

RHINOMAXILLARY MUCORMYCOSIS IMPERSONATING AS CHRONIC OSTEOMYELITIS: A RARE CASE REPORT WITH CONE BEAM COMPUTED TOMOGRAPHY AS DIAGNOSTIC AID

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Abstract. Background: Rhinomaxillary mucormycosis (RMM) is one of the deadliest and quickly spreading types of fungal infection in humans which typically starts in the nose and paranasal sinuses. Radiologically, RMM manifests as sinus opacification with mucosal thickening, involvement of nasal cavity and erosion of maxillary bone. A radiographic study is necessary to confirm a clinical suspicion of RMM. Cone Beam Computed Radiography (CBCT) offers comprehensive details regarding the extent of the lesion and its impact on critical structures. **Case presentation:** This case report aims to highlight a rare case of RMM with osteomyelitis and the significance of CBCT in the early detection, management, and prognosis of RMM. **Conclusion:** CBCT plays an important role in early diagnosis of RMM for timely treatment and better prognosis.

Key words: mucormycosis, maxillary sinus, osteomyelitis

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INTRODUCTION

Mucormycosis is an invasive mycotic infection usually affecting immunocompromised patients and is the third most common mycotic infection globally [1]. It is caused by fungi called Mucoromycetes. Mucormycosis can be classified as disseminated, pulmonary, cutaneous, gastrointestinal, or

rhinocerebral – the most prevalent form. Depending on the tissues affected, the rhinocerebral form can be further differentiated into rhino nasal or rhino maxillary, rhino orbital, and rhino orbitocerebral [2]. Rhinomaxillary mucormycosis (RMM) can undergo intracranial extension by invading the internal carotid artery [3]. Thus, the diagnosis of RMM requires a detailed pa-

tient history to evaluate the medical condition of the patient and complete clinical examination for early intervention. Imaging plays an important role in diagnosing the disease and also to evaluate its extension. As bony resolution is high in Cone Beam Computed Tomography (CBCT) it can even detect minor osseous changes. Thus, CBCT helps to evaluate the osseous and the trabecular changes in the maxilla and the palatal bones more accurately. This case report highlights a rare case of rhinomaxillary mucormycosis mimicking maxillary osteomyelitis with a brief discussion on the role of CBCT as a diagnostic aid.

CASE DESCRIPTION

A 53-year-old male presented to the Department of Oral Medicine and Radiology with a chief complaint of dull, aching pain in the upper left back tooth region for 1 month associated with swelling and a history of pus discharge in the region. A history of intermittent nasal discharge from the right nostril for the past month was also reported by the patient. He had Type 2 diabetes with an HbA1c level of 6.2 and no history of COVID-19 infection or any other systemic condition. There were mobile teeth in the maxillary left

back tooth region, for which he underwent extraction 1 month ago.

There was no extraoral swelling noted. Intraoral examination revealed diffuse swelling on the buccal and palatal aspect from the 24-28 region, causing obliteration of the buccal vestibule and extending up to 1cm away from the mid-palatine raphe. On palpation, the swelling was soft in consistency and tender. There was irregular bone exposure at the alveolar crest with sharp margins in the 24-28 region resembling necrotic bone (Figure 1).

A CBCT was taken with a field of view of 20.3x17.6 cm, and reconstructions were made in coronal, sagittal and axial plane along with 3D reconstruction. Multiplanar reformation with reformatted OPG was also done (Figure 2), which showed missing teeth from the 24-28 region. There was mucosal thickening present in the left maxillary sinus, which resulted in a blocked ostium. The alveolar bone crest in the 24-28 region appeared to be irregular with bony spicules and altered trabeculae pattern. CBCT scan revealed a soft tissue mass in the left nasal fossa, causing a deviation of the nasal septum and extending into the left maxillary sinus and extending to the floor of the



Fig. 1. Exposed necrotic bone in the left maxillary alveolus

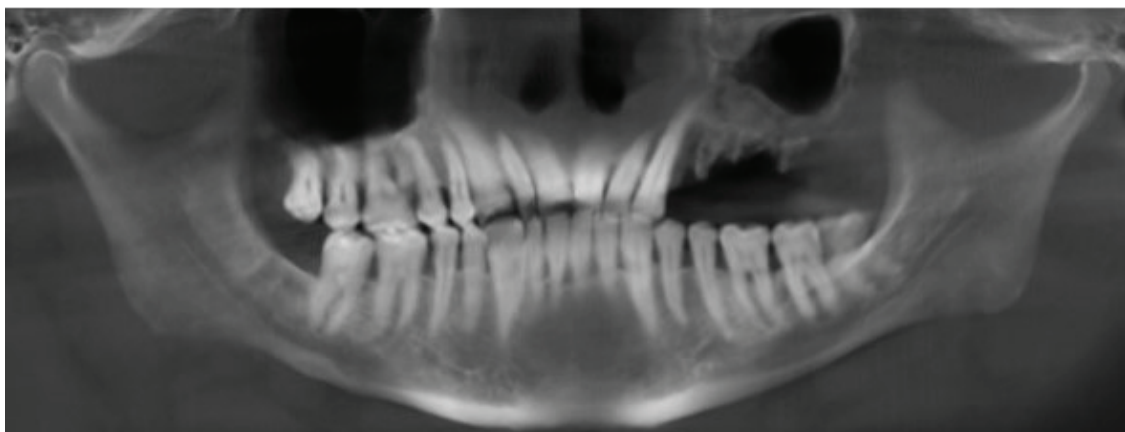


Fig. 2. The Orthopantomogram reveals irregular alveolar bone with missing teeth in the 24-28 region with haziness in the left maxillary sinus

left orbit. Bone destruction was noted in the floor of the left orbit, left lateral wall of the nose, left zygoma up to the frontozygomatic suture, anterolateral wall, posterolateral wall and floor of the left maxillary sinus. In addition, the left zygoma showed areas of sequestration with associated osteolytic areas (Figure 3A-C). 3D reconstruction was also done for better visualization (Figure 4). Radiographic diagnosis of malignancy of the left maxillary sinus was given with differential diagnosis as mucormycosis and avascular necrosis of the left maxillary alveolus.

The patient was then referred to the Department of Oral and Maxillofacial surgery where the necrotic bone was resected, and partial left maxillectomy was done. The patient was prescribed linezolid 600 mg twice daily for 5 days and metronidazole 400 mg thrice daily for 5 days. The specimen was then sent for histopathological investigation. Aspiration was

done from the left maxillary sinus and sent for KOH mount. The pathological report confirmed the diagnosis of mucormycosis with osteomyelitis. Fungal hyphae were noted in the KOH mount. The patient was planned for complete maxillectomy antifungal therapy – amphotericin B (liposomal) for 21 days and oral posaconazole 7.5 ml once daily for the next 15 days. The patient did not report for follow-up or further treatment.

DISCUSSION

Few studies have demonstrated that, compared to Computed Tomography (CT), CBCT scans have a better module transfer function and require less screening time. This aids in assessing the minute modifications to the trabecular pattern or reduction in cortical thickness in cases of RMM [4]. When di-

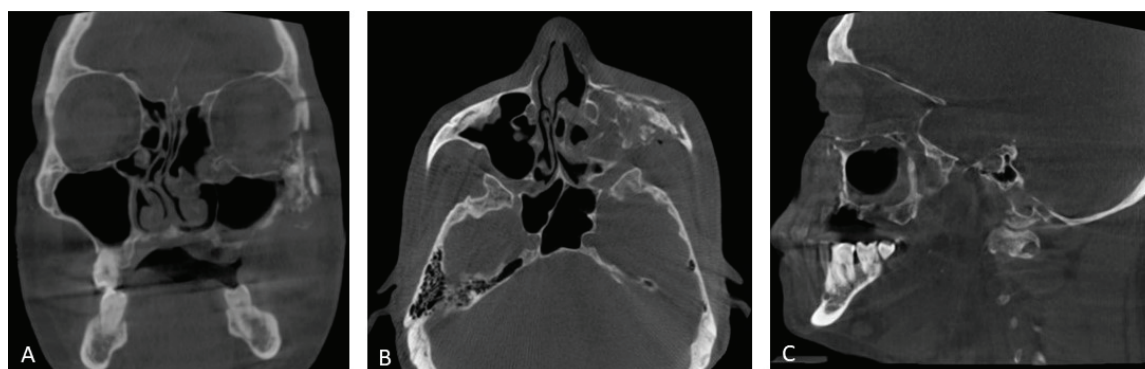


Fig. 3. CBCT images (A) Coronal section showing the destruction of the left lateral wall of the nose, the floor of orbit, and left zygoma, (B) Axial section showing haziness in the left maxillary sinus with sequestration seen in left zygoma, (C) Sagittal section showing mucosal thickening in the left maxillary sinus with destruction of anterior and posterior walls along with the alveolar bone

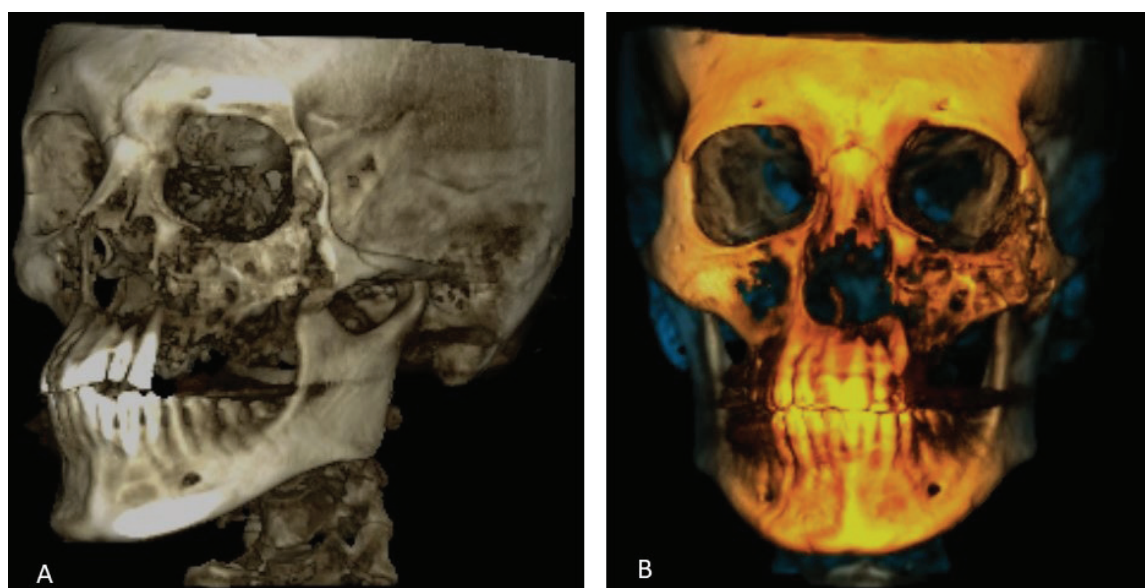


Fig. 4. 3D reconstruction images with volume rendering showing destruction of the left maxilla

agnosing rhinomaxillary mucormycosis, CBCT can be beneficial because it provides specific information on the lesion's extension into surrounding vital structures. However, Magnetic Resonance Imaging (MRI) or Multi Detector Computed Tomography (MDCT) should be used if the lesion extends into the orbit or the cranium [3].

Bony erosion, sinus involvement with mucosal thickening and invasion into the nasal cavity are the common radiographic findings of RMM on CBCT [3]. In a case series presented by Motevasseli et al. [1], radiographic features such as "ground glass" and "salt and pepper" appearance were seen in the infected bones.

Muley et al. [4] developed a diagnostic criteria and grading system for rhinomaxillary mucormycosis based on the evaluation of the CBCT findings in 30 suspected cases (Table 1). The diagnostic criteria included at least one maxillary sinus involvement, osteolytic alterations in the maxillary alveolar bone, either in conjunction with or apart from the other bones such as palatal, nasal or orbital bones and the presence or absence of dental findings [4]. Based on CBCT findings, the above-presented case falls under the category of Moderate Rhinomaxillary mucormycosis.

Table 1. CBCT grading of rhinomaxillary mucormycosis

Grade	Radiographic Features
Mild	At least one maxillary sinus is completely or partially occluded. Reduced bone density and localized alveolar bone osteolysis
Moderate	Opacification of one or more maxillary and ethmoidal sinuses, either full or partial osteolysis of the nasal septum, lateral wall of the nasal cavity, unilateral alveolar bone, and/or palate, as well as the turbinate's
Severe	All paranasal sinuses, including the frontal and sphenoid sinuses, may be involved unilaterally or bilaterally. Osteolysis of more than two bones, including the orbital walls, palate, zygoma, nasal cavity, sphenoid, and maxillary alveolar bone

Mucormycosis associated with osteomyelitis of the maxilla is a rare condition due to the rich blood supply of the maxilla. Motevasseli et al. [1], in a case series of 3 patients, reported post covid osteomyelitis involving the maxillary and other facial bones. In 2019, Arani et al. [5] reported a case of mucormycotic osteomyelitis of the left maxilla in a 48-year-old diabetic male. In a prospective study done by Urs et al. from 2011 to 2013, it was found that only five cases showed fungal osteomyelitis, which was primarily intraosseous and showed radiographic changes in the bone [6].

A ten-year study by Niranjana et al. [7] evaluated the prevalence of fungal osteomyelitis of jaws in diabetic patients. They reported that 52% of cases were fungal osteomyelitis, more commonly seen in males above 40 years of age. They concluded that fungal osteomyelitis associated with diabetes mellitus most commonly affects the maxilla. This was in accordance with the above-presented case. Selvamani et al. [8] reported a case of chronic osteomyelitis of the right maxilla in a patient with uncontrolled diabetes, which involved the zygomatic arch, maxillary sinus and pterygoid plates.

Parenteral antifungal therapy with Amphotericin B and rigorous debridement of the infected hard and soft tissue are standard therapies for mucormycosis. Newer antifungal drugs like Posaconazole, Voriconazole and Itraconazole have been used against mucor as they have a low risk of adverse effects as compared to Amphotericin B and do not require parenteral administration or hospitalisation [9].

CONCLUSION

CBCT is an important diagnostic aid in the diagnosis of rhinomaxillary mucormycosis as it gives complete details about the involvement of the maxilla, nasal cavity and alveolar bone. Thus, by combining and correlating the clinical and radiographic features, early diagnosis can be made. In case of any intracranial extension, the extent of invasion is incompletely evaluated in CBCT, and in such cases, CT and MRI need to be done. Thus, CBCT plays a crucial role in the diagnosis of mucormycosis at early stages, which helps with early intervention and a better prognosis.

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Authors contribution

Conception and design: Vidya Ajila

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Collection of data: Smrithy Sivadas K

Drafting of article: Yashika Jain

Critical revision: G Subhas Babu

Final approval of article: Vidya Ajila

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