CASE PRESENTATION

Tongue tuberculosis, a rare extrapulmonary tuberculosis

Mohammad Yahya Abdulrazaq[a], Hiba Adnan Hammed[b]

ABSTRACT

Tuberculosis is a communicable disease, and nowadays is considered one of the leading causes of death from a single infectious agent, just after COVID-19. Tuberculosis can involve any organ in the body; however, the oral cavity and tongue can get rarely involved. Tongue tuberculosis has many pathological varieties ulcer, tuberculoma, fissure, papilloma, or tuberculous cold abscess. The diagnosis of the patient with tongue tuberculosis is usually delayed. Primary tuberculosis of the tongue is usually presented secondary to pulmonary tuberculosis, and primary tuberculosis of the tongue is very rare. Here, we presented a 46-year-old male from Baghdad with a painless tongue swelling of six months duration associated with non-healing laceration of the oral cavity not responding to local antibiotic treatment. The pathological examination has shown that this lesion is tuberculosis and responded well to the full oral anti-tuberculosis regimen.

Key words: Diseases of the tongue, tongue tuberculosis, granulomatous diseases, extrapulmonary TB.

INTRODUCTION

Tuberculosis (TB) is one of the communicable diseases that cause significant ill health. It is considered one of the leading causes of death worldwide, just second to COVID-19 and above HIV/AIDS. In 2021, 10.6 million people were estimated to fall ill with TB. Between 2020 and 2021, The burden of drug-resistant TB (DR-TB) was estimated to have increased, with 450 000 new cases of rifampicin-resistant TB (RR -TB) in 2021. [1]

TB is caused by the bacillus Mycobacterium tuberculosis (MTB), which is spread when sick people with TB expel bacteria into the air by coughing, sneezing and talking. About a quarter population of the earth is estimated to have been infected with TB during their life. [2] Still, most people will not develop TB disease, and even some will clear the infection with their immunity. [3,4]

Nearly 90 % of tuberculosis affects adults, and men are more commonly affected than women. The disease typically affects the lungs of patients causing pulmonary TB. Still, it can involve other sites leading to extrapulmonary TB.[1]

TB bacilli can reach any organ, including the oral cavity and the tongue, by the hematogenous route. [5]

Oral cavity tuberculosis represents 0.2% to 1.5% of all cases of extrapulmonary TB, and the infection is mainly secondary to primary pulmonary TB. Oral cavity TB is rare because of the continuous showering of oral mucosa with saliva and the natural resistance of oral mucosa to Mycobacterium bacilli. The relative scarcity of lymphoid tissue in the tongue is another protective factor from developing TB. In 1761, Morgagni first described a case of lingual TB. TB of the tongue is commonly seen in immunocompromised patients, smokers and the male gender. Other risk factors are bad oral hygiene and breech of tissue because of trauma. Clinically, the commonest form of TB of the tongue is ulceration which involves the lateral border of the tongue. Fine needle aspiration to biopsy the lesion is needed to confirm the diagnosis. It

a CABM, FIBMS, FRCP; Consultant pulmonologist & Internist, the National Tuberculosis Institute. Baghdad, Iraq.
b MBChB, FIBMS; Family physician. National Tuberculosis Institute. Baghdad, Iraq.
***Corresponding Author**: **Mohammad Yahya Abdulrazaq,** the National Tuberculosis Institute. Baghdad, Iraq. **E mail:** ntp.mohammad@gmail.com.*

usually shows granulomatous changes. [6]

A granuloma is a form of chronic inflammatory disorder which can occur in almost every body organ. The tubercle bacillus is one of the most common causes of granulomatous lesions. Tuberculous granuloma of the tongue is a rare form and constitutes a very low percentage of all inflammatory lesions of the tongue. [7]

The ultimate confirmation is a complete response to anti-tuberculous medication. Nowadays, nucleic acid amplification techniques and polymerase chain reactions are used to reach the correct diagnosis. [6]

Aird described five pathological types of tuberculous lesions of the tongue (a) Tuberculous ulcer (b) Tuberculous Fissure (c) Tuberculoma (d) Tuberculous cold abscess, and (e) Tuberculous papilloma. [5]

CASE PRESENTATION

A 46-year-old male from Baghdad presented to the maxillofacial department with a six months history of tongue swelling associated with a foul mouth smell and continuous non-healing laceration of the oral cavity, not responding to local treatment with antibiotics and mouthwash. The swelling of the tongue was painless, non-tender, and not associated with difficulty in swallowing or affecting the movement of the tongue.

The patient reported a history of low-grade fever, mainly at night. The patient has smoked about 40 cigarettes for the past 30 years and did not drink alcohol. He has no significant past medical or surgical history and did not recall being contact with a patient with tuberculosis.

Examination of the mouth showed a firm nodular swelling measuring 2.5 cm in diameter within the anterior part of the tongue. The swelling was not tender and not ulcerated. The lesion did not restrict the movement of the tongue. A poorly defined ulcerative lesion was found on the interior aspect of the lower lip, associated with foul mouth odour. A cervical examination revealed no palpable lymph nodes. Chest examination was within normal limits. Routine biochemical and haematological laboratory blood investigations were within normal limits except for a raised white blood cell count and high erythrocyte sedimentation rate. Chest X-ray was within normal, and head, neck and abdominal ultrasound were within normal.

The patient was admitted to the surgical department as a case of tongue mass with chronic mouth ulceration and prepared for surgical excisional biopsy. Two pieces were excised and sent for histopathological examination

The histopathological exam showed acanthotic and keratotic oral epithelium with granulation tissue proliferation and chronic inflammation in the submucosa. The connective tissue stroma showed numerous granulomas of Langhans type of giant cells with an area of focal necrosis involving the whole thickness of the epithelium, connective tissue stroma and the muscle layer associated with focal endarteritis. No malignancy was seen in the sections examined.

The histopathologic report was compatible with TB granuloma. The patient was referred to the National TB Institute as a case of tongue TB, recorded as an extrapulmonary TB case (tongue TB) and started on anti-TB medications.

The patient started on the initial phase with four tablets daily of a fixed-dose formula of rifampicin 150 mg, isoniazid 75 mg, pyrazinamide 400 mg, and ethambutol 275 mg for two months. After that, he continued on four tablets daily of a fixed-dose formula of rifampicin 150 mg and isoniazid 75 mg for four months.

The patient showed a dramatic improvement; the tongue lesion and mouth ulceration were resolved within the first month of treatment, and his mouth hygiene has remarkably improved. The treatment was continued for six months as scheduled, with an excellent medical response.

DISCUSSION

TB is a chronic infectious disease usually caused by MBT that is transmitted by expelled infectious aerosol droplets of active TB pa-

Figure 1 | Laceration of the oral cavity with swelling of the tongue and poorly defined ulcerative lesion in the interior aspect of the lower lip.

tients. However, in most cases, TB infection is suppressed by an effective host immune response; TB often becomes asymptomatic in latent state and is not contagious. Approximately 5% of otherwise healthy adults with latent TB will transform into active TB disease within 2 -10 years. TB is classified clinically as pulmonary and extrapulmonary, depending on the site of infection. The lung is the predominant site of TB, but any body organ can be involved.[8]

Oral TB is a rare condition reported only in 0.05 to 5% of total extrapulmonary TB.[9] Oral manifestations usually present as superficial ulcers, papillomatous lesions, patches, or indurated soft tissue lesions. [10] In our case, the patient presented with nodular swelling of the tongue with ulceration in the oral cavity. Martins-de-Barros has reported a similar presentation. [11]

Oral tuberculosis may be associated with cervical lymphadenopathy, as reported by Sareen;[12] however, we did not find any lymph node enlargement in the neck by clinical or ultrasound examinations.

Oral cavity extrapulmonary tuberculosis is usually secondary to pulmonary tuberculosis.[13] Primary oral lesions caused by tuberculosis without pulmonary infection are extremely rare, occurring in less than 1% of extrapulmonary cases. [14] Rafael Pila Pérez [15] and Kumar[16] have reported lingual tuberculosis secondary to a primary TB in the lung. Our patient Chest X-ray was completely normal.

Despite being rare, TB of the tongue should be kept in mind when dealing with refractory tongue lesions not responded to treatment, and biopsy should not be delayed. [17] In our patient, the diagnosis was delayed for six months and only established by a histopathological examination. Sareen, [12] Parul Jain [7] and Kumar [16] have also reported similar delays in their cases.

Without treatment, the death rate from TB disease is nearly 50%.[18] With currently-recommended treatments, about 85% of people can be cured. Regimens of 1–6 months are available to treat TB infection.[1] our patient showed dramatic improvement with full six months of treatment

CONCLUSION

Tuberculosis of oral cavity is a rare condition and a presentation in the tongue is even rarer. Usually presented as a secondary to pulmonary TB

Tuberculosis should be kept in the differential diagnosis of any tongue mass and pathological examination plays very essential role in reaching the correct diagnosis in such a situation.

REFERENCES

1. World Health Organization. Global tuberculosis report 2022. Published on 27 October 2022. Available from: https://www.who.int/teams/global-tuberculosis-programme/tb-reports/global-tuberculosis-report-2022#:~:text=The%20WHO%20Global%20Tuberculosis%20Report,global%2C%20regional%20and%20country%20levels. Accesses on 10 November 2022.
2. Hershkovitz I, Donoghue HD, Minnikin DE, May H, Lee OY, Feldman M, et al. Tuberculosis origin: the Neolithic scenario. *Tuberculosis*. 2015;95 Suppl 1:S122–6. Available from: https://www.ncbi.nlm.nih.gov/pubmed/25726364. Accessed 15 August 2022
3. Sakula A. Robert Koch: centenary of the discovery of the tubercle bacillus, 1882. *Thorax*. 1982;37(4):246–51.
4. WHO consolidated guidelines on tuberculosis. Module 3: Diagnosis – rapid diagnostics for tuberculosis detection 2021 update. Published on 7 July 2021. Available from: https://www.who.int/publications/i/item/9789240029415. Accessed on 7 November 2022.
5. Khatri BK, Jhalla GS. Tuberculosis of tongue: A case report. *Indian J Tuberc* 1971;18:58-9.

6. Jain P, Jain I. Oral manifestations of tuberculosis: Step towards early diagnosis. *J Clin Diagn Re*s 2014;8:ZE18 21
7. Jain P, Adhikari A, Mandal PK, Minz RS. Tuberculosis of tongue, an enigma: Report of two cases. *CHRISMED Journal of Health and Research*. 2019 Apr 1;6(2):117.
8. Kim SY, Byun JS, Choi JK, Jung JK. A case report of a tongue ulcer presented as the first sign of occult tuberculosis. *BMC Oral Health*. 2019 Dec;19(1):1-5.
9. Wang W, Chen J, Chen Y, Lin L. Tuberculosis of the head and neck: a review of 20 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2009;107(3):381–6.
10. Miziara ID. Tuberculosis affecting the oral cavity in Brazilian HIV-infected,patients. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2005;100(2):179–82.
11. Martins-de-Barros AV, Silva ED, Araújo FA, Carvalho MD. Primary tuberculosis of the tongue. *Revista da Sociedade Brasileira de Medicina Tropical*. 2021 Dec 17;54.
12. Sareen D, Sethi A, Agarwal AK. Primary tuberculosis of the tongue: a rare nodular presentation. *British dental journal*. 2006 Mar;200(6):321-2.
13. Iype EM, Ramdas K, Pandey M, Jayasree K, Thomas G, Sebastian P, Nair MK. Primary tuberculosis of the tongue: report of three cases. *British Journal of Oral and Maxillofacial Surgery*. 2001 Oct 1;39(5):402-3.
14. Kakisi OK, Kechagia AS, Kakisis IK, Rafailidis PI, Falagas ME. Tuberculosis of the oral cavity: a systematic review. *Eur J Oral Sci*. 2010;118(2):103-9.
15. Pérez RP, Prieto VA, Peláez RP, Torres PR, Hernández DC. A Case Report of Pulmonary and Lingual Tuberculosis. *Rev. Col. Gastroenterol.* 2014;29(2):183-7.
16. Kumar RD, Udagatti VD. Tuberculosis of Tongue (Oral Cavity): A Case Report. *Tuberculosis*. 2018;9(4).
17. Umesh Kumar Chandra & Atul Shende. Tuberculosis of the tongue: A rare presentation of common disease. *Indian J Med Res* 152 (Supplement), November 2020: 212-213. DOI: 10.4103/ijmr.IJMR_2345_19
18. Tiemersma EW, van der Werf MJ, Borgdorff MW, Williams BG, Nagelkerke NJ. Natural history of tuberculosis: duration and fatality of untreated pulmonary tuberculosis in HIV negative patients: a systematic review. *PL oS One*. 2011;6(4):e17601

Abbreviations Acquired immunodeficiency syndrome (**AIDS**), Corona Virus disease 2019 (**COVID-19**), Drug-resistant TB (**DR-TB**), Human immunodeficiency virus (**HIV**), Mycobacterium tuberculosis (**MTB**), Rifampicin-resistant TB (**RR-TB**), Tuberculosis (**TB**).

Conflict of interest: Author has nothing to disclose.

Funding: Nothing apart from self-funding.