


## Oral manifestation of aspergillosis in an immunocompetent child: case report

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### Abstract:

Fungi is a kingdom present in nature that has low virulence, occasionally they are capable of causing diseases when there is a commitment on host's defenses. There are reports of increased incidence of the disease in immunocompetent patients; however, oral manifestations are rare. A 5-years-old male patient was referred to the Maxillofacial Surgery service for evaluation of a vegetating, sessile a vascularized lesion with granulomatous areas, without reports of systemic changes. Computed tomography showed mucosal thickening in the associated maxillary sinuses and area of bone erosion on the floor of the left nasal cavity, and after that, incisional biopsy was performed obtaining the diagnosis of aspergillosis. Hospitalization of the patient was necessary and the treatment with intravenous liposomal amphotericin B and oral Voriconazole lasted six weeks, successfully treating disease and remitting the lesion. Since then, patient remains asymptomatic and without signs of the disease after 3 months of hospital discharge. The authors emphasize the importance of early diagnosis and the appropriate antifungal agents selection for successful treatment.

**Keywords:** Invasive Fungal Infection, Aspergillosis, Aspergillus.

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## INTRODUCTION

Fungi is a kingdom present in nature that has low virulence, occasionally they are capable of causing diseases when there is a commitment on host's defenses.<sup>1</sup> *Aspergillus* is a fungal genus with a great diversity of species found saprophyte in soil, water and decomposing organic materials. However, few species are able to develop at room temperature causing an opportunistic infection in humans, known as Aspergillosis.<sup>2,3</sup> The disease affects most frequently upper airways, with *Aspergillus* being the fungus most commonly associated with paranasal sinuses infections, especially the maxillary sinus.<sup>4</sup> Aspergillosis pathogenicity depends on two factors: the strain of the fungus involved and the host's immune system status.

Fungal rhinosinusitis has a classification of invasive and non-invasive forms, depending on the invasion of the mucosal layer and bone destruction. Non-invasive forms are allergic sinusitis, which can damage sinus mucosa and cause bone atrophy. Invasive forms, on the other hand, can be chronic or acute, the latter progressing rapidly, destroying the nasal cavity, the sinuses, palate and adjacent structures such as orbit and central nervous system, which can lead the patient to death in a few days.<sup>4,6,7</sup>

Tissue invasion is uncommon and occurs frequently in immunocompromised patients (chronic use of glucocorticoids, presence of malignancies, those undergoing bone marrow transplant and presence of neutropenia); however, there are reports of an increase in the incidence of the disease also in immunocompetent patients.<sup>4-8</sup>

In the present study, authors aim to report the case of an immunocompetent child diagnosed with invasive Aspergillosis presenting with oral manifestation.

## CASE REPORT

A 5-years-old male child from rural area with a history of progressive hypertrophy of the hard palate - noticed 15 days ago by his caregivers - referred to the Maxillofacial Surgery and Traumatology service.

On physical examination, we noticed a vegetating, sessile and vascularized lesion with granulomatous areas (Figure 1 - Intra-oral aspect of injury). Laboratory tests did not show any changes that indicated any suspicion of immunosuppression. Computed tomography revealed a mass of density similar to soft tissue on the left side of the palate and mucous thickening in the maxillary

sinuses associated with an area of bone erosion on the floor of the nasal cavity on the same side (Figure 2 - Coronal and axial tomographic section showing soft tissue mass in the palate region and thickening of the nasal mucosa). The main diagnostic hypotheses were fungal infections with a granulomatous aspect, such as paracoccidioidomycosis and blastomycosis.

As it is a child and an extremely vascularized lesion, for patient comfort and better bleeding control, the patient was referred to the operating room for an

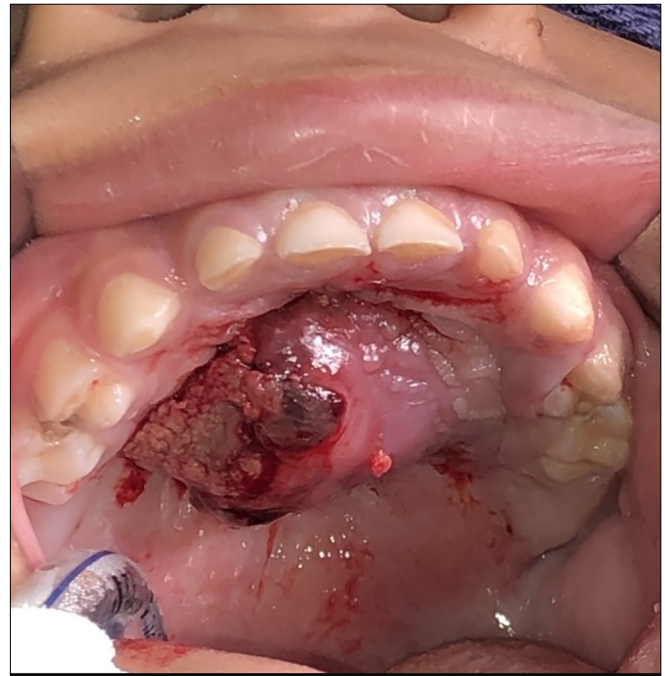


Figure 1. Intra-oral aspect of injury.

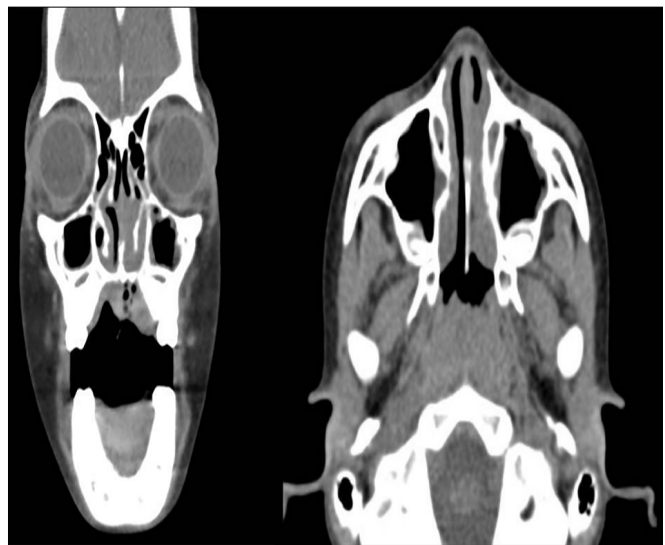
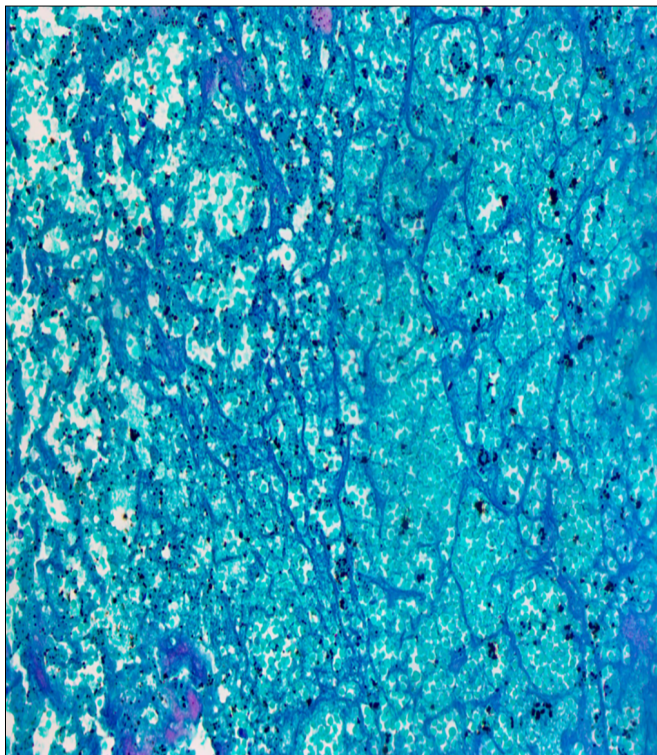


Figure 2. Coronal and axial tomographic section showing soft tissue mass in the palate region and thickening of the nasal mucosa

incisional biopsy of the lesion in the palate region under general anesthesia and the sample was sent to histopathological examination. The specimen received in the department of Oral Pathology of Federal University of Goias for histopathological examination (in formalin) consisted of small black bits of soft tissue of irregular sizes and shapes. Histological sections showed granulation tissue with a mixed inflammatory infiltrate associated with numerous fungal structures consisting of hyaline hyphae septated at a 90° angle, suggesting infection caused by *Aspergillus* spp (Figure 3 - Fungal structures consisting of hyaline hyphae septa at an angle of 90°, suggesting infection by *Aspergillus* spp). Grocott's methenamine silver stain allowed the organisms to be visualized more clearly.

With the diagnosis of aspergillosis, medical treatment started with administration of intravenous 5mg/ml liposomal amphotericin B (20ml daily in a single dose) for 4 weeks, followed by oral Vociconazole 200mg (100mg twice a day) for 2 weeks. After 6 weeks of treatment, the patient presented remission of the lesion on the palate and a new tomography showed a decrease in the thickening of the mucosa of the maxillary sinuses. After three months of hospital discharge, the patient remains asymptomatic and has no clinical or imaging evidence of disease recurrence (Figure 4 -



**Figure 3.** Fungal structures consisting of hyaline hyphae septa at an angle of 90°, suggesting infection by *Aspergillus* spp. (Grocott's methenamine silver).



**Figure 4.** Postoperative appearance showing absence of infectious and inflammatory signs, as well as lesion remission.

Postoperative appearance showing absence of infectious and inflammatory signs, as well as lesion remission).

## DISCUSSION:

It is common to find *Aspergillus* species in decomposing organic materials and in the soil. Therefore, it is a very common infection in people from rural areas and farmers.<sup>1</sup>

Invasive fungal infection of the paranasal sinuses in immunocompetent patients are rare and require an accurate diagnostic so that the appropriate treatment can be started early in order to avoid sequelae and even death. Aspergillosis originating in the upper airways can cause invasion of the orbital cavity as well as intracranial, in these cases associated to a poor prognosis, reaching high mortality rates when there is involvement of the central nervous system.<sup>4,7,9</sup>

Symptoms of maxillary sinus infections include headache, nasal congestion, fever, pain around the eyes and in the face. Aspergillosis must be a diagnostic hypothesis in cases where the condition is resistant to conventional therapies or in episodes of recurrent sinusitis.<sup>3-5</sup>

Aspergillosis invasive form can extend to oral cavity, causing a perforation in the palate. Clinically, it can be noted a gray or violet diffuse hypertrophy of the

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oral mucosa, being associated with ulcers and necrotic tissue.<sup>3,5</sup> The differential diagnosis includes tuberculosis, necrotizing sialometoplasia, syphilis, malignant ulcer and other fungal infections.<sup>5</sup>

Histopathological examination is essential for establishing the definitive diagnosis. The histopathological characteristics found are a variable amount of branched and septate hyphae, with a tendency to branch at acute angles and invasion of small blood vessels. In immunocompetent patients, it is possible to verify granulomatous inflammatory reaction and necrosis, while in immunocompromised patients, there is a presence of great tissue necrosis.<sup>1,3,4</sup>

Aspergillus infections treatment suggests the immediate administration of antifungal treatment, control of predisposing conditions and surgical interventions if necessary. Strongly recommended, Voriconazole is the drug of choice for the treatment of invasive aspergillosis, as it has better tolerance, increased efficacy, improved survival rate and lower toxicity when compared to liposomal amphotericin B, which for decades has been the first-line medication for treating the disease.<sup>1,4,5,7,10</sup>

In the present case, a combination of liposomal amphotericin B for 4 weeks and Voriconazole for 2 weeks was the performed treatment.

Due to the invasive characteristic of the lesion and its ability to leave sequelae or lead the patient to death, our case highlights the importance of mouth assessment, the need for early diagnosis and adequate

treatment, enabling clinical improvement and quality of life for patients.

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