

Case Report
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Cutaneous Nocardiosis in Immunocompetent: About a Case in Morocco

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Introduction

Nocardiosis is a granulomatous and suppurative infection, localized or disseminated, which generally results from inhalation of the germs and, more rarely, contamination of a wound. It affects mainly the immunosuppressed subjects [1].

Therefore, it seems interesting to present a case of cutaneous Nocardiosis in an immunocompetent patient to pay attention to its existence in Morocco.

Case report

A 35-year-old man from northeastern Morocco, consulted for a painless swelling slightly itchy located on the knee and inguinal level evolving for 6 months with alteration of the general state. The cutaneous examination revealed a tumor of the right knee about 15 cm in diameter, of firm consistency, surmounted by multiple ulcero-budding nodules whose surface was covered with melliceric and hemorrhagic crusts (Figures A,B). An homolateral inguinal adenopathy was noted about 5 cm, fistulized in places (Figure C). The biological assessment was correct. An MRI of the knee objectified a tumoral process depending on the muscle and the skin, extended from the lower 1/3 of the thigh to the proximal end of the tibia. The ultrasound examination showed a large inguinal lymphadenopathy. The diagnosis evoked were sarcoma and infection disease like tuberculosis. In histology of skin we found a granuloma with micro-abscesses organized around small grains, with basophilic periphery and slightly eosinophilic center, and they are composed of fine, short, spindly filaments sometimes entangled on the periphery Gram (+) pointing to nocardiosis (Figures F, G,H, I, J). The mycological examination was negative and bacteriological specimens were found on direct examination filamentous branched Gram-positive bacilli, acido alkalo resistant in favor of Nocardia. The complete hemogram, liver, renal functions and immune status were found to be normal. An examination of human immunodeficiency virus, syphilis, hepatitis B, and hepatitis C proved negative. The diagnosis of lymphocutaneous nocardiosis in immunocompetent was retained. The extension assessment was normal and the patient was treated with oral cotrimoxazole (800mg/j) while one year with regression of lesions (Figures D, E). No recurrences was objectified after 20 months.



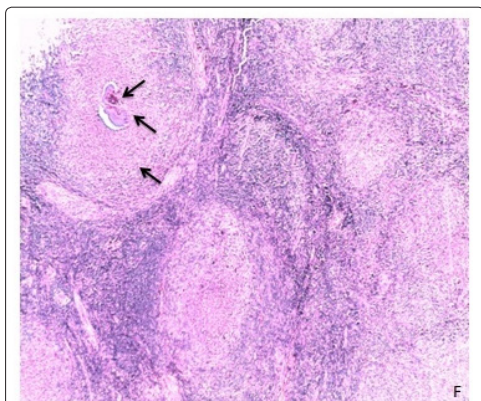
Figures A, B : a tumor of the right knee about 15 cm in diameter, of firm consistency, surmounted by multiple ulcero-budding nodules



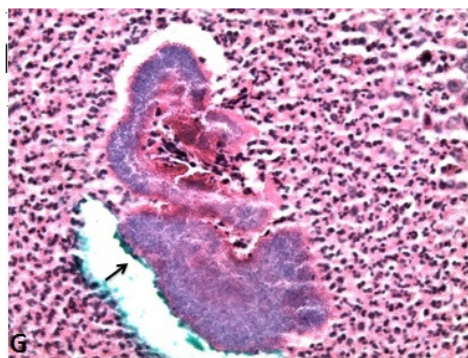
Figures C: inguinal adenopathy, about 5 cm, fistulized in places



Figures D, E: of control after one year of treatment



G x 50, HES coloring: in the dermis: microabscess made of PNN around grains, surrounded by histiocytic crown.



G x 400, HES Staining => Details of the grain
Small grains, +/- multilobulated ovoids with basophilic periphery and slightly eosinophilic center

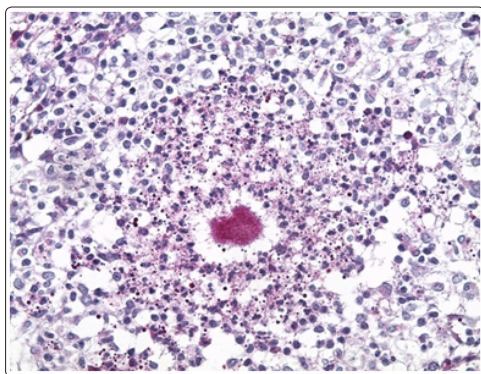


Figure H: PAS staining (+), G x 400



Figure I: Gomori Grocott staining (+) G x 200

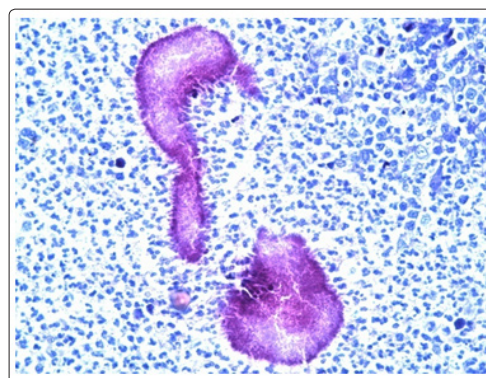


Figure J: Fite Farraco staining (+) G x 400

Discussion

Nocardiosis is a rare infection in humans, caused by bacteria of the genus *Nocardia*, ubiquitous in the environment, especially in terms of water, soil, plants and dust. They are strict, filamentous, pleomorphic and septate aerobic gram-positive bacilli. Currently 107 species of *Nocardia* are identified, especially thanks to molecular biology. Of these species, 33 of them are recognized as pathogenic for humans. They are mainly opportunistic infectious agents, most often reaching immunocompromised patients [2] such as transplant recipients, cancer patients or those with autoimmune diseases [3]. Nocardiosis is classified into pulmonary, systemic and primary cutaneous nocardiosis, this last form being the least common [4]. Cutaneous involvement in *Nocardia* spp. May develop as one of four types: superficial, mycetoma, lymphocutaneous or sporotrichoid, and systemic disease may occur with secondary cutaneous involvement. The brain is the most common non-pulmonary site involved in disseminated nocardiosis, the cerebral involvement of nocardiosis is sometimes asymptomatic and many authors recommend its systematic screening [5,6].

The MRI is useful but no specific and could cause a confusion for a sarcoma diagnosis, especially for young people in the knee.

The diagnosis of nocardiosis remains bacteriological, based on microscopy and isolation of the culture, but nocardia can be confused with mycobacterium, mainly because of their clinical and bacteriological similarity: growth on Löwenstein-Jensen medium (LJ), the presence of acid bacilli through Ziehl-Neelsen staining and colony morphology [7]. At the direct examination of nocardia, the bacilli are fine, branched, gram positive, the character of acid-alcohol resistance of *Nocardia* is absent by the technique of Ziehl-Neelsen classic, but partial with the modified technique of Kinyoun [1]. The histology remains a diagnostic means often solicited as is the case of our patient. Histopathological

examination found abscessed pyogranulomatous lesions surrounded by PNN corona and a second crown of histiocytes in fibrous tissue, nocardia filaments are evidenced by Gram and Grocott staining [8]. The diagnosis of nocardiosis is a challenge for all healthcare providers for three reasons. Firstly, because pathologists are not usually familiar with the histopathological aspects of mycetomas. Secondly, the acid-fastness of the grains, considered of primary importance for the histological identification of *Nocardia* sp, is often difficult to ascertain [5, 9, 10]. Thirdly, *Nocardia* is difficult to cultivate, because positive results can only be obtained after 1–2 weeks [4]. For this reason, a biopsy of the lesion and assessment of grain acid fastness are the main diagnostic tools available to pathologists [4]. Microbiological culture and PCR techniques can improve species identification, but it is always advisable to obtain material for culture and to refer any suspicious aspect of the case to a mycologist so that appropriate culture techniques can be initiated [11].

Therapeutically, nocardia infection requires prolonged systemic antibiotic therapy for 3 to 12 months depending on location, severity. The trimethoprim-sulfamethoxazole combination is the most prescribed treatment given the sensitivity of several species to this treatment. However, the classic combination amoxicillin-clavulanic acid plus amikacin, imipenem plus amikacin or cefotaxime plus amikacin have been shown to be effective on several strains, then an oral relay by trimethoprim-sulfamethoxazole may be proposed [8].

Conclusion

Nocardiosis is a rare disease in immunocompetent, whose complex long-term treatment is poorly codified because of the scarcity of prospective studies to evaluate it. From diagnosis to treatment, the management of nocardiosis is complex. A long term treatment by trimethoprim-sulfamethoxazole allowed a good evolution in our case. This requires strengthening collaboration between microbiologists, anatomopathologists and clinicians. Further study of this infection is important to improve its management.

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