Bilateral Parotid Tuberculosis

JS Thakur, A Thakur[1], NK Mohindroo, S Mohindroo[2], DR Sharma

Departments of Otolaryngology-Head and Neck Surgery, [1]Pharmacology and [2]Pathology, I G Medical College, Shimla, India

ABSTRACT

Tuberculosis of parotid is a rare clinical entity, and cases of bilateral tubercular parotitis are even rarer. We present a case of bilateral primary parotid tuberculosis in a 49-year-old female. The patient received anti-tuberculosis treatment for six months, resulting in complete resolution of the disease. We also review the theories related to the pathogenesis of tubercular parotitis, and propose a novel hypothesis about greater involvement of parotid gland as compared to other salivary glands in primary tuberculosis.

Key words: Mycobacteria, Parotid, Pathogenesis, Tuberculosis

INTRODUCTION

Tuberculosis of head and neck region comprises about 10% of all the cases of extra pulmonary tuberculosis,[1,2] and cervical lymph node is the most common site of extra pulmonary tuberculosis.[1,3] Head and neck tuberculosis can present in skin, oral cavity, oropharynx, nose, ear, thyroid, larynx, salivary glands, mandible, and neck spaces.[1-4] Tuberculosis of parotid is extremely rare even in the endemic areas[3-6] and bilateral tubercular parotitis is even rarer.[3,7] Although parotid gland is the most common salivary gland involved in tuberculosis but its exact etiopathogenesis is still unknown. We present a case of bilateral parotid tuberculosis, review the literature and hypothesize on its pathogenesis.

CASE REPORT

In May 2008, a 49-year-old Indian female presented with swelling of bilateral parotid region since past two months [Figure 1]. The swelling was insidious in onset, painless, and progressively increased in size. Initially, it was noticed in the right parotid area, and within two weeks, a similar swelling was noticed in the left parotid region as well. There was no history of fever, cough, dyspnea, weight loss, loss of appetite, or exposure to tuberculosis. Patient was a housewife and had an insignificant medical history.

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On examination, there was a 4×4 cm non-tender, firm, and mobile swelling just in front and below the ear lobules in both parotid regions. The overlying skin and facial nerves were normal. Oral cavity, oropharynx, parotid ducts, TM joints, ears, and cervical lymph nodes were found normal. Systemic examination did not reveal any significant finding. Due to bilateral manifestation, a possibility of granulomatous disease (sarcoidosis or tuberculosis) was raised and patient was subjected to further investigations.

Fine needle aspiration cytology (FNAC) with 18G needle on both sides was done. Slides were prepared, dried in air, and stained as per the May-Grunewald-Giema's staining procedure. Microscopic examination showed epithleoid cell granuloma [Figure 2a] and Langhan's type of giant cells [Figure 2b]. These findings were consistent with granulomatous disease and tubercular parotitis was kept as

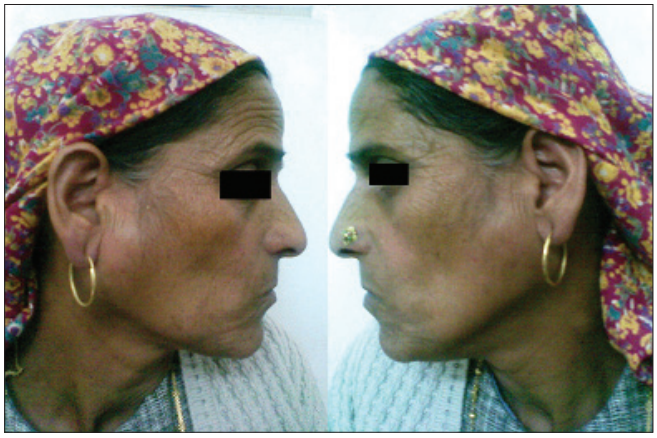

Figure 1: Patient with bilateral parotid swelling present just in front and below the ear lobule

Address for correspondence:
Dr. Jagdeep S Thakur, E-mail: anujagdeep@yahoo.co.in

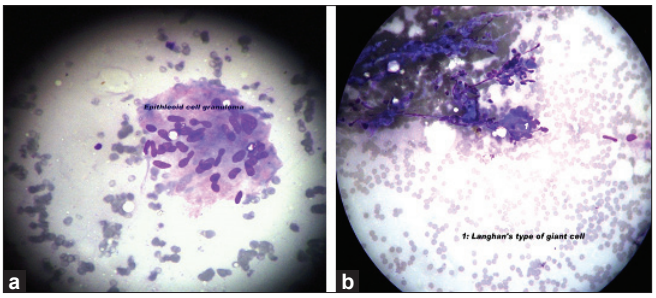

Figure 2: Microphotographs showing epithleoid cell granuloma (a) and Langhan's type of giant cells (b) in hemorrhagic background (MGG 400x)

first possibility. Hematological, biochemical investigations (blood and urine), immune status, and chest X-ray were found normal. Samples of sputum taken on three consecutive days were examined for mycobacteria by Ziehl-Nielsen staining procedure.[8] Polymerase chain reaction (PCR) test was done by extracting DNA from the parotid aspirate, and it was confirmatory for tuberculosis.[9] Due to absence of any other focus of tuberculosis, diagnosis of primary parotid tuberculosis was kept. The patient was put on Revised National Tuberculosis Control Programme (RNTCP) Category III treatment regimen for six months. This consists intensive phase of two months in which isoniazide 600 mg, rifampicine 450 mg, pyrazinamide 1500 mg are given thrice a week. In the next four months (Continuation phase), pyrazinamide was withdrawn and other two drugs are continued in same dose. Within two months of treatment, patient had complete remission of symptoms [Figure 3]. Patient is on regular follow-up and had no relapse.

DISCUSSION

Tuberculosis is endemic in the Indian sub-continent, and India had reported high incidence of tuberculosis in the world for the year 2009 (168/1,00,000 population/year).[10] However, even with this very high incidence, tubercular parotitis is rarely encountered.[3]

Two theories explain the pathogenesis of tubercular parotitis. One hypothesis presumes that it occurs by direct spread of mycobacteria from a nearby infected source. The sources may be tonsils or teeth and the spread may occur through direct inoculation by sputum, retrograde spread of bacilli through duct or by afferent lymphatics. The other possibility is that the gland may be infected by hematogenous or lymphatic spread from the lungs. It is hypothesized that a hypersensitivity reaction on initial infection leads to the bacilli being covered in the scar tissue. These bacteria get reactivated during low immunity after months to years.[11]

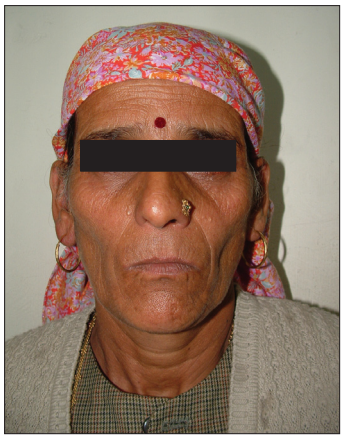

Figure 3: Patient with completely resolved bilateral parotid swelling after two months of anti-tubercular treatment

Parotid glands are the most common salivary glands involved in primary tuberculosis, whereas submandibular glands are the most commonly involved in systemic tuberculosis.[12,13] The local spread of the mycobacteria should be more to the submandibular gland as its duct is present at dependent part of the mouth and the lymphatic drainage of mouth occurs mainly to the submandibular lymph nodes. We believe that parotid glands may be affected more in localized spread because of the sluggish flow of the saliva. The parotid glands contribute about 26% saliva (submandibular glands: 69%; sublingual and minor salivary glands: 5%) at rest and it reaches to more than 70% in stimulatory conditions.[14] Hence, the retrograde spread of mycobacteria may preferentially occur to the parotid glands due to the low flow of saliva at rest. Further, the parotid gland secretes amylase a major protein fraction of parotid saliva, parotin hormone, and IgA immunoglobulin.[14] We believe that abundance of proteins and other metabolic products in the parotid glands as compared to other salivary glands, may be another cause but this hypotheses/statement needs validation by further research.

Tubercular parotitis manifests in two clinical forms.[15,16] In the localized nodular form, the intra or extra glandular lymph nodes are frequently affected from the drainage site. The other, diffuse parenchymatous form is very rare and considered to be an acute pathology involving whole of the gland. Initially, mycobacterium manifests in the nodes of the preauricular or lower pole area. It presents as localized, slow growing, non-tender localized swelling in front and below the ear.[17] The pain, abscess, fistula, and facial nerve involvement are the late features. The constitutional symptoms of tuberculosis like cough, fever, weight loss, loss of appetite may be present but are rare.[12,13]

The diagnosis of tubercular parotitis is very difficult because of the absence of symptoms and may often be

misdiagnosed as a benign parotid tumor.[5,6,12,15,16,18] The past history may reveal tuberculosis exposure or treatment. The provisional diagnosis needs high index of suspicion. The radiological investigations are of limited value in the diagnosis. Although radiographic findings may provide clues to the diagnosis but accurate diagnosis cannot be ascertained without histological evaluation.[6,19-25] These scans usually show central necrosis in the swelling which is not diagnostic as other malignant conditions also present with central necrosis. Ultrasonography (US) is highly sensitive for the 70–80% of tumors within the superficial parotid when compared with computed tomographic scan (CT) or magnetic resonance imaging scan (MRI), though has limitations when imaging the deep lobe. US shows diffuse parotid echo pattern changes with or without hypoechoic or nearly anechoic zones and with or without peri parotid lymphadenopathy.[26] US-guided FNAC gives 100% accuracy compared with CT (77–89%), MRI (88%), and US alone (83–98%).[27,28] CT scan has non-specific finding in the tubercular parotitis. It usually shows homogenously enhancing parotid with or without contrast enhancing round areas.[20,23,24,29-33] MRI has a sensitivity of about 75% for identifying benign features; this can be improved by using contrast enhancement. The MRI may define the disease better than CT and US.[17] MRI shows hypointense lesion on T1-weighted images, and hyperintense on T2-weighted images with homogenous contrast enhancement.[6,34] A detailed history, examination and FNAC have been advocated for the diagnosis of tubercular parotitis.[13,17] The Ziehl-Nielsen staining and culture for the mycobacterium is usually found negative. In tubercular lymph node, the FNAC has high sensitivity (80%) and specificity (93%)[12] and both increases up to 100% in parotid lesions.[17] As we have a protocol of performing FNAC with 18G needle in all lumps, we were able to diagnose this rare case without any surgery. The anti-tubercular drugs should be started early for cure. Earlier, combined medical and surgical intervention has been advocated to avoid recurrence,[13] however, now trend is towards conservative management rather than surgical intervention, which has been reserved for residual enlarged parotid.[6,20-25]

CONCLUSION

Tubercular parotitis is rare but can be encountered in endemic areas. It can be easily diagnosed by FNAC, and treated by standard anti-tubercular regimen. Bilateral manifestation of tuberculosis in parotid is very rare, and further research on the proposed hypothesis will make us to understand its etiopathogenesis better.

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REFERENCES

1. Rinaggio J. Tuberculosis. Dent Clin North Am. 2003;47:449-65. V.
2. Menon K, Bem C, Gouldesbrough, Strachan DR. A clinical review of 128 cases of head and neck tuberculosis presenting over a10-year period in Bradford, UK. J Laryngol Otol 2007;121:362-8.
3. Prasad KC, Sreedharan S, Chakravarthy Y, Prasad SC. Tuberculosis in the head and neck: Experience in India. J Laryngol Otol 2007;121:979-85.
4. Williams RG, Douglas-Jones T. Mycobacterium marches back. J Laryngol Otol 1995109:5-13.
5. Bhat NA, Stansbie JM. Tuberculosis parotitis: A case report. J Laryngol Otol 1996;110:976-7.
6. Birkent H, Karahatay S, Akcam T, Durmaz A, Ongoru O. Primary parotid tuberculosis mimicking parotid neoplasm: A case report. J Med Case Reports 2008;2:62.
7. Erkan AN, Cakmak O, Kayaselcuk F, Koksal F, Ozluoglu L. Bilateral parotid gland tuberculosis. Eur Arch Otorhinolaryngol 2006;263:487-9.
8. Forbes BA, Sahm DF, Weissfeld AS. Mycobacteria. In: Bailey and Scott's Diagnostic Microbiology. 12th ed. Missouri: Mosby Elsevier; 2007. p. 478-509.
9. Forbes BA, Sahm DF, Weissfeld AS. Nucleic acid-based analytic methods for microbial identification and characterization. In: Bailey and Scott's Diagnostic Microbiology. 12th ed. Missouri: Mosby Elsevier; 2007. p. 120-46.
10. Tuberculosis: Cases and Incidence. Global Health Observatory Data Repository. Geneva (Switzerland): World Health Organization. c2011. Available from: http://apps.who.int/ghodata/ [Last cited on 2011, July 19].
11. Cantrell R, Jensen J, Reid D. Diagnosis and management of tuberculosis cervical adenitis. Arch Otolaryngol 1975;101:53-7.
12. Süoğlu Y, Erdamar B, Cölhan I, Katircioğlu OS, Cevikbas U. Tuberculosis of the parotid gland. J Laryngol Otol 1998;112:588-91.
13. Stanley RB, Fernandez JA, Peppard SB. Cervical mycobacterial infections presenting as major salivary gland disease. Laryngoscope 1983;93:1271-5.
14. Nachlas NE, Johns ME. Physiology of the Salivary Glands. In: Paparella MM, Shumrick DA, Gluckman JL *et al.* editors. Otolaryngology, vol. 1. 3rd ed. Philadelphia: Saunders; 1991. p. 391-405.
15. O'Connell JE, George MK, Speculand B, Pahor AL. Mycobacterial infection of the parotid gland: An unusual cause of parotid swelling. J Laryngol Otol 1993;107:561-4.
16. Bhargava AK, Shenoy AM, Kumar RV, Nanjundappa, Rao CR. Parotid tuberculosis simulating malignancy. J Laryngol Otol 1993;107:561-4.
17. Iserì M, Aydìner O, Celik L, Peker O. Tuberculosis of the parotid gland. J Laryngol Otol 2005;119:311-3.
18. Choudhury N, Bruch G, Kothari P, Rao G, Simo R. 4 years' experience of head and neck tuberculosis in a south London hospital. J R Soc Med 2005;98:267-9.
19. Day TA, Buchmann L, Rumboldt Z, Joe JK. Neoplasm of the neck. In: Flint PW, Haughey BH, Lund VJ, *et al.* editors. Cummings Otolaryngology Head and Neck Surgery China: Mosby Elsevier; Available from: http://www.mdconsult.com/books[Last cited on 2010 Sep 12].
20. Mastronikolis NS, Papadas TA, Marangos M, Karkoulias KP, Tsamandas AC, Goumas PD. Tuberculosis of the parotid gland. Tuberk Toraks 2009;57:84-8.
21. Nag VL, Singh J, Srivastava S, Tyagi I. Rapid diagnosis and successful drug therapy of primary parotid tuberculosis in the pediatric age group: A case report and brief review of the literature. Int J Infect Dis 2009;13:319-21.
22. Seeley M, Waterhouse D, Shetty S, Gathercole J, Seeley C. Two cases of parotid tuberculosis.
23. Anuradha K, Satish MG, Revadi PS, Muley PR. Parotid gland tuberculosis-a case report. Indian J Pathol Microbiol 2004;47:277-9.
24. Oktay MF, Aşkar I, Yildirim M, Topçu I, Meriç F. Tuberculous parotitis: A review of seven cases. Kulak Burun Bogaz Ihtis Derg 2007;17:272-7.
25. Chintamani, Daniel R, Manu S, Bhushan V, Gupta K. Parotid TB. Trop Doct 2006;36:119-20.

26. Chou YH, Tiu CM, Liu CY, Hong TM, Lin CZ, Chiou HJ, *et al*. Tuberculosis of the parotid gland: Sonographic manifestations and sonographically guided aspiration. J Ultrasound Med 2004;23:1275-81.
27. Rout J, Brown JE. Dental and Maxillofacial radiology. In: Adam A, Dixon AK, Grainger RG, Allison DJ, editors. Grainger and Allison's Diagnostic Radiology. China: Churchchil Livingstone; 2008, Available from: www.mdconsult.com/books[Last cited 2010, Sep 12].
28. Thakur JS, Sharma ML, Mohan C, Mohindroo NK, Kaushik NK. Clinicopathological and radiological evaluation of cervical lymph nodes metastasis in head and neck malignancies. Indian J Otolaryngol Head Neck Surg 2007;59:327-31.
29. Bhargava S, Watmough DJ, Chisti FA, Sathar SA. Case report: Tuberculosis of the parotid gland-diagnosis by CT. Br J Radiol 1996;69:1181-3.
30. Caldart AU, Adriano CF, Caldart AU, Mocellin M. Primary tuberculosis of the parotid gland. Braz J Otorhinolaryngol 2007;73:720.
31. Alex L, Balakrishnan M, Ittyavirah AK. Tuberculosis of parotid gland: A case report. Ind Radiol Imag 2006;16:689-90.
32. Wei Y, Xiao J, Pui MH, Gong Q. Tuberculosis of the parotid gland: Computed tomographic findings. Acta Radiol 2008;49:458-61.
33. Kim YH, Jeong WJ, Jung KY, Sung MW, Kim KH, Kim CS. Diagnosis of major salivary gland tuberculosis: Experience of eight cases and review of the literature. Acta Otolaryngol 2005;125:1318-22.
34. Erdoğan B, Uzaslan E, Demirdöğen E, Balaban Adim S, Salan A, Cakir U. An unusual reason of parotid gland enlargement; parotid gland tuberculosis. Tuberk Toraks 2006;54:182-4.

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