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Primary mycobacterial infection of the uvula

A. MURRAY, F.R.C.S., D. S. GARDINER, M.R.C.PATH. M.D.*, R. J. MCGUINESS, F.R.C.S.

Abstract
Tuberculosis, and non-tuberculous mycobacterial infections are becoming more common thus it is more likely that otolaryngologists will encounter these conditions. We describe an otherwise well patient, with symptoms and signs from chronic uvular inflammation, who proved to have a primary mycobacterial infection. This is an unique presentation in the literature and reminds clinicians of the need, where uncertainty exists in diagnosis, to consider mycobacterial infections.

Key words: Uvula; Infection; Chronic disease; Mycobacterium, tuberculosis

Introduction

The incidence of the commonest mycobacterial infection, tuberculosis (TB) continues to rise throughout the world (Centers for Disease Control 1991; Nakajima, 1993) with a third of the world's population now believed to be affected (Sudre *et al.*, 1992). Various reasons for this rise in incidence have been proposed, including drug resistant mycobacteria (National MDR-TB Task Force, 1992), human immunodeficiency virus (HIV) infection and a reduction in disease control programmes (Advisory Council for the Elimination of TB, 1989; Williams and Douglas-Jones, 1995). Infections caused by non-tuberculous mycobacteria (NTM) are also believed to be occurring more frequently (Waldman, 1982; Pransky *et al.*, 1990). It, therefore, becomes increasingly likely that practitioners will encounter patients with mycobacterial infections and, in otolaryngology, this will be either primary tuberculosis originating in the head and neck region, or, more likely, manifestations secondary to active pulmonary disease. In children the most frequent infections are NTM involvement of the cervical glands (Pransky *et al.*, 1990; Starke, 1992).

Oral cavity, or oropharyngeal infection with mycobacteria is usually tuberculosis, and is seen secondary to active pulmonary disease, with primary infection only very rarely occurring (Tyldesley, 1978; Haddad *et al.*, 1987). The commonest sites affected by tuberculosis are the tongue, gums and hard palate (Komet *et al.*, 1965; Prabhu *et al.*, 1978; Hashimoto and Tanioka, 1989). Although uvular involvement with anterior tonsillar pillar tuberculosis has appeared in the literature in the past (Komet *et al.*, 1965), no previous reports of an isolated mycobacterial infection of the uvula have been described.

Case report

A 40-year-old man was referred from his GP with an enlarged uvula. This had begun several months previously with isolated episodes of enlargement, which were painful and distressing. Gradually the uvula had become more constantly swollen, and courses of broad spectrum antibiotics were unhelpful. Clinical examination revealed a reddened, enlarged, brawny uvula, sparing the soft palate. No cervical lymphadenopathy was present. Initially, mycobacterial infection was not considered a likely diagnosis, and, given the symptoms the patient was experiencing, an excision biopsy of the uvula was performed as the initial management. Histology revealed granulomatous inflammation within the subepithelial tissue (Figure 1) and stains for AAFB were positive (Figure 2). Subsequent chest radiographs, and general clinical examination were able to exclude any other mycobacterial foci. HIV testing was negative. Further throat swabs were taken, which failed to culture mycobacteria, but, in the light of positive stains, empirical anti-tuberculous therapy in the form of pyrizinamide, rifampicin and pyridoxine, was initiated. One year after surgery he remains well.

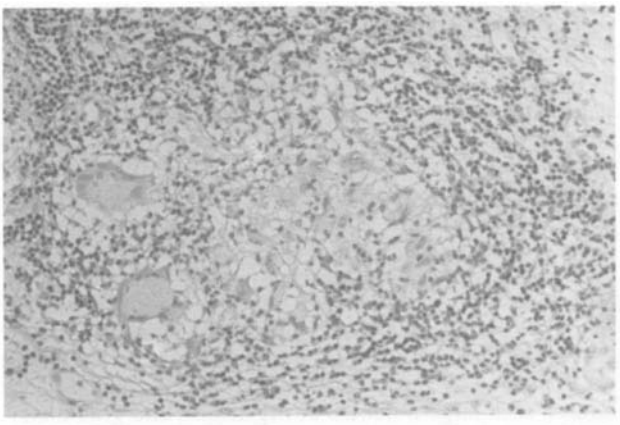

FIG. 1
Granulomatous inflammation within subepithelial tissues of uvula (H & E; × 75).

From the Department of Otolaryngology, Royal Alexandra Hospital, Paisley and the Department of Pathology/Cytology*, Monklands Hospital, Airdrie, UK
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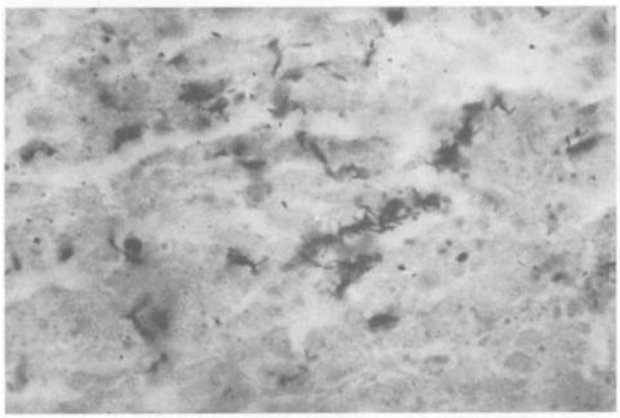

FIG. 2
Positive stains for AAFB (Ziehl-Neelson; × 350).

Discussion

Tuberculous mycobacterial infections of the oral cavity are said to occur with a frequency of less than one per cent of pulmonary disease (Brennan and Vrabec, 1970). Primary infections specifically are even more uncommon (Haddad *et al.*, 1987; Williams and Douglas-Jones, 1995) and are often the result of poor oral hygiene or local trauma, such as dental treatment (Rhoderick-Smith *et al.*, 1982; Dimitrakopoulos *et al.*, 1991) which is thought to allow direct inoculation into damaged tissue. The tongue is the most frequently reported site (Lathouwer *et al.*, 1975; Prabhu *et al.*, 1978; Hashimoto and Tanioka, 1989) but cases affecting most other oral and oropharyngeal sites, have been published (Mitchell, 1914; Rauch and Friedman, 1978; Tyldesley, 1978; Haddad *et al.*, 1987). There are no previous reports of isolated uvular involvement in tuberculous mycobacterial infections, although, interestingly, a published case of tuberculosis of the soft palate commented on the sparing of the uvula (Haddad *et al.*, 1987). Likewise there are no reported cases of an isolated NTM infection of the oropharynx or oral cavity. It is important, therefore, either to establish why the uvula may have become involved by mycobacterial infection, as in our patient, or to decide whether it was a contaminant.

Trauma is known to occur to the uvula during snoring, with chronic inflammatory changes found in histological specimens of patients with obstructive sleep apnoea (Sekosan *et al.*, 1996), and this would therefore be a possible mechanism of tissue inflammation and subsequent inoculation by random exposure to mycobacteria. Potentially pathogenic mycobacteria have been identified in environmental water sources in large numbers, previously (Katila *et al.*, 1995; Schulze-Robbecke *et al.*, 1995). This is obviously a theoretical mechanism and one that would be difficult to substantiate and, although our patient did give a history of snoring, it did not clearly predate the onset of the uvular symptoms. Presumably as the patient's uvula enlarged, snoring could result from the more oedematous tissue, so this explanation is not entirely satisfactory.

It seems unlikely that the mycobacteria were present in the specimen as a contaminant, as our histology demonstrated such large numbers of organisms, and granulomatous reactions. This fulfils at least one of the criteria suggested by microbiology texts, as being necessary to exclude opportunistic isolates of mycobacteria from clinical specimens (Shanson, 1995).

The histopathological diagnosis of this patient was completely unexpected and his management therefore differed from other patients in whom mycobacterial infection is considered clinically likely. If our patient's infection of the uvula had been due to NTM then it is possible that local excision would have sufficed (Rappaport *et al.*, 1990; McGovern *et al.*, 1994; Stewart *et al.*, 1994) but because this could not be assumed, it was felt necessary to treat him with anti-tuberculous chemotherapy as outlined above. Clearly, if the patient had developed any further evidence of mycobacterial disease, it would have been possible to utilize newer methods of identifying the precise species from paraffin-embedded tissue, such as polymerase chain reactions (Richter *et al.*, 1995), but given the patient's lack of symptoms, this was unnecessary.

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Address for correspondence:
Mr A. Murray,
Department of Otolaryngology,
Glasgow Royal Infirmary,
80 Castle Street,
Glasgow G4.