

MUCORMYCOTIC OSTEOMYELITIS-A CASE REPORT

**Dr. Ilayanila. C^{1*}, Dr. Yuvashree Murugan², Dr. Yogasri.A³, Dr. Janarthanan. P⁴,
Dr. Arunachalam. M⁵, Dr. Sathishkumar M.⁶**

^{1*} Postgraduate Oral And Maxillofacial Pathology And Oralmicrobiology Karpaga Vinayaga Institute Of Dental Sciences

²Cri Oral And Maxillofacial Pathology And Oralmicrobiology Karpaga Vinayaga Institute Of Dental Sciences

³Cri Oral And Maxillofacial Pathology And Oralmicrobiology Karpaga Vinayaga Institute Of Dental Sciences

⁴Cri Oral And Maxillofacial Pathology And Oralmicrobiology Karpaga Vinayaga Institute Of Dental Sciences

⁵Reader Oral And Maxillofacial Pathology And Oralmicrobiology Karpaga Vinayaga Institute Of Dental Sciences

⁶Head Of The Department Oral And Maxillofacial Pathology And Oralmicrobiology Karpaga Vinayaga Institute Of Dental Sciences

***Corresponding Author:** Dr. Ilayanila. C,

* Karpaga Vinayaga Institute Of Dental Sciences, Chengalpet , Tamilnadu-603308.

Abstract

Mucormycosis, which is also known as phycomycotic or zygomycotic, is less frequent in the mouth. It is, yet, an aggressive, invasive, and potentially fatal fungal infection. It primarily targets people who have diabetes and altered immune system¹. The mutation causes the virus to continuously change its characteristics, such as virulence, pathogenicity, clinical symptoms, and the rate at which the disease spreads³. The disease usually progresses quickly, and diagnosis is frequently delayed, Life-saving surgical and medical intervention is necessary.² The fungal infection that typically affects the maxilla is also affecting those afflicted with the coronavirus illness 2019 (COVID-19) in this recent pandemic scenario¹. In this case study, we describe the unusual circumstance of a 48-year-old male who developed mucormycosis in his maxilla. The case report includes a brief review of the literature along with clinical findings, radiographic examinations, and histological information. Since mucormycosis is an aggressive fungal infection, treating it with a multimodal approach might lessen an individual's morbidity and mortality as well as systemic liability.

Keywords: Mucormycosis, mutation, systemic liability, multimodal approach, coronavirus 2019, fungal infection.

INTRODUCTION

In the global environment, the coronavirus illness 2019 (COVID-19) has resulted in about one million deaths. Despite the current vaccination system, an appropriate supportive struggle against COVID-19 heavily relies on care. The cause of COVID-19, known as severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2), is primarily linked to a range of opportunistic bacterial and fungal infections. It can also be related to preexisting comorbidities, such as diabetes mellitus, lung disease, or any infections acquired in a hospital. Aspergillus and Candida are the two major microorganisms thought to be the significant coinfection in COVID-19 patients among fungal infections. In addition to these fungal infections, mucormycosis is currently seen in a higher proportion of COVID-19-affected persons, particularly in India¹. The most common types of mucormycosis in patients who are immunocompetent are cutaneous and soft-tissue mucormycosis, which usually develops following skin disruption driven about by traumatic damage (such as that caused by improvised explosive devices in war zones, natural disasters, auto accidents, or iatrogenic origins), burns, or surgery. Characteristic facades include eschars, dry ulcers, necrosis, skin swelling, and abscesses². comorbidities have the highest survival rates, while those with haematological malignancies, HSCT recipients, or extensive burns have the poorest prognosis. Disseminated disease, particularly to the CNS, is often linked to mortality rates exceeding 80%. Conversely, lower mortality is associated with localized sinus or skin infections, where early tissue-based diagnosis and surgical debridement can lead to a potential cure². Mucormycosis has a proclivity to attack ancestry vessels, making loss of consciousness from blockage in vein or artery and tissue loss as a result. Angioinvasion concede possibility exist supported by the endothelial cell

association accompanying fungal spores⁴. The competence to treat mucormycosis efficiently depends on the chance of the surgical approaches and antifungal drugs².

CASE REPORT:

A 49-year-old male patient first reported to our department with the chief complaints of painful swelling in the left side of the face for past 3 months. Patient gives history of swelling which is of sudden onset, slowly progressed to attain its present size. Also gives history of pain which is dull, throbbing and continuous in nature, aggravates during movements and on mastication. Medical history revealed that he was diabetic for the past 5 years and is under medication. Insulin insulated 15-15-10-8 s/c. Patient had undergone FESS surgery before three months. Past dental history revealed that Patient underwent uneventful extraction before 3 months and consultation before 10 days. Patient is a known alcoholic for past 25 years.

Extraoral examination revealed a single diffuse facial swelling of size 4x5 cm seen on the left middle third of the face extending superiorly up to left infraorbital line, anteriorly 1 cm in front of left ala of nose, inferiorly 1 cm above left border of mandible and posteriorly 2 cm before tragus. Blackish discoloration was evident over the swelling on skin. Intraoral examination revealed necrosed bone exposed in relation to 23,24 and 25, tenderness evident over right maxilla, obliteration of right upper buccal vestibule and no discharge was evident.



FIGURE:1



FIGURE:2



FIG:3- Mucormycosis involving palatal region

RADIOGRAPHIC INVESTIGATION:

Radiographic investigation computed tomography CT facial bone reveals Features which are suggestive of residual fungal sinusitis involving left maxillary sinus with extensive bone erosions involving left maxillary antral wall, left zygomatic complex, maxillary alveolus and hard palate with chronic osteomyelitis changes.



FIGURE:4

Based on history, clinical examination and radiographic finding, a provisional diagnosis of fungal osteomyelitis of left maxilla was arrived suggestive of mucormycosis.

Surgical approach was planned with extraction of I1 excising the frontal process of maxilla, ethmoid, palatine bone and zygoma. The entire half of maxilla along with the lesion to be removed by a procedure called subtotal maxillectomy.

HISTOPATHOLOGICAL EXAMINATION:

Gross pathological examination: Received a subtotal left maxillectomy specimen in three necrotic bony fragments. Largest fragment with attached four teeth measuring 6.5 x 6 x 4 cm. Another fragment with mobile one tooth measuring 4.5 x 2.5 x 2.5 cm and smallest fragment measuring 3.8 x 3 x 2cm. On serial slicing shows grey brown friable necrotic material. Also received in the same container multiple friable grey black material amounting to 2cc.

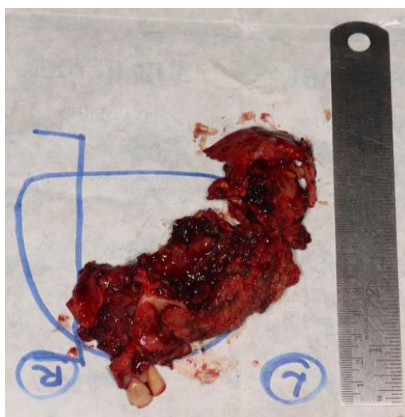


FIGURE:5

MICROSCOPY:

Necrotic tissue: Sections studied show fragments of extensive necrotic tissue and bony spicules enclosing numerous broad aseptate ribbon like hyphal elements exhibiting right angle branching, at places surrounded by fibrous tissue with dense infiltration of neutrophilic collections, lymphocytes, plasma cells, foreign body giant cells and few foamy histiocytes. Admixed are seen few fragments of exuberant granulation tissue rimmed by hyperplastic squamous mucosa exhibiting reactive changes. Angioinvasion is also evident.

Bony fragment: Decalcified sections studied show bony trabeculae with empty lacunae without osteoblastic rimming. Interspersed fibroconnective tissue shows necrotic debris with scattered numerous fungal elements of above-described morphology.

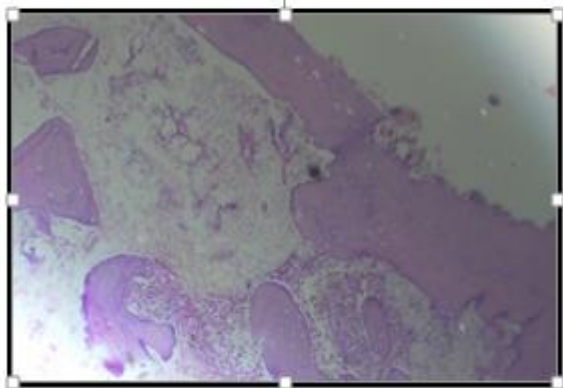


FIGURE: 6A (4X) - Extensive necrotic tissue and bony spicules

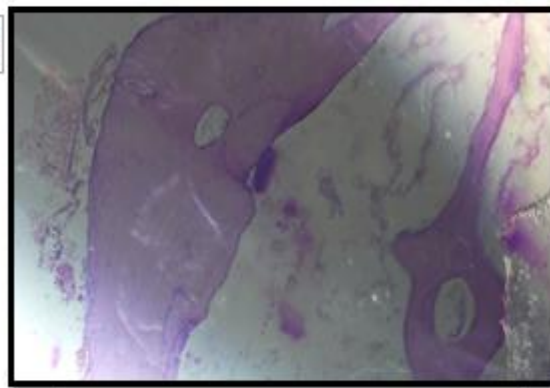


FIGURE: 6B (4X)- Bony trabeculae with empty lacunae without osteoblastic rimming

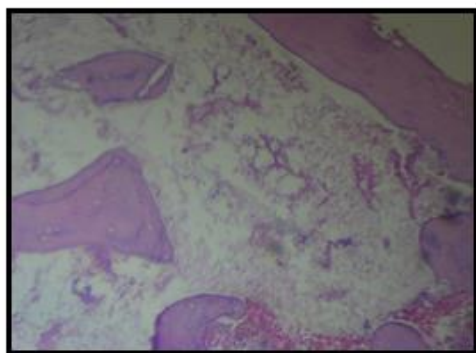


FIGURE :7 (10X) – Bony trabeculae and hyphal elements

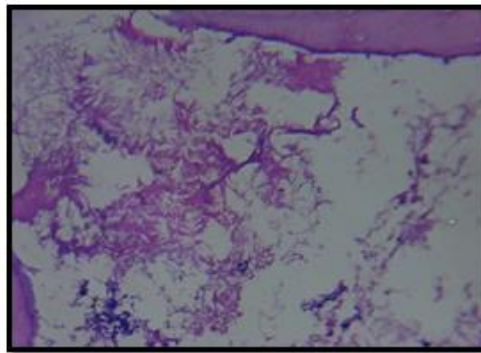


FIGURE: 8 (40X) - Numerous broad aseptate ribbon like hyphal elements exhibiting right angle branching, at places surrounded by fibrous tissue with dense infiltration of inflammatory cells.

Histological features suggestive of MUCORMYCOTIC OSTEOMYELITIS-Left maxilla was given as Final diagnosis.

DISCUSSION

Mucormycosis is a fungal infection that are induced by fungal organisms. It is commonly an opportunistic disease of fungal origin that most usually imitates individual who are immunocompromised, uncontrolled diabetes, lymphomas, leukaemia's, renal deficiency, organ transplant, corticosteroids, immunosuppressive therapy and exquisite in strong individuals, and at the rundown, COVID-19 is again considered in view of this list that causes mucormycosis directly or indirectly¹. The rhino cerebral type of mucormycosis is more prevailing in diabetics accompanying ketoacidosis. The sinuses are the most conspicuous fundamental beginning of connection, and they can even stretch to the revolution, brain, and bony process. In diabetes-associated pulmonary, mucormycosis, or disseminated contaminations are rare⁴.

In worldwide synopsis, the incident concerning this fungal contamination changes from 0.005 to 1.7/heap state and that is 80 opportunities containing regard to India, flag stoning habit for India to have the chief stated cases of mucormycosis that is known as diabetes capital of the world. Concerning the Indian synopsis, diabetes mellitus is most important universal related risk component, but haematological malignancies and organ transplant are the risk determinant in the diverse provinces¹.

The nose and/or maxillary sinuses seem to be the primary locations where respiratory tract infections occur in immunocompetent people. Larger than 10 mm sporangiospores have the potential to remain isolated in the

upper respiratory tract, taking a particular form of sinuses. If not, they may colonise the pulmonary system's distal alveolar spaces. If the infection in question is not identified and treated right away after it has colonised the nose and paranasal sinuses, it may spread to the base of the skull through blood vessels and then circulate to the central nervous system, resulting in the rhino-orbito-cerebral form of the infection, or it may spread throughout the body, resulting in the disseminated form⁵.

In the initial stages of the fungus's spread, the diseased tissue may appear normal upon clinical evaluation. After that, the infected tissue goes through an erythematous phase, either with or without edema, before taking on a violaceous appearance. Eventually, a black, necrotic eschar appears as a result of the thrombosis of the blood vessels and tissue infarction. When illness directly extends from the maxillary sinus and affects the distribution of the sphenopalatine and larger palatine arteries, it typically results in palatal involvement. Oral ulceration is preceded by pain and swelling, and palatal perforation may arise from the tissue necrosis that follows. Sometimes an infection from the sinuses can spread to the mouth, resulting in hard palate perforations and painful, necrotic ulcerations⁵.

The primary factors that promote the growth of fungal spores in individuals affected with COVID-19 are hyperglycemia, hypoxia, diabetic ketoacidosis, high iron levels, and decreased steroid-mediated, phagocytic, or other pre-existing comorbidities. There have been reports of hyperglycemic state in COVID-19, SARS-CoV-1, and pneumonia unrelated to SARS-CoV-1 cases. In individuals infected with COVID-19, lymphopenia is typically observed. In cases of severe infections, high viral replication stimulates the inflammatory response and the influx of neutrophils and monocytes into the bloodstream, leading to an imbalance between neutrophils and lymphocytes and an increased susceptibility to systemic fungal infections. Regarding the current instance, the patient has been taking medicine for diabetes for the previous five years¹.

Osteomyelitis is typically diagnosed in routine maxillofacial practice when there is intraoral exposed bone and maxillary necrosis. Trauma, bacterial osteomyelitis, herpes zoster, iatrogenic infections, or fungal infections such as aspergillosis and mucormycosis can all result in maxillary necrosis⁵. A pulmonary CT scan is advised for patients with suspected pulmonary mucormycosis and a haematological malignancy in order to detect the reversed halo sign, vessel occlusion on CT pulmonary angiography, or an area of ground glass opacity surrounded by a ring of consolidation on thoracic CT. To find out if sinusitis is present in diabetic patients with facial pain, proptosis, ophthalmoplegia, recently diagnosed amaurosis, or both, cranial CT or MRI is highly advised. In the event that sinusitis is identified, endoscopy is highly advised in order to identify mucormycosis. Because MRIs have significantly higher sensitivity than CT scans, they should be used instead of the latter if disease of the eye or brain is suspected. It is highly advised to perform a biopsy if mucormycosis is suspected. After a patient's mucormycosis has been established, moderately strong recommendations are made for cranial, thoracic, and abdominal imaging studies to assess the extent of disease and rule out underlying malignancy. Given the speed at which mucormycosis progresses, weekly CT scans are highly advised, especially for patients who are unstable².

Broad, irregularly formed, nonseptate hyphae with right or obtuse angle branching are typically seen in the histopathology of mucormycosis, but they can also be highly valued in the presence of PAS or silver spots. The organism is typically located close to the necrotic vessel walls, particularly in the vicinity of the necrosis area. For additional confirmation, utilize PAS and Grocott's methenamine silver stains, which typically display wide, aseptate, irregular, and ribbon-like folding of fungal organisms and branching at an obtuse angle. The PAS staining reveals magenta-coloured hyphae at obtuse angles and a filamentous organism that resembles a ribbon¹. Mucorales hyphae differ from septate hyaline molds by virtue of their wider hyphae and atypical branching patterns. Nevertheless, no information is available to indicate how well Mucorales can be distinguished from other molds using these traits. Therefore, at centre's where such assays are available, it is highly advised to confirm the diagnosis of mucormycosis in tissue by culture or by applying molecular or in-situ identification techniques².

On culture and microscopy, it is highly advised to culture specimens in order to identify the genus and species and to test for antifungal susceptibility. Before culturing, tissue homogenization should be avoided. Separate incubation at 30°C and 37°C is highly suggested. It is highly advised to perform direct microscopy using fluorescent brighteners on clinical specimens, primarily concentrating on septation, branching angle, and hyphal width².

Effective treatment of mucormycosis is dependent on the availability of surgical methods and antifungal medications². The affected area will heal more quickly if early intervention is combined with the right medical and surgical techniques, including full debridement and clear tissue margins. In the current COVID-19 scenario, using radiographic aids like OPG, MRI, computed tomography scan, and appropriate clinical examination will also assist in providing patients with the right treatment care¹.

CONCLUSION

Hence to conclude, the case report in this journal reveals the blackish discolouration that was evident over the swelling on skin. Intraoral examination revealed necrosed bone exposed in relation to 23,24 and 25, tenderness evident over right maxilla, obliteration of right upper buccal vestibule and no discharge was evident. CT facial bone reveals Features which are suggestive of residual fungal sinusitis involving left maxillary sinus with extensive bone erosions and hard palate with chronic osteomyelitis changes. Gross examination reveals serial slicing showing grey brown friable necrotic material. Histological features suggestive of mucormycotic osteomyelitis-Left maxilla .and the procedure done in this case is subtotal maxillectomy.

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