Deep Invasive Fungal Infection of the Hand in a Child Mimicking a Local Gigantism

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Abstract

Subcutaneous and deep fungal infections in the hand are rare among children. These are usually found in immunocompromised adults or in persons engaged in soil handling activities, due to direct exposure, especially in the tropics. Delay in diagnosis is usual because pyogenic and other granulomatous infections are considered first.

The authors present the case of a healthy, immunocompetent 2½-year-old child who presented with progressive swelling of the right hand mimicking a localized gigantism of the entire hand. Multiple operative drainage procedures done previously had failed to resolve the condition. A biopsy established the presence of fungal hyphae, thus confirming the diagnosis of deep fungal infection of the hand and guided proper therapeutic intervention. A strong index of suspicion needs to be maintained in cases not responding to conventional antibacterial therapy, and both microbiologic and histopathologic samples need to be obtained to establish the diagnosis.

Keywords

- pediatric hand infection
- deep fungal infection
- local gigantism

Introduction

Chronic hand infections can present to the hand surgeon occasionally and pose a diagnostic dilemma. Fungal infections of the hand in children are usually confined to the superficial layers of the skin and nails, especially to the tropics. Rash, itching, and redness characterize such infections. Deep fungal infections in the hand have been reported in immunocompromised hosts as opportunistic infections.^{1,2} The authors report a case of a deep fungal infection in an immunocompetent child mimicking a local gigantism. Delay in diagnosis occurs, as bacterial and fungal infections can mimic each other. A strong index of suspicion is necessary, and tissue samples for both histopathologic and microbiologic studies must be obtained to establish the diagnosis.

Case Report

A 2½-year-old male child presented with massive swelling of the right hand, which had been progressively getting worse over the previous 3 months.

His parents claimed that it possibly started about 3 months previously when they noticed a pustule over the dorsum of the hand. It was then attributed to a possible insect bite. He was treated by local physicians with antibiotics, both oral and parenteral, and finally by surgical drainage followed by regular wound dressings. Previous wound swabs had come positive for coliform bacteria with varied sensitivity. His blood picture revealed leukocytosis and a raised erythrocyte sedimentation rate (ESR). Mantoux test was negative. A fine needle aspiration cytology (FNAC) (done elsewhere) from the dorsum of the hand showed dense infiltrates of neutrophils, macrophages, and lymphocytes, with no evidence of tuberculosis or malignancy. However, the swelling persisted and kept on increasing despite medication.

At the time of his presentation to the clinic, both the dorsal and volar surfaces of the right hand were swollen to the extent that it mimicked a localized gigantism or a rapidly growing neoplasm. The entire hand was tender, inflamed with minimal serous discharge from previous incision and drainage sites (►Fig. 1). The swelling extended proximal to the wrist joint

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Fig. 1 Clinical picture of the hand showing the massive swelling, scars of previous surgical drainage, and a satellite lesion proximal to the wrist crease (arrow).

with small nodular swellings over the distal radius. The peripheral pulsations were well felt, and there was no motor or sensory deficit in the hand. There were a group of matted lymph nodes palpable in the ipsilateral axilla.

Plain radiographs did not reveal any bony destruction (►Fig. 2). A magnetic resonance imaging (MRI) scan of the hand and wrist region also confirmed the sparing of the

skeletal elements of the hand with diffuse infiltration within the soft tissues, within the palmar and dorsum areas of the hand, finger, and thumb. There was no collection or bone or joint involvement (►Fig. 3). The possibility of a fibromatous neoplastic lesion was suspected. In view of the clinical and radiologic features, a possibility of mycetoma of the hand was considered.

Fig. 2 (A, B) Anteroposterior and lateral radiographs of the hand showing soft tissue swelling, but without any bone or joint erosions.

Fig. 3 Coronal and axial cuts of MRI scan showing diffuse soft tissue infiltration in dorsal and palmar aspect of the hand and thumb infiltrating the muscles. No intermuscular collection or bone or joint infiltration evident.

The child was admitted. The hand was dressed and kept elevated. An elective deep wedge biopsy from the advancing margin of the lesion was performed along with an axillary lymph node biopsy. There was no evidence of frank pus, but only granular material from the sinus. A fresh wound swab was collected from the discharging sinus region at the same time. Smears were sent for Gram stain, acid-fast bacilli (AFB) stain, a potassium hydroxide (KOH) mount, and fungal culture.

The biopsy showed hyperkeratosis and irregular acanthosis in the epidermis. The dermis was densely infiltrated by inflammatory cells—predominantly eosinophils admixed

with neutrophils, lymphomononuclear cells, and histiocytes. Scattered microabscesses were present. The microabscesses showed fragments of fungal hyphae enveloped by eosinophilic material (Splendore-Hoepli phenomenon) (**Fig. 4**). Periodic acid–Schiff (PAS) stain failed to reveal any eumycotic fungal elements, nor did the granules reveal any eumycetoma. The lymph node showed features of nonspecific reactive follicular hyperplasia. The wound swab grew *Enterococcus faecalis* with sensitivity to amoxicillin+clavulanic acid, gentamicin, imipenem, vancomycin, and linezolid. The AFB stain and KOH mount were negative.

Fig. 4 Histopathology slide, hematoxylin and eosin stain. (A) Low-power magnification showing dense infiltration of inflammatory cells with scattered microabscesses. (B) Splendore-Hoepli phenomenon showing fungal hyphae fragment enveloped by eosinophilic material.

A diagnosis of deep invasive fungal infection of the hand was thus confirmed. The child was started on intravenous fluconazole (3 mg/kg/d, once daily) and amoxicillin + clavulanic acid (200 mg/12 hourly) after consultation with an infectious disease specialist. Blood tests (viral screen) for human immunodeficiency virus (HIV), hepatitis B surface antigen (HBsAg), and hepatitis C virus (HCV) came negative, thus ruling out either of these as a possible cause of immunosuppression.

Within 1 week of starting fluconazole, the swelling of the hand showed regression. There was progressive reduction in the swelling over the next 6 weeks, and the hand resumed its normal size and texture by the end of 8 weeks. Fungal culture reports came negative at 6 weeks. All medications were stopped after 6 weeks, and the child was kept on regular follow-up. An exercise and rehabilitation program was started within 1 week of starting antifungals to restore the function of the hand. At the latest follow-up (18 months), there was no recurrence of the swelling, and the child is using the hand for all activities of daily living with a normal pinch, grasp, and grip strength (►Fig. 5).

Discussion

Fungal infections of the hands in children are quite common. These are usually superficial (dermatomycosis), occurring in the interdigital spaces, nails, and are often associated with a rash and itching.² These are usually transferred from similar infection in other areas of the body such as the groin.

Soil handling and contact with household pets have been implicated as the likely source of such infection.¹ The chances of infection increase in immunodeficient or immunosuppressed children.²

Deep fungal infection, involving the subcutaneous tissue, muscles, tendon sheaths, and even bones and joints of the hand are uncommon, especially in children.^{1,2} These have been reported as opportunistic infections in immunocompromised individuals. In this case though initial presentation was with a pustular lesion, it later presented with a global swelling of the entire hand mimicking a localized soft tissue gigantism. This case is also unique because the child was not immunocompromised.

Organisms likely for deep infections that grow hyphae include mucormycosis, aspergillosis, and coccidioidomycosis. Mucormycosis affecting the forearm with ischemia necessitating amputation in an immunocompromised woman has been reported.³ Blastomycosis, endemic coccidioidomycosis, histoplasmosis, and mycetoma have been implicated as causative fungi in deep infections.⁴

The clinical presentation of some of these deep fungal infections may resemble a bacterial infection. Confusion is likely as some bacteria are known to produce similar clinical picture. Diagnosis can be confirmed by identifying yellow sulfur granules in the purulent material in cases of mycetoma.⁵

Plain radiographs may seldom show bony erosions or destruction except in infections caused by mycetoma

Fig. 5 Clinical picture of the hand at 18 months follow-up.

or blastomycosis. MRI can provide information of the deeper tissue involvement, including muscles, tendons, and bone. In this case, diffuse infiltration in the soft tissues was found. Lobulated mass in the adult hand due to chromomycosis with its MRI features has been reported.⁶ Small localized lesions around the wrist, due to histoplasmosis, have also been reported in immunocompromised individuals.⁷

The mainstay of diagnosis is a combination of microbiology and histopathology.^{4,5} This is necessitated by the fact that a negative fungal culture is frequently obtained. Evidence of fungal elements and associated tissue response on histopathology may help in clinching the diagnosis.^{4,5} The Splendore-Hoeppli phenomenon, observed in the histopathology slides of this patient, describes the eosinophilic, pseudomycotic structures composed of necrotic debris and immunoglobulin. Though more commonly seen with fungal infection, it has also been reported around helminthic infection.^{8,9} In this case, the identification of fungal hyphae clinched the diagnosis in favor of deep fungal infection. A nodular lesion with lymphocutaneous spread in the hand of a child has been reported, which was ultimately diagnosed on fungal culture and responded to antifungal therapy.¹⁰

Treatment requires administration of appropriate antimicrobial agents.¹¹ In this case, a variety of broad-spectrum antibiotics was given prior to presentation to the authors, to cover both gram-positive and gram-negative organisms, but with poor response. Even though fungal cultures eventually came negative, the identification of fungal hyphae on histopathology confirmed the diagnosis of deep fungal infection, and antifungal agents were added leading to a dramatic therapeutic response. This highlights the importance of obtaining material for both microbiology and histopathology, establishing a tissue diagnosis and administering appropriate antimicrobial agents.

Conclusion

It is imperative for the attending physician to have a high index of clinical suspicion for deep fungal infection in cases of chronic hand infections, especially those not responding to standard antibiotics and those mimicking a chronic granulomatous lesion even if there is no evidence of immunocompromise.

Conflict of Interest

None.

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