Diffuse tuberculous parotitis

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Abstract

Parenchymatous parotid tuberculosis diffusely affecting the entire gland is very rare. We present a case, associated with a primary pulmonary focus, that was confirmed after positive identification of alcohol and acid-fast bacilli in gastric washings. Both sites of infection resolved with quadruple anti-tuberculous chemotherapy.

Introduction

Diffuse, infective parotitis is common, usually resulting from the mumps, echo or coxsackie A viruses, or staphylococci in the elderly and debilitated. Tuberculosis in the parotid region however is very uncommon. It nearly always presents as a localized mass, resulting from infection of intra-capsular or peri-glandular lymph nodes. Diagnosis is therefore often only made after operative excision of the presenting mass.

Parenchymatous tuberculosis affecting the entire gland is extremely rare. Only two cases are recorded in the Salivary Gland Register of Germany for the period 1965–1981 (Seifert *et* al., 1986) and one further case in the English literature in the last 10 years (Stanley *et al.*, 1983). In the patient reported here the condition was associated with a primary pulmonary focus.

Case report

A 19-year-old Eritrean refugee, resident in the United Kingdom for seven months, presented with swelling of the left parotid gland (Fig. 1). This had been increasing in size for two months and was associated with pain on mastication, weight loss and occasional night sweats. Examination demonstrated tender enlargement of the entire gland causing trismus and an associated pyrexia of 38.3°C. There was no facial nerve weakness or skin change.

Investigations revealed a white cell count of $10.4 \times 10^9/L$ and an erythrocyte sedimentation rate of 110 mm per hour. Viral serology was normal. Ultra-sound scan of the parotid demonstrated uniform enlargement of the gland with reduced echogenicity but no focal lesion, in particular no abscess. A chest X-ray showed consolidation of the left lung base. Mantoux test was positive at 1:10,000 dilution but sputum and saliva specimens failed to reveal alcohol and acid-fast bacilli. These were found in gastric washings. The procedure to obtain them is simply performed on the ward; the patient is allowed clear fluids only from midnight, on the following morning a naso-gastric tube is passed and 10 to 20 mls of water are instilled into the stomach, and subsequently aspirated for Ziehl-Neelsen staining.

Treatment with quadruple anti-tuberculous chemotherapy comprising rifampicin, isoniazid, pyrazinamide and ethambutol was commenced. The parotid swelling clinically resolved over three months as did the chest X-ray appearances of consolidation.

The patient completed two months of quadruple therapy and a further five months of rifampicin and isoniazid alone and has had no signs of recurrence.

Discussion

Mycobacterial parotitis is very uncommon. It was first described in 1894 by Von Stubenrauch (Talmi *et al.*, 1990). Since then only 43 cases have been reported in the English literature and of these the majority were seen before 1941 (Coen, 1987; Ubhi *et al.*, 1988; Talmi *et al.*, 1990). Two pathological



FIG. 1 Diffuse swelling of the left side of the face resulting from tuberculous, parenchymatous parotitis.

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types occur: a commoner focal form resulting from lymph node infection and a rarer diffuse form resulting from parenchymatous involvement. Of 13 reported cases in the last 10 years, 12 are of the focal form (Levin-Epstein and Lucente, 1982; Burrow *et al.*, 1983; Stanley *et al.*, 1983; Coen, 1987; Ubhi *et al.*, 1988; Talmi *et al.*, 1990) and one only of the diffuse form (Stanley *et al.*, 1983). Before pasteurization *Mycobacterium bovis* was the most likely infecting organism but it has now been replaced by *Mycobacterium tuberculosis*.

In the localized form, resulting from infection of intra- or periglandular lymph nodes, the pathogenesis is probably similar to that for cervical node tuberculosis, thereby representing a primary focus. However the organism may have arisen from an initial site in the tonsil (Wilmot *et al.*, 1957). Allen-Mershe and Forsyth (1958) have also suggested infection may be related to antecedent dental sepsis. As with a cold abscess diagnosis may be difficult to make (Levin-Epstein and Lucente, 1982). The patient is often systemically well and the chest X-ray may be normal. In 75 per cent of cases there is no family or personal history of tuberculosis (Donohue and Bolden, 1961). The mass may frequently be mistaken for a parotid neoplasm resulting in fine needle aspiration or surgical excision, with the risk of subsequent fistula formation (Stanley *et al.*, 1983).

The second form, with diffuse involvement of the gland parenchyma, is extremely rare (Donohue and Bolden, 1961; Epker, 1972; Stanley *et al.*, 1983). In Stanley's case the patient developed a parotid abscess and histological examination revealed caseating granulomas of the peri-glandular nodes as well as of the parenchyma. There was no evidence of active disease elsewhere and so it is probable that this case was of primary nodal disease with secondary spread to the surrounding gland. Of 28 cases of parotid tuberculosis reported to the German Salivary Gland Register over a 16 year period only two were of parenchymatous disease (Seifert *et al.*, 1986). In these patients, as in our case, the pathology is thought to be a result of lymphatic or haematogenous spread from a primary focus elsewhere. More usually however systemic disease is associated with submaxillary and sublingual gland involvement (Batsakis, 1974).

The diagnosis is difficult to make given its extreme rarity and similar clinical picture to other forms of inflammatory parotitis. Inappropriate surgical exploration and delay in instituting correct treatment may result in gland destruction and fistula formation (Stanley *et al.*, 1983; Seifert *et al.*, 1986) or possible cross-infection from an open pulmonary focus.

Although saliva and sputum samples were negative in our case, gastric washings provided a further simple and non-invasive method of tapping a possible secondary tuberculous reservoir of swallowed organisms. We were therefore able to avoid surgical exterpation of the parotid gland with its attendant risk of fistula formation (Stanley *et al.*, 1983). Fine needle aspiration cytology may have been helpful and revealed granulomatous, epithelial cells but this is not pathognomonic in ruling out other granulomatous diseases. Ziehl-Neelsen stains may also only be positive for bacilli in less than 50 per cent of patients with positive pathology findings (Stanley *et al.*, 1983).

Parenchymatous tuberculosis must always be considered in the differential for diffuse inflammatory swelling of the parotid

Key words: Parotid salivary gland; Tuberculosis

gland. Just as other authors have warned (Appling and Miller, 1981), cervicofacial mycobacterial infections are not a disease of the past and their incidence in this country will rise with increasing travel from the Third World.

Conclusion

Tuberculosis of the parotid gland is rare, especially the diffuse form. The diagnosis is difficult to make and may initially be incorrect in as many as 91 per cent of cases (Donohue and Bolden, 1961). Gastric washings are a simple and non-invasive method of identifying alcohol and acid-fast bacilli as the pathogen. In parenchymatous disease, search for a primary focus should also be undertaken.

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