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A Case Report of a Misdiagnosed Mycormucosis: Uncommun Clinical Presentation

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Abstract **Case Report**

Background: Invasive mycormucosis is a rare opportunistic fungal disease with high morbidity and mortality rates, predominantly affecting immunosuppressed patients. An increasing incidence has recently been described particularly among corona virus epidemic. Dentists should be aware of this possible diagnosis instead of just instituting some useless and devastating treatments, and to ovoid the delay that can cost the patient 'life juste like our case. ***Clinical presentation:*** An unusual clinical manifestation of mucormycosis in a 42–year-old _man, diabetic who was referred by his Otorhinolaryngologist, to our dental surgery unit of the university hospital FARHAT HACHED Tunisia, suspecting a facial cellulitis related to dental infection. Before consulting his Otorhinolaryngologist, the patient was suffering from severe dental pain for two months, without any signs of relief despite dental treatments and the pain killers prescribed by his dentist**.** He was successfully treated by a combination of surgical intervention and antifungal treatment: Amphotericin B. ***Conclusion:*** Invasive mycormucosis is an emerging infection and is probably related to an increase in immunosuppressed patients. Accurate oral examination may lead to early diagnosis avoiding further damages of this agressive disease.

Keywords: Invasive mycormucosis, opportunistic fungal, immunosuppressed, case report, opportunistic fungal disease.

INTRODUCTION

Mucormycosis is a life-threatening angioinvasive infection caused by the ubiquitous filamentous fungi of the Mucorales order of the class of Zygomycetes.

It is a rare opportunistic fungal disease with high morbidity and mortality rates, predominantly affecting immunosuppressed patients. It represents the third most common opportunistic mold infection after invasive aspergillosis and invasive fusariosis. An increasing incidence has recently been described particularly among corona virus epidemic [1, 2].

CASE REPORT

A 42-year-old male patient diabetic, was referred from the ENT department, to our dental surgery unit of the university hospital FARHAT HACHED Tunisia. The patient was complaining from soft tissue swelling and filling of the left nasolabial fold that appeared one month ago.

Before consulting his Otorhinolaryngologist for the swelling,, the patient was suffering from severe dental pain in the upper left side for two months (one month before the apprerence of the swelling), without any signs of relief despite dental treatments: not indicated extraction of the upper left premolars and molars , and the pain killers prescribed by his dentist**.**

Intra-oral examination at this point, showed nothing special (Fig 2). Periapical radiograph showed insufficient endodontic treatment of the 23 (axial percussion was painful) (Fig 3). Based on the clinical examination, the diagnosis of facial cellulitis related to dental infection was suspected. Our patient has been already on antibiotics for 3 days without any improvement (prescribed by his Otorhinolaryngologist).

As complementary exams a panoramic radiograph and glycosylated hemoglobin value were demanded. Panoramic X-Ray showed complete unilateral opacification of the left maxillary sinus with osteolytic lesions in the Floor and alveolar ridge (Fig 4).

Based on these data, another diagnosis was suspected which is osteitis following sinusitis. Therefore, we asked for a CT scan for further exploration. Meanwhile, Extra oral discharge in the naso labial area was conducted by his otorhinolaryngologist.

The next day, the patient reported complaints from severe disseminated pain in the oral cavity. Intra oral examination revealed a violaceous edematous palatal soft tissue and a 2mm necrotic ulceration of the hard palate.

CT scan showed fluid filling the maxillary sinus, destruction of orbital floor and bone margins (Fig 7).

White blood cell counts were elevated, glycosylated hemoglobin was 12,8 in favour of uncontrolled diabetis.

Clinical and radiological confrontation was strongly suggestive of the diagnosis of rhinocerebral mucormycosis. Especially that the patient was a farmer.

Surgery was scheduled as soon as possible to avoid any further ostolytic and tissue lesions. Aggressive surgical debridement of the affected structures and restorage of the maxillary sinus drainage were conducted.

Biopsy of sinus mucosa was warranted to make the positive diagnosis. In additon to surgical debridement, control of underlying diseases, and systematic amphotericin B therapy were performed as soon as possible.

The patient was diagnosed with mucormycosis by pathological examination, and categorized under the rhinocerebral form by clinical manifestations.

When the wound healed without evidence of residual disease and the patient was clinically in stable condition, he was considered to have survived the disease and was discharged.

Fig 1: Extra oral view shows soft tissue swelling and filling of the nasolabial fold

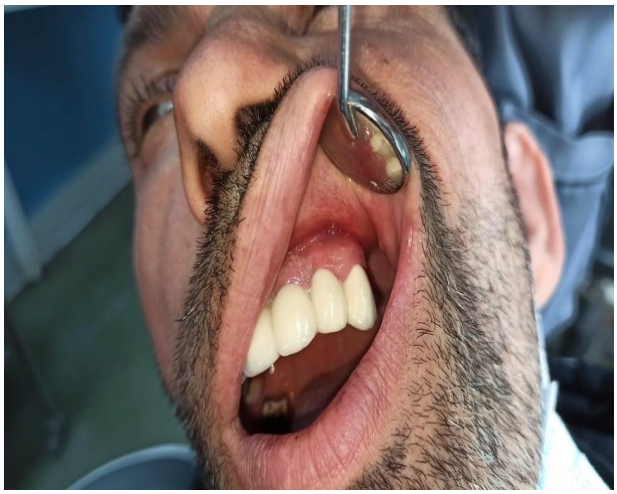

Fig 2: Intra oral view

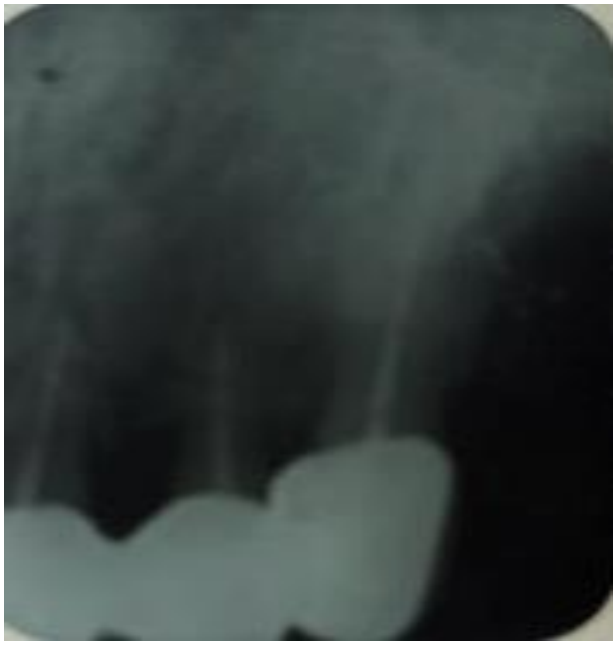

Fig 3: Periapical radiograph shows insufficient endodontic treatment of the 23

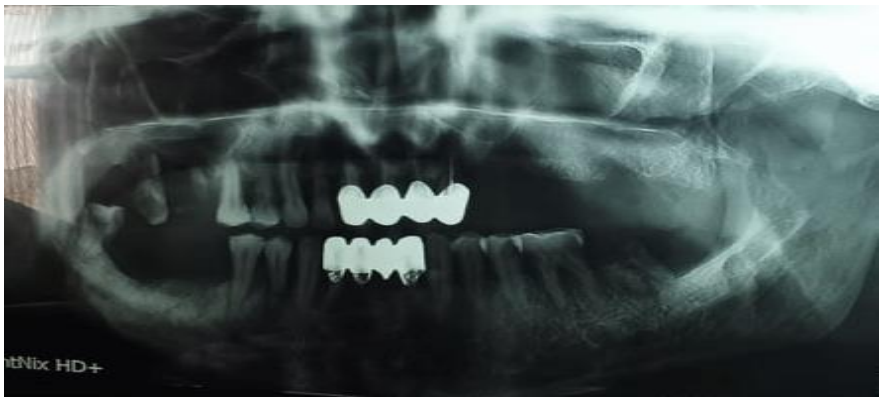

Fig 4: Panoramic radiograph shows complete opacification of the left maxillary sinus and osteolytic lesions in the Floor of the maxillary sinus and alveolar ridge

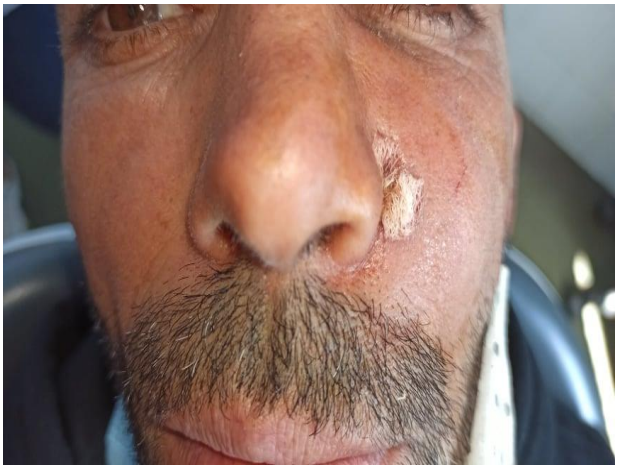

Fig 5: Extra oral drainage

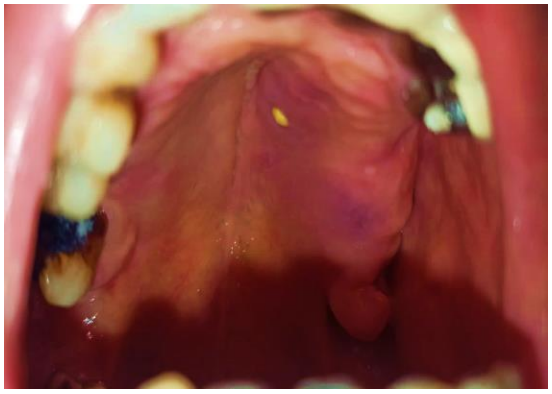

Fig 6: Violaceous infected tissue and a 2mm necrotic ulceration of the hard palate

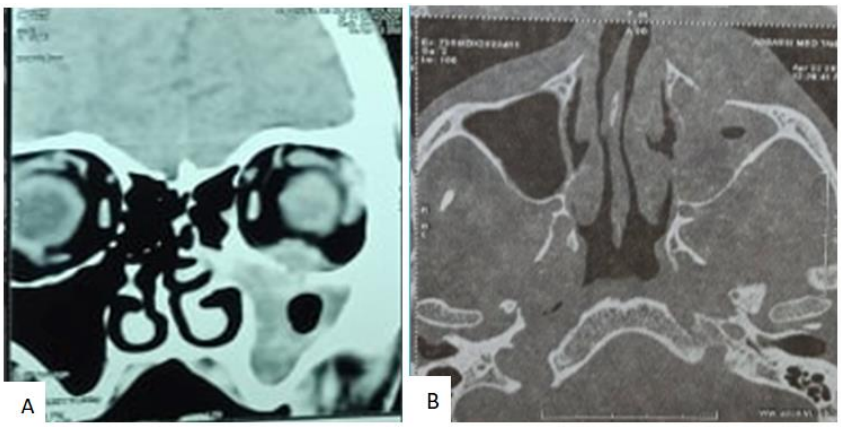

Fig 7: CT scan shows edematous mucosa, fluid filling the maxillary sinus, destruction of orbital floor and bone margins. A: coronal view, B: axial view

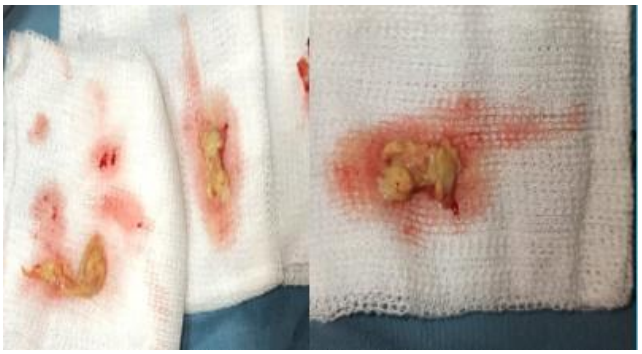

Fig 8: Necrotic tissue

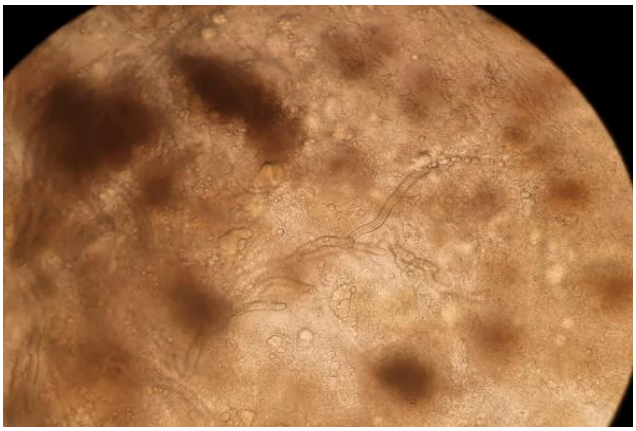

Fig 9: Direct microscopic examination

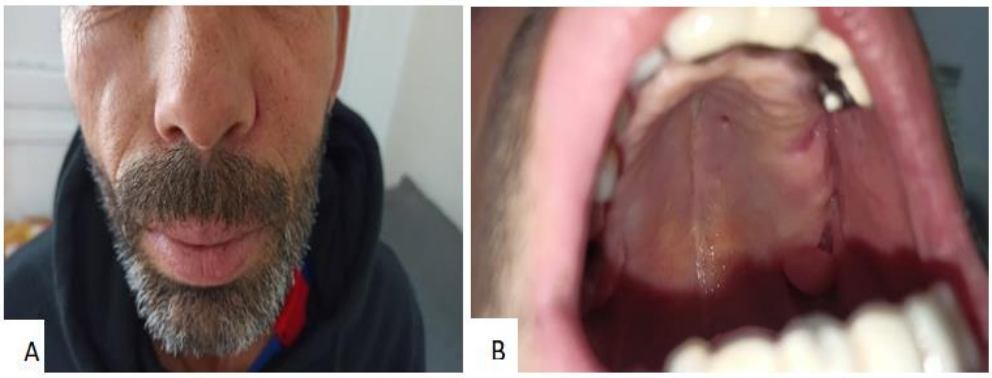

Fig 10: Control: one month later A: extraoral view, B: intraoral view

DISCUSSION

- **Epidemiology**

Mucormycosis represents the third most common opportunistic mold infection after invasive aspergillosis and invasive fusariosis, always occuring in immunocompromised patients. Although rare cases have been reported in apparently normal hosts [1-3].

It is hard to accurately determine the exact incidence of mucormycosis due to several factors such as the rarity of this infection and differences in the epidemiology between developed and developing countries. In US the annual incidence of mucormycosis has been estimated as 1.7 infections per million populations (500 cases per year) [1, 4]. unfortunately, this number seems to increase and it can be related to the rise of cancer incidence, the use of immunosuppressive therapies, including organ transplantations andthe resistance to the commonly used antifungal agents which result in increasing of the number of immunocompromised patients [5, 6].

- **Predisposing Conditions**

According to various studies, The most frequent predisposing conditions are malignant hematological malignancies with or without stem cell transplantation, prolonged and severe neutropenia, iron overload, poorly controlled diabetes mellitus with or without diabetic ketoacidosis, trauma, prolonged high-dose of corticosteroids therapy, HIV infection, neonatal prematurity and malnourishment. Antifungal agents with no activity against Zygomycetes [1, 7-10].

Clinical Manifestations

Mucormycosis is classified 6 major clinical forms based on its clinical presentation and anatomic site: rhinocerebral, pulmonary, cutaneous, gastrointestinal, disseminated, and uncommon rare presentations, such as peritonitis, endocarditis, osteomyelitis, and renal infection 8,11. Infection at these sites may be caused by any of the species of the Mucorales.

The mortality rate varied with [8, 12]:

*Underlying conditions of the host: 35% in patients with no underlying conditions, 44% in diabetics, and 66% in patients with malignancies

*The site of infection: 96% of patients with disseminated infections, 85% with gastrointestinal infections, and 76% with pulmonary infections, 62.5% in rhino-cerebral form

Rhinocerebral mucormycosis (ROCM) is the most common form of mucormycosis (39%) especially in patients with diabetes mellitus [8,10] particularly those with ketoacid.

In fact, Both mononuclear and polymorphonuclear phagocytes of normal hosts kill Mucorales by the generation of oxidative metabolites and the cationic peptides defensins. Otherwise, hyperglycemia and acidosis are known to impair the ability of phagocytes to move toward and kill the organisms by both oxidative and nonoxidative mechanisms.

Besides, patients with diabetic ketoacidosis have an elevation in available serum iron during diabetic ketoacidosis which may increase the susceptibility to mucormycosis 1. The infection develops after inhalation of fungal sporangiospores from the atmosphere which is the most common route for rhino-orbitocerebral and pulmonary infections.

The infection may rapidly extend from the paranasal sinuses into adjacent tissues. The fungus may invade the base of the skull and spread inferiorly to invade the palate, posteriorly to invade the sphenoid sinus, laterally into the cavernous sinus to involve the orbits, or cranially to invade the brain. This invading fungus may also disseminate through blood vessels to the central nervous system, giving the rhino-orbito-cerebal form, or everywhere in the body, giving the disseminated form [4, 13].

The clinical hallmark of mucormycosis is vascular invasion resulting in thrombosis and tissue necrosis.

The initial symptoms of rhinocerebral mucormycosis are similar to either sinusitis or periorbital cellulitis and it includes facial pain and numbness, eye pain followed by blurry vision and soft tissue swelling. Fever is inconstant ; A high white blood cell count, as long as the patient is immunocompetent [1, 14, 15]. Although, in our case, the patient had just dental pain for month without any other signs.

Infected tissue may appear normal during the initial phase then becomes erythematous, with or without edema, followed by the onset of a violaceous appearance, at a later stage, the blood vessels become thrombosed and tissue necrosis occurs resulting in the development of a black, necrotic eschar [1, 15]. Infection may extend to the oral mucosa and produce painful, necrotic ulcerations of the hard palate, sudden Tooth mobility and dental pain.

No reference standard for therapy has yet been established. It usually requires an antifungal treatment, surgical intervention and control of the underlying risk factors 4. The most active agent is amphotericin B (AmB). However, therapy with this drug have a high risk of toxicity and therefore alternative therapies are needed such as lipid formulations of AmB. Posaconazole is the second most active agent and has shown good results in vitro, in animal models and also in patients. Combination therapies with azoles or echinocandins may also represent alternatives to improve the survival of patients infected with mucomycosis [4].

CONCLUSION

Early diagnosis is very important because it allows quicker initiation of adequate management. Obviously, it relies on the patient promptly looking for medical help, alert suspicion of the disease by the doctor, and definitive confirmation of the diagnosis by the pathologist. Delayed diagnosis makes immediate, appropriate treatment impossible.

Declarations

Ethics approval and consent to participate: Not applicable

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Availability of data and materials: Not applicable

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Authors' contributions

AM, GB : Manuscript preparation
AM, GB, DJ : Manuscript editing
NBM, SBY : Manuscript review
All authors : Manuscript final approval

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