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Malakoplakia of the oropharynx

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Abstract
We present a case of malakoplakia presenting as a lesion in the oropharynx of an elderly smoker. In this case a clinical diagnosis of carcinoma of the pharynx was made, biopsy revealed the nature of the lesion and management was radically altered.

Key words: Malakoplakia; Oropharynx

Case report

A 70-year-old male smoker was referred to the Ear, Nose and Throat Department with a six-week history of an irritation of his throat. Clinical examination revealed a 3.5 × 2 cm mass in the right lateral oropharynx at the base of tongue. There was no associated regional cervical lymphadenopathy and the upper aerodigestive tract was otherwise unremarkable. Histological examination of a biopsy revealed the diagnosis of malakoplakia. The lesion was subsequently completely excised in March 2000. No antibiotics were used and there has been no recurrence to date. This patient remains symptom-free.

Histology

Paraffin-embedded sections of both biopsy and resection specimens showed polypoidal tonsillar tissue (Figure 1) with a submucosal diffuse proliferation of macrophages with eosinophilic cytoplasm.

The infiltrate also included neutrophil polymorphs, lymphocytes and plasma cells (Figure 1). Within the macrophages were occasional cytoplasmic calcified concretions that are known as Michaelis–Gutmann (MG) bodies (Figure 2). These are characteristics of malakoplakia. The lineage of the macrophages was confirmed by positive immuno-histochemical staining with the monoclonal antibody CD68.

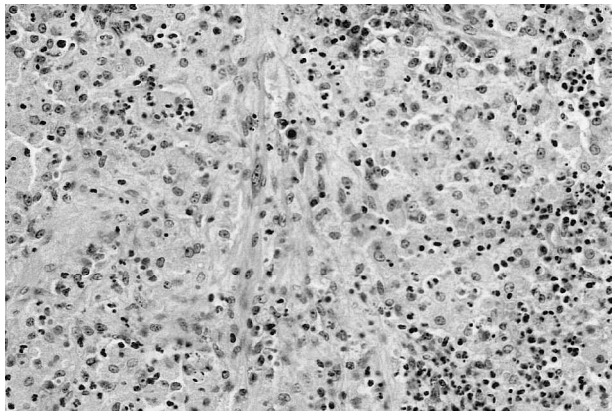

Fig. 1
Diffuse infiltrate of foamy macrophages admixed with acute and chronic inflammatory cells. (H&E; ×120).

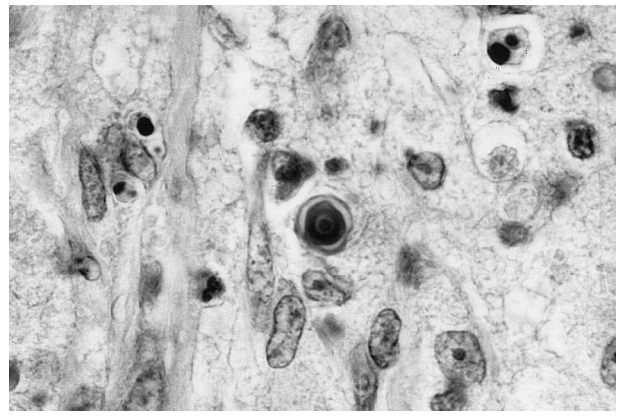

Fig. 2
Foamy macrophages with one bull's eye calcified concretion – the characteristic Michaelis-Gutmann body. (H&E; ×500).

Discussion

Malakoplakia is more frequently reported in the genito-urinary tract, and to a lesser extent the retroperitoneum and colon,[1] but is uncommon in the head and neck region. Within the head and neck region, there are reports of malakoplakia of palatine tonsil[2,3] and tongue base,[4] larynx,[5] middle ear,[6] parotid gland,[7] temporal bones,[8] and nasopharynx.[9,10]

First reported in 1902 by Michaelis and Gutmann,[11] the term malakoplakia (Greek; *malakos*–soft, *plakos*–plaque) was coined in 1903 by Professor Von Hansemann. Malakoplakia is a distinct chronic inflammatory tissue reaction of unknown origin with characteristic histopathological and ultrastructural features.[12–15] These include an infiltrate of lymphocytes, plasma cells and large macrophages called Von Hansemann cells which contains laminated calcified structures called Michaelis–Gutmann bodies.

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The pathogenesis of malakoplakia was thought by Michaelis and Gutmann to be benign neoplastic in origin but that view was rejected by Professor Von Hansemann.[16] The currently held view is that of an immunological associated defect involving macrophage inability to destroy and eliminate phagocytozed bacteria, particularly Gram-negative bacilli and, in particular, *Escherichia coli*. Evidence for such a view was demonstrated by Abou *et al.*[17] and Lou and Tiplitz[13] who concluded that effective microbial response of macrophages to phagocytozed *E. coli* was associated with large intracytoplasmic lysosomal granules, poor lysosomal enzyme release after phagocytosis and low levels of cyclic guanosine monophosphate (GMP). They then postulated that the basic defect was low level of cyclic GMP. Stauton *et al.*[18] successfully treated a case using the cholinergic agonist bethanechol chloride, which acts by increasing the level of cyclic GMP.

Collado Serra *et al.*,[19] treated a case of malakoplakia of the bladder by transurethral resection plus antibiotic. It recurred after four years; the lesion was similarly retreated and yearly endoscopic evaluation for the next ten years showed no recurrence. The cases reported by Love and colleagues[8] and Kalfayan and Seager[7] were treated satisfactorily with antibiotic without excision.

There is no sex preponderance when sites outside of the urinary tract are affected but females exhibit more in urinary tract involvement. Adults are affected much more than children. Malakoplakia is often associated with concomitant diseases such as sarcoidosis, lymphoma, cachexia, and carcinoma, all of which cause disturbances in T lymphocytes or cell-mediated immunity. There is an increased incidence of malakoplakia in immunosuppressed or immunodeficient patients[20] but the localized nature of the disease argues against generalized immunodysregulation.

The cellular structure of malakoplakia changes as the disease process progresses, which leads to three histological phases, postulated by Smith[3] in a study of malakoplakia of the urinary bladder.

1. *Early prediagnostic*. Infiltrate of plasma cells, Von Hansemann-type macrophages with the absence of MG bodies and eosinophils.
2. *Classical phase*. Sheets of large macrophages containing granular cytoplasm along with lymphocytes and plasma cells. MG bodies present in macrophages.
3. *Fibrosing stage*. Islands of macrophages with MG bodies set in a fibrous stroma formed by collagen bundles.

Histologically, the case reported here would be considered to be in the classical phase and demonstrates the existence of the condition in the upper aerodigestive tract, particularly in the differential diagnosis of malignancy.

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