

# Consequences of late diagnosis paracoccidioidomycosis: case report

## *Consequências do diagnóstico tardio de paracoccidioidomicose: relato de caso*

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### ABSTRACT

**Introduction:** Paracoccidioidomycosis is an endemic fungal infection of great prevalence in Latin America. **Case report:** A 68-year-old male patient was referred to the Universidade Federal do Maranhão (UFMA) due to an ulcerated lesion with moriform appearance throughout the palate, with symptoms. The clinical diagnosis was squamous cell carcinoma and the incisional biopsy confirmed the diagnosis of chronic paracoccidioidomycosis. Grocott-Gomori and periodic acid Schiff (PAS) were used as special stains. **Conclusion:** It is important to observe the clinical characteristics of the lesion, as well as to clarify the population about this infection since health education is a fundamental step to prevent and diagnose the lesion.

**Key words:** paracoccidioidomycosis; early diagnosis; health education.

### RESUMO

**Introdução:** A paracoccidioidomicose é uma infecção fúngica endêmica de grande prevalência na América Latina. **Relato de caso:** Paciente do sexo masculino, 68 anos de idade, foi encaminhado à Universidade Federal do Maranhão (UFMA) devido a uma lesão ulcerada de aspecto moriforme em toda a extensão do palato, com sintomatologia. O diagnóstico clínico foi de carcinoma epidermoide, e a biópsia incisional confirmou o diagnóstico de paracoccidioidomicose crônica. Grocott-Gomori e ácido periódico Schiff (PAS) foram utilizados como colorações especiais. **Conclusão:** É importante observar as características clínicas da lesão, bem como orientar a população acerca dessa infecção, uma vez que a educação em saúde é um passo fundamental para prevenir e diagnosticar a lesão.

**Unitermos:** paracoccidioidomicose; diagnóstico precoce; educação em saúde.

### RESUMEN

**Introducción:** La paracoccidioidomycosis es una infección fúngica endémica de gran prevalencia en Latinoamérica. **Reporte de caso:** Paciente del sexo masculino, 68 años de edad, fue referido a la Universidade Federal do Maranhão (UFMA) debido a una lesión ulcerada de aspecto moriforme en toda la superficie del paladar, con síntomas. El diagnóstico clínico fue carcinoma epidermoide, y la biopsia incisional confirmó el diagnóstico de paracoccidioidomycosis crónica. Grocott-Gomori y ácido periódico de Schiff (PAS) fueron utilizados como tinciones especiales. **Conclusión:** Es importante observar las características clínicas de la lesión, así como orientar la población sobre esa infección, una vez que la educación en salud es un paso fundamental para prevenir y diagnosticar la lesión.

**Palabras clave:** paracoccidioidomycosis; diagnóstico precoz; educación en salud.

## INTRODUCTION

Paracoccidioidomycosis (PMC) is a chronic granulomatous disease caused by the dimorphic fungus called *Paracoccidioides brasiliensis*. It is the mycosis most frequently observed in patients living in South America; Brazil is the main affected area<sup>(1-4)</sup>. The fungus from contaminated soils can reach the upper airways through inhalation, and the lungs are the first affected sites; then, it can spread to organs such as skin, mucous membranes, gastrointestinal region, spleen and lymphatic system. Chewing sticks contaminated with the fungus is another form of contagion very common among farm workers<sup>(1,4,5)</sup>.

PMC is clinically classified as acute, subacute and chronic. The characteristics depend on factors such as: virulence of the *P. brasiliensis* strain, host immune response, affected anatomical sites, and particularity of each patient<sup>(2,6)</sup>. There is a significant predilection for males, in the age group between 30 and 50 years, and their occupation as a risk factor, which is related to soil management<sup>(4,5)</sup>.

The diagnosis of PMC can be performed by isolating the fungus in culture, serological tests, skin and mucous membranes histopathological tests, in addition to chest X-rays to observe pulmonary involvement<sup>(1, 2, 5)</sup>. Histologically, hematoxylin and eosin (HE) staining often does not allow the recognition of parasite morphology, requiring the use of Grocott-Gomori staining and the periodic acid-Schiff (PAS) method<sup>(7)</sup>, which allows visualization of multiple budding daughter-cells linked to mother-cells, resulting in an appearance similar to the “Mickey Mouse ears” or “boat steering wheel”<sup>(8)</sup>.

The treatment of patients depends on the severity of the infectious process in which it is found. Drugs derived from sulfonamides have been used since the 1940s and are still a great treatment option for cases with little or medium impairment. Intravenous amphotericin B is the most suitable drug for the most severe cases and at risk of death. In cases with marked severity, but with no risk of death, antifungal such as itraconazole or ketoconazole can be used for long periods and show good results<sup>(1,5)</sup>.

## CASE REPORT

Male patient, 68 years old, brown, resident in the Maranhão state's countryside, sought medical care at the health center due to an oral lesion extending along the entire length of the hard and soft palate, with moriform ulceration feature. After clinical examination, he was referred to dental care in the state capital city, where he was redirected to the Dentistry course at the Universidade Federal do Maranhão (UFMA) due to the presumed clinical diagnosis of

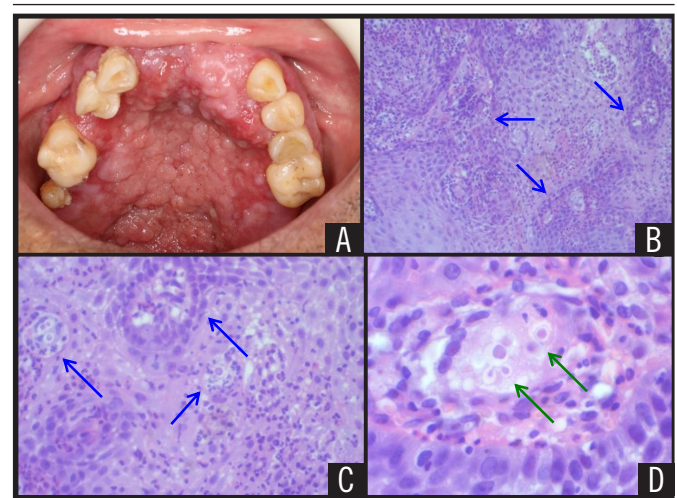
squamous cell carcinoma. In the anamnesis, he did not report any systemic changes; he spends most of the day at work (agriculture) and has the habit of putting sticks on his mouth to chew.

In the extraoral examination, he had similar lesions in the region of the index toe, severe weight loss, breathing difficulties, and physical weakness; all these changes were noticed by him more than three months ago. In the intraoral examination, he presented tooth loss, unsatisfactory oral hygiene, remaining dental calculus, and an extensive ulcerated lesion on the hard palate with extension to the soft palate (**Figure 1A**). In view of these clinical findings, the diagnostic hypothesis was squamous cell carcinoma or PMC. Incisional biopsy of the lesion was performed in four different areas to elucidate the diagnosis. The material was sent to the oral pathology laboratory of the UFMA dentistry course.

The processing was carried out in the laboratory, and the slides were HE stained. The lesion was granulomatous, characterized by foci of necrotic cells or infected with a specific agent, moreover, surrounded by a necklace-like of lymphocytes and plasmocytes interspersed with dense connective tissue. The fungus was identified in the examined sections (**Figure 1B**).

For better clarification of the case, special stains (PAS and Gomori-Grocott) were performed to visualize the yeast phase of the fungus from the collected material (**Figure 1C and D**). The definitive diagnosis was chronic PMC, since the patient had systemic changes. Then, he was referred for treatment and medical follow-up – due to his weakness and the seriousness of the case.

The initial treatment was administration of intravenous amphotericin B. The patient was admitted to the university hospital and, after 15 days of treatment, he showed no evolution of the respiratory condition and died.



**FIGURE 1** – Clinical and histopathological aspects

A) clinical aspect; B) presence of granulomas (blue arrows); C) presence of granulomas (blue arrows) with visualization of the yeast; D) yeast form of the fungus (green arrows).

## DISCUSSION

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In Brazil, systemic mycoses are not included in the compulsory notification list, which makes it difficult to establish the correct prevalence of PMC<sup>(6,9)</sup>. As it is not a notifiable disease, information about its existence is reduced. Due to the absence of regular recording of epidemiological and clinical information on this mycosis, knowledge, unfortunately, is based only on case series studies and isolated studies on the microorganism and its lineages<sup>(9)</sup>, which precludes more adequate analyzes in relation to the real prevalence of cases and the implementation of preventive campaigns to help the population, both in non-contamination and in the search for prior care.

The absence of campaigns aimed at providing guidance on this disease makes early diagnosis even more difficult. The misinformation of health professionals generates a low rate of identification of patients in the initial stage of the disease<sup>(10)</sup>. The diagnosis is made only late and makes it impossible for patients to cure and survive, which was observed in the case reported.

In order to make the definitive diagnosis and appropriate treatment, it is necessary to visualize the fungus. In this case, the methods of choice for staining – PAS and Gomori-Grocott<sup>(8)</sup>,

the latter detects the presence of mucin and glycogen –, enabled the release of aldehyde groups, resulting from the pretreatment with chromic acid. Then, the detection of reduction of the alkaline complex by silver nitrate occurred, since the background cell wall is rich in polysaccharides<sup>(5,11)</sup>. When identified, appropriate treatment is implemented immediately and patients in severe stages are more likely to cure.

The treatment of choice is high-doses drug therapy, in addition to nutritional support, managing the sequelae and maintaining the patient in good health with strict monitoring<sup>(8)</sup>. Our report demonstrates that the delay in diagnosis, and consequently in therapy, decreases the recovery chance of the infected.

## CONCLUSION

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Given the above and based on the analysis on the literature, we conclude that it is very important to observe the clinical features of the lesion and make the recommendation for the correct and rapid diagnosis. In addition, it is necessary to stimulate education and promote information and communication to the population on the various forms of infection of the disease, aiming at reducing the occurrence of cases.

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