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Case Report

Giant cervico-facial mycetoma caused by Streptomyces somaliensis in a 14-year-old girl

Laouali Salissou[1], Saidou Mamadou[2], Rachid Sani[3], Nouhou Hassan[4]

[1]Department of Dermatology and Venereology, National Hospital of Niamey, Niamey, Niger, [2]Laboratory of Microbiology, National Hospital of Lamorde, Niamey, Niger, [3]Department of Surgery, National Hospital of Niamey, Niamey, Niger, [4]Laboratory of Pathology, Faculty of Health Sciences, Abdou Moumouni University, Niamey, Niger

Corresponding author: Dr Salissou Laouali, E-mail: danmata@yahoo.com

ABSTRACT

Mycetomas are inflammatory pseudo-tumors in multiple locations that affect the skin, the subcutaneous tissues and, sometimes, the bones. Their treatment depends on the type of parasite. Fungal mycetomas, also called eumycetomas, are treated mainly through surgery, while actinomycotic mycetomas are treated primarily with drugs. We report here the case of a 14-year-old girl afflicted with a giant cervico-facial mycetoma. The patient was born to poor and illiterate parents in a rural area of the Diffa province, at 1500 km from the capital city of Niamey. Histological examination of a biopsy specimen allowed a diagnosis of actinomycetoma due to *Streptomyces somaliensis*. The patient showed a remarkable sensitivity to ketoconazole, but she ultimately died due to a lack of sufficient medication.

Key words: Cervico-facial mycetoma, *Streptomyces somaliensis*, ketoconazole, Niger

INTRODUCTION

The mycetoma is a pathological condition in which fungal or actinomycotic exogenous agents produce parasitic buds [1]. It is a chronic infectious disease that mostly affects the foot and, more rarely, other parts of the body [2-4]. The rural population is most exposed to the infection due to small injuries sustained in contact with thorny shrubs harboring the infectious agents [3,5]. Gill, was first to recognize mycetoma as a disease entity in 1842 when he worked at a dispensary in the southern province of Madura [6]. The treatment is primarily surgical in the case of fungal mycetomas and medicinal in the case of actinomycotic mycetomas. Both types of treatments are problematic, having rather variable results [5,6]. This paper reports on the case of a 14-year-old girl afflicted with a giant cervico-facial mycetoma due to *Streptomyces somaliensis*. Despite a favorable development was observed, the disease, unfortunately, lead to the death of the patient after the parents, because of poverty and neglect, interrupted the treatment.

CASE REPORT

The patient, a girl aged 14, was born in a rural area of Niger at 1500 km to the east from Niamey, the capital city, to poor and illiterate parents. The parents brought her to medical attention after trying during four years an unspecified traditional medication. When we examined her in April 2011, she had a giant swelling at the cervico-facial area that has deformed the right side of the face, with sores and crusted lesions on the eyelids. This swelling reached the posterior cervical area and formed a block with an occipital swelling. The whole affected area was covered with watery sores that emit yellowish buds. A yellowish purulence was oozing from the right ear (Fig. 1). The remainder of the physical examination turned out nothing remarkable. Radiography of the head made from the face, showed diffuse sclerosis and osteoporosis on the skull and the right maxillary in particular (Fig. 2). An anatomical and pathological examination was carried out on a tissue sample after a hematoxylin and eosin (H&E) staining. This examination showed in a PNN

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Figure 1: A view of the swelling before treatment: from the face (A), the left profile (B), the right profile (C) and from the back (D).

infiltrate several greyish buds of actinomycetes from the *Streptomyces somaliensis* species, thus confirming the actinomycotic mycetoma diagnosis (Fig. 3). The pre-therapeutic assessment (notably transaminases, NFS, creatinine and glucose tests) revealed nothing remarkable. In May 2011 the treatment was started based on ketoconazole at 200 mg per day. The girl's father declined hospitalization, opting for an outpatient regime. We examined the patient after three months and noticed an improvement, despite an array of malnutrition symptoms. In particular, most lesions have dried and healed (Fig. 4). The same tests as in the pre-therapeutic assessment were conducted but turned out nothing remarkable. The ketoconazole treatment was renewed at 200 mg per day and an examination appointment was set for December 2011. Unfortunately, the patient died in June 2012, after six months without medication.

DISCUSSION

Mycetomas mostly affect the feet or other parts of the lower limbs. However, the manifestations of the disease on other body parts are the result of a direct inoculation or are due to parasite metastasis [7]. People are generally ignorant of contamination risks following injuries against shrubs, and this is the case for our patient, who was aged 14, in spite of the fact that she and her parents live in an area known to have a lot of thorny shrubs [5,6]. The clinical diagnosis of mycetomas is usually easy, but other chronic conditions must be ruled out, such as other deep fungal infections, tuberculosis and benign or malignant tumors [6,8,9]. Generally, people in Niger have such confidence in traditional treatments that when these fail, they tend to think that medical treatments, too, will fail. As a result, they do not seek treatment or come in only when it is very late [3]. In the case of our patient, the parents waited four years before coming to the hospital and this led to the development of a tumorous swelling with multiple sores, as is characteristic of the actinomycotic forms of the disease [10,11]. Mycetomas are prevalent

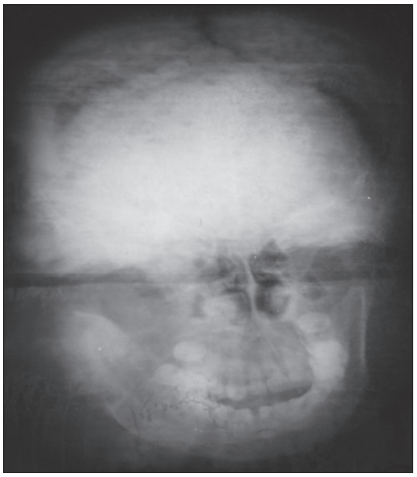

Figure 2: Diffuse sclerosis and osteoporosis (noticeable despite the poor picture quality) in the entire head skeleton (skull and jaws).

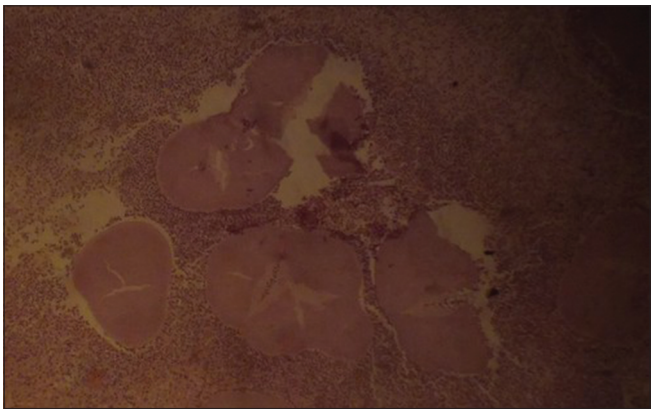

Figure 3: H&E stain (x40) of a PNN infiltrate: Numerous greyish buds *(Streptomyces somaliensis* organisms*)*.

in Niger, both in their fungal and actinomycotic forms [5]. Pathological and anatomical examinations are necessary in order to specify the pathological agent, as noted by some authors [6,12-14]. In our case, these examinations allowed the identification of *Streptomyces somaliensis* organisms. *Streptomyces Somaliensis* appears to be equally prevalent in Niger, Mauritania and Yemen, when compared with other actinomycetes [2,7], but it is absent in other areas despite the important variations in their geography [4]. Just as is the case with our patient,

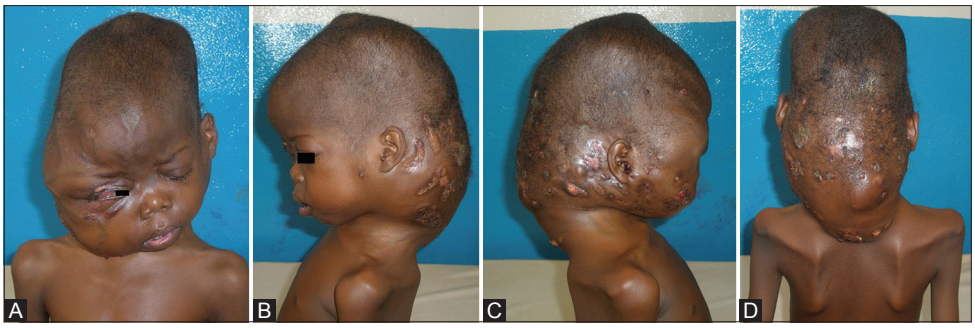

Figure 4: Improvement after a 3-month treatment based on ketoconazole (drying and healing of the lesions) from 4 perspectives (A : Face, B : Left profile, C : Right profile, D : Back).

Streptomyces Somaliensis seems to affect areas other than the foot, in particular the facial and cervical areas [15,17]. Its parasitic effects on the bone are, as is the case with our patient, sclerosis and osteocondensation [15,16]. *Streptomyces Somaliensis*, just like *Nocardia farcinita*, has become resistant to certain antibiotics, which explains the failure of many treatment protocols [18,19]. Despite the fact that the imidazole family of compounds is not indicated for the treatment of actinomycetomas, a complete remission with ketoconazole was observed on a similar case of head and neck mycetoma caused by *Streptomyces somaliensis* [15]. Similarly, a favorable development was observed in the case of a knee actinomycetoma due to the *Nocardia otitidiscaviarum* after 4 months of treatment, as reported from the Comoros [20]. We believe that in our case, the favorable course observed after 3 months of treatment could have also lead to a complete recovery with the ketoconazole, had the treatment been followed for at least one year. Unfortunately, poverty and neglect decided on the case, and the patient died after six months without drugs.

CONCLUSION

We believe that at least a one-year antifongic treatment based on ketoconazole would be effective, no matter the size of a tumor. Given the endemic nature of the pathological fungus in mycetomas, an awareness campaign is necessary so that newly affected people seek early medical treatment. The imidazole family of compounds must also be tested in further studies for minimal one year, in order to confirm their effectiveness on actinomycotic mycetomas, especially those due to *Streptomyces somaliensi* organisms.

Statement of Informed Consent

Informed consent was obtained from the patient's father for being included in the study.

Written informed consent was obtained from the patient's father for publication of this article and any accompanying images.

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