The lesion is usually ill-defined and can be

trans-spatial whereas neoplastic lesions are generally more well defined. The presence of low T2 signal with nonenhancing areas with the lesion represents Actinomycotic granules. Lack of significant neck adenopathy in the presence of a large lesion should prompt consideration of a nonmalignant differential diagnoses including abscess, vascular malformation, chronic granulomatous disease such as tuberculosis or syphilis (Table 2).

There is lack of high quality evidence supporting use a first-line antibiotic regimen. Successful resolution after 4-6 weeks of penicillin therapy, as in our case, was seen in most studies [21]. Clindamycin, macrolides, and doxycycline are alternative options [22,23]. The addition of metronidazole as adjunctive to a beta-lactamase inhibitor is of proven benefit with recurrent and polymicrobial Actinomycosis infections [24].

Cervicofacial actinomycosis has tendency to recur. Recurrent infections were seen mainly in patients who either

Table 2 – Differential diagnosis table for tongue actinomycosis.

Disease	Radiological findings
Lingual abscess	CT: thick-rim enhancing fluid-attenuation lesions, MRI: T1 hypointense–T2 hyperintense lesion surrounded by a T1 hyperintense–T2 hypointense rim enhancing diffusely postcontrast
Squamous cell carcinoma	Contrast-enhanced CT: Moderately enhancing heterogenous mass lesion MRI: Tend to be isointense to muscle on noncontrast T1WI heterogenous increased signal on T2WI. postcontrast enhancement in most cases
Vascular malformations	CT: Enhancing hypoattenuating or heterogeneous lesions MRI: Isointense on T1WI but hyperintense on T2 WI. Usually heterogenous on contrast enhancement.
Tuberculosis	CT: Areas of low attenuation and rim enhancement MRI: Relatively homogeneous intermediate signal intensity on T1-weighted image. Heterogeneous high signal intensity mass on T2-weighted image.
Syphilis	CT: Nonspecific, enlargement of the tongue, central necrosis of lesion, occasionally cystic changes MRI: Nonspecific T1: variable enhancement; T2: high-signal intensities

had insufficient duration or incomplete treatment with antibiotics. Our literature search did not reveal any case report describing local or distant recurrence of lingual actinomycosis after being treated successfully with antibiotics over a prolonged time. Huang et al. (2018) reported actinomycotic brain abscess which developed 15 months after 6 weeks of successful treatment of nasopharyngeal actinomycosis [25]. Long-term surveillance, therefore, is still necessary for patients suffered with actinomycosis in any particular area in cervicofacial region. Published reports reveal that patients have been monitored over variable period of time ranging from 1 to 5 years at different intervals. There exists as much difference in follow-up pattern and duration as there is variation in the organs affected by actinomycosis. Overall, there is lack of uniform strategy to follow these patients up once they have achieved clinical cure from actinomycosis. Based on the afore-reported recurrence of actinomycosis after 1 year in the brain initially treated for nasopharynx actinomycosis, we propose an annual combined clinical and radiologic follow-up for at least 2 years. Further research is needed to validate such suggestion and to set guidelines for continued surveillance in patients diagnosed with actinomycosis in head and neck region.

Conclusion

Actinomycosis of the tongue remains a diagnostic challenge. The correlation of clinical, radiological, and histopathological findings is important to establish the diagnosis and avoid inappropriate treatment. Long-term clinical and imaging follow up is indicated because of the risk of relapse.

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