

A Rare Case of Tubercular Osteomyelitis of Mandible in a 5-year-old Child

Mohd Aswad Khan¹, Sajjad Abdur Rahman², Mohammad Danish³, Ruqiya Afrose⁴

ABSTRACT

Tuberculosis (TB), also known as Koch's disease, is a chronic granulomatous disease typically caused by *Mycobacterium tuberculosis* (*M. tuberculosis*). On 24th March 1882, Dr Robert Koch discovered *M. tuberculosis* that caused TB. In humans, *M. bovis* and atypical mycobacterium may also cause this disease. According to the World Health Organization (WHO) Global Report 2022, published on 27th October 2022, the incidence of TB in India for the year 2022 is 210/100,000 population. Primarily, TB affects the pulmonary region in humans, whereas secondarily, it may affect extrapulmonary sites such as the bones and intestines via lymph nodes. In this article, we are reporting a rare case of tubercular osteomyelitis of the mandible, in which the patient reported swelling of the cheek, mimicking an odontogenic infection that led to mandibular osteomyelitis. The definitive diagnosis of tubercular osteomyelitis was made by cartridge-based nucleic acid amplification test (CB-NAAT) when the cheesy material was found during surgical debridement and curettage of the mandible. Following that diagnosis, antitubercular treatment (ATT) was started immediately for the patient, which led to the complete resolution of the disease. The patient has been on regular follow-up for the last six months with no evidence of relapse of disease. Primary TB of the mandible is very rare, with only a few reported cases in the literature.

Keywords: Case report, Cartridge-based nucleic acid amplification test, Odontogenic infection, Tuberculosis, Tubercular osteomyelitis.

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INTRODUCTION

Tuberculosis (TB) is a chronic granulomatous disease typically caused by *Mycobacterium tuberculosis* (*M. tuberculosis*).¹ In humans, *M. bovis* and atypical mycobacterium may also cause TB. According to the World Health Organization (WHO) Global Report 2022, published on 27th October 2022, the incidence of TB in India for the year 2022 is 210/100,000 people, and a total of 21.4 lakh cases were reported, which is 18% more than the year 2020. Meanwhile, globally, 10.6 million people were diagnosed with TB in the year 2021, which caused 1.6 million deaths. It primarily affects the lungs in humans, whereas secondarily, it may affect extrapulmonary sites such as bones and intestines via lymph nodes.² Oral cavity lesions are rare, amounting to only 0.1% of cases. The tongue has been reported to be the most common site in the oral cavity.^{3,4} A secondarily affected mandibular TB shows the classical features of pulmonary Koch's, whereas a primarily affected mandible is rare and observed in only <2% of cases of skeletal TB.^{5,6} In this article, we report an unusual case of tubercular osteomyelitis affecting the right side of the body of the mandible, where the primary presentation of the patient was favoring odontogenic infection.

CASE DESCRIPTION

A 5-year-old female child was referred to our department for a complaint of swelling on the right lower side of the face for 1 month (Fig. 1).

Upon interview, the patient's father reported that he noticed swelling 1 month ago, which rapidly increased from 1 × 1 to 5 × 6 cm. The swelling was associated with high-grade fever without chills and rigor; however, there was no associated pain in the teeth. They consulted pediatric dentistry outpatient department (OPD), where a provisional diagnosis of dentoalveolar infection with a carious right lower second primary molar was made. An orthopantomogram revealed a carious right lower second primary

¹⁻³Department of Oral and Maxillofacial Surgery, Dr Ziauddin Ahmad Dental College, Aligarh Muslim University, Aligarh, Uttar Pradesh, India

⁴Department of Pathology, Jawaharlal Nehru Medical College, Aligarh Muslim University, Aligarh, Uttar Pradesh, India

Corresponding Author: Sajjad Abdur Rahman, Department of Oral and Maxillofacial Surgery, Dr Ziauddin Ahmad Dental College, Aligarh Muslim University, Aligarh, Uttar Pradesh, India, Phone: +91 8265894741, e-mail: sajjadar1979@gmail.com

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molar (Fig. 2), for which pulpectomy was performed; however, the swelling did not subside, and then extraction of the right lower second primary molar was performed, but even after extraction, the symptoms did not resolve. The patient was then referred to our OPD for further management. On examination, she was found to be healthy and had no history of any systemic illnesses or treatment of chronic infections. There was no loss of weight or diet. On extraoral examination, a solitary swelling of approximately 5 × 6 cm, which was oval in shape having diffuse borders, was noted on the right body of the mandible. The swelling extended to the right submandibular region, which was soft and fluctuant in consistency and tenderness on palpation. The overlying skin appeared normal in color with the localized rise in temperature. Palpation also revealed an enlarged lymph node in the right submandibular region, which was also tender. On intraoral examination, similar swelling was noted



Fig. 1: Swelling present on the right lower side of face since 1 month

in the right buccal vestibule. The overlying mucosa appeared normal in color without any abnormal discharge. A provisional diagnosis of buccal space infection of the right half of the lower jaw was made.

For confirmation of diagnosis, ultrasonography was done, which revealed pus collection of size 35 × 35 mm and volume 5–8 cc with a few enlarged lymph nodes in the body of the right half of the mandible with some cortical bone destruction, suggesting chronic osteomyelitis of the right side of the mandible with acute symptoms.

Incision and drainage were performed to drain the pus through a right mandibular buccal vestibular. Pus of approximately 10–15 mL in volume having yellowish green in color was collected and sent for culture and sensitivity (C and S), which showed no aerobic growth. The patient was advised to use empiric antibiotics, and contrast-enhanced computed tomography (CECT) scans were used to evaluate the extent of bony destruction. On the next appointment, cleaning and debridement were repeated, and approximately 10 mL of reddish–yellow pus was drained and collected. Culture and sensitivity were repeated, which showed growth of *Pseudomonas aeruginosa*, which was sensitive to piperacillin, tazobactam, and amikacin. CECT of the face revealed destructive lesions in the posterior half of the body of the right mandible extending from the first permanent molar region with associated cortical erosion and collection of pus along the buccolingual aspect, which suggested osteomyelitis (Figs 3A and B). Reactionary lymphadenopathy of level 1b was present, showing central area necrosis.

The patient was taken up for intraoral surgical debridement and curettage of the right body of the mandible under general anesthesia. During surgical debridement, cheesy material was noted, and destruction of bone was present involving both the buccal and lingual cortical plates along with the inferior border of the mandible, as noted on the CECT face (Fig. 3C). Four lymph nodes were also found in the body of mandible which were removed during surgical procedure (Fig. 3D). Lymph nodes along with granulation tissue were sent for histopathological examination. Upon looking at the cheesy and slimy material, tubercular infection was suspected; hence, it was sent for the cartridge-based nucleic acid amplification test (CB-NAAT) analysis, which came back positive for *M. tuberculosis*.

The patient was further evaluated systemically and radiologically by X-ray of the chest and USG abdomen to establish a primary lesion. CB-NAAT of Sputum was also performed, but it was found to be negative for *M. tuberculosis*.

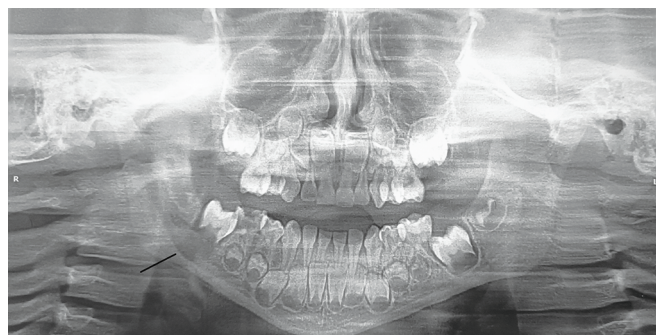


Fig. 2: Optical parametric generator (OPG) reveals carious right second primary molar along with the destruction of bone on the right angle of the mandible and ramus of the mandible

Histopathological examination revealed numerous Langhan's giant cells with caseating necrosis and numerous well-formed epithelioid cell granulomas, which were suggestive of chronic granulomatous lesions favoring TB (Figs 4A and B).

On confirmation of the diagnosis, antitubercular treatment (ATT) category I for 6 months of rifampicin 225 mg, isoniazid 150 mg, and ethambutol 300 mg was started along with pyridoxine hydrochloride 10 mg once daily. On regular follow-up, the patient is doing well, and the swelling has subsided (Fig. 5) with no further evidence of destruction of bone shows the bone formation after 1 month of surgery at the right angle of the mandible (Fig. 6).

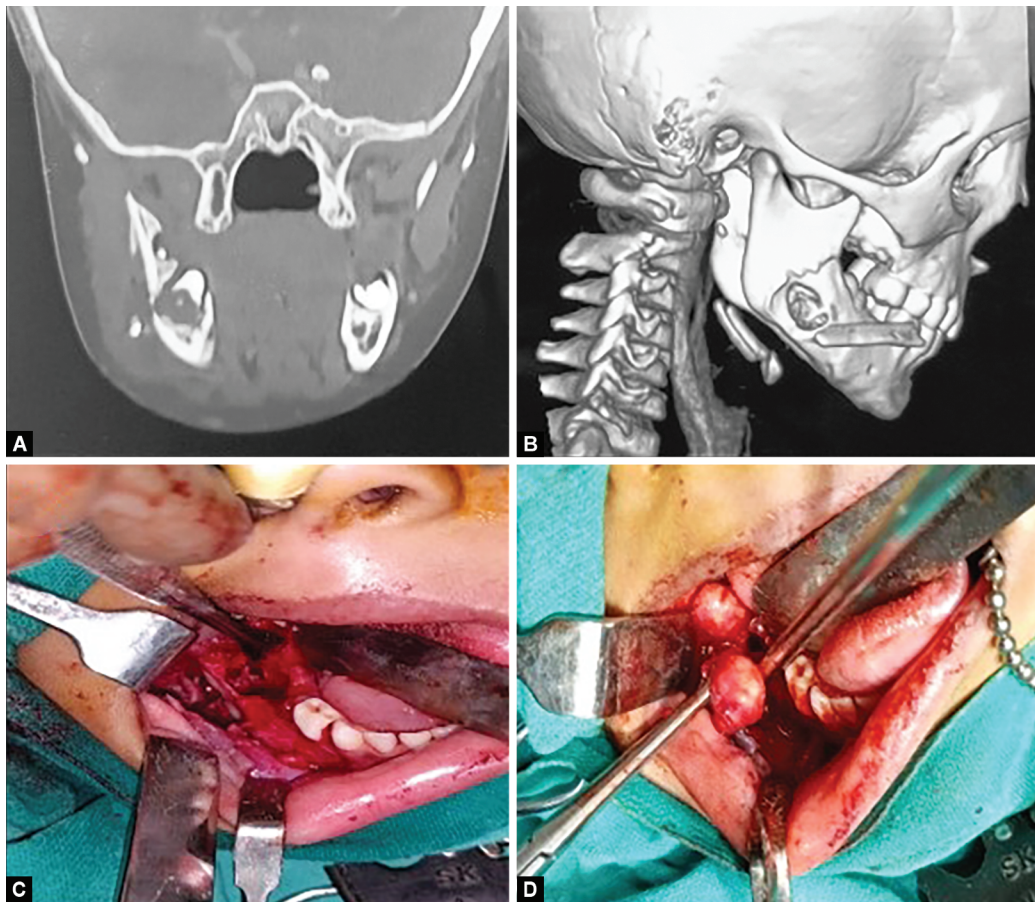
DISCUSSION

According to the National TB Prevalence Survey India 2019–2021—central TB division, the incidence of TB in India is 1.94 million in 2021 against 1.63 million cases in 2020, which accounts for a 19% rise in cases, which may be because of a pandemic.⁷ The prevalence of TB is 312 per lakh population in India, with the highest prevalence in Delhi, followed by Rajasthan and Uttar Pradesh. India alone accounts for one-third of the global blunder of TB.⁸ According to WHO, every year, about 3 million people die because of TB, and it ranks among the top 10 causes of death.⁹

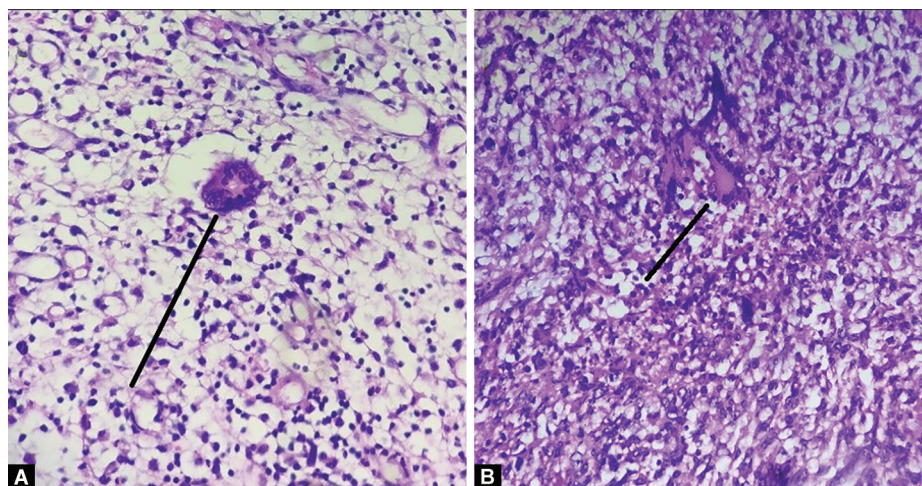
Primary TB normally affects the lungs, intestine, lymph nodes, skeletal structure, etc. Involvement of the oral cavity as a primary lesion of TB is also reported in the literature, but it is rare. The most common oral structures affected are usually the tongue, lips, cheeks, and gums. The patients usually present with an ulcer of the affected area.^{2–4} However, the involvement of the mandible as the primary lesion is a very rare occurrence. The primary lesion of TB in the oral cavity usually affects younger age-groups, whereas the secondary lesion affects older age-groups.⁵

Multiple pathways have been explained by which TB can affect the oral cavity. It can affect the mandible because of direct exposure to the infected sputum or ingestion of infected raw milk through the open pulp of a carious tooth or postextraction socket or mucosal wound of an injury or erupting tooth.¹⁰ In our patient, there is the presence of a carious primary molar, which may be the inlet for tubercular bacilli. The exact reason for infection is difficult to pinpoint.

The lesion of TB in the oral cavity usually presents with swelling, trismus, and pain, which may be associated with weight loss, episodes of fever, and discharge, which are usually the same as the presentation of the odontogenic infection.^{11,12} Our patient presented pain along with fluctuant swelling of the buccal region, which mimicked odontogenic buccal space infection. Odontogenic space infection was suspected because it is more common, and the



Figs 3A to D: (A) CECT face shows destruction of buccal and lingual cortical plate of mandible; (B) CECT face with 3D RECON shows destruction of the right angle of mandible with corrugated rubber drain *in situ*; (C) Surgical finding shows similar destruction of the mandible at the right angle of mandible region; (D) Shows enlarged lymph node in the destroyed region of angle of the mandible



Figs 4A and B: Histopathological examination of the debride tissue shows: (A) Langhan's giant cell with central area of caseating necrosis at 40x; (B) Multiple Langhan's giant cell with central caseating necrosis at 10x

patient did not give any relevant history, which can lead to TB. In the author's experience, complete bone loss in the destroyed structure with no sequestrum and slimy granulation tissue has always given a positive result of TB. Hence, CB-NAAT was done, which diagnosed TB, and the correct treatment was started accordingly.

The diagnosis of primary TB of the mandible was made after the patient was evaluated systemically to rule out TB in other more commonly affected regions.

After the diagnosis was established, the patient was sent to the department of pediatrics and pulmonary medicine, following



Fig. 5: Patient on follow-up after 1 month of surgery shows reduced swelling on the right lower side of the face

which ATT was started (rifampicin, isoniazid, pyrazinamide, and ethambutol).

CONCLUSION

This case highlights the importance of keeping TB in the differential diagnosis of oral infections. Though the patient presented with the signs and symptoms of odontogenic space infection, appropriate treatment could not have been done if the patient had not been evaluated by CECT. Hence, we feel that if the facility is available, advanced diagnostic tools should be used. This case also highlights the importance of surgical intervention, as the sole reason for suspecting TB was the surgical finding of complete destruction of bone and slimy granulation tissue during the operation. CB-NAAT test is highly sensitive for TB and should be applied as it is quick and reliable. In our patient, we did a CB-NAAT test, which confirmed our suspicion, and the treatment was started accordingly. Further, this case also highlights the importance of early treatment of infected teeth so that these complications can be avoided and the child can lead a healthier life.

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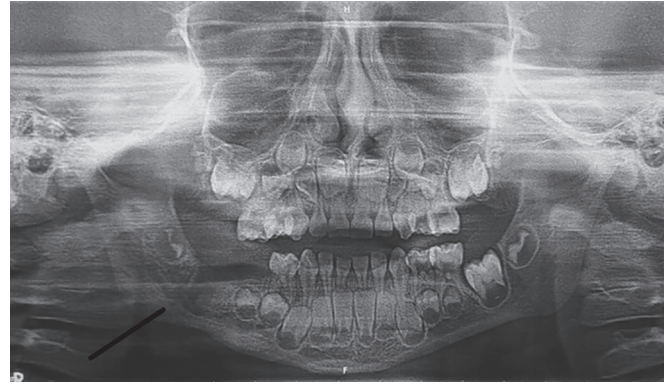


Fig. 6: Follow-up OPG shows bone formation after 1 month of surgery in the region of angle of mandible and extraction region of right first permanent molar and right primary second molar

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