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CASE REPORT/CAS CLINIQUE

A manifestation of cutaneous aspergillosis in immunocompetent host: A rare presentation as forearm mass lesion

Manifestation d'une aspergillose cutanée chez un hôte immunocompétent : une rare présentation par une tuméfaction de l'avant-bras

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Summary The *Aspergillus* species is a ubiquitous fungus, which can cause pathogenic and opportunistic fungal infections in the immunocompromised. This is an atypical occurrence in the host with an otherwise normal immune status. We report a case of an immunocompetent 45-year-old patient who developed cutaneous aspergillosis with a very benign course presenting simply with a gradually enlarging mass and none of the classical signs and symptoms. All prior laboratory examinations failed to detect or reproduce the organism or establish a diagnosis. Surgery was both diagnostic and therapeutic, to remove the mass which causes the patient pain and limitation of activity. This was to our advantage because the fungal elements were very well encapsulated and the mass was a well-organized conglomeration of cystic abscesses that even prolonged chemotherapy alone might not succeed in eradicating the infection.

MOTS CLÉS
Aspergillose ;

Résumé *Aspergillus* sp. est un champignon ubiquitaire qui peut être responsable d'infections opportunistes chez l'hôte immuno-compromis. Sa survenue est inhabituelle chez un hôte au statut immunitaire par ailleurs normal. Nous rapportons un cas chez un sujet âgé de 45 ans qui a

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développé une aspergillose cutanée à évolution favorable se présentant uniquement avec une masse augmentant progressivement de volume sans aucun des signes et symptômes classiques. Tous les examens de laboratoire ont échoué pour déceler ou cultiver l'organisme responsable et établir un diagnostic. La chirurgie a permis le diagnostic et le traitement en enlevant la masse responsable de la douleur et de la limitation d'activité. Par chance, les éléments fongiques étaient bien encapsulés et la tuméfaction était un agglomérat d'abcès enkystés bien organisés que même une chimiothérapie seule et prolongée n'aurait sans doute pas réussi à éradiquer.

Introduction

Mycetoma is a chronic, granulomatous, subcutaneous, inflammatory disease caused by a true fungi (eumycetoma) or filamentous bacteria (actinomycetoma). Mycetoma is categorized as a subcutaneous infection, because it is primarily limited to the subcutaneous tissue and dermis with rare systemic spread [2,4]. More common eumycetoma infections include Madurellamycetomatis, Nocardiabrasilienses and *Streptomyces*, while infections with the *Aspergillus* species is less common [6]. Among *Aspergillus* species, the subcutaneous tissue and dermis infections is mostly caused by *A. flavus, A. fumigatus*, and rarely, by *A. niger* [5].

In this report, we described a case of a chronic cutaneous infection caused by *A. niger* with an unusual presentation in an adult patient who has no known comorbidities.

Case report

A 45-year-old laboratory chemical analyst came into our institution with a large mass on the left forearm. The patient recounted that the mass started as a slowly growing soft nodule over the area of the ulnar styloid ten years ago. He could not recall a specific instance of trauma to the area. Previous consult at a local medical center and excision of the 3×3 cm mass was done. The patient was not aware of the operative findings and a biopsy was not done. The mass then recurred after a few months, beginning around the surgical excision site, but no consult was done. During this period, there was no history of pruritus, bullae or blisters, or drainage from the mass. Furthermore, there were no febrile episodes noted. The patient, however, complained of left wrist pain that limited his activities.

At our clinic, a 10×7 cm cystic, lobulated, nontender, nonerythematous, well-circumscribed mass was fixed at the lateral border of the middle to distal left forearm, with extensions volarly and dorsally. (Fig. 1) Skin was supple and smooth and no wounds or actively draining sinuses were noted. Hyperpigmented macules were noted over the mass, but on further inquiry, the patient denied history of pustules or purulent discharge. There was no note of any sensory or motor deficits to the left hand, and radial and ulnar arteries were both patent. Range of motion of the fingers and wrists were full, and no contractures were noted. Lymph nodes were neither tender nor palpable.

Aspiration of the mass was initially done which yielded a mucoid and bloody aspirate. Bacterial and fungal cultures were negative, and qualitative and quantitative fluid analysis was within normal range. Routine hematologic exam, blood chemistry, immunologic parameters, including HIV test, were normal. Plain radiographs showed a soft tissue shadow over the ulnar side without any bony erosion or periosteal reaction; bony anatomy was normal. MRI findings revealed multilobulated conglomerated cystic lesions with fibrotic thick wall. The lesion had high T1 signal intensity, with variable high and low signal intensity on T2. Peripheral enhancement was noted in the dorsomedial aspect of forearm, mainly in the subcutaneous fat layer, extending to the intramuscular layer of the extensor muscle and tendon.

Excision biopsy of the mass was done. Intraoperatively, we noted a 14×5 cm yellowish-tan, firm, fibrous, lobulated mass which was adherent to the subcutaneous tissue and surrounding muscle fascia and tendons (Fig. 2). It encircled the ulnar artery and nerve, however there was no vascular, neural, or tendon invasion. Extension deep into the flexor digitorum profundus volarly and into the extensor retinaculum of the 4th, 5th and 6th dorsal compartments was observed. The mass was also adherent to parts of the periosteum of the ulna. Pockets of whitish to purulent discharge were occasionally encountered.

Laboratory examination of the discharge yielded no bacterial or fungal growth. The lobulated mass grossly measured $10.5 \times 7 \times 5.4$ cm. On section, the cut surface shows multilocular cystic change with thick yellowish walls; the cysts contained blood clot and whitish to brown mucous (Fig. 3). Histologic examination revealed many fungus balls showing septated and acute forming hyphae on Grocott's Methenamin Silver stain and Periodic Acid-Schiff stains, with the presence of Splendore-Hoeppli phenomenon (Figs. 4 and 5). The biopsy sample were inoculated in Sabouraud dextrose agar with and without cycloheximide in duplicate and incubated at 37 and 25 °C, respectively. After 72 h, all samples were noticed with cottony white mycelium growth, which was covered with abundant black spores. Diagnosis was Mycetoma infection, consistent with *Aspergillus* species and also the fungal isolate was confirmed to be *A. niger* by the above-mentioned features. After antifungal susceptibility testing, at the lowest minimum inhibitory concentration, Amphotericin B was administered intravenously for 2 weeks. After receiving the fungal culture report, intravenous amphotericin B was administered, initially 0.5 mg/kg/day, and then increased to 0.75 mg/kg/day for 2 weeks. The patient was advised to report for regular follow-up. Final follow-up at 1 year there was significant improvement without pain, swelling or limitation of activity.