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#### Correspondence

Sarik Kumar Shrestha Dept. of Orthopaedics & Trauma Surgery, Patan Hospital, Patan Academy of Health Sciences, Lalitpur, Nepal Email: sarikkumarshrestha@gmail.com

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# Primary cutaneous aspergillosis presenting as a cystic lesion in the foot

Sarik Kumar Shrestha<sup>1</sup> Sarik Kumar Shrestha<sup></sup>

<sup>1</sup>Resident, <sup>2</sup>Asst. Prof., <sup>3</sup>Medical Officer, <sup>4</sup>Prof., Dept. of Orthopaedics & Trauma Surgery; <sup>5</sup>Resident, Dept. of Pathology, Patan Hospital, Patan Academy of Health Sciences, Lalitpur, Nepal

#### Abstract

Aspergillosis is a fungal infection, usually affecting the lungs, mostly in immunocompromised patients. Musculoskeletal or cutaneous involvement is extremely rare and when it does occur, the diagnosis and treatment is delayed. We present a case of primary cutaneous aspergillosis presenting as a cyst in the foot of a 54-year-old immunocompetent female. She presented with the complaint of painful lump over right foot at the region of the fourth webspace. Magnetic Resonance Imaging (MRI) was suggestive of infected ganglion cyst. However, histopathological examination showed granulomas with areas of caseous necrosis and silver stain showed multiple fungal hyphae with morphology suggestive of Aspergillus species.

Keywords: aspergillosis, Aspergillus, cutaneous cyst

#### Introduction

Aspergillosis refers to a wide variety of diseases caused by fungi in the genus Aspergillus.<sup>1</sup> Most people breathe in Aspergillus spores every day without getting sick.<sup>2</sup> Invasive aspergillosis is seen in immunocompromised states including glucocorticoid neutropenia, therapy, advanced acquired immunodeficiency syndrome (AIDS), chronic granulomatous disease, hematopoietic stem cell or solidorgan transplantation.<sup>1</sup> Aspergillosis causes pulmonary infection in the majority of cases.<sup>3</sup> Cutaneous aspergillosis is а rarely encountered form of aspergillosis.<sup>1</sup>

Here, we present a case of primary cutaneous aspergillosis presenting as a cystic lesion in the foot of a 54-year-old female without any known risk factors.

#### **Case Report**

A 54-year-old female presented to the orthopaedics outpatient clinic with the complaint of a painful lump over her right foot for two months. She was a known case of hypertension and dvslipidemia under medication. On local examination, a cystic lump was present over the region of the fourth metatarsophalangeal joint extending from the dorsal to the plantar surface which was compressible, fluctuant, non-tender, and not associated with increased temperature or surrounding skin changes, Figure 1. Systemic examination did not reveal any abnormal findings.

Ultrasonography revealed features suggestive of chronic abscess, with a collection of about 3.5 ml at the space between the third, fourth, and fifth rays extending from the dorsal to the plantar region. Magnetic resonance imaging (MRI) was done which showed a well-defined lobulated 'U' shaped lesion in the plantar region of the fourth metatarsophalangeal joint and proximal phalanx, extending into the third and fourth interphalangeal region with T2 high and T1 low signal intensity. MRI also revealed subcutaneous edema in the dorsum of the foot at the level of the third and fourth interphalangeal joint with no intra-articular extension or erosion of bone. MRI findings suggested a provisional diagnosis of an infected ganglion cyst, Figure 2.



Figure 1. Clinical photograph showing cystic lesion over dorsum of foot at the fourth webspace

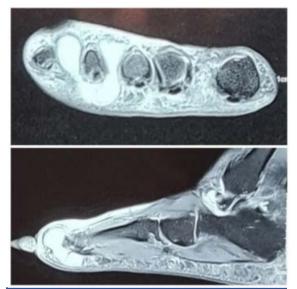


Figure 2. MRI showing a 'U' shaped lesion in plantar region of fourth metatarsophalangeal joint and proximal phalanx, extending into third and fourth interphalangeal region

Pre-operative investigations including complete blood count, prothrombin time, random blood sugar, renal function test, liver function test, serology, urine routine examination, electrocardiogram, and chest radiograph showed no abnormal findings.

Surgical excision of the cyst was done and sent for biopsy. Histopathological

examination showed numerous well-formed granulomas composed of foamy histiocytes, foreign body, and Langhan's type giant cells along with areas of caseous necrosis. Silver stain showed multiple fungal hyphae with morphology suggestive of Aspergillus species, Figure 3.

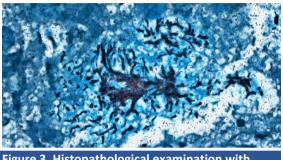


Figure 3. Histopathological examination with Grocott Methenamine Silver stain showing septated acute angle branching hyphae

Examination and investigation findings did not reveal any immunocompromised state. In addition, the patient had no symptoms corresponding to pulmonary involvement or systemic pathology, and her chest examination and radiograph exhibited no evidence of aspergillosis. Hence, a diagnosis of primary cutaneous aspergillosis was established. The patient was then prescribed Itraconazole for 12 weeks. She was followed up regularly for 12 weeks and had no further issues or recurrence.

## Discussion

Cutaneous aspergillosis is а rarely encountered form of aspergillosis. Cutaneous lesions usually develop secondary to hematogenous dissemination from the underlying infected organ. Primary cutaneous aspergillosis is caused by direct implantation of Aspergillus following trauma.<sup>4</sup> Our patient denied any history of injury, however, latent skin cracks following unnoticeable microtrauma leading to primary cutaneous aspergillosis cannot be ruled out. Cutaneous manifestations are non-specific and can present with violaceous patches with central necrotic ulcers, subcutaneous abscesses, or vegetative papules and patches.<sup>5</sup>

Among Aspergillus species, the most common causative agent of infections in humans is Aspergillus fumigatus.<sup>2</sup> It is a filamentous saprophytic fungus that grows in multicellular filaments called hyphae.<sup>6</sup>

Diagnosis of cutaneous aspergillosis is performed by microbiologic examination, culture, and histopathologic examination.<sup>7</sup> Skin biopsy specimens should be taken from the center of the lesion and should reach the subcutaneous fat tissue because Aspergillus species tend to invade blood vessels of the dermis and subcutis, resulting in an ischemic cone above it.<sup>7</sup>

If aspergillosis is diagnosed, it is important to determine whether the patient has a primary infection or secondary dissemination from a primary focus such as the lung.<sup>7</sup> Special attentions to pulmonary symptoms and/or signs may determine whether an evaluation for pulmonary aspergillosis is needed.<sup>7</sup>

Pulmonary primary focus of infection was found in a case report of cutaneous aspergillosis in 2011.<sup>8</sup> Another case report of tracheobronchial aspergillosis following primary cutaneous aspergillosis in a lungtransplant recipient was reported.<sup>9</sup> However, we have not found any reported case of primary cutaneous aspergillosis in an immunocompetent patient after searching in PubMed and Google Scholar.

Hyphal invasion in an immunocompromised patient may lead to angioinvasion that induces ulcer and necrosis, a further extension to adjacent osseous structure and soft tissue, and even hematogenous systemic dissemination to any organ.<sup>3</sup> Therefore, rapid diagnosis and treatment are important. Early and appropriate diagnosis surgical debridement and antifungal agent therapy significantly reduced mortality and morbidity in these patients.<sup>3</sup> For this reason, in the presence of soft-tissue abscesses and other cutaneous lesions with immunosuppressive patients, Aspergillus infections should be considered as a reason.

Aspergillus infections are treated with surgical debridement and antifungal therapy. Amphotericin B has been the classical antifungal drug of choice in aspergillosis. Voriconazole, itraconazole, and caspofungin are among the other alternative treatment options.

Here, we have presented a rare case of primary cutaneous aspergillosis of the foot which presented as a cystic lesion in a patient with no known risk factors. Although MRI findings of primary cutaneous aspergillosis have not been described, a well-defined lobulated lesion with T2 high and T1 low signal intensity gave the impression of an infected ganglion cyst in our case. However, there was no communication between the lesion and the joint capsule which is often seen in cases of ganglion cysts. We were able to treat the patient successfully without further complications and recurrence with surgical excision and antifungal medications.

## Conclusion

Primary cutaneous aspergillosis is extremely rare and usually presents in immunocompromised states. Early diagnosis and treatment can prevent local spread and hematogenous dissemination. Hence, primary cutaneous aspergillus although rare, should be kept as a differential diagnosis of cystic lesions, and such lesions should be sent for microbiological and histopathological examination to avoid missing such cases.

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Department of Orthopaedics and Trauma Surgery, Patan Hospital, Patan Academy of Health Sciences, Lalitpur, Nepal.

## **Conflict of Interest**

None

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#### **Author Contribution**

Concept, design, planning: SKS, RRP, YB, NMSP, RT; Literature review: SKS, YB; Data collection/analysis: SKS, RT; Draft manuscript: SKS, RRP, YB, NMSP, RT; Revision of draft: SKS, RRP, NMSP; Final manuscript: SKS, RRP, YB, NMSP, RT; Accountability of the work: SKS, RRP, YB, NMSP, RT

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