Tuberculosis of the Cheek: A Rare Presentation

Namdev R*/ Jain M**/ Jindal A***/ Bodh M ****

Tuberculosis (TB) typically attacks the lungs. The oral lesions either primary or secondary are rarely seen and often overlooked by the clinician. More so, their atypical presentations make the diagnosis challenging; especially when they are present before the systemic symptoms become apparent. We report a case of primary tuberculosis in a 4 year old female child in a very uncommon location, the cheek. The timely diagnosis and antitubercular therapy resulted in complete resolution of the swelling within 6 months.

Key Words: Mycobacterium Tuberculosis, Swelling of cheek, Histopathological examination, Antitubercular Therapy

INTRODUCTION

Pediatric population presents with facial swelling or facial mass as a common clinical problem. The causes of such swellings vary from congenital to acquired infections. Most important factors to evaluate them are clinical history and their physical manifestations. Swelling combined with lymphadenitis or painless enlarged lesions of the neck can indicate an infection due to *Mycobacterium tuberculosis.*

M. tuberculosis is an acid fast bacillus and transmitted mainly via the respiratory route by inhalation of infected airborne droplets causing a chronic granulomatous disease in human beings called tuberculosis (TB). The disease primarily affects the lungs and can involve meninges, intestine, joints, bones, skin, lymph nodes and other tissues of body.

Oral tuberculous lesions range from 0.05-5% of total TB cases suggesting its oral manifestation to be rare[1]. Primary TB of the oral cavity and its non-manifestation elsewhere in the body is even rarer. These lesions mainly present with an ulceration of the mucosa but may also manifest in other forms such as nodules, peri-apical granulomas and tuberculomas[3]. Tongue is the most commonly involved site followed by soft palate, uvula, gingiva, lips, and salivary glands in the descending order[2]. In the past, studies have reported extra-oral involvement of cheek with formation of sinus[4], fistula[5] or ulceration of the cheek[6]. An adult case of tubercular infection manifesting as a nodular swelling of the cheek has also been reported[7]. To the best of our knowledge, we report the first paediatric case of primary tuberculosis presenting as a swelling of the cheek.

From the Department of Pedodontics & Preventive Dentistry, Post Graduate Institute of Dental Sciences, Rohtak.
*Ritu Namdev, Associate Professor
** Mitali Jain, PG Student
*** Ayushi Jindal, PG student
**** Meenakshi Bodh, PG Student

Send all correspondence to:

Ritu Namdev
Post Graduate Institute Of Dental Sciences
Department of Pedodontics and Preventive Dentistry,
Rohtak , Haryana , India
Phone : 09996005867
E-mail: ritunamdev@rediffmail.com

Case report

A 4 year old female child presented with the chief complaint of a gradually increasing painless swelling of the right cheek and neck region since 3 months (Fig 1). Her past medical and dental history was non-contributory and had normal milestones. Patient's parent denied any history of fever, cough, night sweats, loss of weight, loss of appetite or any discharge from swelling. Patient had been taking antibiotics prescribed by local practitioner since 2 weeks but no gross reduction in the swelling was observed. On clinical examination, a firm, non tender, non fluctuating swelling (7 cm x 3 cm) over right cheek region extending below the inferior border of mandible was observed. There was no intraoral focus of infection. On bilateral palpation, multiple firm, enlarged, and tender lymph nodes were present in the submandibular and cervical region.

Sonographic examination revealed multiple enlarged submandibular lymph nodes (up to 1.5 cm in diameter) but no evidence of any suspicious mass. Blood investigations were normal. Panoramic radiograph (Fig 2) and lateral oblique view of skull (Fig 3) showed no bony pathology. Fine-needle aspiration cytology (FNAC) of the swelling revealed epitheloid cell granulomas and giant cells suggestive of a granulomatous lesion. Multiple rods like acid-fast bacilli (AFB) were seen on the stained section (using Zeihl-Neelson stain) which confirmed mycobacterial infection (Fig 4). An incisional biopsy specimen from lower right buccal vestibular region revealed a granulomatous lesion and focal areas of necrosis with abundant neutrophils and plasma cells (Fig 5a, 5b). Large multinucleated giant cells of langerhan type were also seen. These reports were confirmatory of tuberculosis. Patient was subjected to rule out pulmonary tuberculosis after this unusual diagnosis of the oral

tissue. The sputum test for AFB was negative. The tuberculin test was also negative. The patient was started on Category I treatment under Revised National TB Control Program as directly observed therapy comprising of:

For First 2 Months (Intensive Phase) (2HRZE): Tab Isoniazid 5mg/kg/day(H), Tab Rifampicin 10mg/kg/day(R), Tab Pyrazinamide 25-30 mg/kg/day(Z), Tab Ethambutol 20mg/kg/day(E).

For Next 4 Months (Continuation Phase) (4HR): Tab Isoniazid 5mg/kg/day (H), Tab Rifampicin 10mg/kg/day(R).

The swelling gradually regressed in size and resolved completely in six months with no residual infection or recurrence during one year follow-up. (Fig 6).

DISCUSSION

Tuberculosis annually infects 8 million people globally and results in death of 3 million people due to its complications[8]. India alone bears the load of nearly one-fifth of its global burden[9]. The incidence is increasing in underdeveloped countries due to poor hygiene conditions and greater prevalence of HIV infection and AIDS. .

Clinically, tuberculosis is classified into pulmonary and extra-pulmonary. Pulmonary tuberculosis remains the most common form of the disease. Extra-pulmonary involvement accounts for approximately 10% to 15% of all the patients making it an uncommon form[10]. This occurs more commonly in immunosuppressed persons and young children. Extra-pulmonary infection sites include the pleura in tuberculous pleurisy, central nervous system in meningitis, lymphatic system in scrofula of neck, genitourinary system in urogenital tuberculosis and bones and joints in Pott's disease of the spine. An especially serious form is disseminated TB, more commonly known as miliary tuberculosis.

Head and neck involvement can include cervical lymph nodes, middle ear, nasal cavity, larynx, nasopharynx, oral cavity and parotid gland. In some cases, an oral lesion may be the initial manifestation of TB[11]. Tubercular lesions usually manifest as superficial or deep, irregular ulcers and are typically described as ulcerations and/or exophytic growth. These ulcers tend to slowly increase in size and are very painful[6, 12]. The non ulcerative tubercular lesions resembling non specific chronic infections are uncommonly seen.

Tuberculosis of the oral cavity occurs infrequently which can be because of an intact squamous epithelium of the oral mucosa making the penetration of tubercle bacilli difficult, thus providing protection against the infection[13]. The cleansing action of saliva, presence of tissue antibodies, salivary enzymes, oral saprophytes and the thick epithelial covering are the other significant protecting factors. Mycobacterium gains entry into the oral cavity due to a breach of this natural barrier which may be due to trauma, extraction of tooth, inflammatory conditions or poor oral hygiene.

Primary tuberculosis of the cheek is an unusual occurrence, and thus the differential diagnosis should include sarcoidosis and diseases of the parotid gland, such as suppurative parotitis, mumps, Sjogren's syndrome, and sialosis[14].

Conventional diagnostic procedures like microscopic examination, radiology (commonly chest X-rays), tuberculin skin test, microbiological culture of body fluids like sputum play a significant role in the initial diagnosis of the disease. Special stains such as Ziehl-Neelsen or other acid-fast stains successfully demonstrate the AFB in only 27% - 60% cases. This may be due to its relative scarcity within the tissue. Identification of the AFB in tissues is even more difficult in extra-pulmonary lesions; more so, in skin biopsies as bacillary load is relatively low. Mycobacterial culture can be done using solid or liquid media and can detect as few as 10 to 100 bacteria/ ml of specimen[15]. It is more sensitive than microscopic examination. FNAC examination is a minimally invasive simple technique that can be performed bedside and is well tolerated by children. However, these traditional methods are time-consuming, cumbersome and high concentrations of bacteria must be present in the sample to be detected.

Over the last few years, new molecular methods have been introduced as mycobacterial diagnostic tools, including PCR- Restriction Fragment Polymorphism, real time PCR, spoligotyping, DNA sequencing, DNA strip assays leading to a considerable improvement of both speed and accuracy of identification. PCR plays a valuable role in offering both rapid species identification and detection of drug resistance patterns[16]. These systems not only permit the diagnosis of TB in a comparatively shorter span of time but with high sensitivity and specificity approaching that of culture. These tests are useful in confirmation of both pulmonary and extrapulmonary TB. But due to cost restraints, their usage in low-income countries remains limited. So, it is better to utilize a combination of all the available tests for the diagnosis that can provide maximum useful information to the clinicians.

CONCLUSION

Primary or secondary oral lesions of tuberculosis are often ignored by the clinician because of its non specific presentation making it difficult to diagnose. This leads to delayed treatment and may result in devastating consequences. Thus, it is important for the clinician to exclude tuberculosis from the list of differential diagnosis of all suspicious ulcerative and granulomatous lesions of oral cavity.

Figure 1: Pre-treatment photographs (Lateral and front view of patient).

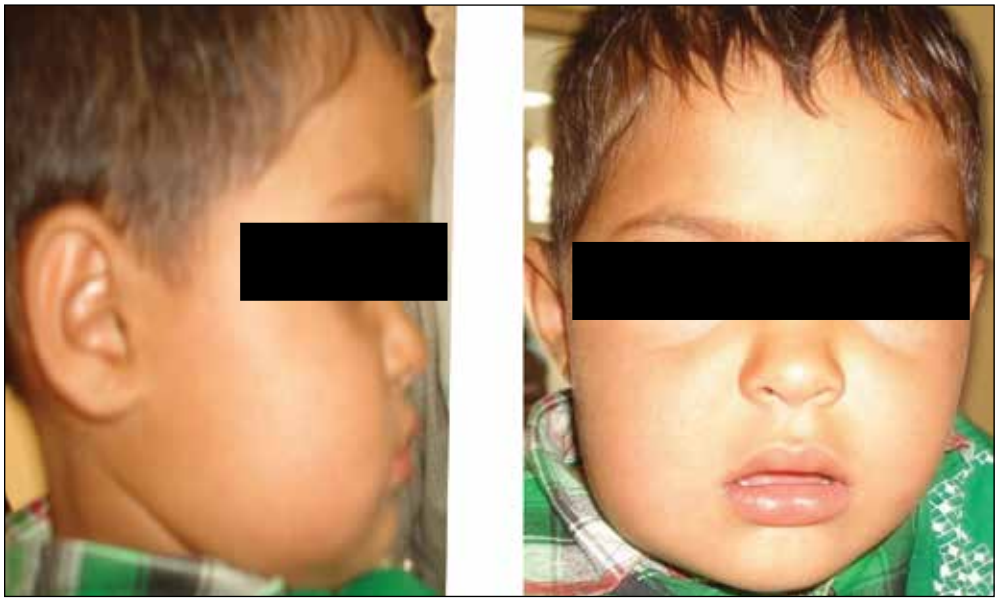

Figure 2:Pre treatment OPG.

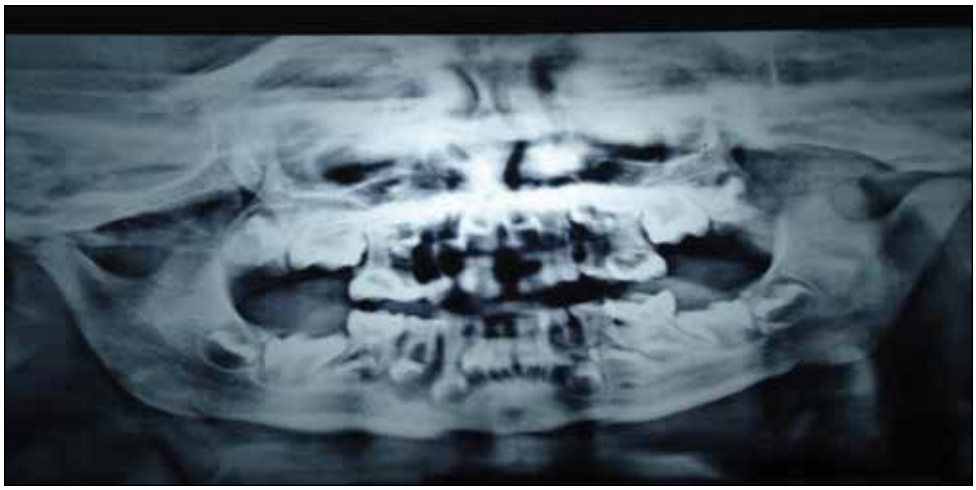

Figure 3: Pretreatment Lateral oblique view of skull

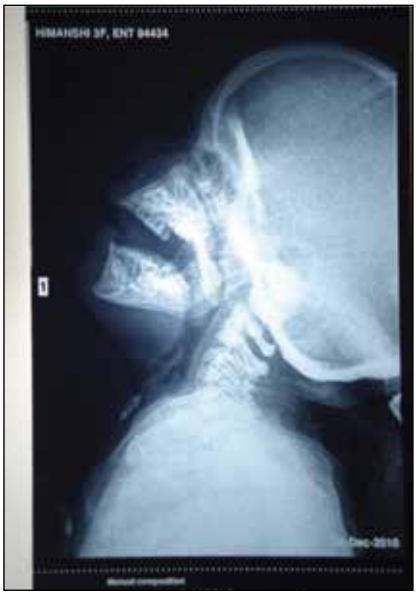

Figure 4: Ziehl-Neelsen stained section revealing acid-fast bacilli as red stained rod like structures.

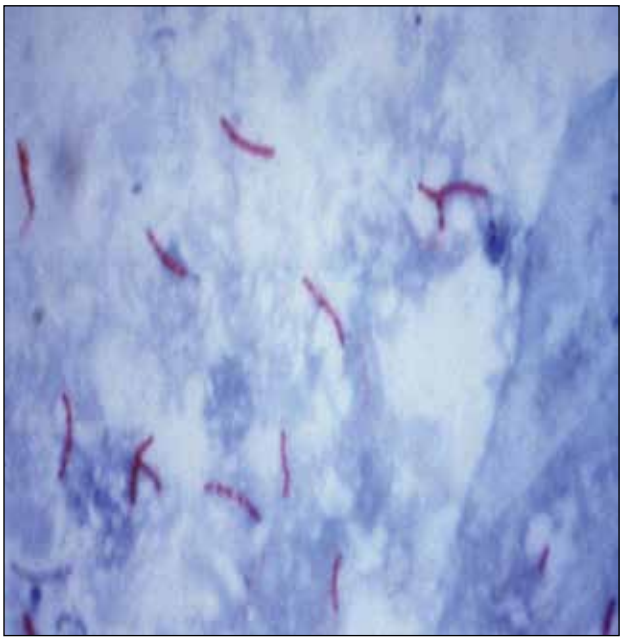

Figure 5 (a): Photomicrograph revealing area of granuloma formation containing chronic inflammatory infiltrate with focal areas of necrosis. (H & E, 4X)

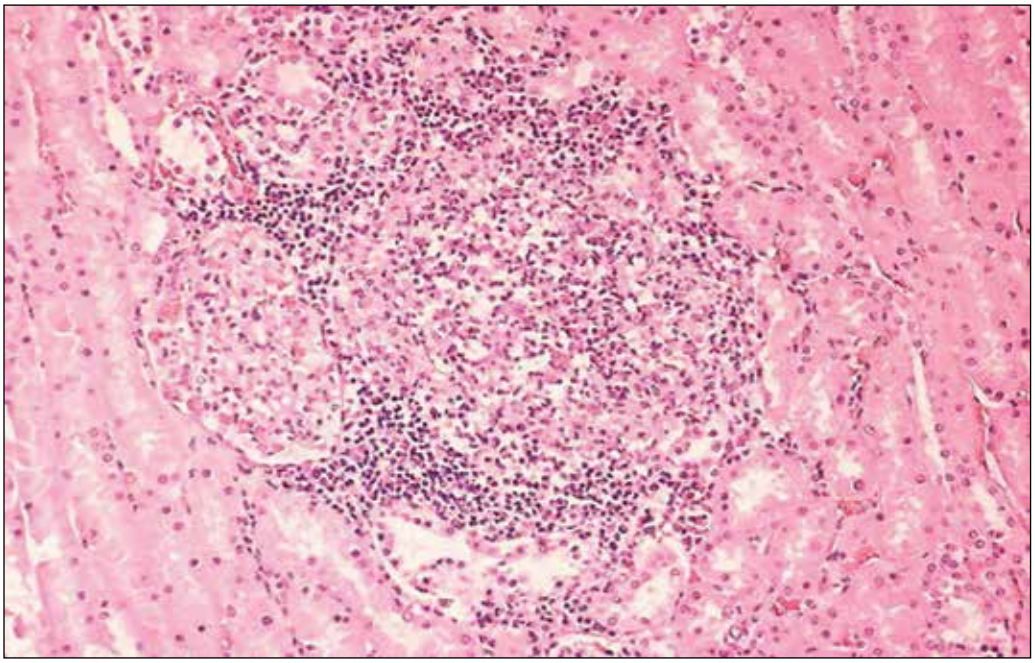

Figure 5 (b): Photomicrograph revealing langhans type giant cells along with epitheloid cells. (H & E, 10X)

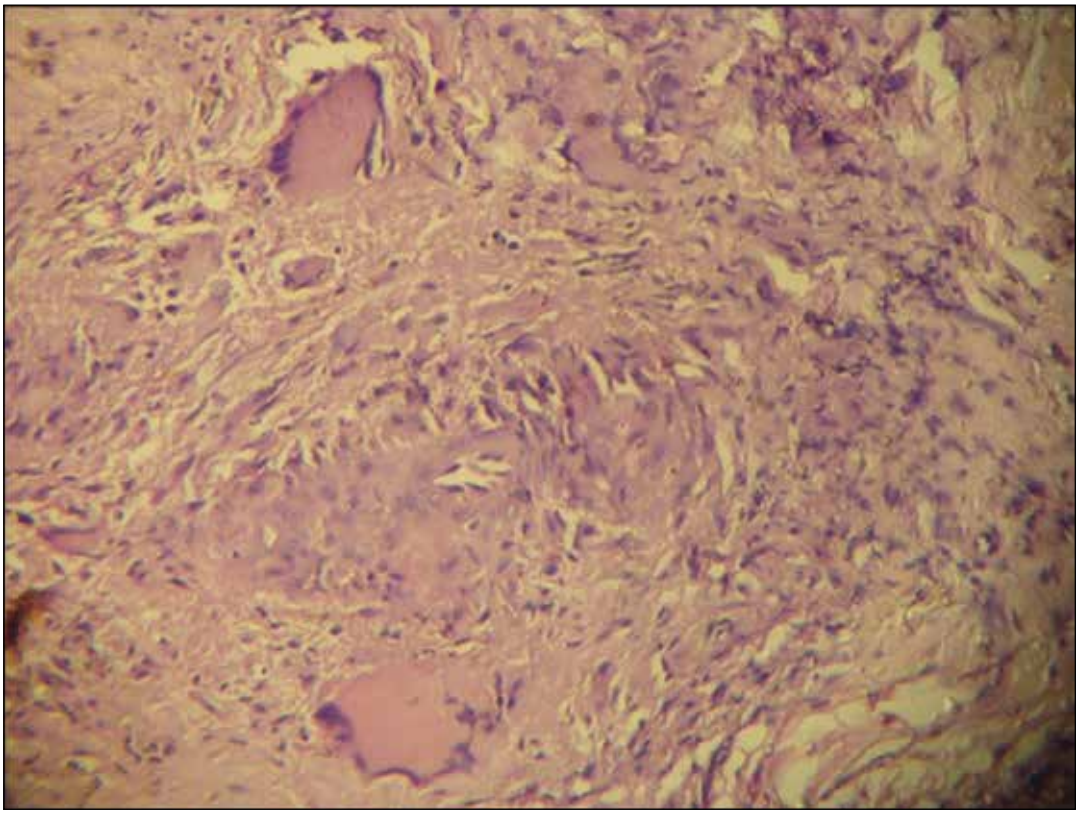

Figure 6: Pre and Post Treatment Photographs

REFERENCES

1. Sezer B, Zeytinoglu M, Tuncay U, Unal T. Oral mucosal ulceration: a manifestation of previously undiagnosed pulmonary tuberculosis. J Am Dent Assoc. ; 135(3):336–40. 2004.
2. Shafer WG, Hine MK, Levy MB. 4th ed. Philadelphia (US): WB Saunders Co; 1983. A textbook of oral pathology; pp. 340–440.
3. Carnelio S, Rodrigues G. Primary lingual tuberculosis: a case report with review of literature. J Oral Sci. ; 44(1):55–7. 2002.
4. Gillbe GV. A Tuberculous sinus of the cheek. Br J Oral Surg.;5(3):203-5. 1968.
5. Purohit SD, Mathur BB, Gupta PR, Agarwal KC, Hathi HH. Tuberculous fistula of the cheek. Report of a case. Oral surg Oral Med Oral Path. 1985;60(1):41-2
6. Hathiram BT,Grewal DS,Irani DK,et al:Tuberculoma of the cheek:A case report.J Laryngol Otol; 111:872.1997.
7. Chaudhary N, Gupta DK,, Chauddhary SR, Dawson L. Primary tuberculosis of the cheek: Acommon disease with rare presentation. Malays J Med Sci. Jan;21(1): 66-8 .2014.
8. Yepes JF, Sullivan J, Pinto A. Tuberculosis: medical management update. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. ; 98(3):267–73. 2004.
9. WHO. Tuberculosis-the global burden. Trends in TB incidence in 9 regions of the world 2005.
10. Umadevi M, Ranganathan K, Saraswathi TR, Uma R, Elizabeth J. Primary Tuberculous Osteomyelitis of the Mandible. Asian J Oral Maxillofac Surg. ; 15:208–13. 2003.
11. Brown RS, Farquharson AA, Nasseri S: Bilateral bone loss of the maxilla. Oral Surg Oral Med Oral Pathol Oral Radiol Endod; 96:6.2003.
12. Memon GA,Khushk IA:Primary tuberculosis of tongue.J Coll Phys Surg Pak; 13:604.2003.
13. Pekiner FN, Erseven G, Borahan MO, Gumru B. Natural barrier in primary tuberculosis inoculation: oral mucous membrane. Int J Tuberc Lung Dis. ; 10(12):1418. 2006.
14. Seifert G:Tumour – like lesions of the salivary glands : The new WHO classification.Pathol Res Pract;188:836.1992.
15. Yeager H Jr, Lacy J, Smith LR et al. Quantitative studies of mycobacterial populations in sputum and saliva. Am Rev Respir Dis; 95:998-1004. 1967.
16. Marais BJ, Pai M. Recent advances in the diagnosis of childhood tuberculosis. Arch Dis Child; 92:446-52. 2007.