

Case Report

MDR Tubercular Parotid Abscess: A Rare form of Extra-Pulmonary TB

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Abstract

Mycobacterium tuberculosis causes Tuberculosis (TB) in humans. It involves the lungs primarily; tuberculous involvement of the head and neck region is extremely rare, except cervical lymph node involvement which is more common. Due to continual flow of saliva, salivary gland involvement is even rarer as it prevents tubercular bacilli from accumulating there and saliva also possesses antibacterial properties. The parotid glands are more likely to be affected than the other salivary glands because of the sluggish flow of saliva. However, even in an endemic country like India, parotid tuberculosis is a rare form of extra-pulmonary tuberculosis. Here, we discuss a case of 35 year old male presented with gradually increasing left parotid swelling associated with dull pain, difficulty in eating and facial asymmetry. There was a discharging wound present over the skin surface of the swelling. Ultrasonography of the left parotid swelling showed a hypochoic lesion with 50 cc volume, with multiple septations and debris, suggestive of left parotid abscess. Pus was evacuated through the discharging sinus. Sample was sent for Pus CBNAAT that came positive for MDR-TB. Chest X-ray was clear. Hence, diagnosis of primary MDR Tubercular parotid abscess was made. The patient was started on shorter oral bedaquiline regimen and on follow-up patient is progressing satisfactory.

Introduction

Mycobacterium tuberculosis causes Tuberculosis (TB) in humans. It involves the lungs primarily, thus pulmonary symptoms are the most common manifestation [1]. TB can involve the gastrointestinal system, musculoskeletal system, lymphoreticular system, central nervous system, and reproductive systems, in addition to the dermis and hepato-biliary system, but respiratory system is most commonly affected in TB [1-3]. Tuberculous involvement of the head and neck region is extremely rare, except cervical lymph node involvement which is more common. Due to continual flow of saliva, salivary gland involvement is even rarer as it prevents *tubercular bacilli* from accumulating there. Salivary glands are more resilient to infection as saliva also possesses antibacterial properties. The parotid glands are more likely to be affected than the other salivary glands because of the sluggish flow of saliva [4]. However, even in an endemic country like India, parotid tuberculosis is a rare form of extra-pulmonary tuberculosis [5]. Surprisingly, only about hundred cases of parotid tuberculosis has been reported in the literature. Sjogrens syndrome, malignancy, HIV, sarcoidosis, TB, and Polyarteritis nodosa should be considered as differential diagnosis for chronic parotitis. The diagnosis of parotid gland involvement with TB has traditionally been made after superficial parotidectomy [6,7]. Here, we discuss a case of unilateral parotid swelling with an underlying abscess.

Case Presentation

A 35 year old male presented with gradually increasing left parotid swelling during a period of 4 weeks, associated with dull pain, dysphagia and facial asymmetry (deviation of angle of mouth towards right). There was no history of evening rise of temperature, cough, recent weight loss, trauma, or any other systemic symptoms and no history of contact with patients diagnosed with tuberculosis. On physical examination, the patient was thin built; his weight was 48 kg and had severe pallor. On local examination, unilateral swelling of 6 cm x 5 cm was found, obliterating the left angle of jaw. It was discharging wound, soft to firm in consistency, mildly tender and discharging sinus was present over the skin surface. There was left facial nerve palsy and multiple cervical lymph nodes were palpable about 1 cm in size, mobile, and non-matted. Right angle of jaw was normal, with no lymphadenopathy found anywhere else. Routine blood tests revealed hemoglobin 6.2g/dl and total leucocyte count 4100/cumm with 61.1% neutrophils and 11.7% lymphocytes. ESR was 60 mm/hr. Ultrasonography of the left parotid swelling showed a hypochoic lesion with 50 cc volume, with multiple septations and debris, suggestive of left parotid abscess. Pus was evacuated through the discharging sinus. Sample was sent for Pus CBNAAT that came positive for MDR-TB. X-ray chest was clear. There were no pulmonary foci or other foci seen. Hence, we made the diagnosis of primary MDR tubercular parotid abscess. The patient was started on Shorter Oral Bedaquiline Regimen i.e., Bedaquiline 400 mg, Moxifloxacin 600 mg, Clofazimine 100 mg, Pyrazinamide 1500 mg, Isoniazid 400 mg and Kanamycin 750 mg daily. On follow-up patient is progressing satisfactory (Figure 1).

Discussion

The World Health Organisation (WHO) has released the global tuberculosis report 2023, highlighting the high burden of TB in 2022. India accounted for the highest number of TB cases in the world in 2022, with 2.8 million TB cases, representing 27% of the global burden [8]. Though the most common presentation of TB being pulmonary

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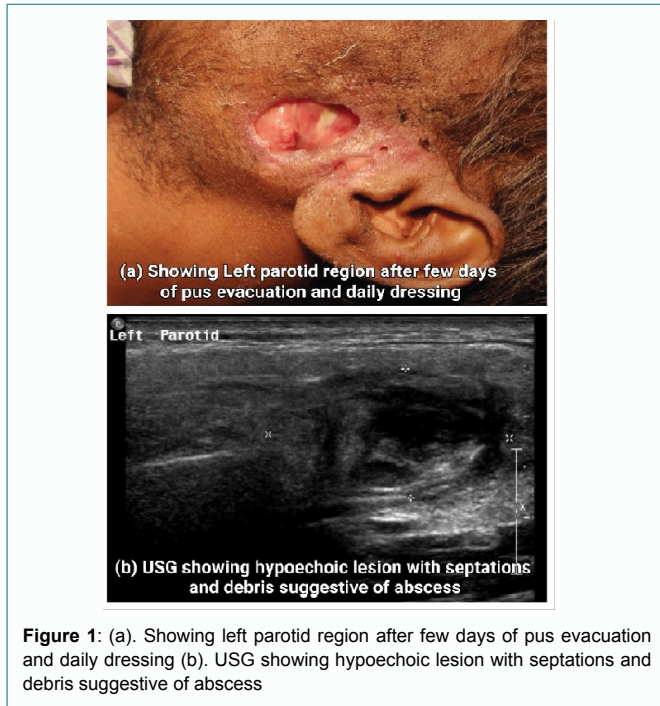


Figure 1: (a). Showing left parotid region after few days of pus evacuation and daily dressing (b). USG showing hypoechoic lesion with septations and debris suggestive of abscess

tuberculosis, salivary gland tuberculosis is very rare, even in endemic countries like India.

The salivary gland tuberculosis is rare because of the presence of thiocyanate ions and proteolytic enzymes, such as lysozyme in the saliva that has bactericidal action. Also, the continuous flow of saliva prevents the accumulation of mycobacteria within the parenchyma of the gland [9].

The pathogenesis of tuberculosis of parotid glands remains unclear and there are various postulates regarding its source. In one of the earliest reports of tuberculosis of salivary glands, Van Stubenrauch postulated that the extension of infection along Stenson's duct from the oropharynx is the important mode of infection [10]. Bockhorn proposed that there is hematogenous spread from any primary focus in body [11]. Berman and Fein postulated that infection can spread to parotid gland, particularly from infected tonsils *via* lymphatics [12]. Commonly tuberculous involvement of the parotid gland is seen secondary to systemic dissemination of pulmonary tuberculosis rather than primary extrapulmonary tuberculosis, which is less common. In primary tuberculosis of the salivary glands, the parotid gland is involved in 70% of the cases [13].

The tuberculosis of parotid gland is a rare form of extra pulmonary tuberculosis. Parotid tuberculosis presents as a unilateral swelling or abscess involving the parenchyma of the gland. It may also involve both the parotids [14]. In 1893, C De Paoli reported the first case of tuberculosis of parotid gland [15]. Since then, only about one hundred cases of parotid tuberculosis have been reported in the literature [16]. There is an unusual form in which peri-parotid and intra-parotid lymph nodes get affected, either through lymphatics from the oropharynx or hematogenous spread from a pulmonary focus. Parotid tuberculosis mostly presents as a localized progressive chronic swelling and symptoms vary from an acute infectious process to an indolent chronic presentation.

Fine Needle Aspiration Cytology (FNAC) is the most simple and reliable technique for the diagnosis of parotid tuberculosis [17]. The

sensitivity and specificity ranges from 81%-100% and 94%-100% respectively in case of parotid gland lesions [18]. Hence, for evaluation of a parotid mass, FNAC should be performed first along with other haematological and radiological investigations. Other benefit of doing FNAC is that it is also possible to culture the aspirate if the report being no conclusive. Similarly AFB staining of the aspirate may be performed if needed. However, due to necrosis at center of the swelling, FNAC is not always contributory in large parotid swellings. A total of 20.6% of FNAC are non-diagnostic, this could be due to the complexity of the tissue Architecture [19]. This can be overcome by repeating FNAC. Cytological studies combined with AFB staining of the aspirated material gives better results.

USG, CT and MRI are important modalities for studying parotid parenchyma. They can identify signs suggestive of malignancy and distinguish lesions of parenchyma of parotid gland from extra-parotid lesions; and identify those entities that may not require surgical intervention. Ultrasound is the initial imaging modality of choice for the assessment of palpable parotid gland abnormalities. The parotid glands are superficial structures and are readily amenable to high resolution ultrasound examination.

Sonographically, there are two types of parotid tuberculosis, namely parenchymal and peri-parotid type. The parenchymal type of parotid tuberculosis presents as a diffusely enlarged, comparatively hypo-echoic, with or without focal intra-parotid nearly anechoic areas in ultrasonography, which might have a cavities within it as seen in our case. The peri-parotid type of tuberculosis appears as hyper-echoic parotid gland surrounded by hypo-echoic nodules on sonography, consistent with peri-glandular lymph node enlargement. USG-guided FNAC correlates well with postoperative histological findings and has an overall accuracy of 86%-89% [20].

Surgical intervention becomes important for obtaining tissue for histopathological examination when imaging and FNAC are inconclusive. Usually, an excisional biopsy is performed but when whole parotid is involved, total parotidectomy should be done. As there is definitive risk of facial nerve injury during the procedure, surgical intervention must be done only after detailed informed consent.

Caseating granulomas and staining for acid-fast bacilli are histological features of tuberculosis. Because of the abscess formation, classical histopathological picture of caseating granuloma was not found in our case. The diagnosis was confirmed by demonstration of acid fast bacilli by Ziehl-Neelsen staining and detection of mycobacterial DNA in Cartridge Based Nucleic Acid Amplification Test (CBNAAT) also known as GeneXpert. CBNAAT is more sensitive than microscopy and it provides result in about 2 hours. It also detects resistance to rifampicin [21]. A study done in India reported that CBNAAT is positive in 77.7% of all suspected tuberculosis cases with lesions in the head and neck region, whereas microscopy with ziehl-neelsen stain detected only 16.6% of cases. The sensitivity is 28.6% and specificity is 100% compared to culture which is gold standard test [22]. Various studies of extrapulmonary tuberculosis have reported a sensitivity ranging from 25% to 91.5% compared to culture [23].

Multidrug-resistant tuberculosis (MDR-TB) is defined as disease caused by *Mycobacterium tuberculosis* which is resistant to at least both rifampicin and isoniazid with or without resistance to other anti-tubercular drug. Rifampicin-resistant Tuberculosis (RR-TB) is defined as resistance to rifampicin detected using genotypic or phenotypic

methods with or without resistance to other anti-tubercular drugs. Multidrug-resistant or Rifampicin resistant tuberculosis is emerging as a major problem due to poor management of drug-sensitive and drug-resistance Tuberculosis as well. MDR-TB is treatable but its treatment is very expensive and it requires long duration of treatment (usually two years) and contains potentially toxic drugs. In December 2022, WHO has released updated guidelines for the treatment of drug-resistant tuberculosis suggesting major improvements in treatment options for people with multidrug-resistant or rifampicin-resistant tuberculosis. There is new recommendation on the use of all-oral 6 month regimen composed of bedaquiline, pretomanid, linezolid and moxifloxacin (BPALM) in people suffering from MDR/RR-TB or MDR/RR-TB with additional resistance to fluoroquinolones (pre-XDR-TB). The newly recommended BPALM regimen gives better outcomes, remarkably shortens the duration of treatment, and thus significantly improves quality of life for people suffering from MDR/RR-TB [24].

Conclusion

While evaluating a solitary parotid mass, diagnosis of tuberculosis should be kept in mind, even in the absence of history of tuberculosis in the patient, as it is a medically treatable condition, and surgery can be avoided. Parotid gland tuberculosis has a good prognosis with drug therapy. The anti-tubercular medication is the important treatment option whether the diagnosis is made clinically, radiologically or even after parotidectomy. Early diagnosis with a high level of clinical suspicion is essential to avert the need for surgery which may be a hazardous procedure in a medically treatable condition.

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