#### **CASE REPORT**

# An Uncommon Parotid Abscess: Localized Mucormycosis

Sheetal Thakur<sup>1</sup>, Sachin Kumar Sharma<sup>2</sup>, Jai Minhas<sup>3</sup>, Jagdeep Singh Thakur<sup>4</sup>

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## **A**BSTRACT

**Background:** Mucormycosis affects immunocompromised and diabetic patients most of the time. Clinically, mucormycosis has six well-known forms based on systemic involvement and rarely occurs in localized form.

Case presentation: We describe a case of parotid mucormycosis in an elderly diabetic male patient. Radiological investigations were suggestive of abscess. Mucormycosis was detected on histopathology and fungal culture. The patient had a complete recovery with liposomal amphotericin B and oral Posaconazole.

**Conclusion:** Mucormycosis should be considered as a possible diagnosis in case of any spontaneous abscess in the parotid. Prompt management improves the outcome of mucormycosis.

Keywords: Cancer, Case report, Mucormycosis, Parotid diseases, Parotid gland.

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#### BACKGROUND

Mucormycosis, a dreadful disease, is associated with an immunocompromised state like diabetes mellitus, cancer, organ transplantation, neutropenia, burns, and trauma. Last year, mucormycosis had an unusual association with severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) (COVID-19) that led to another pandemic over the COVID-19 pandemic in India and other countries.<sup>1-3</sup> Rhino-orbital cerebral mucormycosis (ROCM), and pulmonary, cutaneous, and gastrointestinal are the common presentations of mucormycosis, which rarely presents as an isolated lesion after trauma.<sup>4</sup> Parotid mucormycosis is an unusual and rare presentation.<sup>5-10</sup> Here, we present another unusual case of parotid mucormycosis.

# Case Presentation

A diabetic male in his early 80s presented with progressive and firm swelling in front of the left ear for the past month. The swelling was associated with occasional pain during chewing. The patient was taking oral hypoglycemic drugs regularly. The examination found a  $4 \times 4$  cm diffuse, firm, non-tender swelling over the zygomatic region in the left parotid area. The external ear and tympanic membrane were normal. Further examination was non-contributory. A probable diagnosis of a parotid tumor was made, and the patient was investigated further to clinch the diagnosis.

An ultrasound revealed a  $3 \times 3$  cm cystic lesion in the parotid gland, just superficial to the left zygomatic bone, with increased peripheral vascularity. The parotid gland appeared normal. Ultrasound findings were suggestive of an intraparotid lymph node abscess. Computerized tomography revealed a heterogeneously enhanced area with a centrally hypo-intense area in the left parotid region with mild erosion of the zygomatic arch (Fig. 1). The left masseter, temporalis, and lateral pterygoid muscles had inflammatory features. The paranasal sinuses, orbits, and brain were normal. The possibility of an abscess caused by tuberculosis or cancer was kept in mind. On reverse transcription-polymerase chain

<sup>1,4</sup>Department of Otolaryngology – Head and Neck Surgery (ENT), Indira Gandhi Medical College & Hospital, Shimla, Himachal Pradesh, India

<sup>2</sup>Department of Pathology, Indira Gandhi Medical College & Hospital, Shimla, Himachal Pradesh, India

<sup>3</sup>Department of Otolaryngology – Head and Neck Surgery (ENT), Dr. Ram Manohar Lohia Hospital, New Delhi, India

Corresponding Author: Jagdeep Singh Thakur, Department of Otolaryngology – Head and Neck Surgery (ENT), Indira Gandhi Medical College & Hospital, Shimla, Himachal Pradesh, India, Phone: +91 9418477400, e-mail: anujagdeep@yahoo.co.in

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reaction (RT-PCR), the patient tested negative for COVID-19. The hematological and biochemical investigations were grossly normal except for fasting glucose levels of 172 mg/dL with 7.4% of glycosylated hemoglobin and a serum ferritin value of 598 ng/mL.

Under general anesthesia, the patient was taken to the operating room for abscess drainage. Taking care of the course of facial nerve branches, an incision was made over the swelling, and 5 mL of thick pus was drained and sent for microbiological examination. A tissue biopsy was also taken to rule out malignancy and tuberculosis. The postoperative period was uneventful. The microbiological examination did not find any microorganisms on Gram, Ziehl-Neelsen staining. Similarly, microbiological cultures did not show any bacterial growth. Histopathology revealed necrotic bone studded with aseptate, broad ribbon-like hyphae with wide-angled branching (Fig. 2). A few areas of granulation tissue and hemorrhage were also found. These hyphae were highlighted by Periodic Acid-Schiff and Grocott's Methenamine Silver Stain. These features were supportive of the mucormycosis.

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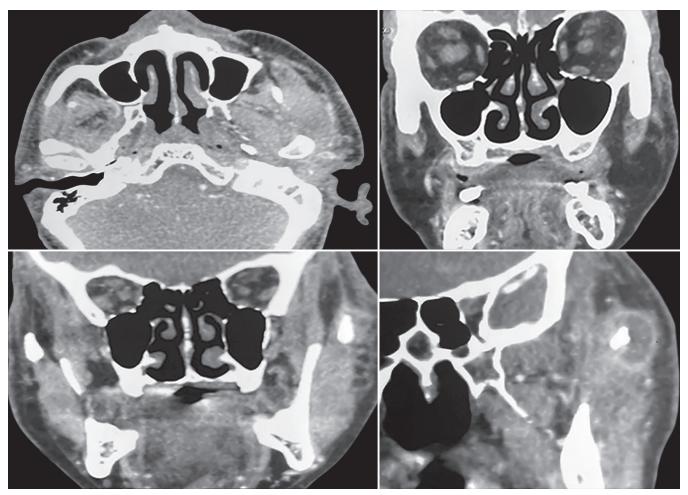


Fig. 1: Computerized tomography shows a hypo-intense with increased peripheral vascularity lesion in the parotid area just adjacent to the zygomatic arch

Rhizopus species grew in the fungal culture. For 3 weeks, the patient received liposomal amphotericin B at a dose of 5 mg/kg, with weekly radiological evaluation. The patient had a significant response to the drug, and the disease did not show any progression. Repeat surgical debridement was not performed based on radiological evidence. He was discharged on the drug Posaconazole (600 mg on day 1 and 300 mg for 1 month).

The patient had significant improvement within a month. Posaconazole tablets were prescribed for the patient for another month. The final review found complete recovery (Fig. 3).

## Discussion

Fungi of the genera *Rhizopus*, *Mucor*, *Rhizomucor*, *Cunninghamella*, and *Absidia* of the *Mucorales* order, class *Zygomycetes* causes mucormycosis with the dominance of *Rhizopus oryzae* in humans.<sup>4</sup> *Mucorales* are prevalent in our surroundings, that is, soil, decaying vegetable matter, bread, and dust. Mucormycosis occurs by inhalation of *Mucorales*-containing spores, consumption of contaminated food, and direct inoculation in skin wounds. Clinically, it is classified into rhino-orbital cerebral mucormycosis (ROCM), and pulmonary, gastrointestinal, cutaneous, renal, and disseminated mucormycosis.<sup>4</sup> Individuals with extensive skin injury and immunocompromised conditions like diabetes mellitus, hematological

cancers, chemotherapy, immunosuppressive therapy with iron and human immunodeficiency disease, have a high risk of developing mucormycosis. Diabetes mellitus is the commonest cause of mucormycosis in India while hematological cancer and solid organ transplant recipients dominate in the developed countries.<sup>4</sup>

The ROCM, and pulmonary and cutaneous mucormycosis are the most common forms (in decreasing order) in India. Pulmonary mucormycosis is the most common clinical presentation in developed countries.

Parotid mucormycosis is a rare presentation.<sup>5–10</sup> Extension of ROCM or direct inoculation due to trauma are the most likely causes of parotid mucormycosis.<sup>5</sup> However, the etiopathogenesis of the present case remained unclear as the patient did not have any trauma, and the sinuses were found to be normal on clinical and radiological examination. Most likely, fungi spores in the mouth might have reached the parotid through the parotid duct or lymphatics, but they should have involved the parotid duct and intraparotid lymph node, thereby negating this hypothesis too.

Recently, a COVID-19-associated mucormycosis (CAM) pandemic in a few countries caused high mortality due to a shortage of drugs. The etiopathogenesis of CAM is still unclear, and COVID-19-induced inflammation and excess use of corticosteroids with antibiotics during COVID-19 management are a few hypotheses;<sup>1</sup> however, these need to be validated through large cohort studies. Although



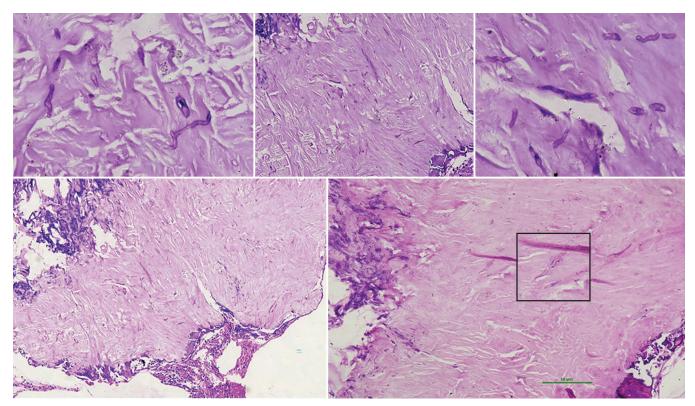


Fig. 2: Microscopic view of tissue showing mucor filaments

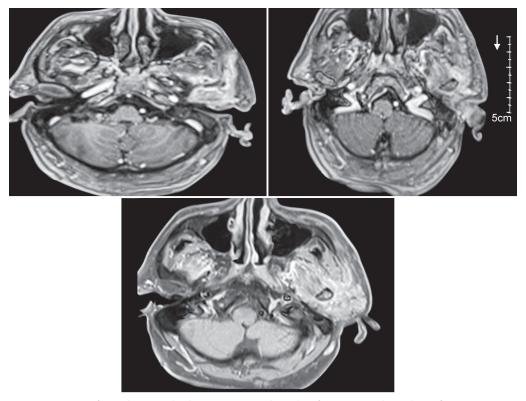


Fig. 3: Magnetic resonance scan performed at 3 weeks showing increased uptake of contrast media and significant improvement in the lesion

the present case did not have a COVID-19 infection, this unusual presentation during the COVID-19 pandemic warrants further

research to better understand the pathogenesis of COVID-19 and mucormycosis.

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