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CASE REPORT

Primary parotid tuberculosis: varied clinical presentations

A Sethi, D Sareen, A Sabherwal, V Malhotra

Department of ENT & Head and Neck Surgery, Maulana Azad Medical College and associated L. N. Hospital, New Delhi, India

Parotid gland tuberculosis is rare and may present in different clinical forms. We present three cases of tuberculosis of the parotid gland that presented to us with different clinico-pathological appearances. The cases were diagnosed on the basis of histopathological evaluation and fine needle aspiration cytology. All the patients responded to four-drug antitubercular chemotherapy.
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Introduction

Tubercular involvement of the parotid gland is extremely unusual even in countries with high incidence of this infection such as India (Handa *et al*, 2001). It most commonly presents as a localized mass, resulting from infection of intracapsular or pericapsular lymph nodes (Rowe-Jones *et al*, 1992). It may also present as parenchymatous tuberculosis (Bhat and Stansbie, 1996), periauricular fistula/sinus (Zheng and Zhang, 1995) or as an abscess (Chatterjee *et al*, 2001). The diagnosis of this condition has been established on the basis of histopathological evaluation of the excision biopsy specimen in the past, but, a diagnosis based on fine needle aspiration cytology (FNAC) and medical management have been shown to be curative without the need of surgical intervention (Handa *et al*, 2001).

Case reports

Case 1
An 18-year-old female presented to us with multiple discharging fistulae in the left preauricular region of 3 months duration. The patient had a history of a painful swelling that ruptured spontaneously producing fistulae 3 months back. There was no history of fever, cough or weight loss. Local examination revealed a diffuse and tender swelling with multiple discharging fistulae over the left parotid and submandibular regions with granulations lining the opening of the fistulae. The patient also had enlargement of posterior triangle lymph nodes that were soft and non-tender (Figure 1). Her routine blood and urine investigations were within normal limits except for a raised erythrocyte sedimentation rate (ESR) and positive Mantoux test. Serum was non-reactive for HIV. Her chest X-ray was normal. The granulations lining the fistulae were biopsied and showed caseating granulomas on histopathological evaluation, with a positive staining for acid-fast bacilli (AFB). The patient received a 6-month course of isoniazid, rifampicin, pyrazinamide and ethambutol with complete resolution of lesion.

Case 2
A 16-year-old female presented to us with a swelling involving the right parotid region of 6 months duration. The swelling was slowly progressive in size, painless and non-tender. There was no history of fever, cough or weight loss. Local examination revealed a firm, cystic, non-tender swelling in the right parotid region. The routine blood and urine investigations were within normal limits except for a raised ESR. The serum was non-reactive for HIV and her chest X-ray was normal. A contrast enhanced computed tomography (CT) scan was done that revealed a round, smooth thick-walled lesion with central lucency involving the superficial lobe of right parotid gland (Figure 2). An FNAC from the lesion showed epitheloid caseating granulomas suggestive of mycobacterial infection. The patient showed complete response to a four-drug antitubercular treatment i.e. isoniazid, rifampicin, pyrazinamide and ethambutol given for 6 months.

Case 3
A 20-year-old male presented to us with a painful swelling in the left preauricular region of 3 weeks duration. There was no history of fever, cough or weight loss. The patient has been receiving different antibiotic regimens from private practitioners without much relief. Local examination revealed a firm and tender swelling measuring 5×3 cm involving the left parotid region (Figure 3). There was left posterior triangle lymphadenopathy. The routine blood and urine investigations were within normal limits. His serum was non-reactive for HIV and chest X-ray was normal. A

Correspondence: Dr Ashwani Sethi, E-80, Naraina Vihar, New Delhi-110028, India. Tel: 91 11 55399725, Fax: 91 11 2323 5574, E-mail: dr_sethi@rediffmail.com
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Figure 1 Showing multiple preauricular and submandibular fistulae along with scarring and posterior triangle lymphadenopathy

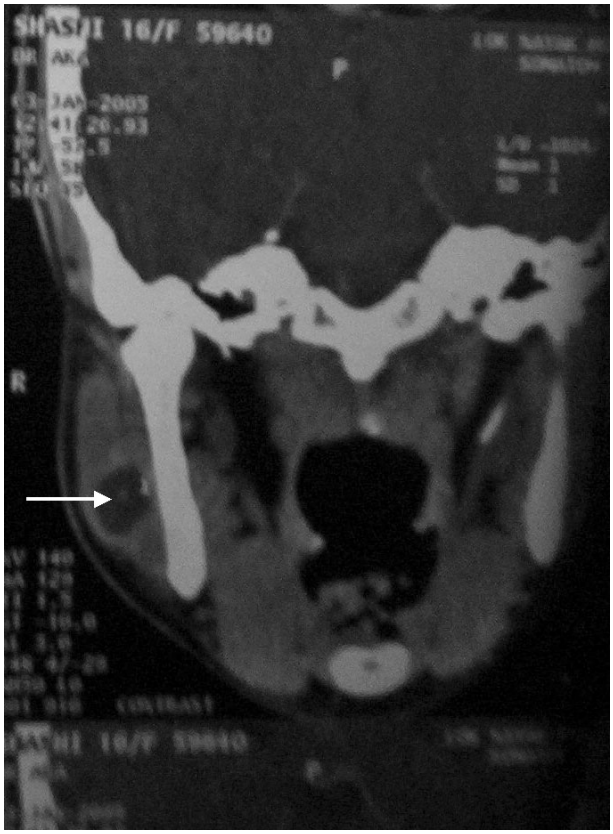

Figure 2 Contrast enhanced computed tomography scan (coronal view) showing tubercular focus in right parotid gland (white arrow)

contrast enhanced CT scan was done that revealed an enlarged left parotid gland. The parotid parenchyma showed vague diffuse enhancement with a cystic lesion suggestive of an abscess (Figure 4). Aspiration of the swelling revealed pus that stained positive for AFB. The patient received isoniazid, rifampicin, pyrazinamide and ethambutol for 6 months with complete cure.

Discussion

Extrapulmonary tuberculosis may present in concurrence with a focus in the lungs or may present primarily without pulmonary involvement. The latter situation may provide a difficulty in diagnosis due to the absence of systemic signs and symptoms of the disease (O'Conell *et al*, 1993). Tuberculosis of the parotid gland is rare and may pose a significant difficulty in diagnosis, as it is difficult to distinguish from the tumors of parotid gland that are comparatively much commoner (Handa *et al*, 2001). It may also occur either secondary to a primary focus in the lung, as a result of hematogenous spread or as an autoinfection from, the oral cavity.

It may occur as one out of the two pathological forms – the localized form, because of the involvement of intra- or periglandular lymph nodes is the more common form with pathogenesis similar to scrofuloderma, whereas, the diffuse form involving the parenchyma may be secondary to the nodal infection, and is rarer (Bhat and Stansbie, 1996).

Clinically, the most common mode of presentation is as a localized swelling with gradual enlargement (Zheng and Zhang, 1995). Our second case presented in a similar manner. The patient may also present with preauricular discharging fistulae as a result of spontaneous or surgical drainage of a fluctuant swelling (Zheng and Zhang, 1995). Our first case presented in a similar manner. The patient may also present with an abscess in the parotid gland (Chatterjee *et al*, 2001), as was the case in our third patient.

The diagnosis of parotid gland tuberculosis is based on the clinical presentation of the patient combined with investigations like a contrast enhanced CT scan, FNAC and histopathological evaluation. In the past, the diagnosis was invariably established on the basis of an excision biopsy (Maynard, 1967). In the recent years, FNAC has become more acceptable as a means of diagnosing the pathology and nature of salivary gland masses without the complication of implantation (Weiner and Pahor, 1996; Handa *et al*, 2001). This has, indeed, obviated the need for surgery. Two of our cases were diagnosed on FNAC. On a contrast enhanced CT scan, the presence of thick walled rim enhancing lesions with a central lucency is suggestive of tubercular pathology (Bhargava *et al*, 1996). One of our cases had similar findings on the scan. This type of a picture may also be seen in a case of lymphoma (Clearly *et al*, 1982). The CT scan may also reveal a diffuse parenchymal enlargement, which was seen in one of our

Figure 3 Showing swelling in the left parotid region because of tubercular mass

Figure 4 Contrast enhanced computed tomography scan (axial view) showing diffuse enlargement with a cystic lesion in left parotid gland (white arrow)

patients. The most diagnostic of all the presentations on a CT scan is the presence of multiloculated nodal mass with nodal lucency and thick, smooth-walled enhancing rims (Rude and Bergeron, 1985). One of our patients was diagnosed on the basis of the histopathological evaluation of the granulation tissue around the preuricular fistulae.

Thus, the diagnosis of the condition requires a high index of clinical suspicion complemented by investigations such as a CT scan and FNAC. Surgery, no longer has a role to play as regards the management of this condition. The mainstay of treatment is medical in the form of anti-tubercular chemotherapy.

Conclusion

In conclusion, tuberculosis of the parotid gland is a rare entity and may present in different clinico-pathological forms. The diagnosis requires alertness on the part of the clinician towards the possibility of this condition. FNAC plays an important role in the diagnosis. Medical treatment alone in the form of anti-tubercular chemotherapy is curative.

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