

## ABSTRACT

**OBJECTIVE:** Mucormycosis is a rare opportunistic infection caused by fungi of the order mucorales, mainly affecting immunocompromised patients with poorly controlled diabetes or hematological malignancies. Rhinocerebral mucormycosis is the most common form, whereas mandibular mucormycosis is rare with only a few cases previously reported in the oral surgery literature. We report a case of mucormycosis of the mandible and adjacent soft tissue.

**METHODS:** Retrospective review of clinical case

**RESULTS:** A 70-year-old female with a history of breast cancer, chemotherapy, myelodysplastic syndrome, bone marrow transplant, and graft-versus-host disease presented with fever and acute left side jaw swelling and pain. CT demonstrated an abscess surrounding the left hemimandible and the left sublingual space, with a foci of air within the left hemimandible. The patient underwent submental incision and drainage without gross purulence. Three days later, the left mental/submental skin became acutely necrotic, and pathology and cultures demonstrated invasive mucormycosis. The patient underwent wide local excision of the submental soft tissue, near-total mandibulectomy, and partial glossectomy. Intravenous and topical antifungal agents, intravenous antibiotics, and filgrastim were given. Multiple debridements including subtotal glossectomy and floor of mouth resection were needed to quell the disease progression. Despite aggressive surgical and medical management, the disease continued to progress and approximately two weeks later, the patient was made comfort care only and expired.

**CONCLUSION:** Although most commonly associated with sinonasal disease, mucormycosis can present in other regions of the head and neck including the mandible. Prompt diagnosis, aggressive medical treatment including correction of underlying immunodeficiencies and appropriate antifungals, and aggressive surgical debridement are important in managing these patients. However, this disease portends poor outcomes, as demonstrated in our patient.

## BACKGROUND

Mucormycosis is a rare life-threatening opportunistic infection caused by fungi of the order Mucorales, commonly found in soil and decaying organic material. The most common species involved clinically are *Rhizopus*, *Lichtheimia* and *Mucor*.

The infection often involves immunocompromised patients with conditions such as poorly-controlled diabetes, leukemia, lymphoma, AIDS, organ transplant, and long-term steroid and immunosuppressive therapy. It has been reported that patients with hematological malignancies with or without hematopoietic stem cell transplantation (HSCT) represent 22-50% of cases. As well, risk factors for *Mucor* infection in HSCT recipients include graft-versus-host disease (GVHD) and high steroid use.

During an active infection, the organisms invade the microvascular, causing thrombi. This leads to decreased local blood flow and tissue infarction. There are a few well-known forms: Rhino-orbito-cerebral aka rhinocerebral, Pulmonary, Gastrointestinal, Cutaneous, Disseminated, CNS, with rhinocerebral being the most common. Oral mucormycosis has been documented in literature, however mandibular involvement is rarer with very few reported cases.

Management of mucormycosis involves a combination of early diagnosis, reversal of underlying predisposing risk factors, surgical debridement and early antifungal treatment. Despite these measures, mortality rate has been reported to be greater than 40% and even higher for those with hematological malignancies or stem cell transplants.

We report a case of mucormycosis of the mandible.

## CASE REPORT

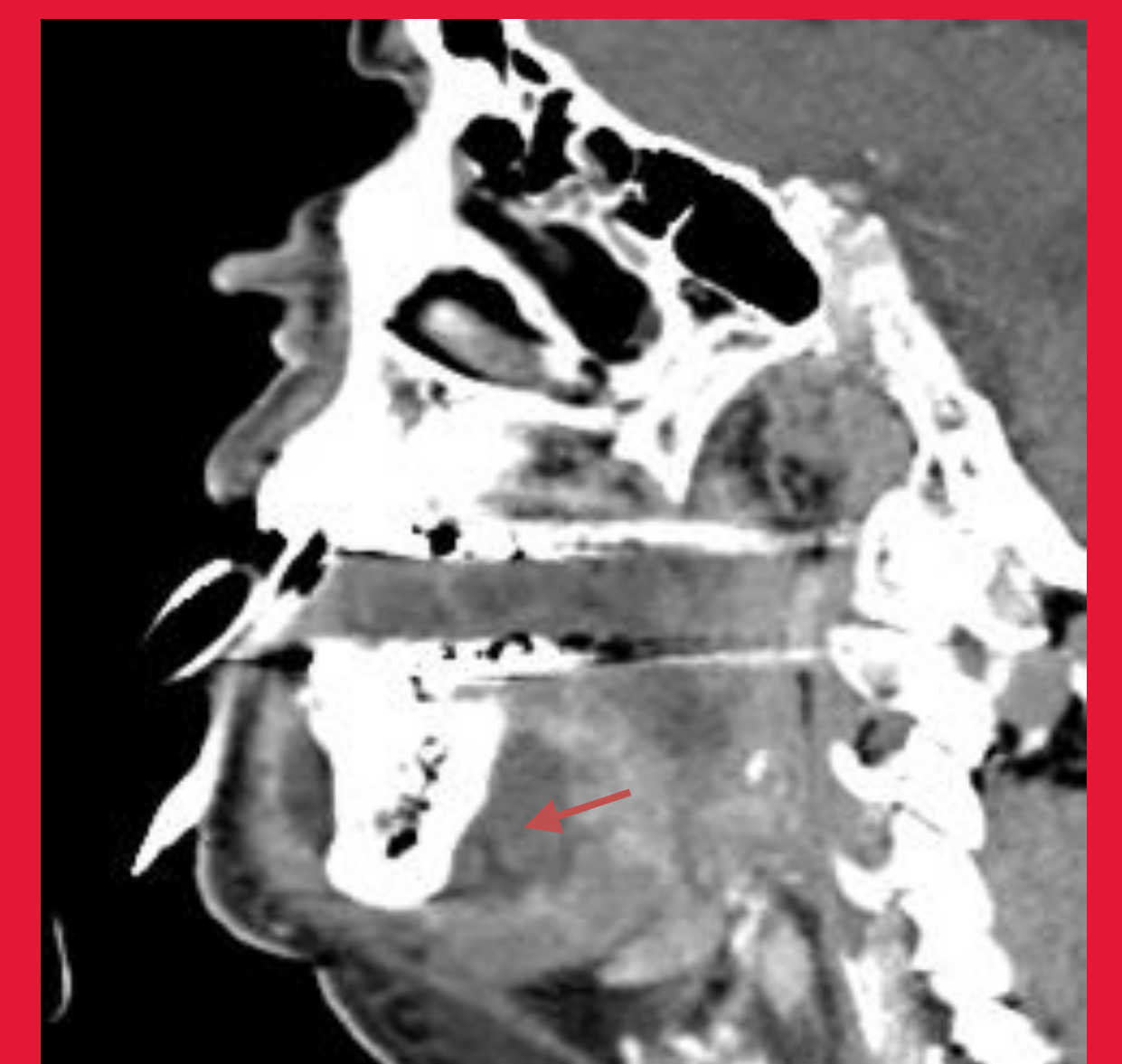
A 70-year-old female, with a history of breast cancer treated with chemotherapy, myelodysplastic syndrome s/p bone marrow transplant complicated by relapse and steroid-dependent graft-versus-host disease of bowel and skin, presented with fever and acute left-sided jaw swelling and pain. She was admitted and started on empiric antibiotics of vancomycin and zosyn. Due to her underlying medical condition, her white blood cell count at presentation was 600/mcL, hemoglobin 6.3 g/dL and platelet count of 14,000/mcL. One day later, the patient developed acute tongue and floor of mouth swelling requiring emergent fiberoptic intubation for airway protection.

CT imaging (Figures 1 and 2) demonstrated an abscess surrounding the left hemimandible, extending into the left sublingual space as well as foci of air in the left hemimandible. The patient underwent submental incision and drainage of the collection, but no gross purulence was found. On oral examination, a small area of black necrotic gingival mucosa along the buccal surface of left mandibular teeth was noted and biopsied.

## PREOPERATIVE IMAGING



**Figure 1.** Axial CT showing collection surrounding the left hemimandible extending along the medial aspect of the mandible in the sublingual space into the inferior left hemitongue and displaces the tongue to the right



**Figure 2.** Sagittal CT with arrow showing low attenuation collection surrounding Left hemimandible

## CASE CONTINUED

Three days later, the left mental/submental skin appeared necrotic (Figure 3), worsening over the course of several hours. Cultures from the previous surgery demonstrated *Rhizopus* and pathology was consistent with invasive mucormycosis. Intravenous antifungals were started (amphotericin and isavuconazole), in addition to filgrastim. The same day, the patient underwent mandibular dental extractions, wide local excision of the submental soft tissue, near-total mandibulectomy and partial glossectomy. The dental extractions were notable for no blood in any of the tooth roots (Figure 4). Post operatively, the wound was irrigated and packed daily with topical amphotericin-soaked gauze.

Due to worsening local disease, the patient was brought back to the operating several times for further debridements. These surgeries entailed near-total glossectomy, partial pharyngectomy and floor of mouth resection. Despite aggressive surgical and medical management, the patient's immune system could not be reconstituted and did not respond to the filgrastim. She had persistent leukocytopenia and an ANC of 265 cells/mm<sup>3</sup>. Approximately two weeks later, the patient's family withdrew all interventions, the patient was made comfort care and she expired two days later.

## INTRAOPERATIVE IMAGES



**Figure 3.** Necrotic mental skin near previous submental incision with penrose.



**Figure 4.** Post extraction of teeth #20-31 revealing necrotic gray-colored sockets with no bleeding

## CONCLUSIONS

Although most commonly associated with sinonasal disease, mucormycosis can present in other regions of the head and neck including the mandible. Prompt diagnosis, aggressive medical treatment including correction of underlying immunodeficiencies, initiation of appropriate antifungals, and performance of aggressive surgical debridement are important in managing these patients. However, this disease portends poor outcomes, as demonstrated in our patient

## REFERENCES

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