

Partial Maxillary Osteotomy In Paranasal Mucormycosis : A Case Report

Dr. Jyotirmay Chakrawarty¹

(Resident Surgical Officer, Dept of Oral and Maxillofacial Surgery , GDC Indore and MY Hospital) ,

Dr. Ankita Barodiya

(Resident Surgical Officer, Dept of Oral and Maxillofacial Surgery , GDC Indore and MY Hospital) ,

Dr. Janhavi Lahane

(Resident Surgical Officer, Dept of Oral and Maxillofacial Surgery , GDC Indore and MY Hospital),

Dr.Srishti Shrivastava

(Postgraduate Trainee, Dept of Orthodontics, KIDS, Bhubaneswar),

Dr.Mayank M. Vagadia

(Postgraduate Student, Dept of Prosthodontics , GDC Indore)

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I. Introduction

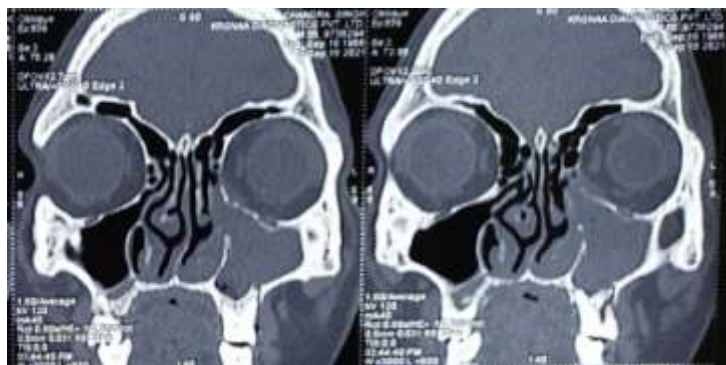
The coronavirus disease 2019 (COVID-19) infection caused by the novel severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) may be associated with a wide range of disease patterns, ranging from mild to life-threatening pneumonia. A wide variety of bacterial and fungal co-infections may exist and may be associated with already existing morbidities (diabetes mellitus, hypertension). The disease even after its cure, is producing wide array of complications such as Mucormycosis, which itself is a grave disease that is one of the leading causes of morbidity and mortality of the patients associated with it. This disease is usually caused by numerous Phycomycetes organisms of the Eumycetes(true fungi) which include *Rhizopus*, *Mucor* and *Absidia*. The Rhinocerebral form of mucormycosis is of great significance and is characterized by the classical syndrome of uncontrolled diabetes, cellulitis, ophthalmoplegia and meningoencephalitis.¹ Here, we present a case report of a 55 year old patient suffering from Mucormycosis of bilateral maxilla recovering after COVID infection 40 days back.

II. Case Report

A 55 year old, male patient reported to the department of Oral and Maxillofacial Surgery, GDC Indore with a chief complaint of pain and mobility in all of his teeth since 20 days. The pain was gradual in onset, dull, aching , boring type which was continuous in duration and not associated with any triggering factors. The pain subsided with over-the counter medications. Patient also complained of numbness in the upper lip and the lateral side of nose bilaterally. On careful history, patient revealed that he was a known diabetic since 10 years(not on regular medications) and the random blood sugar at the time of presentation was 280mg/dl. The patient also gave the history of COVID infection 40 days prior to the presentation for which he was home isolated and took treatment by consulting a local physician over the phone.

Extraoral examination revealed numbness in the upper lip and lateral side of the nose on bilaterally. Intraoral examination revealed Grade II mobility extending from left first molar to right second molar for which was referred for extraction of the following teeth. The patient also complained of pain in edentulous maxilla and heaviness in both the sinuses. Based on the above mentioned findings, a clinical diagnosis of deep fungal infection , bacterial sinusitis and avascular necrosis of maxilla was considered.

Radiographic examination revealed opacification of left maxillary sinus. CT Paranasal sinus scan(fig 1) revealed nonhomogenous opacification of bilateral maxillary sinus (left more than right) causing obstruction of both osteomeatal units extending into middle meatus, sparing the ethmoidal, and frontal sinus causing destruction of walls of left maxillary sinus.



Immediately blood sugar levels were controlled with insulin. The patient was advised and administered amphotericin-B 1 mg/kg body weight/day intravenously, slow infusion over 4–6 hours for 2 weeks, after a test dose of 1 mg in 100 mL of normal saline. The patient blood urea and creatinine levels were monitored as the drug can cause renal toxicity.

III. Surgical Technique

Based on the above mentioned findings, a surgical plan of extraction alongwith anterior maxillary osteotomy followed by secondary closure with roller gauze dressing soaked in local Amphoterecin B gel was planned.

A crevicular incision given extending from Left second maxillary molar to Right maxillary second molar was given, extraction of all the teeth(fig2) to check the inter-radicular bone for necrosis was done. As the necrosis was seen following extraction of the teeth(fig-3) , a plan of Anterior maxillary osteotomy was done and the buccal and palatal flaps were reflected. As the maxilla was mobile, a straight fissure bur in a micromotor handpiece was used to make the necessary cut in the palatal region. After removing the necrosed maxillary bone fragment, the maxillary sinus cavities were debrided thoroughly(fig4). The surgical cavity was thoroughly irrigated with hydrogen peroxide and normal saline. Haemostasis was achieved and Amphoterecin B lipid emulsion packed in the cavities alongwith a roller gauze dressing, followed by 3-0 Silk sutures to hold the dressing in place. The necrosed segment(fig5) along with the lining was sent for histopathological examination which confirmed the diagnosis of Mucormycosis.(fig6)



Fig 2



Fig3



Fig 4



fig5



fig6

IV. Discussion

During the advent of novel coronavirus disease, the prevalence of paranasal mucormycosis is on the rise, probably due to systemic immune alterations due to Coronavirus infection.¹⁰ This may also be contributed to the fact that the medications used to prevent the fulminant spread of Mucormycosis namely Steroids are potent immunosuppressant agents which further decreases the patient's ability to fight off opportunistic infections. Also, debilitating conditions such as Diabetes mellitus (uncontrolled), predisposes the patient to these opportunistic infections and increase the morbidity manifold. Also, it is a known fact that these steroids have an effect of increasing the blood sugar levels through their actions on Gluconeogenesis leading to a vicious cycle of cause and effect.

Mucormycosis is the third invasive mycosis in order of importance after candidiasis and aspergillosis and is caused by fungi of the class Zygomycetes.⁴ The most important species in order of frequency is *Rhizopus arrhizus* (oryzae)⁴. Based on clinical presentation and the involvement of a particular anatomic site, mucormycosis can be divided into at least six clinical categories: (1) Rhinocerebral, (2) Pulmonary, (3) Cutaneous, (4) Gastrointestinal, (5) Disseminated, and (6) miscellaneous.¹³ Chakrabarti et al. observed that rhino-orbito-cerebral type (44.2%) was the commonest presentation followed by cutaneous (15.5%) and renal (14.0%) involvement in their retrospective analysis for ten years in India.⁵

Once the spores have penetrated the lungs or subcutaneous tissues, they are met by the first line of defence, mononuclear and polynuclear phagocytes.⁴ The phagocytes of the healthy host are able to kill the spores of Mucorales by generating oxidative metabolites and defensins (cationic peptides)⁴. Uncontrolled diabetes mellitus, because of ketoacidosis, can alter the normal immunologic response of patients to infections.⁷ Such patients have decreased granulocyte phagocytic ability with altered polymorphonuclear leukocyte response.⁷ In diabetic patients *Rhizopus arrhizus* produce the enzyme ketoreductase, which allows them to utilize the patient's ketone bodies.⁹ The increased risk of mucormycosis in patients with ketoacidosis may also be due to the release of iron bound to proteins.⁴

The term osteomyelitis is derived from the Greek word osteon meaning bone, myelo meaning marrow, and itis meaning inflammation. Nelaton coined the term "Osteomyelitis" in 1844.⁷ Osteomyelitis is inflammation of the bone that begins in the medullary cavity and ends in the periosteum involving the haversian system.⁸ Various factors are involved in the development of the disease such as trauma, surgical therapy, bacterial infiltration in blood (Bacteremia), fungal infection, and systemic diseases decreasing the host defence mechanism such as diabetes, malignancy, anemia, radiation, malnutrition, osteoporosis, osteopetrosis and Paget's disease.⁹ Osteomyelitis involving the facial bones is rare (due to great number of collateral blood supply), and involvement of maxilla is less common than that of mandible due to high vascularity and collateral supply.

As per the guidelines from the 3rd European Conference on Infections in Leukemia (ECIL 3), the **firstline treatment of mucormycosis is as follows**¹¹:

- (1) AmB deoxycholate
- (2) Liposomal AmB 5–10 mg/Kg
- (3) Posaconazole 400 mg bid
- (4) Combination therapy
- (5) Control of the underlying condition
- (6) Surgery
- (7) Hyperbaric oxygen therapy

2nd line of treatment is as follows:

- (1) Posaconazole 400 mg bid
- (2) Combination lipid AmB and caspofungin
- (3) Combination lipid AmB and posaconazole
- (4) Combination deferasirox not recommended
- (5) Maintenance therapy posaconazole

V. Conclusion

As said earlier, fungal osteomyelitis is more invasive than bacterial, if not diagnosed and treated earlier. In recent years, however numerous cases of even fulminant head and neck infection by this organism have been diagnosed, treated and cured. The current survival rate for Rhinocerebral disease in patients without any systemic disease is 75% with diabetes it reduces to 60% and with underlying diseases, about 20%¹. Pulmonary disease is almost fatal. But still, early recognition and prompt surgical and medical intervention has proven successful for majority of the patients with this infection and this case is no exception.

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