

Mycobacterial infection of the parotid gland: an unusual cause of parotid swelling

Janet E. O'CONNELL, F.R.C.S., Ed.,* M. K. GEORGE, B. SPECULAND, M.D.S., F.D.S., F.F.D., F.R.A.C.D.M.S. (OMS),** A. L. PAHOR, M.A., D.M.Sc. (Path), F.R.C.S., D.L.O.*

Abstract

Six cases of mycobacterial infection of the parotid gland are reviewed. All six cases presented solely with a slowly enlarging parotid swelling clinically indistinguishable from a parotid tumour. All of the swellings required surgical removal, superficial parotidectomy in four cases, and enucleation in two cases to obtain a definitive diagnosis.

Key words: Parotid gland; *Mycobacterium tuberculosis*

Introduction

One hundred and eighty seven new cases of mycobacterial infection have presented to this hospital in the last three years. Among these were six which presented solely with a parotid swelling. Sporadic case reports have appeared in the literature in the 1980's (Kuruvillea *et al.*, 1981; Burrow *et al.*, 1983; Coen, 1987; Ubhi *et al.*, 1988) but the last series was presented by Redon in 1955 when the incidence of mycobacterial infection was much higher.

Case reports

Case 1

A 51-year-old Asian lady presented with a slowly enlarging parotid swelling of four years duration. A parotid sialogram showed a normal duct and glandular structure. The swelling was removed by superficial parotidectomy. The histology showed normal salivary tissue, containing lymph nodes which had been completely replaced by caseating epithelioid granulomas, some containing Langerhan's giant cells. The surrounding parotid tissue showed patchy chronic inflammation. Scanty acid-fast

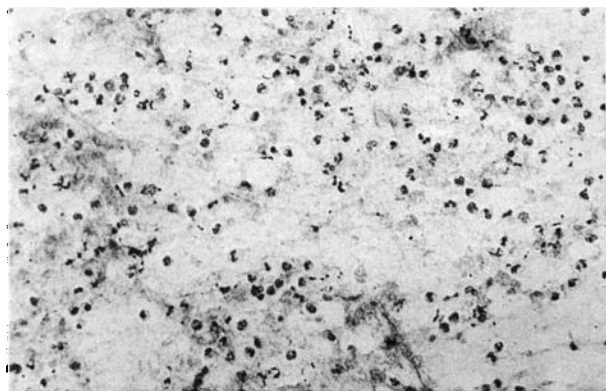


FIG. 1

Case 2. Fine needle aspirate showing an acute inflammatory exudate (mostly neutrophils).

bacilli were seen. Some tissue was submitted for bacteriological investigation but no organisms were identified or cultured.

Case 2

A previously fit, 33-year-old Caucasian man presented with a three-day history of a painful swelling in the right pre-auricular region, preceded one week earlier by the appearance of a furuncle on the right cheek. He was treated with oral flucloxacillin with resolution of the pain but not the swelling. The parotid swelling persisted three months later. The swelling was aspirated and 2 ml of creamy 'pus' was obtained. This was subjected to microscopy and culture but no organisms or acid-fast bacilli were seen or cultured. The smear showed an acute inflammatory response (Fig. 1).

The swelling increased in size despite the previous aspiration. He was treated by superficial parotidectomy where the superficial lobe was found to be replaced by a large fibrotic lesion. The histology showed large groups of coalescent, caseating granulomas with no surviving lymph nodes in the area (Fig. 2). The surrounding parotid tissue showed mild focal inflammation and

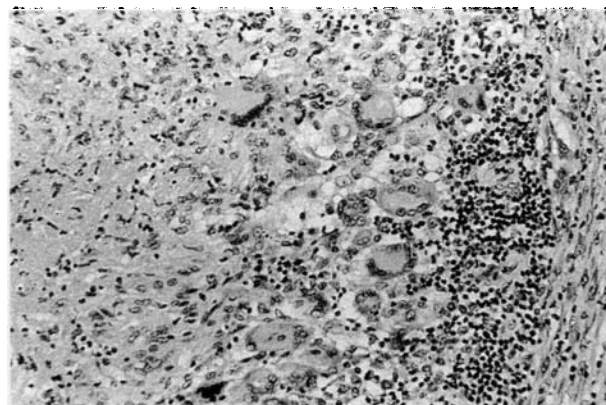


FIG. 2

Case 2. Histology showing epithelioid cells, Langerhan's giant cells, granuloma formation with central necrosis (top left of photomicrograph).

From the Departments of *Otolaryngology and **Oral/Maxillofacial Surgery, Dudley Road Hospital, Birmingham.
Accepted for publication: 30 October 1992.

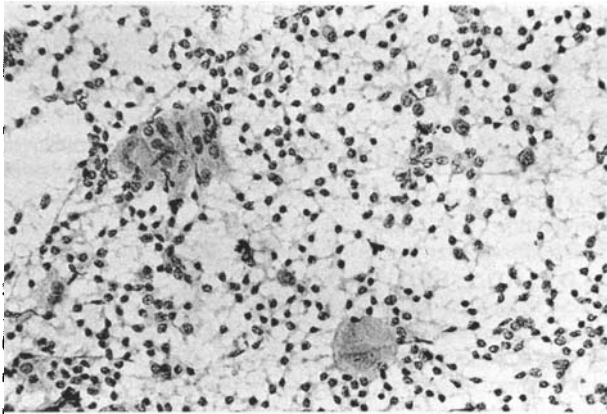


FIG. 3

Case 5. Fine needle aspiration cytology showing epithelioid cells, lymphoid cells and Langerhan's giant cells (large cells in photomicrograph).

scattered 'trapped' lymph nodes showing mild non-specific inflammatory changes only. Tissue obtained at superficial parotidectomy was also submitted for bacteriological investigation but no organisms were identified or cultured. No primary focus of mycobacterial infection was found.

Case 3

A 44-year-old Asian male presented with a painless right parotid swelling of two years duration. The swelling measured 5×4 cm. Fine needle aspiration biopsy revealed numerous lymphocytes but no specific features of mycobacterial infection and no mycobacteria were identified or cultured. Through a superficial parotid incision the swelling was enucleated. Histological examination revealed an enlarged lymph node containing numerous epithelioid granulomas. There were areas showing caseation and central necrosis with adjacent Langerhan's type giant cells. Tissue was subjected to bacteriological investigation but no organisms were cultured. Scanty acid-fast bacilli were seen on Ziehl-Neelson staining. No primary focus of infection was found.

Case 4

A 30-year-old Asian male presented with a left parotid swelling of eight months duration. The swelling was 2×2 cm in size and was tender. Fine needle aspiration did not reveal any specific features. The swelling was removed at superficial parotidectomy and the histology showed epithelioid granuloma formation with areas of central necrosis and giant cells. Tissue was sent for bac-



FIG. 4

Case 5. CT scan with contrast showing enlarged lymph nodes in relation to the medial border of the right parotid gland.

teriological investigation but no organisms were cultured. Scanty acid-fast bacilli were seen on Ziehl-Neelson staining. No primary focus of infection was found.

Case 5

A 59-year-old Asian lady presented with an enlarging swelling over the angle of the right mandible. The swelling had been present for one year and measured 4×4 cm. Fine needle aspiration cytology revealed abundant lymphocytes, epithelioid and giant cells consistent with mycobacterial infection (Fig. 3). A CT scan with contrast (Fig. 4) was reported as showing multiple soft tissue masses closely related to the medial border of the right parotid gland in the region of the internal jugular vein and also infero-medial to the right sterno-mastoid muscle. Some of the lymph nodes showed areas of central necrosis strongly suggestive but not diagnostic of mycobacterial infection. Through a superficial parotidectomy incision several enlarged intraparotid lymph nodes were enucleated (Fig. 5). The lymph nodes varied in size from 2.2×1.3 cm to 0.6×0.5 cm (Fig. 6). Histology showed numerous granulomas, some with central necrosis, numerous epithelioid cells and some Langerhan's giant cells (Fig. 7). Occasional acid-fast bacilli were seen on microscopy with Ziehl-Neelson staining. No primary focus of infection was found.

Case 6

A 29-year-old lady presented with a two-year history of a slowly enlarging swelling at the angle of the right mandible. The swelling measured 2×2 cm. The presumptive pre-operative

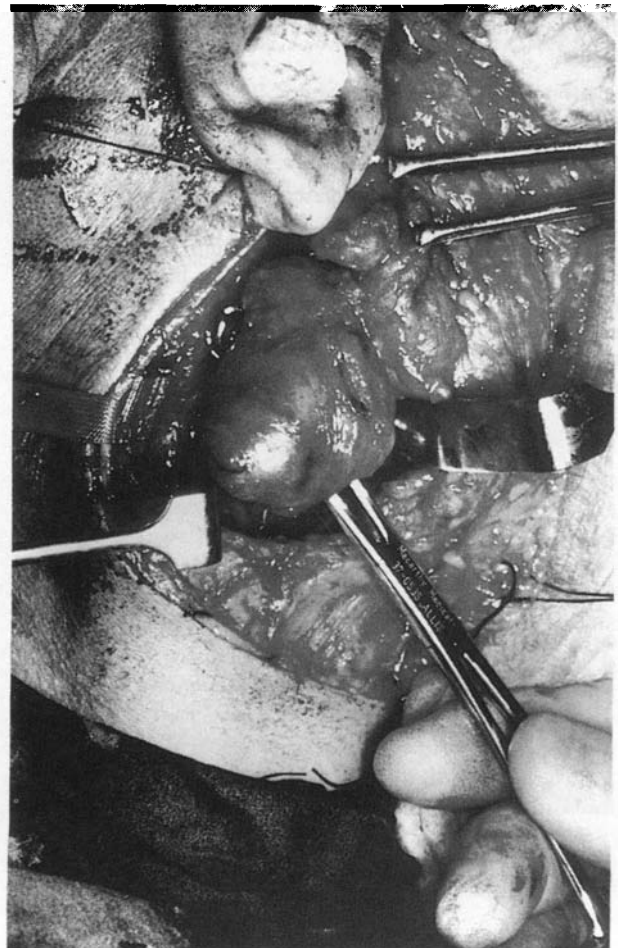


FIG. 5

Case 5. Enlarged lymph nodes found at superficial parotidectomy.

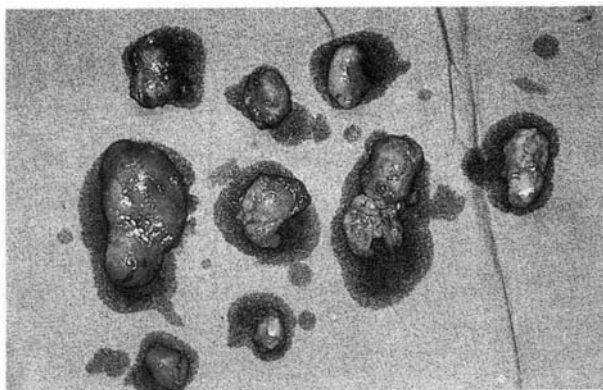


FIG. 6

Case 5. Lymph nodes obtained at superficial parotidectomy.

diagnosis was that of a pleomorphic adenoma. Fine needle aspiration cytology was not performed. At operation the swelling was found to be due to an enlarged lymph node which on histological examination showed many caseating epithelioid cell granulomas. Mycobacteria were not identified nor cultured. No primary focus mycobacterial infection was found although a Mantoux test performed following receipt of the histology was strongly positive.

Investigations

All six patients were systemically well. The chest X-rays were normal in all the patients despite review by a consultant radiologist in the knowledge of the histological diagnosis. Mantoux testing was performed in four patients, three of whom showed a weak positive result and one a strong positive result. Sialography performed in one patient showed a normal duct and glandular structure. CT scanning in one patient showed enlarged lymph nodes with central necrosis consistent with but not diagnostic of mycobacterial infection. Fine needle aspiration biopsy has recently been advocated for the diagnosis of parotid swellings (Webb and Russel, 1982). However we found the results inconclusive in two of the three cases in which it was performed. Mycobacteria were identified by microscopic examination with Ziehl-Neelson staining of caseous material obtained at operation in four cases.

Discussion

Extrapulmonary mycobacterial infection may occur in many organs but usually manifests itself in one of the following ways. Extrapulmonary infection occurs in the miliary process, but usually there are systemic signs and symptoms of more importance than the specific distant organ infection. Organ involvement may occur with simultaneous reactivation in the lung, but an equally common occurrence is the appearance of extrapulmonary mycobacterial infection in the absence of clinical disease in the lung (Betts and Reese, 1986). In most series of extrapulmonary mycobacterial infection less than 50 per cent of patients had chest X-ray evidence of previous infection (Betts and Reese, 1986).

Mantoux testing is a helpful diagnostic aid especially if the patients are previously Mantoux negative. However all our cases had been previously immunized with BCG. A tuberculin test cannot sensitize a non-infected person but it can restimulate or enhance remotely established, and subsequently weakened hypersensitivity (Thompson *et al.*, 1979). In atypical mycobacterial infection skin tests are negative unless a higher dose of purified protein derivative is used, that is 1:100 as opposed to the more usual dose of 1 in 10 000 (Bettes and Reese, 1986).

In order to identify acid-fast bacilli on microscopy of sputum it is thought that concentrations of 10 000 organisms and above, per millilitre of sputum are required (Des Prez and Heim, 1990).

Such heavy concentrations are often not obtained in sites of presumed secondary infection. Identification and culture of organisms in infected lymph nodes therefore can be very difficult if not impossible (Betts and Reese, 1986). Guinea pig inoculation used to be very popular in the identification of *Mycobacterium tuberculosis* and *Mycobacterium bovis*, however Marks (1972) has shown that only very rarely does guinea pig inoculation lead to a diagnosis when cytological and other culture techniques have failed.

CT scanning of affected lymph nodes may show central necrosis but central necrosis sometimes occurs in lymphomas (Clearly *et al.*, 1982) and so the finding of enlarged lymph nodes with central necrosis is not diagnostic.

Fine needle aspiration cytology may well suggest mycobacterial infection, especially if Langerhan's giant cells are present. However Langhan's giant cells may occur in other conditions such as fat necrosis or foreign body reactions. Similarly epithelioid cells in granulomatous lymphadenitis may occur in a wide spectrum of other conditions and are not specific for mycobacterial infection (Koss *et al.*, 1984). Further confusion may occur, as in the fine needle cytology in Case 2, as in the early phases of mycobacterial infection the primary response may be the occurrence of an acute inflammatory reaction. Examination of lymph nodes infected with a wide variety of mycobacterial infections have revealed a broad spectrum of possible inflammatory responses (Sanders and Horowitz, 1990).

Mycobacterial infection of the parotid gland is rare even in those countries where the disease is otherwise common. Mycobacterial infection of the parotid gland usually presents as a slowly growing mass, clinically indistinguishable from a parotid tumour (Redon, 1955). It is usually confined to intraparotid lymph nodes (Maynard, 1967). Two pathological types have been described: (1) diffuse, involving the whole gland with irregular nodules in volume and consistency and (2) localized, with a solid mass corresponding to tuberculosis in the lymph nodes of the parotid. These lesions can progress to caseation with formation of fistulae or fibrosis giving rise to a clinical picture similar to pleomorphic adenoma (Roberi and Montezemelo, 1973).

Maynard (1967) states that there is no way of distinguishing these infections from tumours of the parotid gland and that the diagnosis will always be dependent on the post-operative histological appearances i.e. of granuloma formation, with the presence of Langerhan's giant cells and caseation. Twenty-five years later, despite advances in diagnostic investigations we have found that this statement is still valid.

Summary

Six cases of mycobacterial infection of the parotid gland (five confined to the intraparotid lymph nodes and one with diffuse

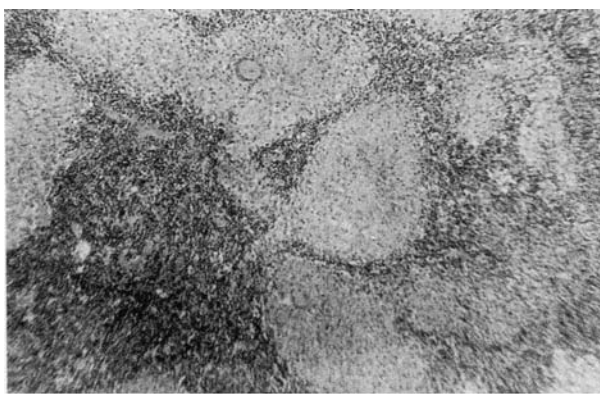


FIG. 7

Case 5. Histology showing epithelioid cells, granuloma formation with central necrosis and Langerhan's cells.

parotid involvement) are described. Despite pre-operative investigations these swellings were indistinguishable from a parotid tumour and in all cases the definitive diagnosis was made on the histological characteristics of granuloma formation, Langerhan's giant cells and central caseation.

Acknowledgements

We should like to thank the Pathology Department, Dudley Road Hospital, especially Dr W. Shortland-Webb and Dr C. Chan, for their help. We should also like to thank Dr Iles and Dr Honeybourne, Consultant chest physicians, for their assistance and advice.

References

- Betts, R. F., Reese, R. E. (1986) Lower respiratory tract infections. In *A Practical Approach to Infectious Diseases*. 2nd Edition. Ch. 9, (Reese, R. E., Douglas, R. G. Jr., eds.), Little, Brown and Co. Boston/Toronto, pp 239–240.
- Burrow, C. T., Haggart, B. G., Thomas, Z. (1983) Tuberculosis of the parotid gland. *Journal of The Royal College of Surgeons of Edinburgh* **28**: 342–344.
- Cleary, K. R., Osborne, B. M., Butler, J. J. (1982) Lymph node infarction foreshadowing malignant lymphoma. *American Journal of Surgical Pathology* **6**: 435–442.
- Coen, L. D. (1987) Tuberculosis of the parotid gland in a child. *Journal of Pediatric Surgery* **22**: 367–368.
- Des Prez, R. M., Heim, R. C. (1990) Mycobacterium tuberculosis. In *Principles and Practice of Infectious Diseases*. 3rd Edition. Chp. 229, (Mandell, G. L., Douglas, R. G., Bennett, J. E., eds.), Churchill Livingstone, Edinburgh, p. 1878.
- Koss, L. G., Woyke, S., Olszewski, W. (1984) The lymph nodes. In *Aspiration Biopsy. Cytological Interpretation and Histological Bases*. 1st Edition. Ch. 5, Igaku-Shoin Ltd, New York, p 109.
- Kuruwilla, A., Saha, N. K., Barton, R. P. E., Zaradawi, I. M. (1981) Tuberculosis of intra-parotid lymph nodes. *Journal of Laryngology and Otology* **95**: 1165–1167.
- Marks, J. (1972) Ending the routine guinea-pig test. *Tubercle* **53**: 31–34.
- Maynard, J. (1967) Parotid enlargement. *Hospital Medicine* **1**: 620–624.
- Redon, H. (1955) *Chirurgie des glandes salivaires*. Masson, Paris, p 101.
- Roberi, P. L., Montezemelo, C. L. (1973) Tubercular parotitis in childhood. *Minerva Paediatrica* **25**: 300–307.
- Sanders, E. W. Jr., Horowitz, E. A. (1990) In *Principles and Practice of Infectious Diseases*. 3rd Edition. Ch. 231, (Mandell, G., Douglas, R. G., Bennett, J. E., eds.) Churchill Livingstone, Edinburgh, p 1915.
- Thompson, N. J., Glassroth, J. L., Snider, D. E. Jr. (1979) The booster phenomenon in serial tuberculin testing. *American Review of Respiratory Diseases* **119**: 587–597.
- Ubhi, S. S., Neoptolemos, J. P., Watkin, D. F. L. (1988) Incidence and diagnosis of parotid gland tuberculosis in Asians in Leicester. *British Journal of Surgery* **75**: 313.
- Webb, A. J., Russel, R. C. G. (1982) *Recent Advances in Surgery Vol. 11*, Churchill Livingstone, Edinburgh, pp 49–52.

Address for correspondence:

Mr A. L. Pahor, M.A., D.M.Sc., (Path), F.R.C.S., D.L.O.,
Consultant Otolaryngologist,
Dudley Road Hospital,
P.O.Box 293,
Birmingham B18 7QH.