



Microbes and Infectious Diseases

Journal homepage: <https://mid.journals.ekb.eg/>

Case report

Hyalohyphomycosis in an orthopedic patient: Case report

Leonardo Henrique Bertolucci ^{*1}, Pedro Yuki Shibuya Moreira Pinto ¹, Alex Oliboni Sussela ², Lauro Manoel Etchepare Dornelles ², Osvaldo André Serafini ²

1- Medicine School, Pontifícia Universidade Católica do Rio Grande do Sul, Av. Ipiranga, 6681, Porto Alegre, RS, Brazil, 90160-092.

2- Department of Orthopaedics and Traumatology, Hospital São Lucas da Pontifícia Universidade Católica do Rio Grande do Sul, Av. Ipiranga, 6681, Porto Alegre, RS, Brazil, 90160-092.

CASE REPORT INFO

Case Report history:

Received 3 May 2021

Received in revised form 8 June 2021

Accepted 11 June 2021

Keywords:

Hyalohyphomycosis

Fungi

Orthopedics

ABSTRACT

Hyalohyphomycosis are opportunistic fungal infections caused by saprobic fungi. Such mycoses usually occur in immunosuppressed patients. The most affected organ is usually the lungs. A male patient has arrived at the Orthopedics and Traumatology Service complaining of a mass on the dorsal surface of his right foot. On physical examination, a subcutaneous, elastic, non-mobile mass was observed on the dorsal surface of the right foot. Ulcerated lesion, without drainage of secretion and with pain on palpation. He reported focal increase in the dorsal surface of the right foot for 7 years. Among other diagnostic possibilities, imaging studies indicated the hypothesis of a giant cell tumor with a tendon sheath. The patient was then submitted to biopsy of the lesion. The anatomopathological study showed chronic suppurative inflammation with granulation tissue and the presence of septate hyaline hyphae compatible with hyalohyphomycosis. Surgical resection of the lesion and systemic treatment were then performed. Patient evolved well after the procedure.

Introduction

Hyalohyphomycosis are opportunistic fungal infections caused by saprobic fungi (which feed on decomposing organic matter) [1]. This group of mycoses are caused by septate filamentous fungi, with hyaline walls (except *Aspergillus* species), therefore, in histopathological examination, the visualization of septate hyaline hyphae, or toroidal hyphae is expected. [1,2]. Such mycoses usually occur in immunosuppressed patients [1].

Although normally its etiological agent is *Aspergillus* spp. (aspergillosis), *Fusarium*, *Penicillium* and *Paecilomyces* fungi can also cause the disease. Any infection caused by hyaline non

pigmented fungus are called Hyalohyphomycosis, which, when parasitic on a tissue, appears as hyaline septate hyphae [1].

Hyalohyphomycosis is becoming more and more frequent in tropical regions such as Brazil and the factors that favor an infection may depend on the individual, such as the pre-existence of serious diseases such as diabetes, leukemia, pernicious anemia (such as serious hematological changes), liver diseases, serious deficiency states, tuberculosis, abscesses, bronchiectasis and emphysema (diseases that can cause sequelae in the respiratory tract). Such basic diseases are aggravated when the individual is exposed to an intense therapy

DOI: 10.21608/mid.2021.76193.1154

* Corresponding author: Leonardo Henrique Bertolucci

E-mail address: leonardohbertolucci@gmail.com

© 2020 The author (s). Published by Zagazig University. This is an open access article under the CC BY-NC-ND license <https://creativecommons.org/licenses/by/4.0/>.

of antibiotics, corticosteroids and cytotoxic drugs, favoring the infection [1,3].

As the infection occurs through the respiratory tract, the most affected organ is usually the lungs, although there are reports of endocarditis, skin, eye lesions and even brain abscesses caused by the fungus. Infection can also occur by direct inoculation. Although rare, the fungus can also cause osteoarticular changes, in which case it is important to perform the differential diagnosis with other pathologies, especially neoplasms, such as Giant Cell Tumor (GCT) [1,3].

The diagnosis is made after collecting the material and processing the mycological study through a culture. Pathological material can be made up of sputum, earwax, eye exudate, pus from gummy lesions. Examination reveal only the presence of hyaline and branched septate hyphae, without being able to affirm the genus that causes mycosis [1]. European Society of Clinical Microbiology and Infectious Diseases Fungal Infection Study Group and European Confederation of Medical Mycology recommendations for diagnosis are direct microscopy and culture examination [3].

The choice of clinical treatment is still unclear. In addition, the treatment also depends on the causative fungus, considering that hyalohyphomycosis can be caused by several fungal etiologies. Thus, among the main treatments are the use of Amphotericin B, Itraconazole, Voriconazole, Posaconazole, Caspofungin, Micafungin, Anidulafungin and surgical resection [1,3].

When the disease is at an early stage, it is important that localized therapy is performed to prevent progression to a more aggressive disease. The use of topical antifungals and surgical resection are options in these cases [3,4].

The purpose of this report is to expose this rare presentation of the disease to the medical community and to evaluate the symptoms of adult patients with cutaneous hyalohyphomycosis, as well as to review the differential diagnosis options of the pathology.

Case report

Male patient, 53 years old, arrived at the Orthopedics and Traumatology Service of Hospital São Lucas da PUCRS, Porto Alegre, Brazil complaining of a mass on the dorsal surface of his right foot. He reported focal increase in the dorsal surface of the right foot 10 years ago, when he

underwent surgical excision of the nodule in a hospital in his hometown, without anatomopathological analysis of the lesion. He reported recurrence in 2014, when he sought care at a medical center and was referred to the current Orthopedics and Traumatology Service. He reports pain when wearing closed shoes and sporadic drainage of sero-hematic secretions. The patient had no other diseases and his job was not related to exposure of hyalohyphomycosis.

On physical examination, a subcutaneous, elastic, non-mobile mass was observed on the dorsal surface of the right foot. The lesion was ulcerated, without drainage of secretion and with tender on palpation. At X-ray, there was no evidence of bone lesions and magnetic resonance imaging revealed a massive expansive lesion with lobulated, multinodular contours, which undergoes intense enhancement after the contrast medium, of a nature to be clarified. The lesion measures approximately 5.9 X 3.6 X 1.8cm in its antero-posterior, lateral-lateral and craniocaudal axis, showing no signs of bone malnutrition or invasion of the other adjacent structures next to the extensor tendons of the second toe.

Lesions with similar characteristics, but with smaller dimensions, could be seen next to the extensor tendons of the third toe, measuring approximately 4.6 X 1.8 X 0.9 cm in their anteroposterior, laterolateral and caudal radius axis.

Among other diagnostic possibilities, computed tomography and ultrasonography indicated the hypothesis of a GCT with a tendon sheath.

The patient was then sent to biopsy of the lesion. The anatomopathological study showed chronic suppurative inflammation with granulation tissue and the presence of hyaline septate hyphae compatible with hyalohyphomycosis.

The surgical resection of the lesion was then performed (**Figure 1**) and systemic treatment was started with Itraconazole 100mg subcutaneously for 2 months [5]. Upon definitive anatomopathological examination (**Figure 2**), fibroadipose connective tissue with chronic suppurative inflammation containing suppurative granules, microabscesses and fibrosis was observed. Positive fungi research, with numerous septate hyphae and conidia.

After a year, the patient evolved well after the procedure, maintaining the extension

movements of the second right toe and showed no new growth in the region or elsewhere.

Figure 1. Hyalohyphomycosis being removed in surgery.



Figure 2. Macroscopic piece demonstrating hyalohyphomycosis.



Discussion

Hyalohyphomycosis is a disease that mainly affects the immunosuppressed and has its greatest presentation in the pulmonary system. However, it is important to understand that this disease manifests itself in other ways, such as cutaneous. In the literature there are cases of subcutaneous infection by hyalohyphomycosis that manifest as subcutaneous nodules in a kidney transplant recipient [6] and as abscesses in a patient with myasthenia gravis [7].

Giant Cell Tumor is a mesenchymal neoplasm, characterized by the proliferation of multinucleated giant cells that resemble osteoclasts, in a mononuclear cell stroma. The main differential

diagnoses, both from a clinical, radiographic and anatomopathological point of view, are aneurysmal bone cyst, telangiectatic osteosarcoma and chondroblastoma [8]. In this case, hyalohyphomycosis manifested similarly to GCT both clinically and on imaging tests. Thus, the differential diagnosis can only be made through the anatomopathological examination.

The limitation of this case report is that the authors do not have photos of the initial lesions. Despite this, the case is relevant to the scientific community due to its rarity and low probability of clinical and radiological diagnosis. Besides that, quick diagnosis is necessary because early therapy of localized disease is important to prevent progression to a more aggressive or disseminated infection [3].

Disclosure of potential conflicts of interest

The authors declare no conflicts of interest and there are no funding sources.

Financial Disclosures: there is no funding sources.

Contributors: All authors approved the final version.

Conception and design: Leonardo Henrique Bertolucci, Pedro Yuki Shibuya Moreira Pinto, Alex Oliboni Sussela, Lauro Manoel Etchepare Dornelles, Osvaldo André Serafini; Data collection: Leonardo Henrique Bertolucci, Pedro Yuki Shibuya Moreira Pinto, Alex Oliboni Sussela; Writing the article: Leonardo Henrique Bertolucci, Pedro Yuki Shibuya Moreira Pinto; Critical revision of the article: Alex Oliboni Sussela, Lauro Manoel Etchepare Dornelles, Osvaldo André Serafini; Final approval of the article: Leonardo Henrique Bertolucci, Pedro Yuki Shibuya Moreira Pinto, Alex Oliboni Sussela, Lauro Manoel Etchepare Dornelles, Osvaldo André Serafini.

References

- 1- **Naggie S, Perfect JR.** Molds: hyalohyphomycosis, phaeohyphomycosis, and zygomycosis. *Clin Chest Med* 2009;30(2):337-53.
- 2- **Londero AT, Ramos CD.** Cutaneous hyalohyphomycosis caused by *Acremonium falciforme* - Report of a case. *An Bras Dermatol* 1990; 65(2).

- 3-**Tortorano AM, Richardson M, Roilides E, van Diepeningen A, Caira M, Munoz P, et al.** European Society of Clinical Microbiology and Infectious Diseases Fungal Infection Study Group; European Confederation of Medical Mycology. ESCMID and ECMM joint guidelines on diagnosis and management of hyalohyphomycosis: *Fusarium* spp., *Scedosporium* spp. and others. *Clin Microbiol Infect* 2014 ;20 Suppl 3:27-46.
- 4-**Nucci M, Anaissie E.** *Fusarium* infections in immunocompromised patients. *Clin Microbiol Rev* 2007 ;20(4):695-704.
- 5-**Zaitz C, Porto E, Heins-Vaccari EM, Sadahiro A, Ruiz LR, Müller H, et al.** Subcutaneous hyalohyphomycosis caused by *Acremonium reeffi*: report of a case. *Revista do Instituto de Medicina Tropical de São Paulo* 1995: 267-270.
- 6-**Keskar VS, Wanjare S, Jamale TE, Mahajan D, Jawale SY, Fernandes G, et al.** Subcutaneous hyalohyphomycosis caused by *Fusarium* in a kidney transplant recipient. *Renal failure* 2014 ;36(7):1129-32.
- 7-**Raghavan R, Chithra G, Fernandez S, Suryanarayana BS, Singh R.** An unusual case of hyalohyphomycosis due to *Purpureocillium lilacinum* in a patient with myasthenia gravis. *Current medical mycology* 2018;4(2):36-39.
- 8-**Montgomery C, Couch C, Emory CL, Nicholas R.** Giant Cell Tumor of Bone: Review of Current Literature, Evaluation, and Treatment Options. *J Knee Surg* 2019 ;32(4):331-336.