

CASE REPORT

Mucormycosis of the Maxillary Sinus in a Healthy Patient Probably Triggered by Dental Implant

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ABSTRACT

Mucormycosis of the maxillary sinus is a rare fungal disease that is characterized by uncontrolled and dramatic proliferation. The disease is an uncommon fungal presentation in healthy individuals.

We describe a 36-years-old female benign *Rhizopus* colonization at the maxillary sinus probably following dental implantation. The patient complained of chronic rhinorrhea. The ENT examination revealed unilateral chronic sinusitis involving the left maxillary sinus, which was confirmed by CT scan. Direct mycological analysis combined to inoculation of sinus fluid drainage on SDA medium isolated *Rhizopus spp.* as causative agent of sinusitis.

Laboratory tests were unremarkable, with no immunosuppression evidence. Unrecognized or inadequately treated mucormycosis has certainly an acute fatal outcome. However, if the disease is detected and treated early, sinus involvement will be limited to the paranasal sinuses and nasal cavity. After surgical debridement, our patient had uneventful postoperative recovery. One year after surgery, she had no clinical and radiological evidence of disease. This case of maxillary sinus mucormycosis is unique in that the patient was healthy and not immunosuppressed.

KEYWORDS

Fungal sinusitis; Mucormycosis; *Rhizopus spp.*; Dental implantation; Maxillary sinus

INTRODUCTION

Mucorales are considered as ubiquitous, and opportunistic organisms widely present in the environment, air, soil, food, animal wastes, and compost [1]. They can infect many sites, namely sinonasal, lung, gastrointestinal tract,

nervous system, and skin [2]. Immunocompromised individuals like diabetics are primarily afflicted by mucormycosis, while immunocompetent hosts can rarely be affected [2]. A literature search analysis showed that Mucormycosis infections in healthy individuals are distributed in 35 countries around the world [1]. Fulminant mucormycosis of maxillary sinuses following dental extraction in patients with uncontrolled diabetic have been described [3], the disease has a rapid and extensive spread, and should be considered as the most acutely fatal fungal infection [2]. Sinuses involvement could be limited to paranasal sinuses and nasal cavity if the condition is recognized and treated early [2].

All endodontic treatments, like tooth extractions, and dental implantations can induce fungal maxillary sinusitis [4].

If Amphotericin B is still a cornerstone in the treatment of fungal invasive infections, a surgical debridement is required in healthy individuals [5].

CASE REPORT

A 36-year-old female patient presented with one-year history of unilateral watery rhinorrhea.

The history revealed allergic rhinitis treated by corticosteroids, and she had also dental implant procedure next to her left maxilla one year ago.

On initial examination, left maxillary sinusitis was confirmed. The systemic physical examination was normal, without exhibiting any clinical signs of immunosuppression. Blood count and serological tests (HIV, HBs) were within normal range.

The diagnosis of Mucormycosis was established on the basis of the clinical results and confirmed by light microscopic examination of sinus drainage liquid.

On initial examination the patient had no history of physical trauma or burn suggesting a cutaneous route for fungal invasive infection. Sinus drainage fluid was addressed to the Laboratory of Parasitology-mycology of Oran university hospital for mycological assessment (Figure 1).



Figure 1: Sinus fluid drainage collection.

Cultures from sinus drainage on SDA medium yielded rapid growth of a fluffy grayish mold within 72 hours (Figure 2). Microscopic evaluation of the mold revealed aseptate pigmented hyphae, identified as *Rhizopus* specie,

characterized by a body of branching mycelia composed of three types of hyphae: Stolons, rhizoids, and sporangiphore (Figure 3 and Figure 4).



Figure 2: Direct examination showing broad hyphae, with right-branching angle.



Figure 3: Macroscopic aspect of the cultures, revealing fluffy white-grey colonies.



Figure 4: Microscopic features of *Rhizopus spp.*

CT scan was evocative of maxillary sinus Aspergilloma, with total filling of the left maxillary sinus and thinning of the wall.

The patient was managed by surgical debridement inducing a clear improvement. The clinico-radiological control examinations were unremarkable, with favorable evolution at one year of follow-up.

DISCUSSION

Mucormycosis is a rare opportunistic fungal infection caused by mucorales. Paultaufin first reported it in humans in 1885. For many years the term mucormycosis has been interchangeably used with zygomycosis in the medical literature, the most common species of medical importance are *Mucor spp.*, *Rhizopus spp.*, *Lichtheimia spp.*, and *Rhizomucor spp.* [6-8].

Mucorales fungi can infect many sites, namely paranasal sinuses, gastrointestinal tract, lungs, nervous system and skin [2]. Progression of initial sinus involvement to the orbit and brain is common, especially in immunocompromised individuals. However, the maxillary sinus mucormycosis is still a rare condition and sporadically infects a healthy host [2], some of the latest cases reported in literature are depicted in table 1.

Table 1: Cases reports of mucorales sinusitis involving healthy patients.

Cases Ref.	Age/Sex	Clinical Signs	Involved Sites	Underlying Diseases	Predisposing Factors	Follow-up
1	46/M	Respiratory Signs Headache	Right Maxillary Sinus	None	None	24
2	47/M	Diplopia Orbital Pain	Left Maxillary Sinus, Nasal Fossa	BPH*	None	60
3	54/M	Rhinorrhea Facial Pain	Left Maxillary Sinus	None	None	14
4	53/F	Rhinorrhea Orbital Pain Headache	Left Maxillary Sinus	None	None	20

*BPH = Benign Prostate Hypertrophy

Due to the rarity of this infection, it is difficult to calculate accurately its incidence [1], especially in healthy patients. The most described clinical presentation in immunocompetent patients is the cutaneous/subcutaneous form, followed by the rhino-orbital-cerebral patients and the genitor-urinary patients [1]. Our patient had no history of physical trauma or burn, and oral examination showed no ulcerations or necrosis.

Diabetes mellitus creates a suitable environment for the Mucorales growth [3]. Invasive Fungal Sinusitis due to Mucor Species was described in a patient on Ibrutinib [9], and recent findings are suggesting that patients who are debilitated by immunosuppression or COVID-19, might develop sino-orbital mucormycosis, although sometimes can happen in healthy patients [10].

A similar benign case induced by Mucor colonization at the sinus maxillary was associated to deviated nasal septum [10]. A patient reported a history of radiotherapy due to rhino-pharyngeal cancer occurred 10 years earlier, which might have made him more inclined to develop a chronic sinusitis facilitating Mucorales grafting [1], another case was described in well-controlled diabetes mellitus (hemoglobin A1c 6.9%) [9].

We do not know when and how our patient contracted this fungal infection. Usually, the upper aerodigestive tract is basically the portal of entry of multiple fungal species [10,11]. However, endodontics treatments like tooth extractions and dental implantations have been suspected of being one of the main sources of maxilla mucormycosis [4]. The principal predisposing factor is sinus mucosal manipulation during a dental procedure; this facilitates primary saprophytic colonization [4], particularly dental implants that partially extend into the maxillary sinus or nasal cavity are known to cause complications. Furthermore, patients with a predisposition to develop sinusitis are prone for complications after dental implant placement in the maxillary sinus area [4,12].

Usually, nasal endoscopy performed in maxilla mucormycosis of healthy patients demonstrated a variably inflamed mucosa with or without the presence of purulent exudates or showed necrotic tissues dotted with black eschar [1]. In addition, no biological abnormalities are detected in the benign forms.

Mucorales are considered as etiologic factor of mucormycosis infection. They are marked by the production of a coenocytic mycelium and the formation of asexual spores (sporangiospores) in a variety of fungal structures [1,8].

Histological literature review of mucormycosis cases showed numerous broad aseptate fungal hyphae, with right angle branching from 45° to 90°, in accordance with the morphology of the order Mucorales [1,8].

Benign Mucorales sinusitis among healthy patients is commonly due to the species, mucor and rhizopus [10]. Dysfunctional sinus remains a pure clinical phenomenon with lack of histological characterization [13], both radiological and mycological assessment should be considered as crucial.

Mucormycosis management still represent a big challenge based on different strategies, which provide diagnose, eliminate or reduce risk factors [1], the surgical debridement in our case was sufficient, in accordance with literature [5]. Maxillary fulminant mucormycosis following uncontrolled diabetes mellitus cases are successfully managed with either amphotericin B monotherapy or combined with posaconazole [3]. Dramatic course can occur if the disease is not treated early or if the underlying immunosuppression is irreversible [2].

Adjuvants therapies can be proposed for refractory or intolerant patients to polyene-based treatments (Posaconazole, recombinant cytokine granulocyte, granulocyte macrophage colony-stimulating factor or interferon-g, hyperbaric oxygen) [1].

As seen in table 1 the follow-up duration of sinus mucormycosis is included between 14 months and 60 months.

CONCLUSION

Maxillary mucormycosis Sinusitis following dental implant procedure is rare in healthy patients and should alert practitioners on the necessity of a mycological follow-up.

Learning Points

1. Mucormycosis is a rare, fulminant, and rapidly spreading fungal infection.
2. Usually, mucormycosis can occurs in patients with compromised immunity and it is rare in healthy patients, as illustrated in our case.
3. Sinusitis following dental extraction, or dental implantations may be mistaken for other benign sinusitis conditions because the similar clinic presentation
4. Due to the huge risk of fungal contamination, maxillary dental implantations should require a least one-year of follow-up.

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