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Journal of Otolaryngology-ENT Research

Case Report

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Cervical mycetoma: an infectious disease that behaves like cancer

Abstract

Introduction: Mycetomes are inflammotory pseudo-tumors that affect the skin, the subcutaneous tissues and sometimes the bones. Their treatment depends on the type of parasite. Fungal mycetomas, also known as eumycetomas, are treated primarily by surgery, while actinomycotic mycetomas are treated primarily with drugs.

Observation: we report the case of a fungal mycetoma with a rare location in a 13-year-old boy from Diourbel, which is an arid and poor area. The diagnosis of fungal mycetoma has been made clinically, confirmed by histology with the presence of black grains. Pan-endoscopy and cervical CT scan had made the extension assessment possible. Surgery was not possible, because of the extent and the depht of the lesion. The treatment consisted of taking Ketoconazole for 6 months but without success. Unfortunately the evolution ended in death.

Conclusion: Mycetoma is diseases that occur in endemic areas. Cervico-facial location is rare. The color of grain makes it possible to make the etiological diagnosis the treatment is surgical and almost carcinological. The dilemma remains the resecability of the lesion because of its location and its extension.

Volume 10 Issue 6 - 2018

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Received: November 01, 2018 | **Published:** December 04, 2018

Introduction

The mycetoma is a pathological condition in which fungal or actinomycotic exogenous agents produce parasitic buds.[1] The rural population is most exposed to the infection due to small injuries sustained in contact with thorny shrubs harboring the infectious agent. Clinically the disease is characterized by swelling, and sinuses in the affected part. Another characteristic feature of mycetoma is the formation of aggregates of the organism (grains) in the tissues, which are visible to the naked eye and are discharged through sinuses in the skin. The grains vary in colour, size and consistency depending on the causative agent.[2] 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. We report a 13 years senegalese boy with cervical fungeal mycetoma ; he has multiple sinus tracts drain granule-containing pus. The aim of this work is to report a rare location of fungal mycetoma and to discuss therapeutic difficulties.

Case report

We report the case of a 13-year-old male patient who received 5-year-old cervico-facial ulcer tumefaction, 6 months after mental trauma, and long-term herbal treatment. The condition began with a sub-mental nodule that progressively increased in volume, followed by fistulization with black grain emission. The patient was followed in a dermatology department where the diagnosis of fungal mycetoma was made. He had benefited from medical treatment for several months without success and given the cervicofacial location, he was sent to our department. The examination at the entrance found an inflammatory swelling under and sus hyoid with extension to the left mandible and the parotid, allowing welding pus with blackish grains (Figure 1). Nasofibroscopy found edema of arytenoids and ventricular bands with bilateral laryngeal hypomobility. The cervical CT injected (Figure 2) found a pseudotumoral inflammatory attack in contact with the right submaxillary gland, extended to the pharyngolarynx and thyroid damage, with lysis of the thyroid cartilage. During hospitalization, the patient had laryngeal dyspnea which required tracheostomy. A panendoscopy performed under general anesthesia found diffuse edema of the entire larynx and hypopharynx, with black grains on the mucosa.

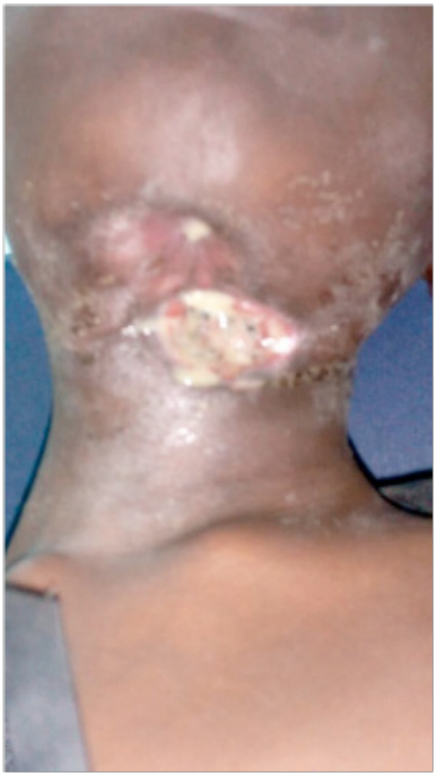

Figure 1 Ulcerated lesion with pus and black grains.

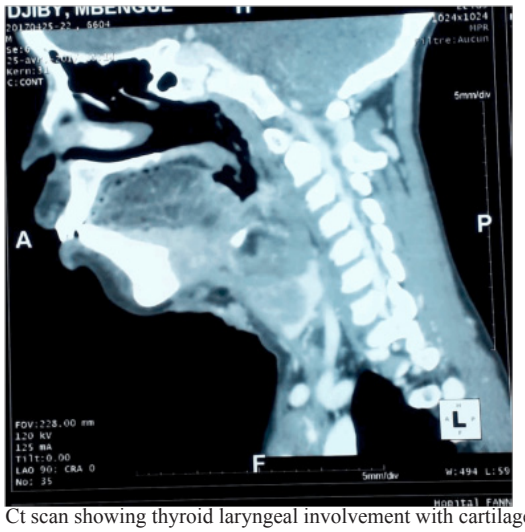

Figure 2 Ct scan showing thyroid laryngeal involvement with cartilage lysis.

There was also infiltrative stenosis of the esophagus, requiring the establishment of a nasogastric tube. Cervical and laryngeal biopsies were performed at the same time. The histological result concluded in a fungal mycetoma with black grains (Figure 3). Considering the extension of the lesion in depth she was inaccessible to surgery, in collaboration with the dermatologists, a medical treatment based on terbinafine was started on an ambulatory basis. We rehospitalized the patient six months later for hematemesis and epistaxis all in a context of general deterioration (Figure 4). After symptomatic treatment, the exit was done after two weeks, the death occurred one month after discharge.

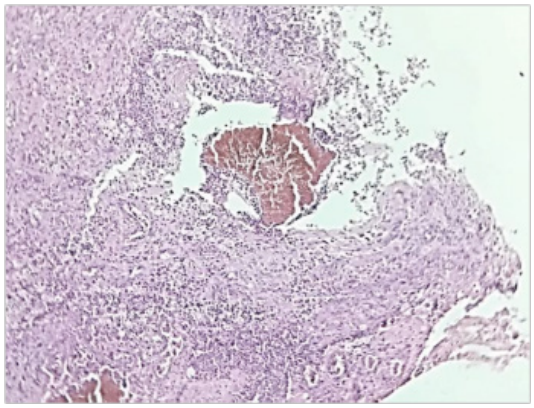

Figure 3 Histology mycetoma fungal granulum surrounded by pus magnification X1OO Hematoxylin eosin staining.

Discussion

Mycetoma is a pathological condition in which exogenous fungal or actinomycotic agents produce parasitic buds.[1] It is a chronic infectious disease that primarily affects the foot and, more rarely, other parts of the body.[2,3] Cervico-facial localization was discovered in 1857 by Lehert.[4] The rural population is the most susceptible to infection because of small wounds in contact with thorny shrubs harboring infectious agents.[5–7] Because the pathogens live as saprophytes in the soil, they are introduced during trauma or microtrauma often forgotten by the patient.[2] This was the case with our patient living in the region of Diourbel which is an arid zone with a notion of cervical trauma and a treatment based on bark. The incidence of mycetoma of the head and neck is low, the most frequent localization is podal and represents 70 to 90%, the ORL localization is very rare, thus on a series of 50 cases of extrapodal mycetoma reported by Dieng et al. only 1% concerns the head and neck.[7] Lynch found 18 out of 1860[8] cases, Gumma et al 15 cases out of 500,[9] whereas Sarr et al in 44 cases of black fungal mycetoma found only 2 cases with cervicofacial location.[10] The male sex is the most concerned as reported in the literature.[2,6,8] Clinically, the incubation is long ranging from several months to several years, explaining the delay of consultation, the signs are discreet at first, gravity type and pain at the point of inoculation, simulating granulomatosis or cellulitis . At an advanced stage the diagnosis becomes obvious in front of the black grain emission which is characteristic of a fungal mycetoma.[2,12] The biological diagnosis allows the direct examination of sampling, the cultivation of grains, but allows above all to determine the fungal species and make an antifongigram, it is an examination requiring a specialized laboratory and an informed biologist.[11,13]

Figure 4 6 month after medical treatment, persistence of black grains.

In our patient the mycological examination on Sabouraud mileu, was not contributive, therefore could not determine the fungal species; in front of the black color of the grains, the diagnosis of a fungal mycetoma is formal, the most involved germs in Senegal are Leptospheria Senegalensis, and Madurela Mycetomatis.[2,7,10] Histological examination is important at the early stage in a patient with no fistula or grain emission and thus allows the identification of grains. Whatever the localization, and the evolutionary stage, the locoregional extension assessment is capital and calls for the investigation of bone damage[2] in our patient we noted a bone lysis. Computed tomography and MRI perform well in the determination of the extension of the process in the soft tissues.[14] the ultrasound is performed at the beginning stage in the absence of visible grains, the granulomas and their capsules give characteristic images of gaps.[15] Panendoscopy remains important in the diagnosis of extension

Citation: Ahmed H, Ndiaye C, Mbaye A, et al. Cervical mycetoma: an infectious disease that behaves like cancer. *J Otolaryngol ENT Res.* 2018;10(6):406–408.
DOI: 10.15406/joentr.2018.10.00391

because it allows to explore all airways and give an idea of the resecability. The treatment is mainly surgical in the case of fungal mycetoma and is medical in the case of actinomycotic mycetoma.[16–18] In our patient the therapeutic difficulties are related to the atypical localization of the fungal mycetoma, its extension in depth to the larynx and the big vessels. If for the podal location, an amputation can be considered, we cannot make a surgical gesture because it would be mutilating and incomplete. Mycetoma surgery is indeed a true cancer surgery. Indeed, if after an intervention, there remain some grains, the recurrence or rather the resumption of the infectious process is inevitable. Antifungal treatment is actually used in addition to surgery and reduces the risk of recurrence; the protocol includes antifungal treatment with Ketoconazole (200 to 400 mg/day) for several months.[18] In our patient seen extension, the lesion was inoperable, and therefore the prognosis dark. In agreement with the dermatologists, a medical treatment based on terbinafine because of 1 tablet per day had been introduced for a period of twelve months, but this treatment was abandoned because of its high cost, explaining the continuation of the disease which is is ended by the death of our patient.

Conclusion

Fungal mycetoma of the head and neck region is a rare pathology of insidious installation; the diagnosis is often made when the lesions are major. The pathway of infection, susceptibility and resistance of mycetoma remains poorly understood and this is compounded by the lack of prevention and control measures. The hope of healing for these patients lies in education for health, raising their standard of living and especially the discovery of new molecules for the destruction of the fungus *in vivo*.

Acknowledgments

None.

Conflicts of interest

The author declares there is no conflicts of interest.

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