Attritional extensor tendon rupture in a patient with *Phialophora verrucosa* tenosynovitis: case report

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Abstract Deep tissue fungal infections of the hand are exceedingly uncommon. We present a case of fungal tenosynovitis caused by *Phialophora verrucosa* that led to extensor tendon rupture in a patient who was on chronic immunosuppressive therapy. Indolent fungal cysts can elude clinical diagnosis until excision is performed with definitive pathologic examination. In immunocompromised patients, antifungal therapy may be warranted after cyst excision even in the absence of acute infection to prevent subsequent progression to tenosynovitis.

Keywords Hand infection · *Phialophora verrucosa* · Phaeohyphomycosis · Tenosynovitis

Hand infections are frequently encountered in the community setting. Greater than 80 % of these cases are caused by bacterial pathogens commonly found in the skin and oral flora. Superficial infections may lead to deep tissue infections that present as tenosynovitis, arthritis, and osteomyelitis that require surgical intervention. In contrast, deep tissue fungal infections involving the hand are exceedingly rare and usually are caused by penetrating trauma or systemic seeding. Fungal infections are typically indolent before becoming clinically symptomatic. Immunocompromised patients are particularly susceptible to fungal infections, which are a distinct concern for this patient population [7]. Given its rarity, an algorithm for diagnosis and treatment of deep tissue fungal infections of the hand has not been established and only a handful of cases have been reported to date [4, 5, 10]. A delay in the diagnosis can lead to local progression of the infection or systemic dissemination [4]. We present a case of tenosynovitis from

Phialophora verrucosa leading to attritional tendon rupture in a patient on chronic immunosuppression.

Case Report

A 77-year-old woman presented with painless dorsal right hand mass. Her medical history was notable for long-standing autoimmune hepatitis that was well controlled on oral prednisone and azathioprine for the past 20 years. Fifteen years prior to her presentation, she sustained a minor penetration injury from a wood splinter over the dorsal right hand while gardening. This subsequently led to localized erythema and pain that subsided completely without further treatment. However, 2 years prior to her initial presentation, she noticed a gradually enlarging mass over the dorsum of the right hand. This mass was clinically diagnosed as a benign epidermal inclusion cyst and excised en bloc with no injury to the extensor tendons. Interestingly, the pathology examination revealed a cyst containing gray-tan gritty material and a foreign body consistent with a wood splinter. Furthermore, microscopic analysis demonstrated giant cell granulomas, fibrosis, and granulation tissue. Paraffin sectioning of the specimen showed suppurative inflammation with fungal elements consistent with phaeohyphomycosis. Because no acute infection was encountered and the cyst was entirely excised as indicated in the pathology report, there was no indication to initiate antifungal therapy. Postoperatively, the patient was seen in our clinic and the excision site healed without signs of infection. The patient was notified of the finding of fungal elements and was advised to present to her primary care physician for routine surveillance.

Five months after the operation, the patient presented to her primary care provider with new gradual onset of swelling over the dorsum of the right hand and wrist with tenderness over the fourth extensor compartment. When conservative

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measures did not resolve the symptoms, she presented to the emergency department for further evaluation. On exam, range of motion was intact and limited only by swelling. No sensory deficits were noted. Laboratory findings showed a normal white blood cell count. The erythrocyte sedimentation rate and C-reactive protein were mildly elevated to 25 and 0.8, respectively. Furthermore, a synovial fluid aspiration of the wrist was performed by a rheumatologist and analysis demonstrated no evidence of inflammatory arthritis. The patient did not have a clinical history suggestive of rheumatoid arthritis, and therefore, further laboratory studies including ANA and rheumatoid factor were deferred by the rheumatologist. Computed tomography revealed diffuse calcifications involving the entire wrist with soft tissue thickening surrounding some of the extensor tendons. Based on these findings, the differential diagnosis included possible reactive tenosynovitis. Hand surgery was consequently consulted, and a diagnostic synovial biopsy was planned in an outpatient setting.

However, the patient returned to hand clinic prior to the scheduled biopsy and demonstrated loss of extensor digitorum communis (EDC) function of the middle, ring, and small fingers. On operative exploration, extensive tenosynovitis in the fourth and fifth compartment was found to be extending

from the distal forearm to the hand. Furthermore, the EDC tendons for the index, middle, ring, and small fingers were ruptured, but the extensor indicis proprius remained intact (Fig. 1a, b). Extensive synovectomy was performed as well as transfer of the extensor carpi radialis longus tendon to the EDC of the middle, ring, and small fingers to restore extension. Synovial specimens revealed extensive fibrosis and granulation tissue with focal giant cell reaction. Culture results were available after 4 weeks and demonstrated *P. verrucosa* phaeohyphomycosis (Fig. 2). Based on culture sensitivities, the patient was initially placed on itraconazole therapy for a planned course of 3 months under the stewardship of an infectious disease specialist. This was later transitioned to voriconazole and finally discontinued after a total of 2 months

Fig. 1 a, b Intraoperative photograph demonstrating extensive tenosynovitis involving the fourth and fifth compartments extending from the distal forearm to the midcarpal joint. The extensor digitorum communis tendons to the index, middle, ring, and small fingers are ruptured (asterisk)

Fig. 2 Microscopic section of extensor tendon. a Hematoxylin and eosin staining demonstrating fibrosis and granulation tissue with focal giant cell reaction (short arrow). b Grocott's methenamine silver staining showing fibrotic changes with septate hyphae (long arrows)

due to persistence of side effects including nausea and vomiting. Postoperatively, she attained full grip strength but had residual extensor lag of the middle and ring fingers. There were no signs of recurrent fungal infection or tenosynovitis at 1 year after the operation.

Discussion

Phaeohyphomycoses comprise a rare group of mycotic infections caused by characteristic pigmented (dematiaceous) fungi that contain melanin in their cell wall and are frequently found in soil and decaying wood. Implantation into subcutaneous tissue commonly occurs through a thorn prick or wood splinter, leading to encapsulation of fungal hyphae within a granuloma. The granuloma may remain indolent as a subcutaneous cyst and persist for several years without symptoms. Retention of an associated foreign body seems to play a critical role in the development of this infection [10]. Phaeohyphomycoses of the hand are exceedingly rare and have only been reported in a few selected cases [1, 4–6, 10]. Infections appear to occur in immunocompromised patients and may present as an isolated mass [1] or multifocal cysts [4]. *P. verrucosa* is a member of this species, and the first human infection was reported in 1915 presenting as subcutaneous nodules [6].

Our patient sustained a penetrating injury to the hand while gardening 15 years prior to her initial presentation with a fungal cyst that was initially mistaken for an epidermoid cyst. Five months after excision, a delayed infection evolved and became clinically evident. We hypothesize that excision of the nidus in our patient resulted in seeding of the fungal hyphae into the surrounding subcutaneous tissues of the dorsal right hand. Although the cyst was entirely excised, it is nonetheless possible that minor tears in the capsule may have allowed seeding of fungal elements into the tissue that subsequently led to fungal infection that caused widespread destruction of the extensor tendons. Fungal inoculation may not necessarily incite an infection in an individual with a competent immune system, but our patient had a long-standing history of immune suppressive therapy and may have been unable to mount a sufficient immune response. In a similar case, Kimura et al. describe an 85-year-old patient who experienced subcutaneous progression of a hand infection with *P. verrucosa* after initial excision of an indolent nodule [4]. These cases suggest that antifungal therapy after excision of a fungal cyst can be critical to prevent the development of a fungal infection particularly in immunocompromised patients [8].

Although fungal tenosynovitis in the hand is associated with more common species including *Cryptococcus* and *Sporothrix*, tendon rupture is a rare complication [2, 9]. Chahal et al. described a case of flexor tenosynovitis with *Phaeoacremonium* leading to subsequent rupture of several flexor digitorum superficialis and flexor digitorum profundus

tendons in a patient who 6 months prior received a steroid injection for treatment of a trigger finger [3]. In contrast, our patient's extensor tenosynovitis resulted in rupture of multiple extensor digitorum communis tendons requiring exploration and tendon transfers.

Phaeohyphomycoses are extraordinary rare causes of hand infections, and there remains a paucity of diagnostic and therapeutic algorithms to guide management. The presentation can oftentimes be indolent and mimic other more common skin lesions. Thus, a prior history of local trauma or endemic exposure to fungal pathogens should alert the clinician to be mindful of this unusual diagnosis. In cases where a fungal cyst is suspected, meticulous excision without gross violation of the outer capsule should be performed. However, small undetected rents made in the capsule during excision can lead to fungal contamination and the hand surgeon should be particularly aware of this possibility in immunocompromised patients. Therefore, patients who are immunosuppressed may benefit from prophylactic antifungal therapy in order to prevent the development of fungal infections resulting from residual hyphae within the surgical site.

Conflict of Interest The authors declare that they have no conflict of interest.

Statement of Human and Animal Rights All procedures were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008.

Statement of Informed Consent Informed consent was obtained from the patient.

References

1. Arakaki O, Asato Y, Yagi N, et al. Phaeohyphomycosis caused by *Exophiala jeanselmei* in a patient with polymyalgia rheumatica. *J Dermatol*. 2010;37:367–73.
2. Bruno KM, Farhoomand L, Libman BS, et al. Cryptococcal arthritis, tendinitis, tenosynovitis, and carpal tunnel syndrome: report of a case and review of the literature. *Arthritis Rheum*. 2002;47:104–8.
3. Chahal J, Dhotar HS, Anastakis DJ. Phaeohyphomycosis infection leading to flexor tendon rupture: a case report. *Hand (N Y)*. 2009;4:335–8.
4. Kimura M, Goto A, Furuta T, et al. Multifocal subcutaneous phaeohyphomycosis caused by *Phialophora verrucosa*. *Arch Pathol Lab Med*. 2003;127:91–3.
5. McDonald LS, Bavaro MF, Hofmeister EP, et al. Hand infections. *J Hand Surg [Am]*. 2011;36:1403–12.
6. Medlar EM. A cutaneous infection caused by a new fungus, *Phialophora verrucosa*, with a study of the fungus. *J Med Res*. 1915;32:507–522.
7. Musial CE, Cockerill 3rd FR, Roberts GD. Fungal infections of the immunocompromised host: clinical and laboratory aspects. *Clin Microbiol Rev*. 1988;1:349–64.

8. Park SG, Oh SH, Suh SB, et al. A case of chromoblastomycosis with an unusual clinical manifestation caused by *Phialophora verrucosa* on an unexposed area: treatment with a combination of amphotericin B and 5-flucytosine. *Br J Dermatol.* 2005;152: 560–4.
9. Stratton CW, Lichtenstein KA, Lowenstein SR, et al. Granulomatous tenosynovitis and carpal tunnel syndrome caused by *Sporothrix schenckii*. *Am J Med.* 1981;71:161–4.
10. Ziefer A, Connor DH. Phaeomycotic cyst. A clinicopathologic study of twenty-five patients. *Am J Trop Med Hyg.* 1980;29:901–11.