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Intraosseous maxillary phaeohyphomycosis mimicking an odontogenic cyst in an immunocompetent patient: a case report.

Intraosseous maxillary phaeohyphomycosis mimicking an odontogenic cyst in an immunocompetent patient: a case report

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Summary

Phaeohyphomycosis is an uncommon fungal infection caused by melanized (dematiaceous) fungi, which can mimic odontogenic lesions when it affects the maxillofacial bones. Intraosseous involvement of the maxilla is exceptional, especially in immunocompetent patients. A 50-year-old male patient, immunocompetent, presented with pain in the region of the upper incisors associated with progressive swelling on the buccal and palatal surfaces. Computed tomography revealed an extensive hypodense intraosseous lesion involving teeth 22 to 12, with destruction of the cortical bone and extension into the nasal cavity, initially suggestive of an aggressive odontogenic cyst. Histopathological examination revealed a granulomatous inflammatory process composed of granular histiocytes, multinucleated giant cells, and cholesterol clefts. Special stains (Grocott, PAS/PAS-D, and Fontana-Masson) revealed pigmented septate hyphae and spherical fungal structures compatible with dematiaceous fungi, confirming the diagnosis of intraosseous phaeohyphomycosis. Treatment through surgical debridement combined with systemic therapy with itraconazole resulted in complete clinical and radiological resolution, without recurrence after two years of follow-up. This case highlights the importance of including phaeohyphomycosis in the differential diagnosis of atypical intraosseous lesions of the maxilla and reinforces the decisive role of histopathological examination with special stains for establishing a definitive diagnosis.

Keywords: Phaeohyphomycosis. Dematiaceous fungi. Maxilla. Intraosseous lesion. Oral fungal infections.

Abstract

Phaeohyphomycosis is a rare fungal infection caused by melanized fungi and may mimic odontogenic lesions when affecting the maxillofacial bones. Intraosseous involvement of the maxilla is exceptional, particularly in immunocompetent patients. A 50-year-old immunocompetent man presented with pain in the maxillary incisors and progressive buccal and palatal swelling. Computed tomography revealed an extensive hypodense intraosseous lesion involving teeth 22 to 12, with cortical bone destruction and extension into the nasal cavity, initially suggestive of an aggressive

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odontogenic cyst. Histopathological examination showed granulomatous inflammation with granular histiocytes, multinucleated giant cells, and cholesterol crystals. Grocott, PAS/PAS-D and Fontana-Masson stains demonstrated pigmented septate hyphae and spherical fungal structures consistent with dematiaceous fungi, confirming intraosseous phaeohyphomycosis. Surgical debridement associated with systemic itraconazole therapy resulted in complete resolution, with no recurrence after two years of follow-up. This case highlights the importance of including phaeohyphomycosis in the differential diagnosis of atypical intraosseous maxillary lesions and reinforces the decisive role of histopathological examination with special stains.

Keywords: Phaeohyphomycosis. Melanized fungi. Maxilla. Intraosseous lesion. Oral fungal infections.

1. Introduction

Phaeohyphomycosis comprises a heterogeneous group of fungal infections caused by melanized (dematiaceous) fungi, characterized by the presence of pigmented hyphae or yeast-like elements in human tissues. Since the original description by Ajello and colleagues in the 1970s, this term has been used to define infections that do not fit the classical morphological patterns of chromoblastomycosis or eumycetoma, particularly due to the absence of muriform cells or compact fungal grains (REVANKAR; SUTTON, 2010; SÁNCHEZ-CÁRDENAS; ISA-PIMENTEL; ARENAS, 2023). Dematiaceous fungi include multiple genera such as *Exophiala, Cladophialophora, Fonsecaea, Curvularia* and *Alternaria,* which are widely distributed in soil, decaying vegetation and organic matter, especially in tropical and subtropical regions (REVANKAR; SUTTON, 2010; SÁNCHEZ-CÁRDENAS; ISA-PIMENTEL; ARENAS, 2023; HE et al., 2022). Human infection usually occurs through traumatic inoculation, inhalation of fungal propagules or, less frequently, hematogenous dissemination from a primary focus (REVANKAR; SUTTON, 2010; SÁNCHEZ-CÁRDENAS; ISA-PIMENTEL; ARENAS, 2023; HE et al., 2022).

Although phaeohyphomycosis has classically been associated with immunosuppression, an increasing number of cases have been reported in immunocompetent individuals, suggesting that local factors and innate immune responses may also play an important role in disease susceptibility (HE et al., 2022; SIGAMANI et al., 2025; QUEIROZ-TELLES et al., 2017). The presence of melanin in the fungal cell wall is considered a major virulence factor, conferring resistance to oxidative stress and contributing to chronic, slowly progressive infections (REVANKAR; SUTTON, 2010; SÁNCHEZ-CÁRDENAS; ISA-PIMENTEL; ARENAS, 2023).

Involvement of the oral and maxillofacial region by melanized fungi is uncommon, and intraosseous infection of the maxilla is exceptionally rare (RAWAL; KALMAR, 2012; ANDRADE et al., 2017; CARDOSO et al., 2007; SHUKLA et al., 2009). When present, these injuries may present as expansile radiolucent defects with cortical destruction, closely resembling odontogenic cysts, tumors or chronic osteomyelitis, often leading to delayed or incorrect diagnosis (RAWAL; KALMAR, 2012; ANDRADE et al., 2017; CARDOSO et al., 2007; SHUKLA et al., 2009; RAI et

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al., 2014; SURESH et al., 2022).

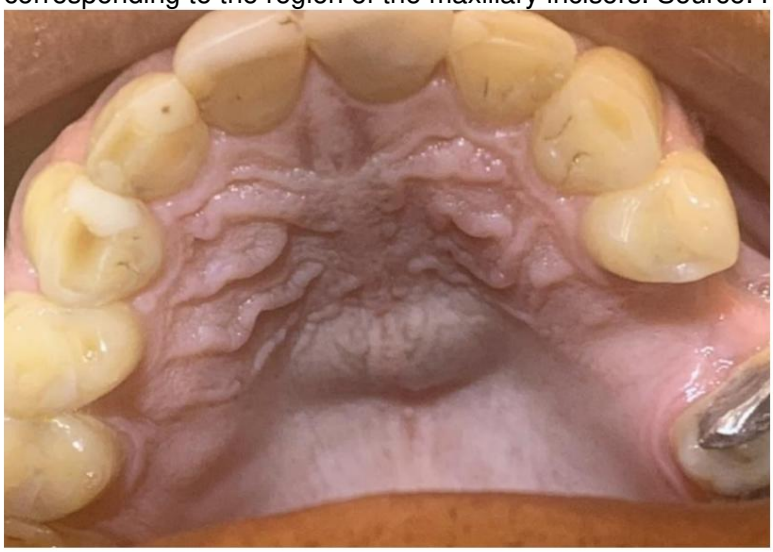
This report describes a rare case of intraosseous maxillary phaeohyphomycosis in an immunocompetent patient, clinically and radiographically simulating an odontogenic cyst, emphasize the importance of histopathological examination with special stains for accuracy diagnosis and appropriate management.

2. Case Report

A 50-year-old male patient with no relevant medical history was referred to a specialized oral diagnosis service with a chief complaint of pain in the anterior maxillary region. According to the patient, symptoms had started approximately three months earlier, characterized by mild to moderate, initially diffuse pain involving the maxillary incisors, partially relieved by the occasional use of nonsteroidal anti-inflammatory drugs. The patient denied any history of local trauma and reported that the teeth involved had undergone endodontic treatment more than ten years previously, without subsequent complications.

Two weeks prior to presentation, the patient noticed progressive swelling in the buccal and palatal regions corresponding to teeth 21 and 11, accompanied by increased pain, particularly on palpation. Extraoral examination was unremarkable. Intraoral examination revealed a firm, poorly defined swelling in the anterior maxilla, tending to digital pressure, without spontaneous drainage, fistula formation or mucosal ulceration (Figure 1).

Figure 1. Intraoral clinical photograph showing a firm, poorly defined swelling in the anterior hard palate, corresponding to the region of the maxillary incisors. Source: Prepared by the author.



Computed tomography of the maxilla demonstrated an extensive hypodense intraosseous lesion involving teeth 22, 21, 11 and 12, with partially defined margins and marked destruction of the buccal and palatal cortical plates. Extension of the lesion toward the nasal cavity was observed, with thinning and focal discontinuity of the lateral nasal wall (Figure 2). Based on these findings, the initial

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diagnostic hypotheses included an infected radicular cyst, an odontogenic keratocyst or another aggressive cystic lesion of indeterminate origin.

An incisional biopsy was performed under local anesthesia. Surgical access revealed an intraosseous cavity containing a brownish-red fluid with thick consistency and mild odor, without purulent material. Fragments of granulation tissue and portions of the cavity wall were submitted for histopathological examination (Figure 2 and 3).

Figure 2. Computed tomography of the maxilla showing an extensive hypodense intraosseous lesion involving teeth 22 to 12, with destruction of the buccal and palatal cortical plates and extension toward the nasal cavity. Source: Prepared by the author.

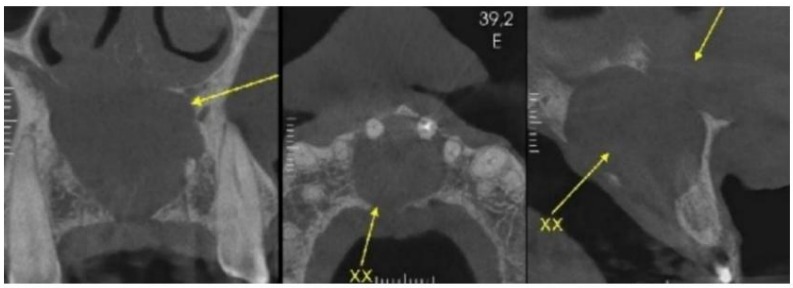
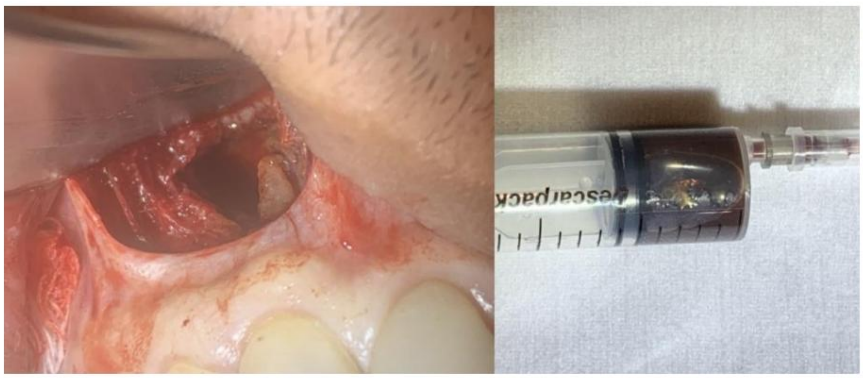


Figure 3. Intraoperative view showing brownish-red fluid content with thick consistency and mild odor within the intraosseous cavity. Source: Prepared by the author.



Microscopically, the lesion exhibited a granulomatous inflammatory pattern composed of numerous granular histiocytes and multinucleated giant cells. Cholesterol clefts were observed throughout the inflammatory stroma, indicating chronicity, with occasional microabscess formation interspersed within the connective tissue. Notably, pigmented spindle-shaped cells were identified infiltrating adjacent skeletal muscle fibers (Figure 4).

Special histochemical stains were decisive for diagnosis. Grocott–Gomori methenamine silver staining demonstrated numerous pigmented tubular and spherical fungal structures, including septate hyphae. Periodic acid–Schiff and PAS–diastase stains revealed diastase-resistant PAS-positive cytoplasmic granules. Fontana–Masson staining highlighted intensely pigmented spherical and filamentous structures, consistent with melanin-containing fungal elements within an area of

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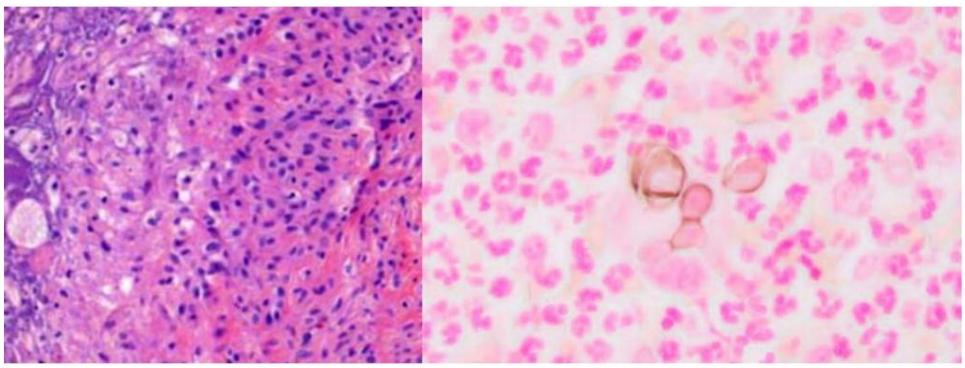
pyogranulomatous inflammation with microabscess formation. These findings confirmed the diagnosis of intraosseous maxillary phaeohyphomycosis (Figure 4).

Figure 4. Histopathological examination showing granulomatous inflammation composed of granular histiocytes and multinucleated giant cells, with cholesterol clefts within the inflammatory stroma. Septate fungal hyphae, occasionally pigmented, are observed embedded within the pyogranulomatous inflammatory infiltrate (hematoxylin and eosin stain) (A). Fontana–Masson staining highlighting intensely pigmented fungal elements, confirming the presence of melanin in the fungal cell walls, within an area of pyogranulomatous inflammation with microabscess formation (B).



Following diagnosis, the patient was referred to the public health system for definitive treatment. Surgical management consisted of wide opening of the lesion, complete removal of infected tissue and thorough debridement of the affected bone. Systemic antifungal therapy with itraconazole was initiated at a loading dose of 200 mg twice daily for three days, followed by 200 mg Once daily for 12 weeks.

Clinical and laboratory follow-up was performed monthly during antifungal therapy, including periodic assessment of liver function. A follow-up computed tomography scan obtained three months after treatment demonstrated complete resolution of the lesion, with partial bone regeneration and no evidence of residual disease. After two years of follow-up, the patient remains asymptomatic, with no clinical or radiographic signs of recurrence.

3. Discussion

The present case illustrates a rare manifestation of phaeohyphomycosis presenting as an intraosseous expansile lesion of the maxilla, radiographically and clinically simulating an odontogenic cyst. This presentation is particularly challenging because expansile radiolucent lesions involving the anterior maxilla are far more commonly associated with inflammatory odontogenic cysts, keratocystic lesions or benign odontogenic tumors, which often leads to an initial non-infectious diagnostic hypothesis (RAWAL; KALMAR, 2012; ANDRADE et al., 2017; CARDOSO et al., 2007; SHUKLA et al., 2009; RAI et al., 2014; SURESH et al., 2022).

Phaeohyphomycosis is caused by melanized fungi whose pathogenicity is strongly

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associated with the presence of melanin in the fungal cell wall. Melanin acts as a major virulence factor by protecting fungal cells from oxidative stress, inhibiting phagocytosis and reducing susceptibility to host immune responses, thereby facilitating chronic infection and progressive tissue destruction (REVANKAR; SUTTON, 2010; SÁNCHEZ-CÁRDENAS; ISA-PIMENTEL; ARENAS, 2023). These characteristics help explain the slow, indolent and locally aggressive behavior observed in many deep-seated phaeohyphomycoses, including those involving bone structures (REVANKAR; SUTTON, 2010; SÁNCHEZ-CÁRDENAS; ISA-PIMENTEL; ARENAS, 2023; HE et al., 2022).

Although phaeohyphomycosis has historically been regarded as an opportunistic infection, recent studies have demonstrated that a substantial proportion of cases occur in immunocompetent individuals, particularly in localized forms of the disease (HE et al., 2022; SIGAMANI et al., 2025; QUEIROZ-TELLES et al., 2017). In the oral and maxillofacial region, several reports describe phaeohyphomycosis affecting the palate, oral mucosa or maxillary sinus in patients without identifiable systemic immunosuppression, supporting the concept that local factors, microtrauma or environmental exposure may play a decisive role in fungal inoculation and establishment (RAWAL; KALMAR, 2012; ANDRADE et al., 2017; CARDOSO et al., 2007; SHUKLA et al., 2009; RAI et al., 2014; SURESH et al., 2022).

Intraosseous involvement of the maxilla by dematiaceous fungi remains exceptionally rare. When present, radiographic findings typically include ill-defined or partially defined radiolucent lesions with cortical thinning, perforation and extension to adjacent anatomical spaces, such as the nasal cavity or maxillary sinus (GAVITO-HIGUERA et al., 2016; ARIBANDI; McCOY; BAZAN, 2007; RAI et al., 2014; SURESH et al., 2022). These imaging characteristics closely overlap with Those of aggressive odontogenic cysts, chronic osteomyelitis and, in some cases, malignant neoplasms, reinforcing the importance of histopathological evaluation for definitive diagnosis (GAVITO-HIGUERA et al., 2016; ARIBANDI; McCOY; BAZAN, 2007; RAI et al., 2014; SURESH et al., 2022).

Histopathologically, phaeohyphomycosis is characterized by a granulomatous or pyogranulomatous inflammatory response, frequently associated with histiocytes, multinucleated giant cells and cholesterol clefts, reflecting the chronicity of the infection (REVANKAR; SUTTON, 2010; SÁNCHEZ-CÁRDENAS; ISA-PIMENTEL; ARENAS, 2023; QUEIROZ-TELLES et al., 2017). In routine hematoxylin–eosin staining, fungal elements may be subtle or inconspicuous, appearing as faintly pigmented hyphae or rounded structures within the inflammatory infiltrate. Therefore, the use of special stains such as Grocott–Gomori methenamine silver and periodic acid–Schiff is essential to highlight fungal morphology, while Fontana–Masson staining plays a critical role in confirming the presence of melanin and differentiating dematiaceous fungi from hyaline fungal

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organisms (REVANKAR; SUTTON, 2010; SÁNCHEZ-CÁRDENAS; ISA-PIMENTEL; ARENAS, 2023; QUEIROZ-TELLES et al., 2017).

From a therapeutic standpoint, the management of deep or intraosseous phaeohyphomycosis generally requires a combined approach. Surgical debridement is recommended to remove necrotic tissue, reduce fungal burden and improve antifungal penetration, particularly in localized disease (REVANKAR; SUTTON, 2010; SÁNCHEZ-CÁRDENAS; ISA-PIMENTEL; ARENAS, 2023; HE et al., 2022; CHOWDHARY et al., 2014). Systemic antifungal therapy with azoles such as itraconazole, voriconazole or posaconazole is frequently indicated, with itraconazole remaining a first-line option for many localized infections due to its efficacy against dematiaceous fungi and favorable safety profile (REVANKAR; SUTTON, 2010; SÁNCHEZ-CÁRDENAS; ISA-PIMENTEL; ARENAS, 2023; HE et al., 2022; CHOWDHARY et al., 2014). The favorable clinical outcome observed in the present case, with complete resolution and no recurrence after two years, is consistent with previous reports involving immunocompetent patients treated with combined surgery and antifungal therapy (RAWAL; KALMAR, 2012; ANDRADE et al., 2017; CARDOSO et al., 2007; SHUKLA et al., 2009; RAI et al., 2014; SURESH et al., 2022).

The main limitation of this report is the absence of fungal culture or molecular identification at the species level, which was not possible because the entire specimen was submitted for histopathological examination. However, the morphological features observed in routine and special stains were sufficient to establish the diagnosis of phaeohyphomycosis, as recommended by current diagnostic guidelines (REVANKAR; SUTTON, 2010; SÁNCHEZ-CÁRDENAS; ISA-PIMENTEL; ARENAS, 2023; HE et al., 2022; CHOWDHARY et al., 2014).

In summary, this case reinforces that phaeohyphomycosis, although rare, should be included in the differential diagnosis of atypical intraosseous maxillary lesions, especially when imaging findings suggest an aggressive cystic process and histopathological examination reveals granulomatous inflammation. Awareness of this entity and appropriate use of special stains are essential to avoid misdiagnosis and to guide adequate clinical management.

4. Conclusions

Intraosseous maxillary phaeohyphomycosis is an exceptionally rare condition that may closely mimic odontogenic cysts or other aggressive intraosseous lesions, particularly in immunocompetent patients. This case highlights the decisive role of histopathological examination with special stains in establishing an accurate diagnosis and guiding appropriate treatment. Early recognition and combined surgical and antifungal management may result in favorable outcomes and prevent unnecessary or inadequate therapeutic approaches.

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Ethical Considerations

Written informed consent was obtained from the patient for publication of this case report and accompanying images. Patient anonymity was preserved.

Conflicts of Interest

The authors declare no conflicts of interest related to this study.

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