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Case Report

CLINICAL CASE STUDY OF TUBERCULOSIS IN PAROTID GLAND FOUND IN CHILDREN AT A MEDICAL SCHOOL IN SOUTH EAST ASIA

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ABSTRACT

Objective: Tuberculous parotitis in children is a rare disease and its pre-operative diagnosis remains difficult because of the low incidence and nonspecific presentation of swellings, mimicking benign and malignant neoplasm of the parotid gland.

Methods: Patients presenting in the pediatric ward and outdoor patient department were screened and diagnosed on a day to day basis. Diagnoses of the disease in three children were made by strong clinical suspicion on morphological observations of developed swellings and were confirmed by ancillary diagnostic measures, ultrasonography (USG), histology, and Ziehl–Neelsen staining of aspirates of lesions obtained with USG-guided fine needle aspiration cytology.

Results: Rare cases of parotid gland tuberculosis were detected and they were properly investigated, photographed after consent and treatment given to them were documented. Tubercular infections of parotid glands in three children are described that were addressed by anti-tubercular treatment.

Conclusion: Etiology confirmed nonspecific presentations of swellings as tuberculous parotitis in three children.

Keywords: Tuberculous parotitis, Ultrasonography, Fine-needle aspiration cytology.

INTRODUCTION

Mycobacterium enjoy a ubiquitous distribution including deserts where, those are found under rocks and dried roots of vegetation even, but are not isolated from polar regions. They are widely distributed in soil-edges nearing fresh water bodies, and a few particularly the notorious *Mycobacterium tuberculosis,* alternatively called, tubercle bacilli (TB) has become obligate parasites with man, while *Mycobacterium bovis* infects animals. *M. tuberculosis* is almost intractable in recent years, because of the emergence of multidrug-resistant (MDR) strains, and less recently a 58% of total isolates in India were reported as MDR strains [1,2]. Taking to surprise, the migration status of *M. tuberculosis,* was reported in 27 countries of Europe at the saturnine figure of 100% [3], with unknown drug sensibility patterns of European TB strains. However, a totally drug-resistant TB-strain resistant to all first and second line TB-drugs had been reported independently from Italy, Iran, and India [4,5]. Moreover, strains of mycobacteria other than tuberculosis (MOTT), *Mycobacterium avium, Mycobacterium kansasii*, *Mycobacterium Fortuitum,* and *Mycobacterium scrofulaceum* are widely distributed in developing and developed countries too [6].

As known, considerable proportions of marginalized slum-dwellers in India develop TB infection and there is utter difficulty in identification of MDR-TB strains due to the lack of facilities in the molecular diagnostic method, the "nucleic acid amplification test," widely. Thus, there is always a dependence on the classical smear test for TB-identification [7,8], which are done for pulmonary TB detection, unwittingly. Further, the widespread of human immunodeficiency virus also promotes the shockingly repellent infection of TB strains by crippling the immune system, in addition to several obvious factors, mainly the multidrug resistance [1]. In unhygienic living conditions, with immunocompromised patients of all of ages, the slow-growing TB gets along with a sizable population [9]. MOTT also get circulated subtly and burgeon as pathogens indolently, as many animals uncompetitively serve as their reservoirs, but these species are generally regarded are non-tubercular, and at times *M. bovis* causes tuberculosis in man [10]. Less frequently in people of all age groups, MOTT causes parotitis, i.e., infection of parotid glands – The largest salivary gland. Moreover, tuberculosis of parotid gland in children is rare, even in India [11]. It manifests as a slow-growing lobulated parotid-swelling, basically indistinguishable from the similar presentation of parotid neoplasm (benign or malignant), the commonly occurring problem of parotid. Anyway, there is scarce in the literature on tuberculous parotitis in the pediatric age group. Three cases of primary parotid tuberculosis in the pediatric age group, presented herein were diagnosed pre-operatively and treated for TB with the standard chemotherapy regimen of the initial administration of four first-line drugs (isoniazid, rifampicin, pyrazinamide, and ethambutol), together followed by a 4 months stabilization phase with isoniazid and rifampicin, that landed at blithesome success.

CASE REPORTS

Case 1

A 4-year-old girl lacking the usual pizzazz was presented with a swelling (size 1.5 cm × 1 cm) of the right parotid, developed since 2 months and another swelling (size 3 cm × 2 cm) of the left parotid, developed since 2 weeks, associated with a low-grade fever and local pain. Swellings were firm and non-tender with an ill-defined border and a bosselated surface (Fig. 1a and b). The oral cavity was normal and the facial nerve was separated on both sides; there were no fistulae or sinuses. Past history was unremarkable and gave no personal or family history of tuberculosis; in addition, a chest X-ray picture was normal that ruled out any prior pulmonary tubercular infection. Hematologic/biochemical investigations revealed only a raised erythrocyte sedimentation rate (ESR), 27 mm/1st hr and the Mantoux test result after 72 hrs had been negative. Ultrasonography (USG) of the parotid region depicted multiple rounds to oval heterogeneous hypoechoic lesions, studded in parotid parenchyma with both superficial and deep lobe involvement. A few enlarged cervical lymph nodes (up to 14 mm size) and necrotic

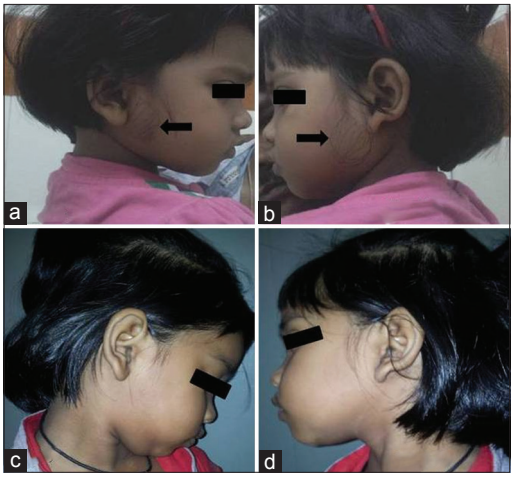

Fig. 1: (a and b) Right and left parotid gland swelling (before treatment), (c and d) resolution of parotid swelling (after treatment)

degenerations in the left intraparotid node (8-10 mm) were noted. USG-guided fine-needle aspiration cytology (FNAC) attempts on both swellings revealed epithelioid granuloma with lymphohistiocytic clusters on an amphophilic necrotic background, suggestive of tubercular pathology (Fig. 2a and b). Further, fluorescent stained bacilli were seen under a microscope with auramine-rhodamine staining that was suggestive of the presence of TB. Test for acid-fast bacilli (AFB) of gastric aspirate was negative. However, there was a marked improvement with anti-tubercular treatment as complete disappearance of swellings at the end of 6 months was noted (Fig. 1c and d), associated with the weight gain of 1.5 kg. She is in the regular follow-up and presently is asymptomatic.

Case 2

A 9-year-old girl was presented with a non-tender and firm swelling of size 5 cm × 4 cm on the left cheek, developed since 2 months. General physical examination including the oral cavity was normal and the facial nerve was not involved. A chest radiograph and blood counts revealed no abnormality, but the result of tuberculin test was positive with a reaction spot size of 11 mm. USG of the left parotid revealed a large heteroechoic lesion with an irregular outline, predominantly involving the deep parotid lobe with a central necrotic area and a few enlarged intraparotid nodes (Fig. 3a and b). USG-guided FNAC revealed clusters of benign salivary epithelial cells on a dense inflammatory background with polymorph, lymphocytes, histiocytes, and epithelioid cells. A good amount of amorphous granular necrotic debris was present in the background. AFB was seen on Ziehl–Neelsen (ZN) staining of the aspirate but, not in induced sputum. After 2 months of anti-tubercular therapy, the size of the swelling decreased to a size of 2 cm × 2 cm with the general well-being, and therapy is in continuation.

Case 3

A 4½-year female child was presented to us with left parotid swelling for 4 months with intermittent fever. She had a history of contact with tuberculosis. She was an underweight child with normal physical and oral health. On investigation, mild pallor (9.4 g/dl), high ESR (58 mm/1st hr), positive Mantoux test (25 mm) was recorded with no abnormality in X-ray picture of the chest. High-resolution USG depicted the involvement of both superficial and deep lobe of left parotid with multiple microabscesses. Few enlarged left neck nodes were seen of size 16-21 mm. FNAC revealed features suggestive of the granulomatous disorder, but AFB in ZN staining and gastric aspirate were absent. She was started with anti-tubercular therapy; the parotid swelling subsided completely with weight gain and general well-being.

Anti-tubercular therapy

All the three cases were treated with the daily regimen of isoniazid (5 mg/kg/day), rifampicin (10 mg/kg/day), pyrazinamide (25-35 mg/kg/day), and ethambutol (15-25 mg/kg/day) for the first 2 months followed by isoniazid and rifampicin for the next 4 months. All the three patients were followed up to 6 months after the anti-tubercular therapy.

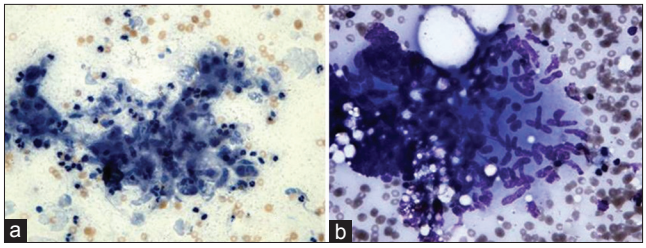

Fig. 2: (a) Degenerated ductal cells with occasional epithelioid cells against background of acute inflammatory cells. (May-Grünwald-Giemsa [MGG] stain, ×200), (b) well-formed cluster of epithelioid cells (MGG stain, ×400)

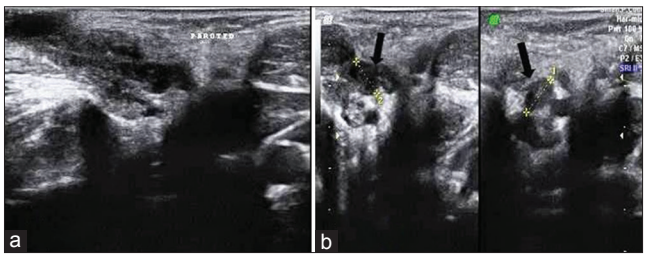

Fig. 3: (a) Irregular hypoechoic intraparotid parenchymal lesion with internal, (b) enlarged intraparotid lymph node

DISCUSSION

Parotid tuberculosis is a rare form of extra-pulmonary tuberculosis. The rarity is because of inhibiting the effect of saliva over the infecting mycobacterium [12]. However, the most common route of the disease is the direct migration of bacilli from the oral cavity via, the Stenson's duct [13]. Uncommonly, intra and periparotid lymph nodes get infected, either by the drainage of lymphatics from the oral cavity or a hematogenous spread from the pulmonary focus, when the latter is infected. Primary focus of an infection on gland should be established at the earliest for parotid tuberculosis, which is mostly misdiagnosed as parotid neoplasm [14]. The most reported cases of parotid tuberculosis were of adults and those were diagnosed by histology, after parotidectomy [15,16]. Further, tubercular involvement of the salivary glands is more commonly seen as secondary to the systemic dissemination of pulmonary tuberculosis, rather than as primary incidences of extra-pulmonary tuberculosis. When the salivary glands are primarily affected, infection of the parotid gland is reported as frequent as 70% [17].

The Mantoux test is the most widely followed method for diagnosing TB, even though it is not totally accurate. Its positive and negative predictive values are affected by a number of factors. The positive test ascertains the patient has been infected with *M. tuberculosis* either recently or in the past. The tubercular population percentage of positive test steadily increases with the age. Thus, the tuberculin reaction is regarded as more specific in younger age groups, i.e., in 0-9 years age group. Children in the highest risk category (contact with an indexed case, HIV infected, or clinical evidence of TB) are considered infected with *M. tuberculosis,* if the indurations is at least 5 mm [18] Furthermore, the effect of Bacillus Calmette–Guérin (BCG) vaccination on the subsequent Mantoux test is highly variable. In BCG vaccinated children, the reaction of tuberculin ranges from 3 to 10 mm. Hence, the presence of the size of post-vaccination tuberculin test does not reliably predict the degree of protection afforded BCG. In a study, it was demonstrated that there was 50-60% waning in the 1st year itself. However, BCG vaccination provides protection against disseminated and meningeal tuberculosis, and against death from tuberculosis [18].

Parotitis can be due to both infectious and non-infectious causes. Suppurative parotitis is caused by bacterial infections, characterized by acute pain, swelling, warmth and induration associated with cervical adenitis and leukocytosis. Viral parotitis causes parotid swelling, pain, and otalgia. Tuberculous parotitis usually is presented as unilateral parotid swelling or abscess, but it may be also involved with parotid glands of both sides [19]. The swelling usually is painless initially without surrounding inflammation, but its slow enlargement is often associated with complications such as, sinus formation and facial nerve palsy. But in a young child, it is often presented acutely with a painful swelling, consistent with a suppurative parotitis [20]. USG can differentiate tubercular infection from neoplasm – either as the lesion is intraparotid or periparotid, but it cannot differentiate accurately tuberculosis from other chronic inflammatory conditions, sarcoidosis or fungal infection [21]. However, USG-guided FNAC corroborates postoperative histologic findings and has an overall accuracy of 86-89%, as it is known [22]. Obviously, an early diagnosis and a prompt/prudent medical treatment yield a good prognosis. Herein, both patients were benefited by ancillary diagnostic procedures, USG and FNAC in the coveted evaluation of parotid swellings, avoiding the hazardous surgical procedure, and anti-tubercular chemotherapy regimen could be started in time, ending at success.

CONCLUSIONS

Radiological investigations, USG, computed tomography, and magnetic resonance imaging are used in detecting intraparotid tubercular developments, but associated findings are not specific as most of the images/findings mimic neoplasm. A chest radiograph is helpful in cases of associated pulmonary tuberculosis, as done with present cases. Particularly in parotid lesions, FNAC were reported to have a sensitivity of 81-100%, and a specificity of 94-100% [23,24]. Eventually, FNAC is the dependable diagnosis in the evaluation of a parotid mass for TB or neoplasm. Although particularly in tubercular endemic areas, children presented with painless unilateral or bilateral parotid swelling with poor response to antibiotic therapy, one of the differential diagnoses for tuberculous parotitis would be mandatory, along with the routine test parotid neoplasm, in order to avoid unnecessary surgery.

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