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Case Report: The Diagnosis, Treatment And Outcome Of A Rare Case Suspected As *Mucormycosis*

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ABSTRACT

Background: *Mucormycosis* is an acute and rapidly progressing infection with high mortality rate unless identified and treated promptly and is caused by a saprophytic fungus. *Rhizopus oryzae* is the predominant pathogen and accounts for 60% of all forms and 90% of the rhinocerebral cases. The infection begins in the nose and paranasal sinuses due to inhalation of fungal spores. The infection spreads by direct invasion or through blood vessels. The fungus invades arteries leading to thrombosis that subsequently causes necrosis of hard and soft tissues. Often there is a history of extraction of a maxillary tooth with pus discharge from an unhealed extraction socket and exposure of necrotic bone or solitary palatal ulcer with exposed maxillary bone as a sole oral manifestation. The aim of this study is to introduce a rare case, with the associated clinical signs and treatment. **Case:** The focus case is 46 year-old immune competent female patient with the damage of gingival maxilla post dental extraction. **Result:** The patient was hospitalized and treated with injectable antibiotic but the swelling could not be reduced. After that trial, she received hyperbaric oxygen (HBO) treatment and had a good outcome. **Conclusion:** It is suspected that this infection was *mucormycosis* because of the typical clinical signs, the progressive course of the disease, the weak response to antibiotics, the location (in the maxilla) and the good response to Hyperbaric oxygen therapy.

Keywords: Post dental extraction, damage, maxilla, gingiva, immune competent, HBO.

Introduction

Invasive fungal infections (mycoses) are uncommon, but when they occur, they are devastating to patients. These infections are opportunistic, they occur when organisms to which we are frequently exposed gain entry to the body due to a reduction in the host defenses, or through an invasive portal, such as a dental extraction (Fogarty C *et al*, 2006).

Mucormycosis (*Zygomycosis, phycomycosis*) is an acute opportunistic infection caused by saprophytic fungus. *Rhizopus* is the predominant pathogen accounting for 90% of the cases of *rhinocerebral mucormycosis*. This microbe maybe cultured from the oral cavity, nasal passage, throat and stool of healthy patients without clinical signs of infection (Madan R *et al*, 2013).

The most common presentation in the head and neck region is maxillary and orbital cellulitis in a person with inadequately controlled diabetes mellitus and immunocompromised. Since *mucormycosis* occur infrequently; it may pose a diagnostic and a therapeutic dilemma for those who are not aware of its clinical presentation (Madan R *et al*, 2013). Pulmonary infection is the most common clinical presentation of this disease. The fact that all the patients had primary *rhinosinusoidal* involvement may reflect the fact that the referring physicians are more aquainted with the use of HBO for this particular site, probably associating it with the few reported cases *rhinocerebral mucormycosis* which were treated with adjunctive HBO. With respect to the delay in initiating treatment, it appears that HBO was initiated later than Amphotericin B (AMB) and surgery. This delay seems to indicate that HBO was used as the last resort in most of the patients (Covarrubias L.,G. *et al*, 2004).

This is a rare case with no known cause. In the case presented here the infection followed by an acute course which eventually caused necrosis of mucous gingiva lining the maxilla in which hyperbaric oxygen (HBO) therapy played an important role in the outcome of the case.

The Case Report

We received a patient as a referral from a private practice back in 2001 at the Oral Surgery Clinic in the Dr. Ramelan Naval Hospital Surabaya. The patient was a 46- year- old female and had a big swelling on her left cheek, which was very hard in consistency. This condition also accompanied by difficulty to swallow, facial paresthesia and trismus. The swelling was very painful and the patient could not sleep for days. Five days before she had undergone dental extraction of the second premolar in the left maxilla. She was in good health before the extraction, no systemic diseases and there were no complications from the dental extraction. One day after the extraction, she suffered severe pain on the cheek and had the above-mentioned symptoms. She came back to the dentist and was given oral antibiotic (Ciprofloxacin 500mg tabs, 3 times daily). Instead of resolving, the swelling enlarged and became more painful. On the left cheek, there was a 2 cm discolored area, which had a red and blackish complexion.

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Fig 1. Extra - Oral Image before HBO

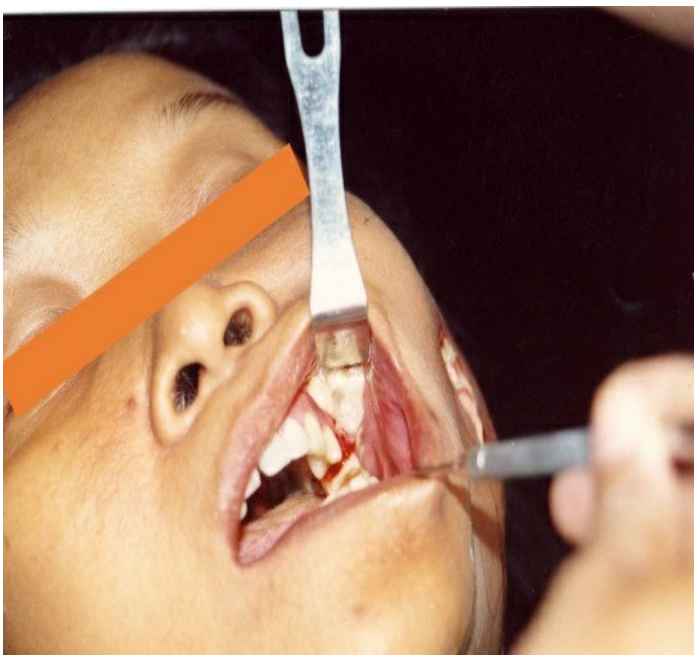

Fig 2. Intra - Oral Image before HBO

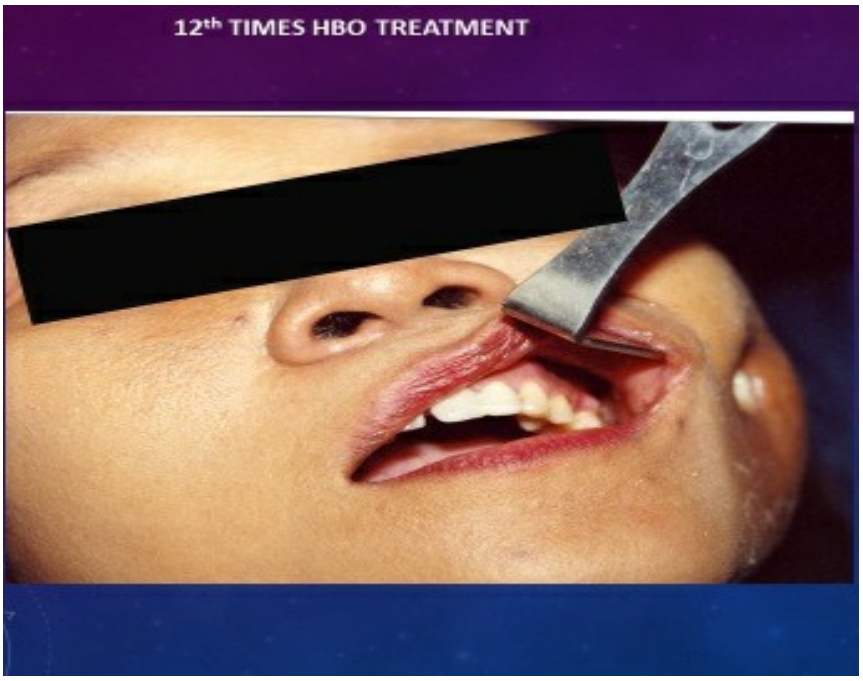

Fig 3. Intra oral and extra oral image after HBO treatment

Case Management

The patient had been hospitalized and treated with high dose intravenous antibiotics, among other drugs (pain killers, anti-inflammatory drugs) and had a microscopy, culture and sensitivity test on samples obtained from the swelling. The test indicated *Escherecia coli* on bacterial culture, which was sensitive to Cefotaxim. The panoramic photo at the time, found no abnormality. After four days of treatment, comprising of Cefotaxim 1gm iv injection td, combined with Metronidazole 500- mg, twice a day, the patient's condition did not improve (in the order of symptoms, swelling size and her vital signs) so HBO therapy was decided upon. The HBO therapy was given once every day in 12 consecutive days. Other drugs was provided to cope with clinical symptoms without antibiotic injection. After HBO treatment through a hole of 1 cm on the cheek at the red/blackish discolored area (because it was a fibrotic tissue), edema was reduced. HBO promoted wound healing with granulation tissues in the buccal and palatal mucosae which demarcated the necrotic area so a greater part of the involved tissue could be salvaged and restored to normal function. The patient felt very comfortable, did not complain of pain anymore and felt a lot better. She had plastic surgery six months after the treatment and overall made an excellent progress.

Discussion

In *mucormycormycosis*, aggressive spread of the infection is due to its angio-invasive nature causing thrombosis, ischemia

and necrosis of nearby tissue (West BC *et al*, 1995). The maxilla rarely undergoes necrosis due to its rich vascularity. Maxillary necrosis can occur due to bacterial infections such as osteomyelitis, viral infections such as herpes zoster or fungal infections such as *mucormycosis*, *aspergillus* among others (Auluck Ajit, 2007). In routine maxillofacial practice, intra-oral exposed bone infection (such as maxillary necrosis) is generally diagnosed as osteomyelitis. Pandey *et al* (2009) reported four cases with exposed bone infection, clinically mimicking bacterial osteomyelitis, but leading to a different picture on microbiological and histopathological examination. All the patients gave a history of previous trauma due to self-extraction of a tooth or injection (Pandey A *et al*, 2011). Bone necrosis can also occur due to extension of an infection from the gingiva to the bone (Auluck Ajit, 2007). Mohanty *et al* (2012), reported four cases of *rhinomaxillary mucormycosis* masquerading as chronic osteomyelitis. Bakathir (2006) also reported other two cases of *mucormycosis* of the jaw after dental extractions. Auluck (2007) reported a case of maxillary necrosis by *mucormycosis*. The patient had undergone extraction of the second and third molars six months prior due to her poor periodontal health. Following extractions, the socket never healed completely and the patient had persistent pain and discomfort for the following 6 months (Auluck Ajit, 2007).

During the first observation of this case, it is suspected that this was the Phlegmon access due to its symptoms and clinical signs. Therefore, the patient was treated with drugs in accordance with this anaerobic infection, culture and sensitivity test. When the patient did not make a significant improvement after 4 days of treatment, we decided to give the HBO therapy as the indication of HBO therapy among others is necrotizing soft tissue infection and soft tissue swelling. It was unclear at that time what the cause of this disease is until recently when there are many cases like this. After reassessing the case that occurred in 2001, it is presumed that this case might be one of invasive fungal infection (*Mucormycosis*). This hypothesis is based on the fact that it was an acute condition seeding from typical area in the maxilla near the maxillary sinus; it was consistently associated with fibrosis, was stimulated by dental extraction, and led to damage to the gingival mucosa, with a little response to antibiotic therapy. This case was a very rare case and the caring team was not familiar with its clinical presentation at that time (Laihad FM, 2010).

Based on literature review, it was found that many cases were in chronic condition where the infection had already entered into the alveolar bone and the only therapy for such cases was surgery, combined with Amphotericin B (AMB) and hyperbaric oxygen (HBO) therapy as an adjunct. Most patients were immunocompromised and just a few were immunocompetent. Invasive fungal infection is difficult to diagnose and therapy can be ineffective due to misdiagnosis (Brown GD *et al*, 2012; Alfano C *et al*, 2006).

Early diagnosis of this kind of infection is typically impractical and is challenging due to the following reasons: 1. Late presentation of the associated symptoms such as pain or fever, as fungal infections does not generally cause an inflammatory process; 2. Extensive tissue and bone necrosis tends to lead the physician to diagnose in terms of osteomyelitis, as it is the most common infection, unless being guide by culture results or by failure of antibiotic therapy; 3. In general, cultures of the fungus take approximately three weeks to confirm and such confirmatory diagnosis is delayed. Hence histopathology of scrape biopsy or tissue biopsy can be promising in early detection of fungi (Ahamed SK & Thobaiti YA, 2014). In this case, the damage affected the gingival mucosa of the maxilla in the buccal, palatal and inside of left cheek mucosae. By performing the above mentioned therapy, it is confirmed that antibiotic had a little response but a hyperbaric oxygen therapy gives positive outcome after the treatment.

Conclusion

This case reinforces the concept that simple procedures such as dental extractions can cause catastrophic complications in patients. It is important to understand how our treatment can affect patients. We must remain vigilant in our efforts to follow patients after we perform procedures to be certain that appropriate healing occurs. Knowledge of potentially devastating complications can help to prevent the unfortunate consequences described here. In this case, accuracy and early diagnosis, prompt and aggressive therapy was essential to reduce the infection because *mucormycosis* can lead to high mortality and morbidity.

With invasive fungal infections like *Mucormucosis*, it is a difficult task to diagnose and treat appropriately. And because of its rapidly progressing nature, associated with its high mortality, the need for early identification and prompt treatment cannot be overestimated. In all the reported cases of *mucormycosis*, HBO therapy was used as an adjunctive therapy after antifungal treatment and surgery trial. In this case report, HBO played an important role as an initial treatment before antifungals and surgery was undertaken.

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