

Does every black necrotic lesion need surgical debridement?

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Abstract

A 4-year-old boy with beta-thalassemia major who underwent a successful bone marrow transplant (BMT), presented with a necrotic looking scalp lesion. Debridement was initially suggested but a punch biopsy showed *Aspergillus fumigatus*. Antifungal therapy was commenced and the lesion healed with minimal scarring. Invasive fungal infections are a major cause of morbidity and mortality in immune-compromised patients. Invasive aspergillosis (IA) has been on the increase since 1980. The most common manifestation of IA after BMT is invasive pulmonary aspergillosis. Primary cutaneous aspergillosis is extremely rare in the paediatric BMT population. There is scanty information regarding management of these lesions in children. Excision and skin grafting of these lesions is not the ideal treatment in the immune compromised and healing by secondary intention may leave a large scar. Suspicious skin lesions should not be subjected to immediate wide surgical debridement leaving a large uncovered area with subsequent extensive scarring and failure to respond to skin grafting.

Keywords

Cutaneous aspergillosis; BMT; invasive aspergillosis.

Case report

A 4-year-old boy with beta-thalassemia major was admitted for haematopoietic stem cell transplantation (HSCT) from his HLA-matched sibling donor. His allogenic graft was uneventful.

On the 19th post-transplant day he developed a small red patch on the scalp 2 days after the area was shaved. A slowly growing papule with minimal reaction in the surrounding skin was noticed over the next few days. Within a week, the lesion evolved into an ulcer and started to discharge dirty grey coloured fluid. A surgical opinion was sought in order to see whether incision and drainage with or without debridement was indicated. Blood tests showed an absolute neutropenia and severe leucopenia with a count of $0.1 \times 10^9/l$ (5.0–14.0) as would be expected after HSCT.

Physical examination revealed a 2×2 cm ulcerated lesion on the scalp with rounded, everted and punched out edges. The base was covered with necrotic tissue which was adherent to the underlying soft tissue. Neither discharge nor tenderness was encountered during the examination, and there was no evidence of fasciitis. Culture swabs taken initially by the referral team were



Fig. 1. Skin lesion with biopsy site.

negative for fungal infection. Surgical debridement was initially considered but was delayed because of the patient's underlying condition.

A punch biopsy was carried out. This was not associated with any bleeding and the necrotic layer was found to be very thin and firmly adherent to the underlying fleshy tissue. The area was dressed and specimens were sent for histopathology and microbiology. The result revealed a growth of *Aspergillus fumigatus* (Fig. 1). The patient had already been receiving amphotericin B (AmBisome) 5 mg/kg from the 15th post-transplant day and voriconazole was added when the histopathology and microbiology results were received. Daily dressings with saline were carried out and no topical antifungal was used. The central dark portion of the lesion became lighter, shrunken, and was replaced gradually with healthy granulation tissue, which started epithelizing. His leukocyte count gradually fell to normal. A second punch biopsy was taken after 6 weeks and no fungal growth was seen. Three months later, the area was completely healed with a small alopecic patch with surrounding normal hairy scalp.

Discussion

Aspergillus species are widely distributed in nature; more than 30 species have been implicated in different human infections. The most important species are *Aspergillus fumigatus*, *Aspergillus flavus* and *Aspergillus niger*^[1]. Invasive fungal infections are a major cause of morbidity and mortality in the expanding immunocompromised patient population. The incidence of invasive aspergillosis (IA) has increased by 357% since 1980. The majority of information about aspergillosis has been obtained from studies conducted almost solely on adult patients^[2]. There is scanty information about aspergillosis in the paediatric population. Immunocompromised patients are particularly susceptible to these kinds of fungal infections because of a number of risk factors including bone marrow transplantation (BMT), immunosuppressive drugs, corticosteroids, chemotherapy, radiotherapy and haematologic malignancies^[2]. The most common manifestation of aspergillosis in these patients is invasive pulmonary aspergillosis. Haematogenous dissemination as a secondary event occurs in 20–50%. Secondary skin involvement due to systemic aspergillosis is extremely rare in BMT patients.

There are less than 10 reported cases of primary cutaneous aspergillosis (PCA). Amongst these reported cases, the appearance of the skin lesions varies. They usually present with red/violet patches which may have a central necrotic ulcer^[1]. The presence of a necrotic lesion in a neutropenic patient suggests an infectious cause. Thus, a host of infectious causes must be

considered, including aspergillosis, candidiasis, cryptococcosis, zygomycoses and various bacterial causes. In addition, a rapid growing ulcerating lesion with punched out borders may be caused by pyoderma gangrenosum.

We were presented with a scalp lesion that looked like a bacterial infection and had a fungus-negative culture swab. This child was promptly investigated and after the presence of PCA was confirmed and successfully treated with aggressive systemic antifungal drugs resulting in a complete cure.

There is an approximate annual incidence of aspergillosis of about 0.4% among hospitalized immunocompromised children, 74% of whom are suffering from an underlying malignancy. The highest incidence was 4.5% in children who had undergone an allogenic BMT^[2,3]. Burgos et al.^[2] found in a series of 139 children with hematologic malignancies that the most commonly affected site was the lungs in 59%, followed by the skin (primary and secondary) in 10%. High mortality rates have been associated with aspergillosis in the paediatric population. Rubio et al.^[1] reported 55% mortality and Burgos et al.^[2] reported 52.5% mortality in their series. In paediatric oncology and patients after HSCT the infecting species seems to be location dependent. *Aspergillus flavus* has been associated mainly with cutaneous cases and *Aspergillus fumigatus* is more commonly found in pulmonary infections^[1,2].

Primary and secondary types of *Aspergillus* skin infection have been described. Primary cutaneous aspergillosis (PCA) results from local trauma or skin maceration, particularly in neutropenic post-transplant patients. Secondary cutaneous aspergillosis infection results from haematogenous spread^[4]. Rubio et al.^[1] reported a 4.57% incidence of aspergillosis in his series of 306 children with hematologic malignancies and 3 of them were PCA. Burgos et al.^[2] described 9 patients with cutaneous involvement out of 139 but unfortunately it is unclear how many of these actually had PCA. Hashmi et al.^[4] described 3 of 154 HSCT patients with PCA.

The description of these lesions is variable throughout the literature. Frequently they begin as an erythematous lesion and progress to a necrotic or ulcerated lesion^[1,2,4]. The patient described in this report had absolute neutropenia after undergoing HSCT during the treatment of beta-thalassemia major. He developed the scalp lesion as a result of skin trauma caused by shaving. Although swab cultures from the wound were negative for fungus, aggressive antifungal treatment was already commenced because of the increasing incidence and higher mortality of aspergillosis in these immunocompromised patients.

Techniques available for the detection of the *Aspergillus* antigen by polymerase chain reaction (PCR) may help in early diagnosis and management of this disease^[3]. Radiologic imaging such as chest radiography and computed tomography scans may be helpful in demonstrating the multiplicity and spread of the disease, but with limited value in isolated PCA^[5]. In the absence of a positive culture, a biopsy may prove to be diagnostic.

Full recovery after medical treatment without relapse is achieved in 2 of 3 cases described by Rubio et al.^[1]. The third case died of a different underlying condition. Generally mortality rates are very low amongst reported cases; delay in diagnosis and treatment is likely to result in a higher mortality.

Wound debridement is usually carried out to expedite wound healing in immunocompetent patients, but healing may be severely impaired in the presence of neutropenia. This patient had an absolute neutropenia and severe leucopenia at the time of diagnosis of primary cutaneous aspergillosis without any evidence of systemic involvement. It would be sensible to delay debridement and wait for the recovery of neutropenia and leucopenia, however if the lesion does not shrink, or if it increases in size, then debridement would be the treatment of choice. This patient responded very well to antifungal therapy and the lesion shrank and became lighter in colour without spreading. The leukocyte and neutrophil counts were slow to normalize but recovered fully at about the time the wound healed completely.

Teaching points

- Fungal infections are increasingly recognized after HSCT in children suffering from malignancy.
- Black skin lesions in immunosuppressed children should be identified by obtaining full depth skin biopsies for histopathologic processing as well as for tissue culture (bacterial, fungal and atypical mycobacteria) before any surgical debridement.
- Early detection is helpful in early management.
- The presence of black tissue does not always mean the presence of necrosis.

References

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