



(CASE REPORT)



Understanding testicular malakoplakia: A rare inflammatory condition and its clinical implications case report

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Abstract

Genitourinary malakoplakia (GUM) is a rare and chronic form of bacterial soft tissue infection, which primarily affects the genitals and urinary organs. The diagnosis is histological. Symptoms may include itching, pain during sex, bleeding, abdominal pain and burning during urination. Although treatment usually includes antibiotics, corticosteroids or anti-inflammatory drugs, and a variety of other medications and procedures may be needed to treat the symptoms. In some cases, surgery is also indicated.

Keywords: Malakoplakia; Genito-urinary; Michaelis-Gutmann; Testis

1. Introduction

Malakoplakia is a rare disease described at the beginning of the 20th century by Hansemann. This disease is part of the macrophage diseases, characterized by an acquired and local disorder of cellular immunity occurring in the context of coliform infection. Histologically, it is characterized by the presence of Hansemann cells and Michaelis and Guetmann bodies. Contrary to this anatomical-pathological specificity [1].

Testicular malakoplakia is a difficult condition to diagnose because it has no specific symptoms. Clinical and radiological examinations are often necessary to confirm the diagnosis [2]. Biopsy can also be helpful in diagnosing the condition and ruling out other possible causes.

The treatment depends on the severity of the symptoms and the presence of other diseases, and is essentially based on antibiotic therapy in the absence of definitive destruction of the testicle, in the opposite case surgery can be proposed [3].

We report the case of a young patient who was diagnosed with testicular malakoplakia and had an orchidectomy.

2. Case Report

A 24-year-old patient, with no medical or surgical history, was initially admitted for treatment of a large left bursa, which had been evolving for 3 years, without any associated signs, in particular no mictionnal burning or hematuria, all evolving in a context of apyrexia and conservation of the general state.

The clinical examination has revealed a left scrotal tumefaction, without inflammatory signs, and palpation revealed a firm, painless, mobile mass. The rest of the examination was normal.

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The patient initially underwent a biological workup, which showed no abnormalities, with tumor markers in the normal range,

An ultrasound of the scrotal contents showed the presence of a left multicompartmental tissue mass, containing anechoic and echogenic zones in places of supracentimetric size, not taking color Doppler, occupying the entire bursa (Fig1), without individualization of the epididymis, or the testicle.

This echographic aspect evoked in the first place a malakoplakia.

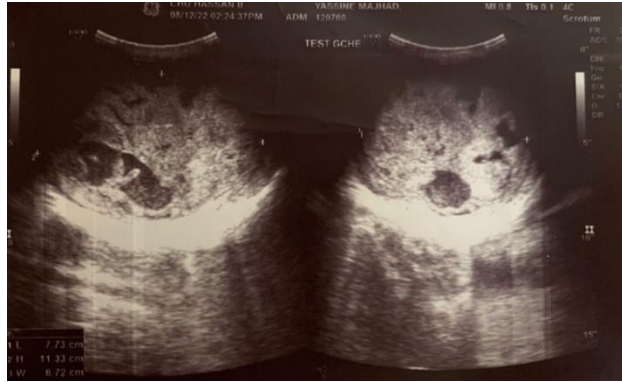


Figure 1 Ultrasound appearance showing heterogeneous, anechoic and echogenic images suggesting malakoplakia

An MRI of the scrotal contents was impossible to perform due to lack of means, we proceeded then to a surgical management, consisting in the realization of an orchidectomy by inguinal way, by respecting carcinological rules.

Frontal surgical exploration reveals a large, firm, rosary-colored mass with no inflammatory signs (Fig2).

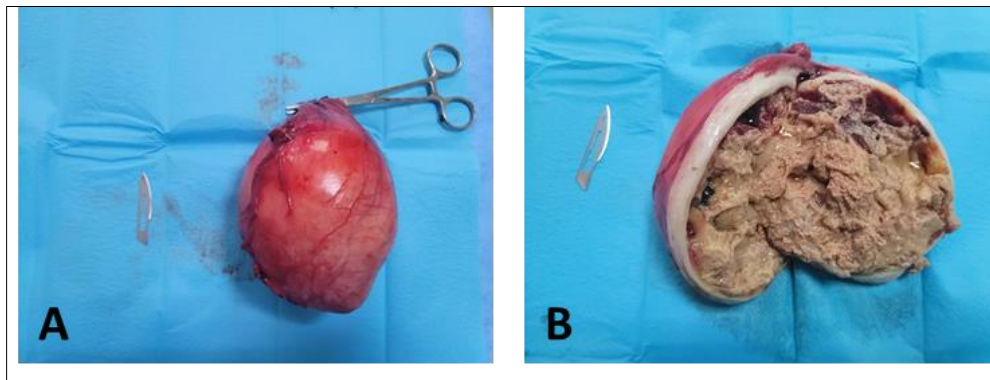


Figure 2 Image showing the macroscopic aspect of the piece before (A) and after opening (B)

Anatomopathological examination found the presence of granulomatous lesions, which are clusters of differentiated and enlarged macrophages containing iron inclusions called Michaelis-Gutmann bodies. These Michaelis-Gutmann bodies appear as circular or oval basophilic structures in the macrophages.

The evolution was favorable at 06 months of treatment without recurrence.

3. Discussion

Malakoplakia is a chronic multisystemic granulomatous inflammatory disease characterized by the presence of single or multiple soft plaques that may affect different organs [3]. It can occur at any age with an average age of 50 years at diagnosis and a female predilection. Pediatric cases are rare. It is more frequent in immunocompromised patients with a history of diabetes, transplantation, lymphoma, corticosteroid therapy or alcoholism. In the majority of cases (60-80%), malakoplakia affects the urinary tract (bladder, ureters and kidney) and cases of locoregional extension (retroperitoneal and lymph node) have been reported [4]. Involvement of other organs is possible and the

gastrointestinal tract (15%) is the second most frequent location (left colon, sigmoid, rectum, stomach). The other locations are rarer (genital, cutaneous, cervical, lingual, pulmonary and central nervous system) [5].

On the physiopathological level, malakoplakia appears to be due to an impaired immune response to bacterial infections. The phagolysosomal activity of macrophages and monocytes is diminished: they phagocytose bacteria without being able to digest them completely. The partially lysed bacteria accumulate in the cytoplasm and cause a reaction granulomatous reaction of the immune cells. The decrease in phagolysosomal activity may be due to a decrease in intracellular cyclic guanosine monophosphate (cGMP) levels [5-6].

The diagnosis of malakoplakia is difficult due to the lack of specific symptoms and characteristic imaging. Isolated testicular localization is rare with about fifty published cases [6]. The diagnosis is only made on the excisional specimen of a complicated orchitis that does not respond to the usual treatments or on a biopsy. Links with granulomatous orchitis have been suggested but the presence of Michaelis Gutmann bodies authenticates malacoplakia. Isolated epididymal involvement is even more exceptional with only 9 cases reported [7].

Differential diagnoses depend on the organ affected, primary or secondary neoplasia, inflammatory diseases (sarcoidosis, Crohn's disease), infections (tuberculosis, Whipple's disease) and skin mycoses [8-9].

Treatment is based on intracellular penetration antibiotics (Fluoroquinolones, trimethoprim-sulfamethoxazole). The duration of treatment has not yet been standardized. Pseudotumor forms require surgical removal [9].

4. Conclusion

Malakoplakia is a rare pathology. It is due to an acquired and local immunity disorder associated with a non-specific germ infection. Contrary to its histological specificity, genitourinary malakoplakia does not present a univocal clinical or radiological picture. The diagnosis is often misplaced and is only made on the anatomical-pathological study of the surgical specimen. The treatment of malakoplakia is essentially based on antibiotic therapy. Excision is performed only in case of total destruction of the affected organ.

Compliance with ethical standards

Acknowledgments

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Disclosure of conflict of interest

No conflict of interest

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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