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Medical Management of an Ankle Mycetoma: About One Case

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Abstract: Mycetoma is a chronic, progressive and destructive disease which affects skin, subcutaneous and connective tissue, muscles and bones. It is usually found on foot, but any part of body could be involved. Infection probably results from traumatic inoculation of subcutaneous tissues with certain funguses or bacterias. The aim of this article is to study the medical management of one case of an ankle mycetoma. These were a 28 year's old married woman. She is Malian and lived in Bourem Fougas in the Northern part of Mali (sahelian area); she showed a wound on her right ankle. The patient presented a hot pain tumefaction associated with a flow of fluid on her right ankle. In biological examination, we noted an increase of both sedimentation rate (47mm) and C - reactive protein (81mg/l); hemogramm revealed an inflammatory type of anemia and a polyclonal Hypergammaglobulinemia. Biopsy revealed single and white-grain mycetoma (Actinomadura pelletieri). Two drugs were prescribed, cotrimoxazole and diclofenac. The dosage of cotrimaxole was 1920 mg a day for a period of 6 months, and the dosage of diclofenac was 100 mg a day for a period of 15 days. Mycetoma is a chronic subcutaneous infection of tropical region. Etiological agents were actinomycetes or fungus. Its frequency is high in sahelian area, but there is an issue of diagnosis. Functional and vital prognostic could be involved. The treatment is first medical. Then, severe cases require surgical treatment.
Keywords: Mycetoma, ankle, medical treatment.

INTRODUCTION

Mycetoma is a chronic, progressive and destructive disease which affects skin, subcutaneous and connective tissue, muscles and bones. It is usually found on foot, but any part of body could be involved. Infection probably results from traumatic inoculation of subcutaneous tissues with certain funguses or bacterias [1].

The first case of mycetoma published in the modern literature was back in 1694. This affection is widely known as "Madura foot", referring to a case which appeared in the mid-19th century in Indian city called "Madura" [1].

Mycetoma has several medical and socioeconomic consequences for patients, communities and health authorities. Mycetoma generally touches young men adult's aged 20 to 40 in developing countries. Low income group and manual workers as farmers, farm workers or breeders are the most affected. There is no accurate data about the incidence, prevalence and distribution of this disease over the world.

This chronic infection affects rural people living in arid tropical regions: it is a disease of poverty which was added to World Health Organization (WHO) list of "neglected tropical diseases" in 2013. Long and expensive medical treatment is not always effective when surgical treatment should be done in case of fungus origin [WHO 2016].

Mycetoma is known to have two distinct etiologies: these etiologies could be funguses or bacterias origins. Infections caused by funguses as Madurella mycetomatis lead to Eumycetoma, and for bacterias as Actinomadura madurae cause Actinomycetoma. Eumycetoma is more frequent than Actinomycetoma with the exception of American continent where the latter is dominant [1].

Microorganisms which are responsible of the disease are present over the world, but the majority of mycetoma cases are found in tropical or subtropical regions. An area called « mycetoma belt » extending from 15 degrees South latitude to 30 degrees North latitude. It includes several countries such as Brazil, Ethiopia, Mauritania, Mexico, Senegal, Somalia, Sudan, Chad, Venezuela and Yemen. The

most geographic areas affected are characterized by a short rainy season and long dry season.

Endemic area is situated in arid and tropical regions on different part of 15 degrees North latitude: Indian hearth, African hearth, and Mexican hearth. Etiologic agents live as saprophyte in external environment. They are sometimes transmitted during minor traumatisms and pass unnoticed. Spike bites contamination is the most classical circumstance. Patients are essentially rural and young adult. Male predominance is evident.

Mycetoma is known in West Africa years ago. In 1894, the first African case was reported by LE DANTEC in Saint-Louis (SENEGAL). Few years later, two etiologic species, madurella mycetomatis and actinomadura pelletieri were described from Senegalese blood samples. Recently, studies of Dakar schools broaching principles of clinical, radiologic, bacteriologic, mycological, anatomo-pathologic and therapeutic aspects of mycetoma have reported a major contribution in knowing the infection [1-4]. Years later, SEGRETAIN and MARIAT have showed different etiologic agents in external environment in Senegal and Mauritania [5]. In these last twenty years, new published articles were described that mycetoma issue remains in West African endemic area [6-9]. This region is found on different part of 15 degrees north latitude where Annual rainfall normally varies from 50 to 800 mm.

Etiological statistic was recently supplied by Niger and Mali. Two endemic countries where data was not available last years.

Transmission result in inoculation of causal agent with organism occurring during minor traumatism or penetrant wound and often by spike bites explain the high frequency of mycetoma in people who walk barefoot and manual workers. In endemic area, the disease particularly touches rural people who live barefoot, but no one is immune to this infection. No animal reservoir seems to be involved in transmitting the disease.

Biological diagnosis comprises several stages [10, 11]. The first stage is collection of grains or pus and their direct examination. Biopy also permits to reveal grains which precise the type of reaction around them. Grains are cultivated and only cultures allow getting specie diagnosis. Molecular biology is only performed by certain specialized laboratories; it compensates for inadequacies of conventional methods in identifying precisely etiological agent. Serology is used a little. All difficulties of biological diagnosing of mycetoma arise from the fact that one of these stages could be remained negative: non-emission of grain by fistula, lack of grains in tissue sample, sterile culture or lack of fructification which could allow identifying fungus agent.

The treatment of actinomycetoma is first medical [1, 13]. It should be extended, because certain failures are due to short-term treatment. For nocardia brasiliensis, Mexican authors have proposed an association of diaminodiphenyl sulfone and cotrimoxazole. The treatment duration should be appropriate to the clinical response. In case of mediocre response or spread risks due to localization, cures of amikacine from 2 to 3 weeks are proposed without extending 5 cycles. Another option is an association of amoxicillin-acid clavulanic from 3 to 6 months. Imipenem associated or not with amikacin have been proved to be efficient in refractory cases. In actinomadura pelletieri cases, cotrimoxazole administration for a period of one year provides satisfactory outcome. On the other hand, cases of A. madurae and S somaliensis are less responsive to conventional treatment. Bone effects are not contraindicated for medical treatment, but prognosis gets uncertain. The aim of this article is to study the medical management of one case of an ankle mycetoma.

OBSERVATION S

She was a 28 year's old married woman. She is Malian and lives in Bourem Fougas in the Northern part of Mali (sahelian area); she showed a wound on her right ankle.

The patient was dressing the wound on the spot in health center. After that, she didn't notice any improvement in her health. She then decided to come to Bamako and carry out an orthopedic consultation and exams.

After examination, an amputation was proposed to her, but she declined this offer. Afterwards, she inquired medical alternative and finally decided to come to military hospital of Kati.

The patient presented a chronic hot pain tumefaction associated with a flow of fluid on her right ankle. Moreover, patient general condition was satisfactory (fig. 1).

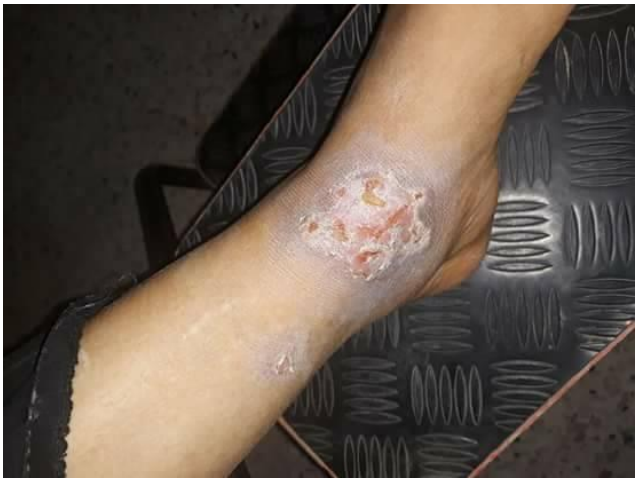

Fig-1: Wound healing after three months of treatment

In biological examination, we noted an increase of both sedimentation rate (47mm) and C - reactive protein (81mg/l); hemogramm revealed an inflammatory type of anemia and a polyclonal Hypergammaglobulinemia. Biopsy revealed single and white-grain mycetoma (Actinomadura pelletieri).

Two drugs were prescribed, cotrimoxazole and diclofenac. The dosage of cotrimoxazole was 1920 mg a day for a period of 6 months, and the dosage of diclofenac was 100 mg a day for a period of 15 days (a non-steroidal anti-inflammatory drug used in purulent hearth during a localized chronic inflammatory). Lastly, we made dressing of the wound.

The first line treatment is cotrimoxazole (800/160 mg per day), it should be maintained for a year. In case of lack outcome, it is recommended to add amikacin 15 mg/kg per day (alternative: streptomycin) for a three-week period. According to the outcome, we can renew cycles to a maximum of 4. Another alternative could be an association amoxicillin + acid clavulanic per os for a ten-month period. The response depends on specie involved; the best outcome is gotten through *Nocardia* sp and *Actinomadura pelletieri* [WHO 2016].

After three months of treatment, symptoms were clearly decreased. By six months, the patient is cured. The treatment was continued two months later. An appointment was scheduled each six months. On scheduled appointment, the patient didn't show any recurrence.

DISCUSSION

It is usually easy to evoke a mycetoma on the clinical aspect. It is a subcutaneous swelling of chronic and fistulized evolution. Fistulas sometimes emit pus containing grains that can be visible to the naked eye. Interrogation often reveals that the pus is intermittent; the patient can be seen for the first time during a non-productive period. The infection generally evolves for several years before the first hospital consultation. Swelling is predominant at foot in approximately 70% of cases.

The fistulised extra-podal mycetoma has clinical aspects sometimes less evocative than those localized on the foot. Other data associated with the clinical aspect will constitute arguments in favor of the diagnosis of mycetoma. The male predominance is expressed: 88 out of 122 (72.1%) in Mauritania [9], 90 out of 109 (82.5%) in Senegal [14], 45 out of 54 (83, 3%) in Mali [6], 106 out of 133 (79.7%) in Niger [17]. The most affected age group is between 20 and 40 years old. Cases observed before puberty are very rare. Finally, the interrogation looks for the notion of exposed profession and trauma. The patients are rural, mainly farmers and breeders. The notion of precise trauma is not always found [14]. There are few differential diagnoses of mycetoma. The most common in our experience is endemic Kaposi's disease. Other infections such as chronic osteitis fistulized to the skin, lymph node or articular tuberculosis, another subcutaneous mycosis may suggest this diagnosis clinically.

Mycetoma cannot be asserted without noting the presence of grains. Although visible to the naked eye sometimes, they will be highlighted by direct and/or anatomopathological examination. If there is emission of pus without visible grains, it is necessary to think of possibility of mycetoma due to Nocardia sp where the small grains will be able to be brought out only by the microscopic examination. This possibility is relatively rare in Sahelian West Africa where Nocardia sp accounts for only 2.7% of etiological agents in Senegal [15], 1.6% in Mauritania [14], and 9% in Niger [17]. Ultrasound has made a major progress in the exploration of none or fistulized swelling [18]. Images made by mycetoma are pathognomonic: single or multiple cavities with thick walls and without acoustic reinforcement; grains produce very reflective echoes.

Tumefactions of foot of other nature do not give such images.

Actinomycetoma must be distinguished from fungal mycetoma; this phase is fundamental, since treatment and prognosis will be completely different whether it is actinomycetoma or fungal mycetoma. Except Mauritania, results of large West African series showed that actinomycetoma predominates; in this case, priority should be given to medical treatment with antibiotics. Too often, mycetoma treatment is exclusively considered as the sphere of activity of surgery because the step of distinguishing the two etiological types is neglected. It is difficult to get rid of this misconception that reflects the well-established habits of the times when we were simply diagnosed mycetoma without specifying the causal agent, due to lack of knowledge and means to achieve it. The clinical presentation may already be a guiding element, especially the appearance of fistulas [14, 19]. Fistulas of black-grains mycetoma are flat or slightly raised, sometimes granulation tissue; the grains are easily visible, often numerous, "pointing their noses through the orifice of fistula". Mycetoma due to A. madurae is very different; fistulas are rare, flat and often dry. Fistulas due to S. somaliensis are also flat and often dry with small diameter. In contrast, mycetoma due to A. pelletieri has fistulas located on top of many multi-lobed and soft nodules. Pus flows spontaneously or it is easily obtained with vaccinostyle by scraping the top of the nodules. The red grains are abundant, but they are difficult to discern with the naked eye because of their small size. Another characteristic of these mycetomas is their tendency to give more extra-podal localizations than those due to other species; among these locations, they note the frequency of involvement of leg, buttock, knee, and trunk [15, 21]. According to our experience in West Africa, a few of mycetoma due to Nocardia sp that we diagnosed presented either highly inflammatory swelling with numerous nodules or swelling with rare flat fistulas. Of course, these clinical aspects are only tendency; red grains mycetoma apparearence should be the best defined. Orientation can also be provided by the patient's region of origin. S. somaliensis is an agent of desert regions like Atar in Mauritania or Agadez in Niger. It is exceptionally involved in Senegal where there is no desert. A. pelletieri is primarily found in the most rainfall area of mycetoma endemic zone where the annual rainfall varies from 500 to 800 mm. it is the case of certain regions of senegal (Thies, Diourbel) and Niger (Niamey southern part). On the other hand, this agent is rarely isolated in Mauritania where the annual rainfall is lower. If clinical aspects can orientate on etiology, absolute certainty can only be brought by data of direct examination of grains; anatomopathology and culture exam when it is possible to make it. Direct microscopic examination of fresh grains between slide and coverslip is simple; it can often distinguish fungal grains from actinomycotic grains. It may be necessary to use potash to lighten the preparation. We will record size, color, consistency of grains and presence of cement. Actinomycotic grains consist of filaments with diameter less than 1 µm, filaments of fungal grains have diameter of 2 to 5 µm with often vesicles. Pathological examination is particularly indicated when patient is seen at a stage of unproductive fistulas; it then allows to diagnose atypical forms such as non-fistulized forms. Culture is long and difficult to obtain; It seems advisable to us to make a biopsy even when there is emission of grains. Direct microscopic examination of grains allows getting an etiological orientation, but anatomopathological examination is more precise [22]. It is possible to make a diagnosis of specy or genus according to the sectional appearance of grains most frequently encountered in the West African endemic zone. This is the case of M. mycetomatis S. somaliensis, A. madurae, A. pelletieri. For Leptosphaeria, we can only give a diagnosis of genus; only culture with subculturing on poor environments would make possible to distinguish L. senegalensis from L. tompkinsii. These four species and Leptosphaeriare genus together account for about 90% of etiological agents in each of four countries in West African endemic zone. Small grains of Nocardia are easily recognizable, but we cannot distinguish different species from the appearance of grains. White fungal grains without cement may be due to Pseudallescheria Boydii, Acremonium sp and Fusarium sp. The fungal and Nocardias white grains are rarely found in the African endemic zone, contrary to what is noted in few statistics from the most rainfall regions of the western part of the continent such as Côte d'Ivoire or Nigeria where mycetoma is less common [23, 24]. The culture is the third stage of etiological diagnosis. actinomycotic grains are cultured on Lowenstein medium or blood, fungal grains on Sabouraud medium supplemented with antibiotics. In daily practice, cultures are rarely practiced, they are often negative because of superinfection germs that overrun cultures despite a prior washing of grain. Grains are composed of major dead filaments which also explain the frequent negativity of cultures. Finally, cultures are long to obtain, sometimes requiring subculturing on poor environments for precise identification of involved agent.

CONCLUSION

Mycetoma is a chronic subcutaneous infection of tropical regions. Etiological agents are either actinomycetes or fungus. Its frequency is high in Sahelian zone, but there is a problem of diagnosis. Functional or vital prognosis may be involved depending on location. The treatment is first medical. In extreme cases and as a second intention, surgical treatment could be carried out.

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