

Rare case of extrapulmonary tuberculosis masquerading as an intraoral sinus in association with a nonhealing extraction socket in a teenage girl—“A case report with Review of literature”

Tribikram Debata¹, Anuroopa Mohapatra², Sujit Ranjan Sahu¹, Upasana Das¹

¹Department of Oral and Maxillofacial Pathology, SCB Dental College and Hospital, Cuttack, Odisha, ²Department of Dentistry, Pandit Raghunath Murmu Medical College and Hospital, Baripada, Odisha, India

Abstract

Tuberculosis (TB) is a serious infectious disease with significant mortality and most commonly affects the pulmonary system and rarely the oral cavity. Because oral tuberculosis is a rare disease, it is often overlooked in the differential diagnosis of oral lesions. Despite the recent decline in the incidence of tuberculosis, it remains a highly contagious and serious public health problem, thus requiring early diagnosis and rapid intervention. Extrapulmonary tuberculosis (EPTB) is defined as any bacteriologically confirmed or clinically diagnosed case of tuberculosis (TB). A 17-year-old girl reported at the outpatient Department of Dentistry with the chief complaint of a swelling at the backside of mouth along with pus discharge. She noticed these symptoms approximately one month after the extraction of the left mandibular first molar. The patient was referred to the Department of Pulmonary Medicine for further investigation and treatment. Subsequently, diagnosis of EPOTB was reached on the basis of the histopathological findings and the previous personal as well as family history. Drastic improvement was observed in the general condition and a complete resolution of the oral lesion after four weeks of ATT and the patient was completely free of all the symptoms after six months of follow-up period. This case demonstrated the importance of oral manifestation of oral tuberculosis for dentist who may be the first healthcare provider to encounter a variety of oral lesions.

Keywords: Nonhealing extraction socket, oral sinus, pulmonary tuberculosis, teenage girl

Address for correspondence: Dr. Tribikram Debata, Department of Oral and Maxillofacial Pathology, SCB Dental College and Hospital, Cuttack, Odisha, India.
E-mail: tribikramdbt@gmail.com

Submitted: 08-Jul-2023, **Revised:** 19-Dec-2023, **Accepted:** 20-Dec-2023, **Published:** 11-Jul-2024

INTRODUCTION

Extrapulmonary tuberculosis (EPTB) is defined as any bacteriologically confirmed or clinically diagnosed case of tuberculosis (TB) involving organs other than the lung parenchyma, e.g. pleura, lymph nodes, abdomen, genitourinary tract, skin, joints, bones, and meninges. It

constitutes 10–15% of total TB cases worldwide. It may be a result of direct inoculation of bacilli into extrapulmonary tissues called primary EPTB. Dissemination of bacteria from lungs into other organs or systems through blood circulation or lymphatics that clinically manifests due to reactivation or reinfection, i.e., lesions occurring in

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

For reprints contact: WKHLRPMedknow_reprints@wolterskluwer.com

How to cite this article: Debata T, Mohapatra A, Sahu SR, Das U. Rare case of extrapulmonary tuberculosis masquerading as an intraoral sinus in association with a nonhealing extraction socket in a teenage girl—“A case report with Review of literature”. *J Oral Maxillofac Pathol* 2024;28:301-6.

Access this article online

Quick Response Code:



Website:

<https://journals.lww.com/JPAT/>

DOI:

10.4103/jomfp.jomfp_304_23

previously sensitized individuals, is called secondary EPTB.^[1,2]

Oral TB is a fairly uncommon entity, constituting 0.2–1.5% of the global load of TB (R). Morgagni was the first to report tuberculous ulcers on tongue along with a pulmonary focal infection in 1761. Extrapulmonary oral TB (EPOTB) is considerably rare, mostly found in older individuals. Such an involvement usually presents as ulcers of tongue, gingiva, and muco-gingival fold or is associated with an extraction socket.^[3] The key to management of these cases is early diagnosis which is challenging as the symptoms are not that of a typical pulmonary infection. It will not be an exaggeration to state that identification of the tuberculous lesion in a dental OPD may potentially serve as its first line of control and effective management of EPOTB. This is factually supported by a systematic review, reporting that timely diagnosis of an oral manifestation of TB has led to the diagnosis of a previously unknown systemic infection in nearly 50% of patients.

The aim of this article is to report the first of its kind EPOTB presenting as an intraoral sinus in association with an extraction socket in a teenaged girl.

CASE REPORT

A 17-year-old girl reported at the outpatient Department of Dentistry with the chief complaint of a swelling at the backside of mouth along with pus discharge from the same. She noticed these symptoms approximately one month after the extraction of the left mandibular first molar. Despite administration of antibiotics and topical mouthwashes, the symptoms did not subside.

On examination, an intraoral swelling was found along with a sinus discharge at the vestibular sulcus with respect to the extracted tooth [Figure 1a]. Palpation revealed the presence of a fibrotic band in the region. The overall oral hygiene was good. Lymph nodes of the neck were not palpable. The patient looked apparently healthy and did not give any history of fever, weakness, or significant weight loss. She, however, was diagnosed with TB lymphadenitis twice previously with an interval of 1 year approximately 2 years ago. However, the particulars of the antitubercular therapy (ATT) with regard to the agents administered and the duration is unclear. The patient also revealed that her father was treated for TB meningitis and completed the entire regimen of ATT.

She was advised an oral panoramic radiograph (OPG) which revealed an ill defined radiolucency along with loss of normal trabecular pattern in the extraction socket [Figure 1b]. On the basis of the clinical and

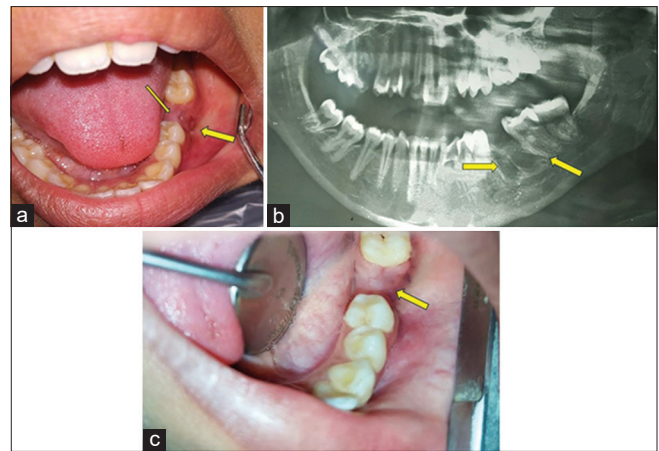


Figure 1: (a) Clinical photograph showing intraoral sinus opening in the alveolus and sulcus region with respect to 36 tooth region. (b) Orthopantomogram showing widening of extraction socket with irregular alveolar bone destruction with respect to distal root socket. (c) Clinical photographs showing the regressed lesions after six months of antitubercular treatment

radiological findings, the provisional diagnosis was a chronic inflammatory lesion, residual cyst, actinomycosis, and osteomyelitis of mandible.

Routine blood investigations revealed a raised ESR. With the provisional diagnosis of chronic inflammatory lesion, an excisional biopsy was performed and the sample of the pus discharge collected on a cotton swab was sent for cartridge-based nucleic acid amplification testing (CBNAAT).

Histopathological examination (H & E stain) of the excised specimen revealed a granulomatous inflammation characterized by Langhans giant cells, abundant epithelioid cells, and lymphocytic infiltrate with areas of caseous necrosis, consistent with findings of TB granuloma [Figures 2 and 3].

The patient was referred to the Department of Pulmonary Medicine for further investigation and treatment. In spite of the raised erythrocyte sedimentation rate, pulmonary TB was ruled out on the basis of the following:

Constitutional symptoms of TB were absent.

CBNAAT was negative.

Chest radiographs of the patient were normal.

Subsequently, diagnosis of EPOTB was reached on the basis of the histopathological findings and the previous personal as well as family history. A chemotherapeutic sensitized regimen of rifampicin, isoniazid, and pyrazinamide was initiated. There was a drastic improvement in the general

condition and a complete resolution of the oral lesion after four weeks of ATT. ATT for bone TB was then initiated. The patient was completely free of all the symptoms in the six-month follow-up period [Figure 1c].

DISCUSSION

Tuberculosis (TB) is a chronic granulomatous infectious disease caused by *Mycobacterium tuberculosis* that has a primary affinity for the parenchymal tissue of the lungs but may affect any part of the body including the oral cavity.^[4] It is a global health challenge that has existed for more than a millennium. Over the years, with an increased understanding of the pathophysiology and clinical presentation, there has been a trend of early diagnosis and efficient management.^[5] Even so, it is still a significant contributor to morbidity and mortality worldwide, especially among people of lower socio-economic status and hence, the developing nations. It afflicts approximately 10 million individuals every year

and is one of the top ten causes of fatality worldwide. By WHO estimates of 2017, India accounts for 27% of the global estimated 10 million cases and 25% of the estimated 1.6 million deaths due to tuberculosis.^[6]

According to all the reviews published before 1950, there was a considerable decrease in the global incidence of TB due to advancements in chemotherapeutics, along with improved public health and hygiene awareness. Oral TB constituted about 0.1–5% of the total cases. The late twentieth century, however, witnessed a re-emergence of all forms of the disease, due to an increasing global resistance to antituberculous drugs and Acquired Immuno-deficiency Syndrome (AIDS).^[3]

EPOTB is called so when there is no pulmonary involvement. Primary EPOTB occurs as a result of primary inoculation and invasion of bacilli into the oral mucosa and is an extremely rare incidence. It is possibly due to the resistance to bacillary invasion provided by the intact oral mucosa aided by the cleansing action of saliva. The presence of tissue antibodies, oral saprophytes, and salivary lysozymes further reduces the chances of disease initiation. However, a breach in the oral mucosa, resulting from deleterious oral habits, traumatizing dentures, or poor oral hygiene or that induced iatrogenically by extractions or surgical manipulations, may lead to localization of the mycobacteria, resulting in infection. More commonly, EPOTB occurs secondary to pulmonary TB due to reactivation or reinfection and occasionally by hematogenous spread.

Part of the enigma surrounding tuberculosis is its myriad presentation and latent period of infection. On primary exposure, the bacilli settle into lung tissue and are subjected to phagocytosis and degradation by the



Figure 2: Photomicrograph of the grossed specimen showing two bits of g/w soft tissue specimen

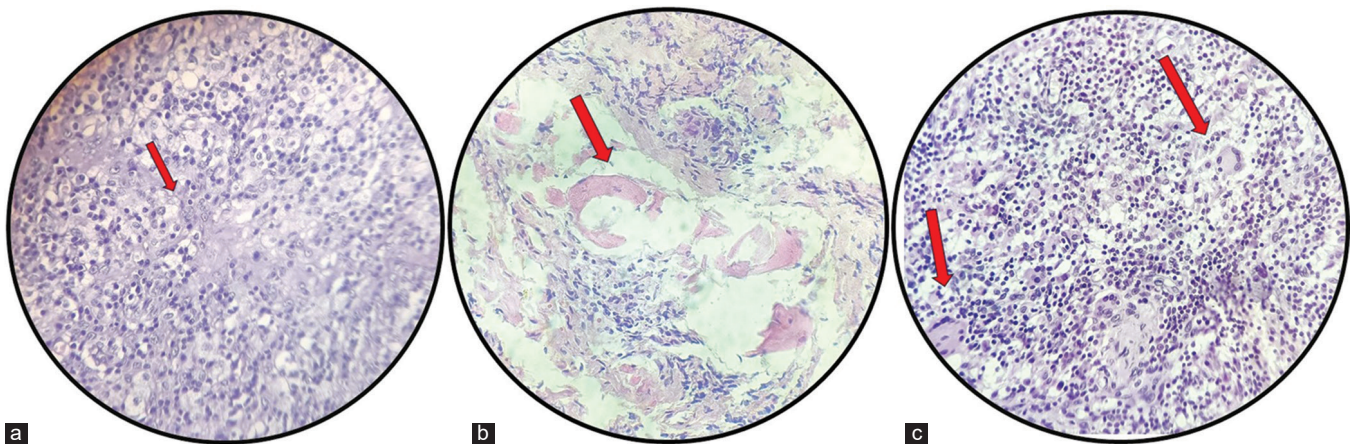


Figure 3: (a) Photomicrograph showing granulomatous inflammation with foci of necrosis. (b) Necrotic bone with chronically inflamed fibrocellular connective tissue. (c) Epithelioid cells with Langhans giant cells. (H & E, 10×)

alveolar macrophages called dust cells. A few bacilli may escape the lysosomal delivery and survive inside the macrophages. They are maintained in a nonpathogenic stage by the formation of granulomas. These granulomas may stay put in lung parenchyma or get disseminated into other tissues through vascular supply. In a majority of cases, infection by the tubercle bacilli does not progress to disease, i.e. virulent mycobacteria exists in a so-called 'dormant' state for as long as the host immune system is effective with granulomas acting as the 'hide-outs', ensuring long-term survival of the bacilli. On the occasion of reinfection or reactivation of latent tubercle due to immune dysfunction, the disease manifests [Figure 4] in

primary lung tissue or the organs housing the dormant granulomas.

In the present case, a careful anamnesis indicated the probability of the bacilli lodged directly into the alveolar socket due to disruption in the oral mucosa caused by the dental extraction. Another possible and more probable route of infection could be the reactivation of latent granulomas previously lodged in oral mucosa via hematogenous or lymphatic spread, leading to the formation of nonhealing, pus-discharging intraoral sinus. This is an extremely rare (incidence of less than 1%) presentation, i.e. oral manifestation of reactivated TB with

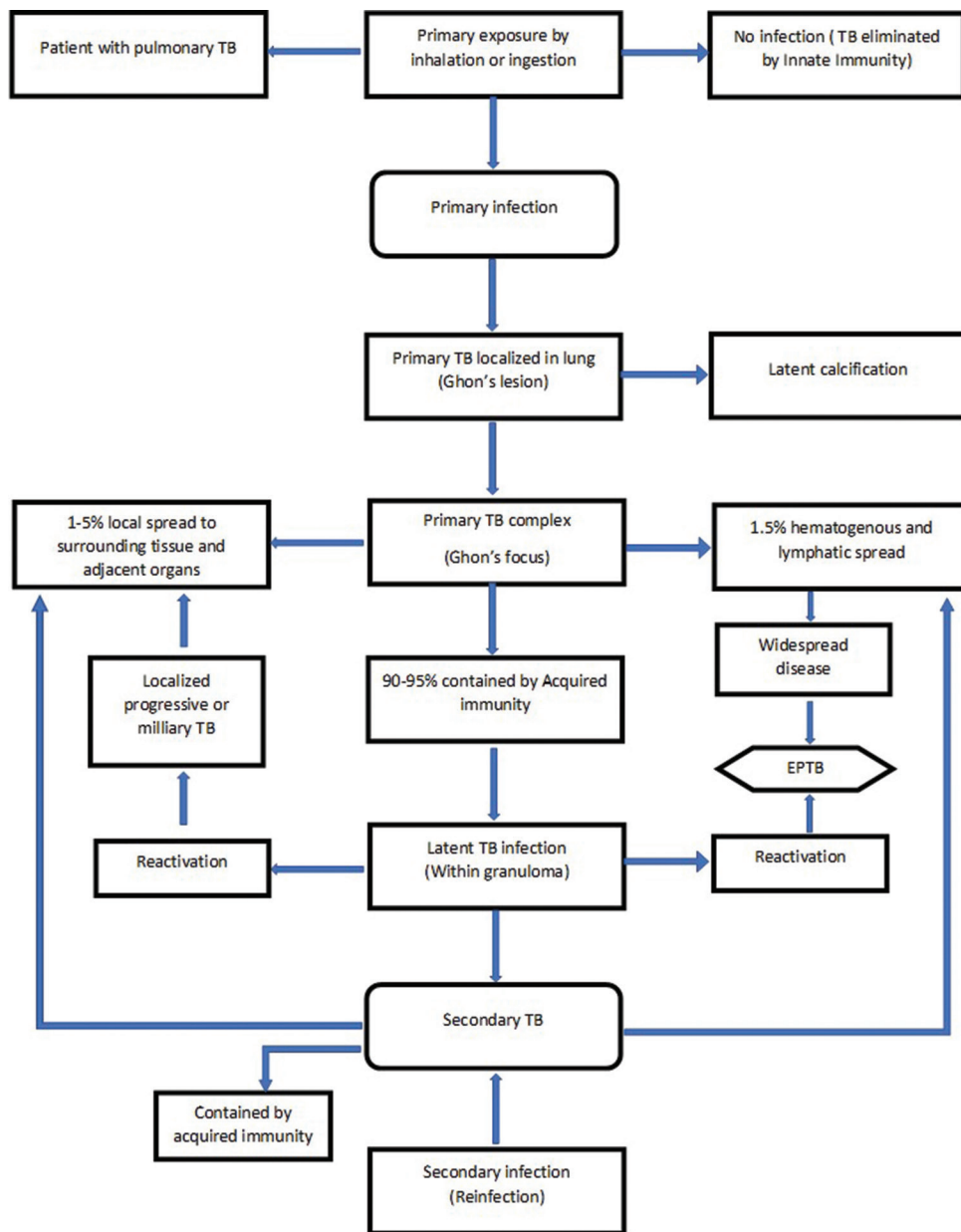


Figure 4: Schematically represent extrapulmonary tuberculosis masquerading as an intraoral sinus in association with a nonhealing extraction socket in a teenage girl

no pulmonary infection. In a similar case report in 2018, Shanahan *et al.*^[7] discussed about a painful nonhealing ulcer of the hard palate in a female patient, aged 56 years. After a series of investigations that confirmed the absence of active focus of infection in the lungs, a diagnosis of reactivated TB was reached. It was confirmed on the basis of mycobacterial culturing of tissue biopsy. After an extensive electronic survey of the existing scientific data of the past three decades (1992–2022), we mostly came across case reports and a few case series of primary EPOTB (42%) or oral TB coexisting with pulmonary TB (58%). There were also a few reports of tuberculous lesions in the oral cavity those were chance findings and helped in diagnosing a pulmonary infection. What is perplexing is that, out of 58 systematic reviews done with regard to oral TB, EPOTB is neither reported nor reviewed as a separate entity. It is imperative to understand that this diagnostic term refers to a tuberculous lesion of the oral cavity presenting without any pulmonary infection. These lesions may be primary or secondary in origin, the latter referring to TB due to reinfections and reactivations.

In itself, the lesion is painful and rarely subsides without intervention. Lynch (1984) described oral tuberculous lesions, as those having unremitting severe pain that increases progressively and interferes with the daily lifestyle of the patient. Once detected, these lesions are fairly straightforward to treat except for those complicated with HIV or multiple drug resistance. The challenge, however, is timely diagnosis owing to its atypical presentation more so in EPOTB. It may present itself as a nonhealing, painful ulcer of tongue or in other sites like the lip, cheek, uvula, and alveolar mucosa. It may be a nodular swelling, fissures, tuberculomas, or granulomas. Constitutional symptoms may not be apparent. The varied presentation increases the risk of overlooking TB in the differential diagnosis of oral lesions. That in turn may lead to an increased spread of disease, especially in immune-compromised patients. According to The WHO estimates, an individual with active TB can infect 10–15 other individuals through close contact over the course of a year.^[8] Thus, a missed case is not just a missed case! What is making matters worse is the tunnel vision acquired due to specialization in medical and dental practice.

An instance of a nonhealing extraction socket could be attributed to several local or systemic causes like nutritional status, use of steroid hormones, age, and metabolic status.^[9] The differential diagnosis in our case ranged from alveolar osteitis, secondary infection due to foreign bodies, residual cyst, and actinomycosis

to debilitating conditions like osteomyelitis of mandible (OM) and neoplasms like oral squamous cell carcinomas (OSCC).^[10] However, the ill-defined radiolucent area with respect to the extracted tooth ruled out the possibility of a dry socket, the presence of secondary infection, and foreign bodies as well as a residual cyst. The inconclusive history of previous antitubercular therapy led us to a high index of suspicion for chronic TB also supported by the raised serum ESR. Although the result CBNAAT of the pus discharge from the site was negative, it did not completely rule out the possibility of mycobacterial infection.^[11] The histopathologic examination of the biopsy specimen confirmed the diagnosis of tuberculosis. Pulmonary involvement was ruled out by chest radiographs, leading us to the confirmatory diagnosis of EPOTB of secondary origin. The lesion resolved within six weeks of commencement of multidrug therapy, further establishing our final diagnosis [Figure 4]. As per the literature search, this is the first published case report of primary EPOTB that presented as an intraoral draining sinus. It is a rare occurrence, so it highlights the importance of in-depth clinical examination and proper investigation for accurate diagnosis and management. The uniqueness of this case is that in spite of uncommon distinctive presentation, the case was diagnosed on time due to which the patient treatment was not delayed.

CONCLUSION

Many times oral conditions are merely a symptom of a systemic disorder that may or may not have been diagnosed and thus are the proverbial ‘tip of the iceberg’. The importance of careful consideration of the overall clinical picture and detailed history-taking can never be undermined. After all, as a health professional, our aim is to treat a patient and not just cure a disease.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. World Health Organization. Global TB Report. 2019. Available from: https://www.who.int/tb/publications/global_report/en/. [Last accessed on 2021 Mar 01].
2. Sharma S, Mohan A. Extra pulmonary tuberculosis. *Indian J Med Res* 2004;120:316–53.
3. Kakisi OK, Kechagia AS, Kakisis IK, Rafailidis PI, Falagas ME. Tuberculosis of the oral cavity: A systematic review. *Eur J Oral Sci* 2010;118:103–9.
4. Pai M, Correa N, Mistry N, Jha P. Reducing global tuberculosis deaths—time for India to step up. *Lancet* 2017;389:1174–6.
5. World Health Organization. Global TB Report. 2019. Available from: https://www.who.int/tb/publications/global_report/en/. [Last accessed on 2021 Mar 01].
6. GBD Tuberculosis Collaborators. Global, regional, and national burden of tuberculosis, 1990-2016: Results from the global burden of diseases, injuries, and risk factors 2016 study. *Lancet Infect Dis* 2018;18:1329–49.
7. Shanahan D, Staines K. Oral tuberculosis: a case report and review of the literature. *Oral Surgery* 2019;12:65-70.
8. Narasimhan P, Wood J, MacIntyre CR, Mathai D. Risk factors for tuberculosis. *Pulm Med* 2013;2013:828939.
9. Henry CJ, Stassen LFA. The non-healing extraction socket: A diagnostic dilemma - case report and discussion. *J Ir Dent Assoc* 2016;62:215-20.
10. Gupta R, Prakash V. Non-surgical management of mandibular molar with multiple intra-oral sinus tracts: A case report. *J Clin Diagn Res* 2013;7:2392-3.
11. Admaja KN, Thomas VA. Typical granulomatous lesion of gingiva: A case report on rare manifestation of tuberculosis in oral cavity. *J Dent Oral Care Med* 2017;3:203.