

## Malacoplakia: Case report in tongue and review of the literature

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### Abstract

Malacoplakia is a relatively uncommon chronic inflammatory reaction of unknown etiology. It usually affects the genitourinary tract but may rarely involve the tongue. There are many theories that explain this reaction but it seems to be the answer to an infectious agent in a patient with immunologic deficiency. Microscopically, malacoplakia is characterized by the presence of foamy histiocytes with distinctive basophilic inclusions, which are known as Michaelis-Gutmann bodies due to a partially ingested bacteria and their posterior calcification. There are many alternatives to treat this entity. We report the only case diagnosed in the tongue, in a 15 years-old male in the Maxillofacial Surgery and Stomatology Service of the Hospital San Vicente de Paúl in Medellín, Antioquia, Colombia.

**Key words:** Malacoplakia, chronic inflammatory reaction.

### Introduction

Malacoplakia is a chronic inflammatory disease of unknown aetiology, described initially to be found in the bladder by Michaelis and Gutmann in 1902. (1-3) The following year, von Hansemann described nodules of yellowish brown soft tissue with central umbilication and coined the term malacoplakia for this condition from the Greek words malakos = soft and plakos = plaque.

It is usually reported in genitourinary tract although it has also been reported in different sites such as the gastrointestinal tract, retroperitoneum, and less common in lungs, bones, mesenteric lymphatic nodules, medium ear, larynx, palatine tonsil, parotid gland, temporal bone, neck (2) and tongue. (4-7) The age of presentation varies between 6 weeks of life to 85 years, being more frequent in adult age than in childhood. The distribution is equal in both sexes although in bladder the predilection is for the female sex. (4)

Histologically, there is a tissue with numerous macrophages (von Hansemann cells) some of which show eosinophilic cytoplasm; these macrophages have partially digested bacteria due to an a defective phagolysosomal activity and lead to the deposition of calcium and iron resulting in a basophilic inclusion structure, the Michaelis-Gutmann body, considered the hallmark for the diagnosis of malacoplakia. (2,3,7,8)

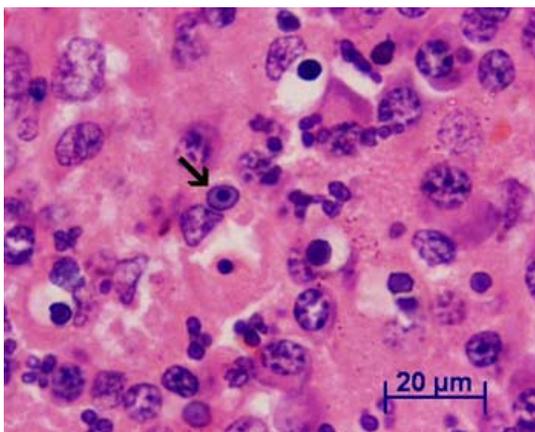
The aetiology remains unknown but three possible theories have been postulated to explain the unusual reaction in this lesion: (3) Microorganisms with unusual toxic properties, immune anomaly that affects the intracellular death of the microorganisms and enzymatic deficiency of the macrophage to destroy phagocytized bacteria.

Therapy with choline agonist (bethanechol chloride) has been used to correct the decreased cGMP levels that are believed to interfere with complete bacterial killing; ascorbic acid has been used to increase cGMP and cyclic adenosine

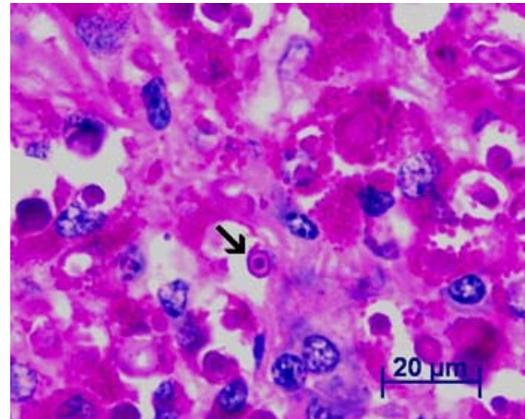
monophosphate (cAMP) levels in monocytes and to increase synthesis of collagen fibers; and antibiotics against Gram negative microorganisms (trimethoprim-sulfamethoxazole, quinolone) because of the possible infectious origin and surgical procedures to excise the mass. (8).

**Case Report**

A fifteen year-old male, presented to the Maxillofacial Surgery and Stomatology Service of the Hospital San Vicente de Paúl in Medellín, Antioquia, Colombia, with a one-month history of a painful left tongue mass, with difficulty chewing and swallowing, fever and decreased subjective body-weight. He had received analgesic and anti-inflammatory drugs for a week without any improvement. Clinical examination revealed a 4x2 cm, well defined, tender mass of the left side of tongue, covered by normal mucosa, with a sinus tract in the central zone and drainage of purulent material. A lingual abscess and a malignant tongue tumour were suspected and an initial treatment with Amoxicillin 500 mg three times per day for 7 days, Acetaminophen 500 mg four times per day and an incisional biopsy was programmed. One week later, it was found no purulent discharge and an improvement in the symptoms of the patient. Under local anaesthesia the mass was deeply biopsied and macroscopically revealed a firm, fibrous and yellow tissue. Histologically, the specimen was composed of skeletal muscle with chronic inflammatory reaction with abundant foamy histiocytes and lymphocytes with presence of eosinophiles and small basophilic nodular structures, with concentric laminations: Michaelis-Gutmann bodies. (Figure 1) Periodic acid-Schiff (PAS) positive small nodular structures were found inside the histiocytes exhibited a targetoid appearance. (Figure 2) The lesion was diagnosed as Malacoplakia based on the histological features.



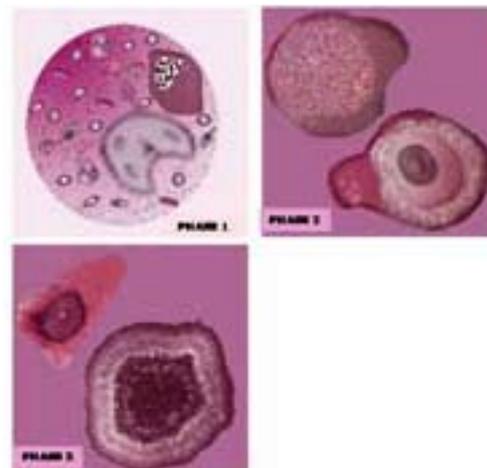
**Fig. 1.** Histological aspect with haematoxylin and eosin stain (40x). Black arrow shows Michaelis-Gutmann bodies.



**Fig. 2.** Periodic acid-schiff (PAS) reaction showing concentric laminations inside the histiocytes. (Black arrow, 40x)



**Fig. 3.** Clinical aspect after 20 days of treatment with Vitamin C.



**Fig. 4.** Stages of development of Michaelis-Gutmann bodies. Phase 1: Black arrows show a cell with numerous phagolysosomes of different sizes and forms. White arrow shows phagolysosomes with intracellular bacteria partially digested. Phase 2: Illustrate Michaelis-Gutmann bodies developing within the phagolysosomes. Partially digested bacteria (white arrow) and with calcium deposition (Black arrow). Phase 3: The end-stage of Michaelis-Gutmann bodies. Typical concentrically lining or “owl-eye” appearance.

The patient continued the intake of antibiotic for a week and did not return to control because of living in a rural area far from the city and being of scarce economic resources. He did not make immunologic tests. Six months later he returned for follow-up, there was a decrease of the swelling in the tongue; he refused any surgical treatment and therefore was initiated with Vitamin C, 2 grs daily for 20 days. At that time, we observed a mass almost imperceptible. (Figure 3) The patient agreed to continue in periodic controls.

## Discussion

Malacoplakia is a disease that rarely involves the mouth. This is an extremely rare case that involves tongue and to the best of our knowledge this is the fifth case reported in tongue after revising the world literature since 1960 and the first one in Colombia. (4-7)

The first case in tongue was reported in 1985 (4) in a Caucasian 9 years-old male with a mass of similar characteristics to the described in this article, the second case was reported (5) in a patient of 68 years-old who was treated with vitamin C, bethanechol and clotrimoxazol, the third case was reported (6) as an asymptomatic lingual malacoplakia occurring in a 57 years-old female, diagnosed during a routine clinical examination who did not need any supporting medical therapy and the fourth case was reported (7) in a 98 years-old woman with a shallow ulcer in the base of the tongue.

Because the aetiology remains unknown, immune deficiencies are tested in these patients (1,3,4) like sarcoidosis, lymphoma, carcinoma, diseases associated to disturbances of the lymphocytes T or immunity mediated by cells, hereditary immunodeficiencies (1) or a history of immunosuppression due to some diseases or long-term therapy with systemic corticosteroids.

Different authors believe that the chronic inflammatory reaction in malacoplakia is an answer to an infectious agent. Electron microscopy studies found intracellular microorganisms forming the matrix of Michaelis-Gutmann bodies. These studies have also shown remnants products of an incomplete bacterial digestion inside the phagolysosomes due to a decreased intracellular cyclic guanosine monophosphate (cGMP) level. (8) Calcification is initiated in the central zone showing a concentric lamination (owl eyes). Inside the macrophage, one or several intracitoplasmatic basophilic round or sometimes irregular inclusions are observed with PAS stain. (Figure 4)

Viruses, fungies, bacteria, tuberculous bacilli and other infectious agents have been implied, but none of them show sufficient evidence to be considered causal agents. (1) Some of the most studied infectious agents have been E. Coli and Klebsiella because they have been cultured occasionally in urine, blood and clinical wounds. The serotyping of the E. Coli, does not show microorganisms with greater virulence. The anecdotic cases of focal

malacoplakia in chronic periapical periodontitis support the opinion that lesions characterized by macrophage accumulation facilitate the local condition for the production of Michaelis-Gutmann bodies. These could be related to local bacterial antigen load due to a pulpar necrosis. (9) Treatments with antibiotics, such as quinolones and trimetoprim - sulphamethoxazol combined with surgery have better results. Also the combination of surgery with bethanechol (cholinergic agonist) has been used, because it increase levels of cGMP; although vitamin C has been used with an insufficient number of patients as a treatment method, it has employed to increase the cGMP and cAMP levels in monocytes and has the ability to induce collagen fibril synthesis and tissue repair.

In this case report, socioeconomic and cultural environment variables, impeded carrying additional exams to verify the patient's immune status. Due to the same reason and because the commitment of the patient with his health was minimal, vitamin C, which was the treatment of choice does not represent any risk.

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