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Aspergilloma of maxilla in an immunocompromised patient: A multidisciplinary approach

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ABSTRACT

Aspergillosis is a rare, invasive, rapidly progressive, and life-threatening fungal disease of the maxillofacial region and what makes the disease sinister is its indolent course and ability to cause death (16%). Early diagnosis and treatment are essential for such infections. We here report a case of an elderly male patient with diabetes mellitus, cardiac disease, and kidney failure who was diagnosed with aspergilloma with osteomyelitis of the right maxilla. Treatment was based on the comorbidities and the clinical, radiological, and histological features of the lesion.

Introduction

Globally, mycotic infections of the paranasal sinuses are on the rise, associated with an increasing incidence of coronavirus disease-19 and the number of individuals with compromised immune systems. Aspergillosis has been recognized as a fungal disease since Katzenstein et al. (1) discovered it in 1983, although its diagnosis, course of treatment, and classification remain unclear. Aspergillus is a part of the phylum Ascomycota.

Although Aspergillus species are diverse, only a few thermotolerant species can infect humans opportunistically and cause aspergillosis. The most prevalent subtype is aspergilloma. It is characterized by a non-invasive chronic fungal sinusitis that frequently occurs in the maxillary antrum of healthy individuals. The disease has an ominous quality because of its slow course and the possibility of mortality. Mortality may be as high as 16% (2). We here present a case of aspergilloma of the maxilla, managed by a multidisciplinary team owing to a compromised immune system.

Case Presentation

A 75-year-old male patient was admitted with a chief complaint of pain followed by pus discharge from the right maxilla for 8 months. The patient had a history of tooth extraction 10 months ago, followed by gradual onset, progressive, and continuous pain. His history was relevant for type 2 diabetes mellitus for 20 years and ischemic heart disease and chronic kidney disease for 5 years. On physical examination, there was pus discharge from a previous extraction socket on the right, upper labial vestibule (Figure 1a). Serum urea (72.7 mg/dL), creatinine (2.2 mg/dL), sodium (134 mmol/L), potassium (5.30 mmol/L), and chloride (104.9 meq/L) were measured. A chest X-ray was normal. A cardiologist, nephrologist, and endocrinologist were counseled. An incisional biopsy revealed ulcerated mucosa lined by granulation tissue and subepithelium with an abundance of mixed inflammatory cell infiltrates with numerous bacterial colonies with sulfur granules and radiating filaments, suggesting actinomycosis. The colonies were observed in dead bony spicules, suggesting osteomyelitis of the maxilla secondary to actinomycosis species. A contrast-enhanced computed tomography scan revealed moderate circumferential thickening with few areas of hyperdensity and erosion involving the anterior and posterior walls of the right maxillary sinus, extending to the right orbital floor. The intramedullary erosion involved the maxillary premolar and molar alveolar processes (Figure 2). The patient underwent aggressive surgical debridement and primary closure under general anesthesia, though with high cardiac risk (Figure 1b). The final histopathological report suggested aspergilloma with osteomyelitis and a bony sequestrum in the right maxilla. Multiple sections of hematoxylin and eosin (H&E) staining and periodic acid-Schiff (PAS) staining showed fibro-collagenous tissue bits lined by respiratory mucosa with seromucous glands, hemorrhage, dead bone fragments, and calcification with clumps of thin, acute-angled branching septate hyphae and an area of necrosis (Figure 3). The post-operative recovery of the patient was favorable.

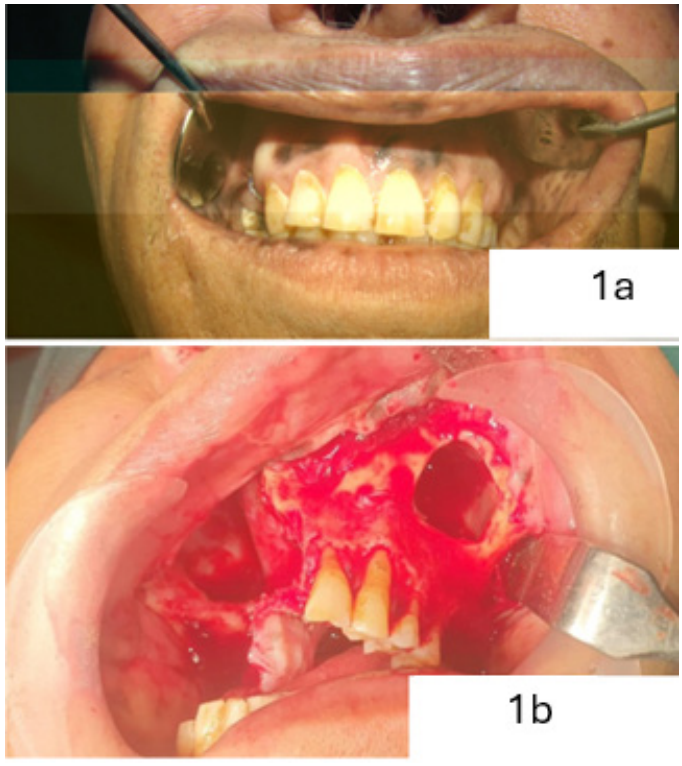

Figure 1. (a) Intraoral sequestrum noted in 16 regions. (b) Intraoperative image

Discussion

The priest botanist Micheli (3) was the first to mention aspergillosis in 1729. In 1893, Mackenzie (4) published the first report of maxillary sinus aspergillosis. Its local invasiveness and types 1 and 3 hypersensitivity responses are linked with its pathogenicity. Of all rhinosinusitis diagnoses, 6-9% are related to fungal sinusitis. In the head and neck area, the maxillary sinus is the most common location (87.8%), followed by the sphenoid sinus coming in second (5).

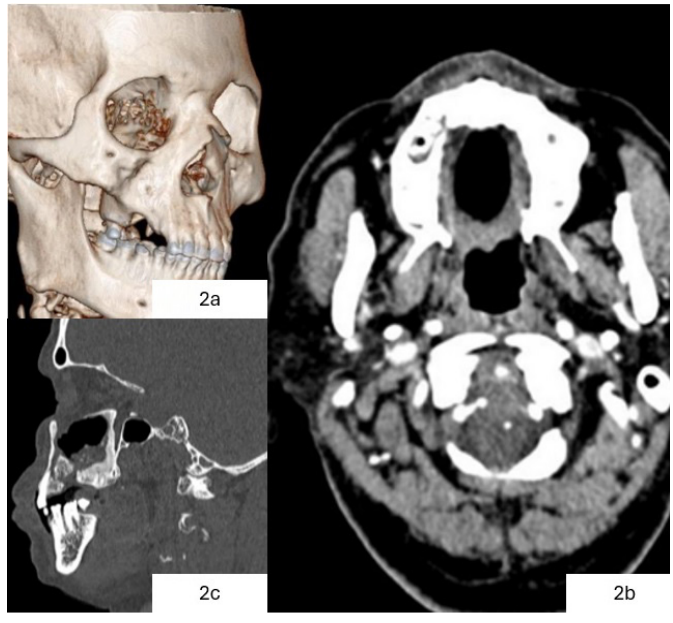

Figure 2. Contrast-enhanced computed tomography images. (a) 3D reconstructed image. (b) Axial section. (c) Sagittal section

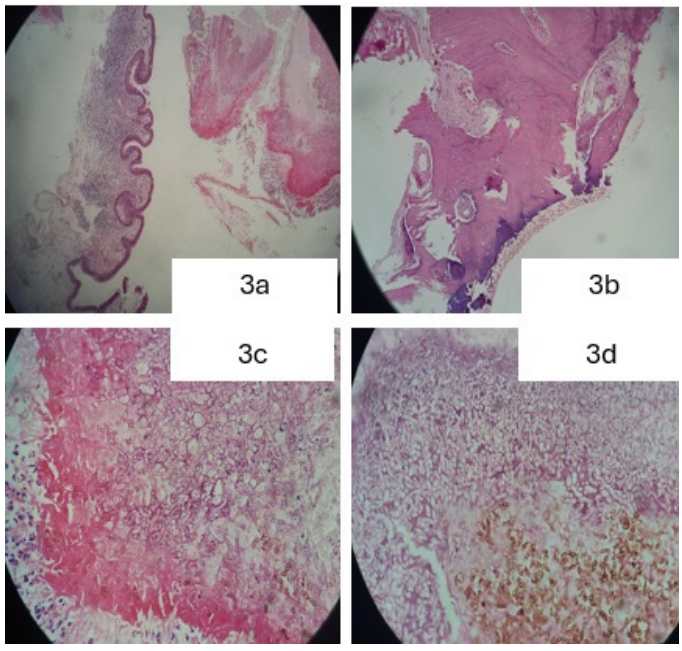

Figure 3. Histopathological images. (a, b) H&E staining in 10X magnification of the respiratory mucosa with seromucous glands, hemorrhage, and dead bone fragments. (c) H&E staining in 100X. (d) PAS staining in 100X
H&E: Hematoxylin and eosin, PAS: Periodic acid-Schiff

Despite its rarity, orofacial aspergillosis can have disastrous effects when the immune system is impaired due to uncontrolled diabetes, lymphomas, leukemia, renal failure, long-term steroid usage, and acquired immunodeficiency syndrome (5,6). The clinical manifestations include nasal congestion, headache, purulent or blood-stained discharge from the nose, and pain in the orbital region. Ocular involvement can occur and presents with chemosis, ptosis, proptosis, conjunctival suffusion, sore eyes, blurred vision, and visual loss associated with retinal artery thrombosis (7). In the present patient, although there was purulent leakage to the oral cavity from a removed socket, there was no sign of ocular involvement.

Radiographically, it appears as a region with a density comparable to iron, resembling a foreign mass within a uniformly clouded maxillary sinus. The affected area may show sinus cavity opacification, mucosal thickening, or erosion of the underlying bone (8). Common histological fungi stains are Grocott methenamine silver (GMS) and PAS. The GMS stain has a signal-to-noise problem because it stains not only the fungi but also the inflammatory cells (lysosomes) and tissue reticulin despite being more sensitive than the PAS stain (9). The small benefit of PAS staining is that it makes the morphology of the tissue next to the fungus easier to see; however, this issue may be overcome by applying an H&E counterstain and GMS stain. When viewed under 10X and 100X, histopathological sections show fibro collagenous tissue bits lined by respiratory mucosa with seromucous glands, hemorrhage, dead bone fragments, and calcification with clumps of thin, acutely angled branching septate hyphae with areas of necrosis (9).

Treatment for fungal infection includes sequestrectomy, wound debridement, and anti-fungal drug regimens, including amphotericin B, voriconazole and posaconazole (10). In aspergilloma of the maxillary sinus, the surgical approach has some advantages compared with functional endoscopic sinus surgery by enabling complete removal of the fungus, drainage of the remnant sinus cavity, and prevention of recurrence (11). However, our patient underwent only aggressive surgical debridement and did not receive any anti-fungal therapy because of his compromised renal condition.

Conclusion

Aspergillosis is a rare, invasive, life-threatening fungal disease of the maxillofacial region, particularly in immunocompromised patients. The present immunocompromised patient was accurately diagnosed and aggressively managed with a multidisciplinary team.

Ethics

Informed Consent: Informed written consent for publication was obtained from the patient.

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Footnotes

Authorship Contributions

Surgical and Medical Practices: D.S., V.A., N.K. Concept: D.S., V.A., N.K. Design: D.S., V.A., N.K. Data Collection or Processing: D.S., T.M., Analysis or Interpretation: D.S., T.M., Literature Search: D.S., T.M., Writing: D.S., T.M.

Conflict of Interest: No conflict of interest was declared by the authors.

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