



**CASE REPORT**

# Oral paracoccidiodomycosis: a case report

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## Abstract

Paracoccidiodomycosis (PCM) is a mycosis caused by fungi of the genus *Paracoccidioides*. Although it is rare globally, PCM is especially prevalent in Latin America, with 10 million infected individuals. Notably, approximately 80% of these cases are in Brazil, where PCM accounts for the highest mortality rate among systemic mycoses. Annual incidence estimates of PCM in Brazil range from 0.71 to 3.70 cases per 100,000 inhabitants. There is an important relationship between PCM and agriculture, since the soil serves as a reservoir for the microorganism. The fungus is primarily contracted through inhalation. In about 5% of patients, the disease may progress to an acute/subacute or chronic state, affecting various organs, including the oral cavity. Oral lesions are observed in about 60% of diagnosed individuals and are more prevalent in middle-aged and elderly males. This report presents a case of a 58-year-old man with a PCM lesion on his soft palate.

**Keywords:** paracoccidiodomycosis; oral manifestations; soft palate.

**How to cite:** Nogueira GMC, Gomes EO, Andrade Filho EKR, Henriques SRM, Rêgo MMB. Oral paracoccidiodomycosis: a case report. Arch Head Neck Surg. 2023;52:e20230017. <https://doi.org/10.4322/ahns.2023.0017>

## Introduction

Paracoccidiodomycosis (PCM) is a deep endemic mycosis caused by fungi of the genus *Paracoccidioides*. Although it is rare globally, PCM is especially prevalent in tropical areas, such as in Latin America, with about 10 million infected individuals. Notably, 80% of these cases are in Brazil, making it the country's most deadly deep mycosis<sup>1</sup>. However, these figures might be underestimated because of a lack of mandatory reporting. In Brazil, for instance, PCM was only recently added to the National Mandatory Notification List of diseases, conditions, and public health events, but only a few states have included this disease in their local records<sup>2</sup>.

Annual incidence estimates of PCM in Brazil vary between 0.71 and 3.70 cases per 100,000 inhabitants. The disease is strongly linked to agricultural activities, like earthworks, soil preparation, and gardening practices, as the soil serves as a reservoir for the causative microorganism. Most affected patients had agricultural exposure in their early life, even though clinical manifestations appeared years later<sup>3</sup>.

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**Financial support:** None.  
**Conflicts of interest:** No conflicts of interest declared concerning the publication of this article.  
**Submitted:** July 05, 2023.  
**Accepted:** September 07, 2023.

The study was carried out at the Liga Norte Riograndense Contra o Câncer, Natal, RN, Brazil.



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PCM is contracted by inhaling the fungus, leading to a transient lung infection. In 5% of patients, the disease is not contained, and the patient can develop either an acute/subacute or chronic condition<sup>4</sup>. The acute/subacute form accounts for about 10% of cases and presents with lymphadenopathy, hepatosplenomegaly, digestive symptoms, skin lesions, and bone and joint involvement. Fever, weight loss, and anorexia are common findings. The chronic form affects approximately 90% of patients, setting in insidiously and lasting over four months. Lung involvement is frequent, as well as involvement of mucous membranes and skin<sup>3</sup>.

Both clinical forms can affect the oral cavity, although this finding is more common in the chronic form. Lesions typically appear on the lips, oropharynx, and oral mucosa<sup>5</sup>.

Microscopic examination and direct mycological testing are crucial for identifying the pathogen in secretions from lymph nodes, sputum, or material collected from lesions<sup>2</sup>. As for treatment, even though a wide range of therapeutic options is available for managing the disease, in clinical practice, itraconazole, cotrimoxazole (sulfamethoxazole/trimethoprim combination), and amphotericin B are most commonly used<sup>5</sup>.

We report a case of a 58-year-old man with a PCM infiltrative lesion in his soft palate.

## Case report

A 58-year-old male patient, a smoker for 30 years (60 pack-years), diagnosed with diabetes mellitus and with a history of psoriasis, was referred to the Head and Neck Surgery Department because of a painful lesion on the soft palate. He reported that the lesion had appeared 40 days ago, was painful and erythematous, and had not exhibited any bleeding or secretions.

Upon physical examination, he displayed an infiltrative lesion on the soft palate measuring about 2.5 cm (Figure 1), and another on the right buccal region approximately 1.5 cm in size, with no cervical lymphadenopathy (Figure 2).

An incisional biopsy was chosen as the course of action, which was performed using a punch biopsy instrument. The removed fragment was irregular, elastic, and whitish, measuring 0.4 x 0.3 cm, and was sent for histopathological analysis.

The histopathological examination ruled out neoplasia and revealed an exacerbated and ulcerated chronic inflammatory process with granulation tissue associated with the presence of fungal yeasts compatible with *Paracoccidioidis* spp. (Figure 3). Moreover, the search for fungal organisms using the Grocott method revealed organisms morphologically consistent with the genus *Paracoccidioidis* spp. (budding yeasts and/or in the shape of a ship's helm).

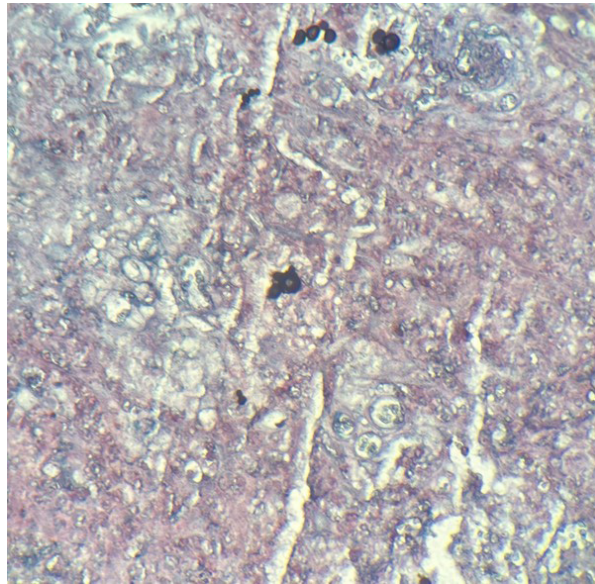
Given the infectious nature of the condition, the patient was referred to infectious disease specialists for treatment with the antifungal drug Itraconazole.



**Figure 1.** Image of the lesion in the oral cavity (palate).



**Figure 2.** Image of the lesion on the buccal mucosa.



**Figure 3.** Histopathological examination with the search for fungal organisms using the Grocott method, showing helm-shaped yeasts.

### Ethical issues

The patient agreed to the publication of this study by signing an Informed Consent Form.

### Discussion

The lungs are the primary site affected by the disease, but about 60% of affected individuals can present with an oral lesion or in the nasopharynx, and this might be the main finding of the disease<sup>1,2</sup>.

Oral cavity lesions most commonly manifest as ulcers, either singular or multiple, located in the gum, palate, lip commissure, and buccal mucosa areas. They are more prevalent in men, with a ratio of about 9.5:1. This might be attributed to hormonal factors, as estradiol prevents the fungus from differentiating into its pathogenic form. Middle-aged and elderly individuals (aged >50 years) are more affected<sup>2</sup>. Smoking is also considered a significant risk factor for developing the oral form of PCM, with smokers having a risk 14 fold higher<sup>5</sup>. These findings align with our case, which involves a middle-aged male smoker.

The clinical diagnosis of PCM can be challenging, given that the lesions can resemble other infectious or neoplastic diseases<sup>2</sup>. Since we are dealing with an oncology service, the primary hypothesis was squamous cell carcinoma (SCC).

Regarding oral PCM, the histopathological examination of biopsied lesions is the most commonly utilized method because of its straightforward evaluation<sup>2</sup>. However, the direct mycological test should be prioritized as it is more cost-effective and less invasive, despite requiring a professional capable of correctly identifying *Paracoccidioides*<sup>5</sup>. Nevertheless, the histopathological exam rules out malignant tumors, which was the initial hypothesis for this case.

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Initiating treatment depends on disease severity, and it is essential to adequately manage the malnutrition-immunosuppression-infection triad<sup>1,2</sup>. The first-line drug is Itraconazole, while Ketoconazole and Amphotericin B serve as alternatives, with the latter being used in severe cases<sup>5</sup>. For our patient, Itraconazole was chosen.

Thus, PCM has a significant incidence of oral lesions, which often serve as the primary sign and diagnostic confirmation site. From this perspective, curing the disease hinges on early identification of the pathogen and patient adherence to effective treatment. In oral lesions, it is essential to consider an infectious profile. A biopsy with histopathological analysis is vital to discard malignant tumors.

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