Blastomycotic extensor tenosynovitis of the hand: a case report

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Introduction

Blastomycosis is an uncommon fungal infection caused by the thermally dimorphic fungus *Blastomyces dermatitidis* [12]. Infections in humans are primarily pulmonary in nature, but disseminated infection can occur. After lung and skin, the bone is the third most common site of infection. The most common musculoskeletal manifestations of the disease process include osteomyelitis or a monoarticular or polyarticular septic arthritis [1, 5, 6, 13, 16]. Blastomycotic infections involving the hand can be seen in the setting of multiple osseous lesions, but isolated lesions of the hand are relatively rare [5–7, 9, 13, 14, 16]. To the best of our knowledge, there has only been one other report of blastomycotic infection causing tenosynovitis and no reported cases involving the extensor tendons [8]. We present a case of an immunocompetent male with recurrent extensor tenosynovitis that ultimately proved to be a blastomycotic infection.

Case Report

A 72-year-old male presented with recurrent dorsal wrist swelling approximately 1 year after undergoing a tenosynovectomy at another institution. MRI obtained prior to the

index irrigation and debridement revealed proliferative extensor tenosynovitis involving the extensor digitorum communis tendons of the index, long, and ring fingers with no evidence of tendon rupture or abscess formation (Fig. 1a, b). The initial histopathology report revealed “chronic fibroproliferative synovitis.” A subsequent serological work-up for inflammatory arthritis was reportedly normal. The patient's symptoms failed to resolve following the index procedure, and an infectious etiology was suspected. He was placed on an oral cephalosporin, but a prolonged antibiotic course did not alleviate the wrist swelling or discomfort.

At the time of presentation to our clinic, the patient denied fevers, chills, night sweats, weight loss, or pulmonary symptoms. There was no history of trauma to the hand or ulceration. He acknowledged that he resided in Florida over the winter season and golfed regularly in Tennessee.

The past medical history was significant only for prostate cancer and a pulmonary nodule detected on chest radiographs for which he had undergone an extensive work-up with no definitive diagnosis. Physical examination revealed a well-healed dorsal longitudinal incision with diffuse extensor tenosynovitis involving the dorsum of the hand and wrist. There was no erythema, ulceration, or adenopathy.

Through a dorsal approach that incorporated the prior incision, the extensor compartments were identified. Proliferative tenosynovitis involving the extensor carpi radialis longus and brevis, extensor pollicis longus, and extensor digitorum communis tendons of the index, long, ring, and small fingers was noted extending from the musculotendinous junction proximally to the mid-metacarpal level distally. Although tendon invasion was noted in several areas, impending or frank tendon rupture was not noted. A complete tenosynovectomy was performed with several samples of tenosynovium sent for histopathological and microbiological analysis including fungal cultures. Histopathology revealed

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Fig. 1 **a** Fat-suppressed T1-weighted magnetic resonance image. The axial sequence show marked edema surrounding the extensor digitorum communis tendons on the dorsal aspect of the wrist and hand. **b** T2-weighted magnetic resonance image. The axial sequence shows marked edema surrounding the extensor digitorum communis tendons on the dorsal aspect of the wrist and hand

scattered non-necrotizing granulomas with rare double-contoured yeast that were Gomori methenamine silver stain positive (Fig. 2). The Kinyoun stain was negative for acid-fast bacilli. The histopathological features were consistent with blastomycosis. Consultation with an infectious disease specialist was obtained and the patient was treated with oral Itraconazole. There was no evidence of recurrence at the 6-month follow-up.

Fig. 2 Gomori methenamine silver stain with a large double-contoured yeast in the center of a granuloma

Discussion

History

The first recorded case of blastomycosis was presented by Gilchrist in June of 1894, with Busse reporting the first case of blastomycotic osteomyelitis in July of that same year [2, 5]. Since that time, the orthopedic literature has infrequent mention of blastomycotic infection, and it is primarily referenced with disseminated disease.

Epidemiology

In endemic areas, blastomycosis commonly infects dogs and, to a much lesser extent, humans. It cannot be transmitted by animals or humans [12]. The disease is commonly referred to as North American blastomycosis because most cases occur in USA and Canada. Outbreaks of North American blastomycosis have occurred primarily along rural river banks [12]. Endemic areas include the southeastern and south central states that border the Mississippi and Ohio Rivers, the Midwestern states and Canadian provinces that border the Great Lakes, and a small area of New York and Canada adjacent to the St. Lawrence Seaway [4]. As the organism is exceedingly difficult to harvest and cultivate from the natural environment, these endemic regions have been determined based on previous epidemics or outbreaks in humans [4].

Pathophysiology

The lung is the primary portal of entry for the organism. Once in the host, *B. dermatitidis* transforms to a large, singly budding yeast cell [4]. It then develops into a granulomatous and suppurative infection in the lungs. It has the ability to disseminate hematogenously to any organ but most commonly infects the skin, followed by the skeletal system and genitourinary system [4]. Central nervous system involvement is common in the immunocompromised patient. Direct inoculation in the laboratory or from animal bites or scratches has also been reported [4, 10]. There has been a reported case of septic arthritis following knee arthroscopy in which the patient had no evidence of pulmonary or cutaneous involvement [17].

The pulmonary presentation of blastomycosis is often that of an acute or chronic pneumonia with clinical manifestations that are indistinguishable from pyogenic bacteria, tuberculosis, other fungal infections, and cancer [3, 4]. Extrapulmonary disease is seen most commonly during the chronic form of illness, in which two-thirds of patients have been reported to have multiple organ involvement [8]. In cases of isolated blastomycosis without pulmonary involvement, a transient antecedent pulmonary infection is usually

suspected. Careful questioning often reveals prior or concurrent symptoms such as cough, fever, and skin lesions that are indicative of systemic involvement [16]. Skin disease is present in three-fourths of patients with two or more organ system involvement [15].

Osteomyelitis is a much more common manifestation of blastomycosis than septic arthritis [5, 15, 17]. Joint involvement can occur with or without adjacent osteomyelitis, most often involving the knee, followed by the ankle, elbow, wrist, and hand [6, 16]. Patients with bone lesions rarely present with bone pain but instead present with contiguous soft tissue abscesses or chronic draining sinuses [4]. There has been previous report of isolated infection involving the synovium of the forearm without bone or skin involvement [2].

Diagnosis

There is no adequate skin or serologic test to diagnose infection [12]. Specimens can be obtained from sputum, serum, urine, exudates, or biopsies of lesions [12]. The Gomori methenamine silver stain is best used for screening tissue for the presence of fungal elements [4]. Histologic examination reveals a distinct pyogranulomatous reaction with Europhiles and concatenating granulomas [12]. *B. dermatitidis* can be grown, usually within 2 weeks, on Sabouraud's agar at room temperature to produce a white or brownish colony with branching hyphae [4]. In host tissue or culture at 37°C, the fungus grows a thick-walled, multinucleated, spherical yeast that usually produces single buds [12]. The conversion to the yeast form at 37°C confirms identification of *B. dermatitidis*. Additionally, confirmation of the diagnosis can be performed by extraction and detection of the *B. dermatitidis*-specific antigen A or by a specific DNA probe [4].

Blastomycosis has a variable appearance on plain radiographs, in which it may be either focal or diffuse and can involve cancellous or cortical bone [7]. The lesions are predominantly lytic, indicative of a non-specific osteomyelitis. The usual site of origin is the epiphyseal–metaphyseal region of the bone, with possible extension into the surrounding soft tissues and joints [7, 11]. A periosteal response is uncommon but may be robust in younger patients [13]. On occasion, pathologic fracture may also be present. Bony lesions may be asymptomatic in patients at the time of initial presentation [11]. In patients with pulmonary or cutaneous disease or with symptomatic bony lesions, a bone scan can be used to determine other asymptomatic areas of involvement. MRI should be carefully interpreted as it may not reliably differentiate infection from neoplasm.

Treatment

In the early literature, mortality from disseminated skeletal blastomycosis approached 90 % [5]. With the advent of

improved antifungal agents such as amphotericin B and the azoles (e.g., ketoconazole, itraconazole), mortality from the disease process has dramatically improved. Medical treatment with chemotherapeutic agents is necessary for all patients with blastomycosis. Resolution of the disease process may occur with or without surgical intervention. In combination with appropriate pharmacologic therapy, irrigation and debridement of infected regions of musculoskeletal blastomycosis in patients who are responding poorly to therapy is an effective treatment for eradication of the disease [13]. Repeat debridements are often needed and new lesions that require surgical intervention may appear during the initial treatment of the sentinel lesion.

For mild to moderate manifestations of extrapulmonary disease, the Infectious Disease Society of America (IDSA) recommends oral itraconazole for 6–12 months. More severe infections require amphotericin B initially, usually for 1–2 weeks or until improvement is noted. Step-down therapy with itraconazole then follows for a total of at least 12 months [3]. The IDSA guideline for patients with osteoarticular blastomycosis is a total of 12 months of antifungal therapy [3].

Conclusion

In this report, we present the case of an immunocompetent male with isolated blastomycotic extensor tenosynovitis. He was successfully treated with surgical irrigation and debridement and appropriate antifungal therapy. Diagnosis was delayed due to the patient's non-specific clinical presentation and postoperative course. Patients that fail to respond to traditional therapy for possible inflammatory or infectious processes require further investigation, and a work-up for atypical organisms should be considered. It would be prudent for clinicians in endemic areas to consider blastomycosis in the differential diagnosis.

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