CASE REPORT

Parotid tuberculosis in a young child causing moth-eaten mandibular osteomyelitis: an elusive diagnosis

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ABSTRACT

Tuberculosis can affect almost any organ in the body and have unusual presentations. We hereby report a case of parotid tuberculosis causing osteomyelitis of the mandible in a 3-year-old child presenting with left-sided facial swelling since 4 months. There was a history of repeated incision and drainage for suspected parotid abscess elsewhere. Initial work-up was inconclusive. Radiology revealed a heteroechoic mass lesion of the left parotid gland with extensive destruction of the adjacent ascending ramus of the mandible suggesting an odontogenic tumor. Biopsy of the left parotid gland finally clinched the diagnosis of a tubercular abscess. The child responded remarkably well to antitubercular therapy. This case highlights the importance of complete work-up, including biopsy to avoid unwarranted surgical interventions.

KEYWORDS

Tuberculosis; Salivary gland; Extrapulmonary; Child.

INTRODUCTION

Tuberculosis is endemic in India and most developing countries. Both pulmonary and extrapulmonary involvement have been extensively described in literature. Amongst the varied presentations, of particular interest to the ENT specialist is the tuberculosis of the head and neck region. Cervical lymph nodes bear the brunt of extrapulmonary disease (95%) followed by the larynx, cervical spine, oropharynx and ear [1].

Mycobacterial infection of the salivary glands, especially the parotid gland, has been described as a rarity even in endemic countries. Less than 200 cases have been reported in literature [2]. Atypical presentations of parotid tuberculosis have been described. We present a rare case report of a young child with parotid tuberculosis mimicking odontogenic tumor.

CASE REPORT

A three-year-old male child presented to our Outpatient Department with history of a swelling

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in the left parotid region since 4 months, preceded 15–20 days earlier by blunt trauma to the cheek. He had an evening rise of temperature in the initial week of onset only. Elsewhere with a suspicion of parotid abscess, he underwent incision and drainage twice. Despite multiple antibiotic courses, he did not improve. There was no history of contact with tuberculosis, loss of weight or appetite. Physical examination revealed a firm, non tender swelling 7 × 6 cm in size in the left parotid region with a well healed transverse scar mark in the overlying skin (Figure 1A). There was no palpable cervical lymphadenopathy or facial nerve involvement. The rest of the ENT examination was non-contributory.

Initial diagnostic work up revealed hypochromic microcytic (iron deficiency) anemia, normal white cell count, raised erythrocyte sedimentation rate (55 mm in first hour), Mantoux test reading of 12 mm induration (48 hours), and nothing definitive on fine needle aspiration cytology (FNAC). An ultrasound guided FNAC was performed. Chest X-ray was normal. A contrast enhanced computed tomography (CT) scan of neck suggested a heteroechoic mass lesion involving superficial and deep lobes of the left parotid gland with extensive destruction of the adjacent ascending ramus of the mandible (Figure 2A).

Keeping in mind the possibility of an odontogenic tumor based on the CT findings, a biopsy was taken without waiting for the results of the ultrasound guided FNAC. The guided FNAC (Figure 2B) meanwhile, showed necrotizing granulomatous lesion with superadded suppuration consistent with tubercular abscess [Ziehl Neelsen stain positive for acid fast bacilli (AFB)]. Histopathology revealed a necrotising granulomatous lymphadenitis. AFB were grown in mycobacterial culture with MPT-64 Ag positivity denoting *Mycobacterium tuberculosis* complex. The child was put on Category I antitubercular therapy (ATT) and responded remarkably well to treatment (Figure 1B).

DISCUSSION

In 2015, there were an estimated 10.4 million incident TB cases worldwide, 1.0 million (10%) being children. India is one of the six countries accounting for 60% of new cases [3].

Figure 1. Clinical image (A) at the time of presentation, (B) after completion of antitubercular treatment.

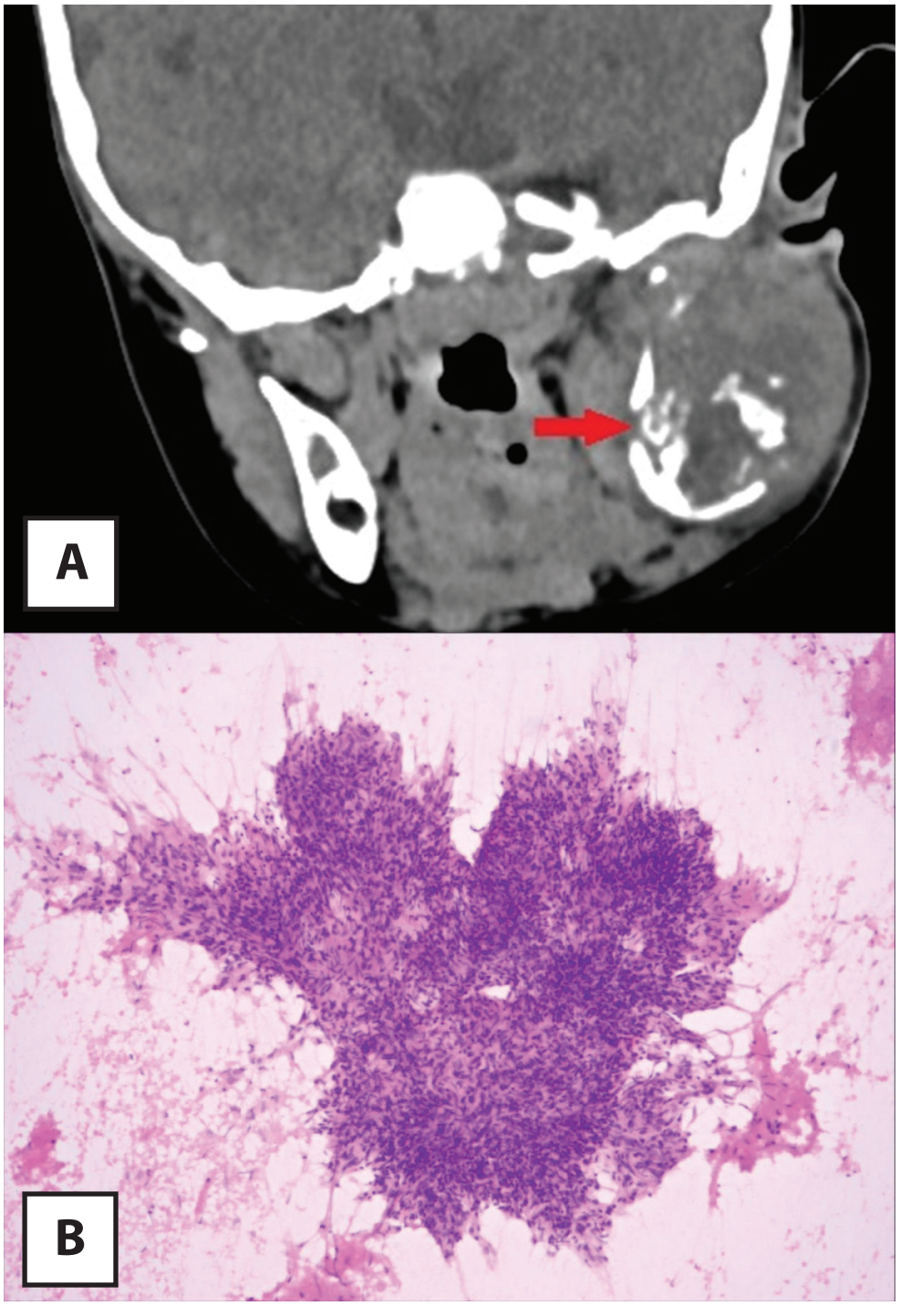

Figure 2. (A) Extensive destruction of ramus of left mandible (red arrow) caused by heteroechoic mass involving superficial and deep lobes of the parotid gland as seen on contrast enhanced computed tomography (CT) scan of neck (coronal image). (B) Histopathological image (10× H&E) showing epithelioid granuloma.

Tuberculosis is a necrotising granulomatous infection, which can affect multiple organ systems, caused by *M. tuberculosis* complex. About 15%–20% of cases have extrapulmonary involvement [4]. Salivary gland tuberculosis per se is rare. This may be due to the antibacterial

action of proteolytic enzymes in saliva along with the inhibitory effect of continuous salivary flow on stagnation and growth of mycobacteria [5]. For cases affecting the salivary glands, 70% involve the parotid glands, 27% the submandibular and 3% the sublingual glands [4]. Older children and adults are most commonly affected. The primary focus is usually in the tonsils, teeth or gingivobuccal sulcus, which ascends through the salivary ducts to involve the respective glands [6]. Other postulated mechanisms include hematogenous spread from a distant focus or ascending infection from an infected cervical lymph node.

Irrespective of the mode of infection, presentation of parotid tuberculosis is quite varied. It may present as an acute parotitis, recurring abscess or a slow growing mass mimicking a parotid neoplasm (pleomorphic adenoma), invoking a parotidectomy or even parotid fistula [7]. Tuberculosis may affect the mandible creating a picture of chronic osteomyelitis, with greater affinity for the alveolus and angle [8]. Thus, diagnosis of parotid tuberculosis in the absence of a primary lung focus can be befuddling.

Important differential diagnoses of a parotid swelling in a child which should be considered include:

1. Viral parotitis (mumps)—It has an acute onset and is usually bilateral in presentation and maybe difficult to differentiate from tuberculosis clinically. One side maybe involved 24–48 hours prior to the other side. However, the disease process being primarily inflammatory does not cause bony destruction as seen in our patient.

2. Sialadenitis—In the acute form, it causes a sudden, painful enlargement of the salivary gland which is aggravated following meals. Local tenderness, warmth, erythema and purulent discharge from the duct may accompany the swelling. Such cases are bacterial or viral in origin. Chronic recurrent parotitis is amongst the most frequent sialadenitis in infancy.

3. Salivary gland tumours —Though uncommon in the pediatric age-group, both benign and malignant tumours may involve the parotid gland. These include pleomorphic adenoma, hemangioma, lymphangioma, muco-epidermoid carcinoma, adenoid cystic carcinoma and embryonic rhabdomyosarcoma, and may present in a similar fashion.

4. Odontogenic tumours and cysts—Ameloblastoma, fibromyxoma, keratocystic odontogenic tumour, adenomatoid odontogenic tumour and odontogenic fibroma may present as cystic lesions around the mandible in a child resembling the clinical picture of our case.

5. Granulomatous diseases—Sarcoidosis presents as a bilateral painless swelling. Histopathology helps to differentiate it from tuberculosis by the presence of non-caseating epithelioid granulomas. Other diagnostic investigations include chest X-ray, high resolution computed tomography scans of thorax and serum angiotensin converting enzyme.

A primary work-up for a parotid swelling in a child includes imaging- ultrasonography followed by CT scan of the neck (when indicated) and FNAC from the lesion. Magnetic resonance imaging of the neck may be of more benefit in case of multiple site involvement in the periparotid region. However, nonspecific imaging features and inconclusive FNAC reports are not uncommon. A high index of suspicion is required to arrive at the diagnosis. In our patient, the age, absence of history of contact with tuberculosis, lack of constitutional symptoms and absence of lung lesions were misleading. The imaging was suggestive of mandibular osteomyelitis raising the more likely possibility of an odontogenic tumor. However, histology clinched the diagnosis.

Other investigations which a patient with suspected extra-pulmonary tuberculosis should undergo include a chest X-ray, sputum examination for AFB, TB-PCR (polymerase chain reaction) of tissue, Mantoux test, ESR, GeneXpert MTB assay and culture studies for *M. tuberculosis* depending upon availability of the tests and financial constraints. Specific investigations to rule out the other possible diagnoses, as listed above, should be carried out on a case-to-case basis.

In 2016, Reddy et al. [8] reported a similar case of a 25-year-old female with a swelling

in the pre-auricular region and submandibular lymphadenopathy. A panoramic radiograph revealed a single, well-defined, 2 × 2 cm, non-corticated radiolucency in the middle third of the ramus. A diagnosis of extrapulmonary tuberculosis was made after complete work up. She responded well to ATT. In an important meta-analysis by Lee and Liu [9], 49 patients with tuberculous parotitis were enrolled. Forty-eight were unilateral cases, more than half presented with a painless parotid mass, and four had draining sinuses. Interestingly, 25% had pulmonary tuberculosis.

Parotid tuberculosis is essentially a histologic diagnosis. In parotid lesions FNAC has a sensitivity of 81%–100% and specificity of 94%–100% [10]. A meta-analysis of 64 parotid lesion FNAC studies showed a sensitivity of 80%, specificity of 97% and positive predictive value of 90% for differentiating benign from malignant parotid lesions [7]. On the other hand, Lee and Liu [9] found that FNAC contributed to diagnosis in only 10 cases, while 34 cases were labelled as tuberculosis only after parotidectomy and histopathology.

To conclude, tuberculosis often presents a clinical and histopathologic surprise. Parotid tuberculosis being rare is often misdiagnosed. Often, such patients are subjected to superficial parotidectomies in the likelihood of a parotid neoplasm. A high index of clinical suspicion is required in order to avoid unwarranted surgery with possible cosmetic morbidity while treating a medical condition.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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None.

ETHICAL APPROVAL

Ethics clearance and approval was taken from the Institutional Ethics' Committee. Duly signed informed consent was taken from the parents of the child in question to include personal case details and clinical images of the child. Confidentiality was ensured at all stages.

REFERENCES

1. Nalini B, Vinayak S. Tuberculosis in ear, nose and throat practice: its presentation and diagnosis. Am J Otolaryngol. 2006;27:39–45.
2. Verma RK, Kaushal D, Bal A, Panda NK. Primary parotid tuberculosis mimicking parotid neoplasm: a rare case report. SEAJCRR. 2013;2:455–62.
3. World Health Organization. Global tuberculosis report. Geneva, Switzerland: WHO [cited 2017 Oct 8]. Available from www.who.int/tb/data
4. Sharma SK, Mohan A. Extrapulmonary tuberculosis. Indian J Med Res. 2004;120:316–53.
5. Guyton AC. Textbook of medical physiology. 11th ed. Philadelphia: Saunders; 1996.
6. Som PM, Brandwein MS. Salivary Glands. In: Som PM, Curtin HD, editors. Head and neck imaging. 5th ed. St. Louis, MO: Mosby Elsevier; 1996. p 2492.
7. Jackson NM, Mitchell JL, Walvekar RR. Inflammatory disorders of the salivary glands. In: Flint PW, Haughey BH, Lund VJ, editors. Cummings Otolaryngology, Head and Neck Surgery. 6th ed. Philadelphia: Saunders Elsevier; 2015. p 1232.
8. Reddy V, Chaudhary A, Wanjari PV, Warhekar AM, Lalawat S, Verma M. Extrapulmonary tuberculosis involving parotid and ramus region: a forgotten diagnosis. J Appl Dental Med Sci. 2016;2:1.
9. Lee I, Liu J. Tuberculous parotitis: case report and literature review. Ann Otol Rhinol Laryngol. 2005;114:547–51.
10. Iseri M, Aydýner O, Celik I, Peker O. Tuberculosis of the parotid gland. J Laryngol Otol. 2005;119:311–13.