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UPPER EYELID SUBCUTANEOUS ZYGOMYCOSIS IN AN IMMUNOCOMPETENT YOUNG NIGERIAN FEMALE: CASE REPORT

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SUMMARY

A 23 year-old female was presented with a year history of gradual, slowly progressive, painless swelling in the left upper eyelid. Examination revealed a well circumscribed, firm, not tender eyelid mass, not attached to the skin or underlying structures which was completely excised. Histopathological examination showed numerous hyphae and spores with Gomori Methylamine Silver stain while culture yielded floccose - whitish fast growing colonies. A diagnosis of zygomycosis probably entomophthoramycosis was made and she was treated with oral Fluconazole. The residual ptosis was corrected by tarsofrontal sling surgery. Zygomycosis should be considered a differential of a circumscribed eyelid subcutaneous mass in an immunocompetent adult.

INTRODUCTION

Zygomycoses are uncommon opportunistic fungal infections, with increasing incidence (1). They rarely affect immunocompetent individuals, and are caused by fungi in the order Mucorales (mucormycosis) or Entomophthorales (entomophthoracomycosis) (1,2). The term "zygomycosis" was proposed by Ajello et al (3) in 1976 to replace the obsolete name "phycomycosis", and, it describes infection by organisms in these two orders. Over the years however, the term "zygomycosis" has been used synonymously with "mucormycosis", which is an infection by fungi in the order Mucorales (1).

Clinically, cutaneous zygomycoses present with varying lesions ranging from non-healing ulcers to proliferating necrotizing fasciitis (4). Infections involving immunocompetent individuals though rare, usually present as single, painful, indurated skin lesions with tissue infarction and necrosis (5). To the best of the authors' knowledge, there has been no report in literature of zygomycosis presenting as a

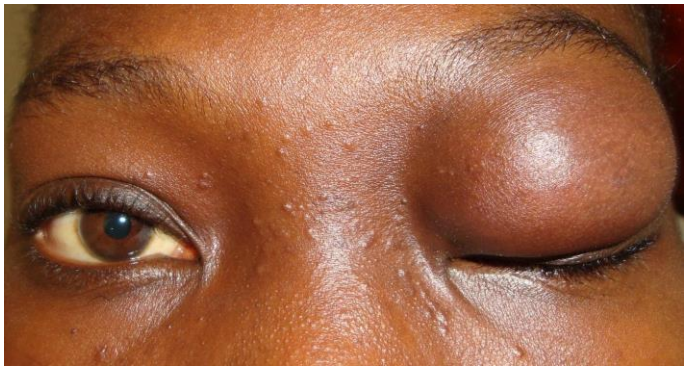
circumscribed subcutaneous mass, hence, this report of an unusual presentation of an uncommon fungal infection in a young, immunocompetent Nigerian female.

CASE REPORT

A 23 year-old female student was presented to the Eye Clinic with a year history of gradual, slowly progressive, painless swelling in the left upper lid. No antecedent history of trauma, or work in an agricultural set up. There was no associated bleeding, ulceration, or visual symptoms. She had no systemic disease.

Examination revealed a healthy lady with visual acuity 6/5 each eye, left upper eyelid mass (about 8 x 6 x 4 cm), well circumscribed, firm, not tender, and not attached to skin or underlying structures. There was mechanical ptosis, however, ocular motility was full and the posterior segment was normal. Computed tomography showed a well circumscribed non-enhancing subcutaneous mass on the left upper eyelid (Figure 1). The right eye was essentially normal.

Figure 1
Clinical picture of the patient showing the well circumscribed subcutaneous mass



She underwent complete excision of the mass (Figure 2a and 2b), but defaulted from follow-up for 11 months due to socio-economic reasons. She re-presented a year later with a recurrent mass in the same location, and, a repeat surgical excision was indicated due to its increasing size.

Figure 2a
Clinical picture of the mass intraoperatively during surgical excision

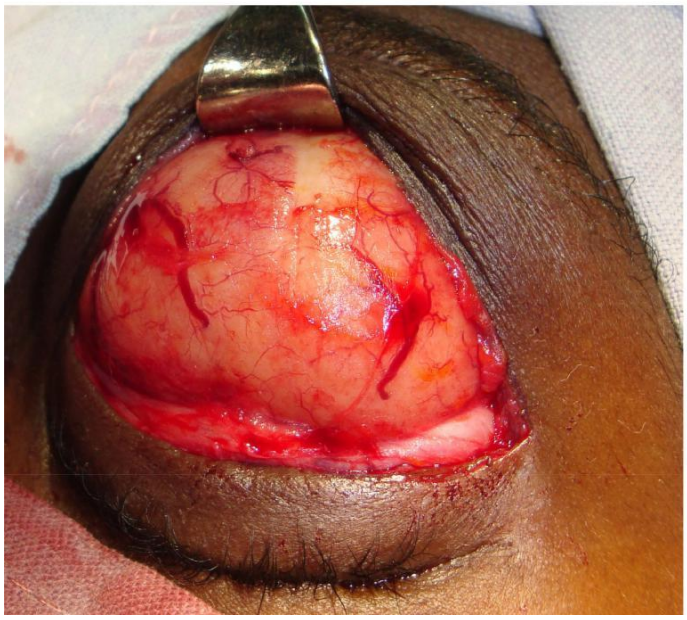
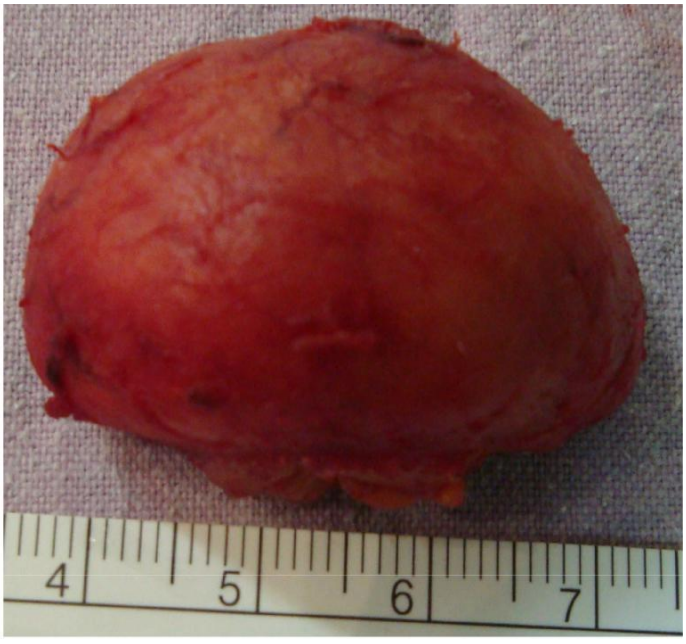


Figure 2b
Clinical picture of the completely excised mass



Histopathological examinations of the specimens showed numerous non-caseating granulomas with foreign body type and multinucleated giant cells (Figure 3). Numerous hyphae and spores were identified with Gomori Methylamine Silver stain (Figure 4). Lactophenol Blue stain of the specimen could not be done. Microscopic demonstration of the specimen showed characteristic non-septate hyphae while culture of the biopsy specimen after direct inoculation onto the Sabouraud dextrose agar yielded whitish fast growing colonies.

Figure 3
Photomicrograph showing:
a. Diffuse non-caseating granulomas with a sprinkling of neutrophil polymorphs at X40
b. Non-caseating granulomas at X40
c. Non-caseating granulomas at X100
d. Fungal hyphae are apparent at high power X400
No invasion of blood vessels was observed in any of the sections

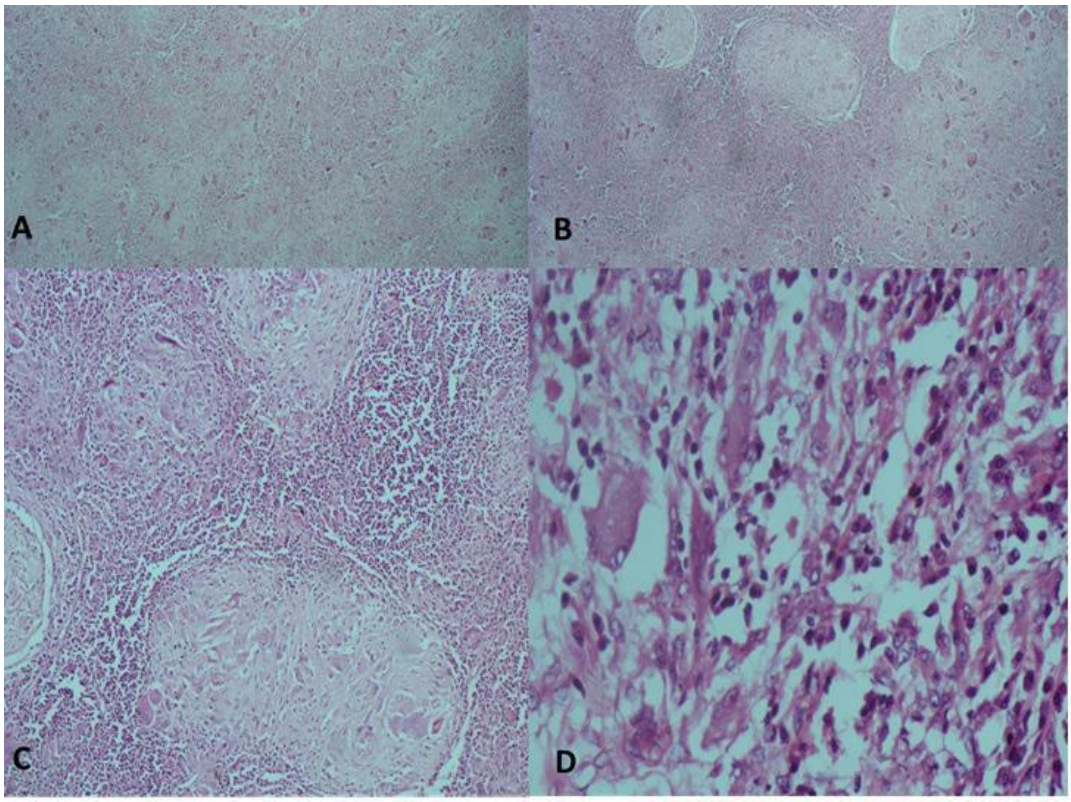
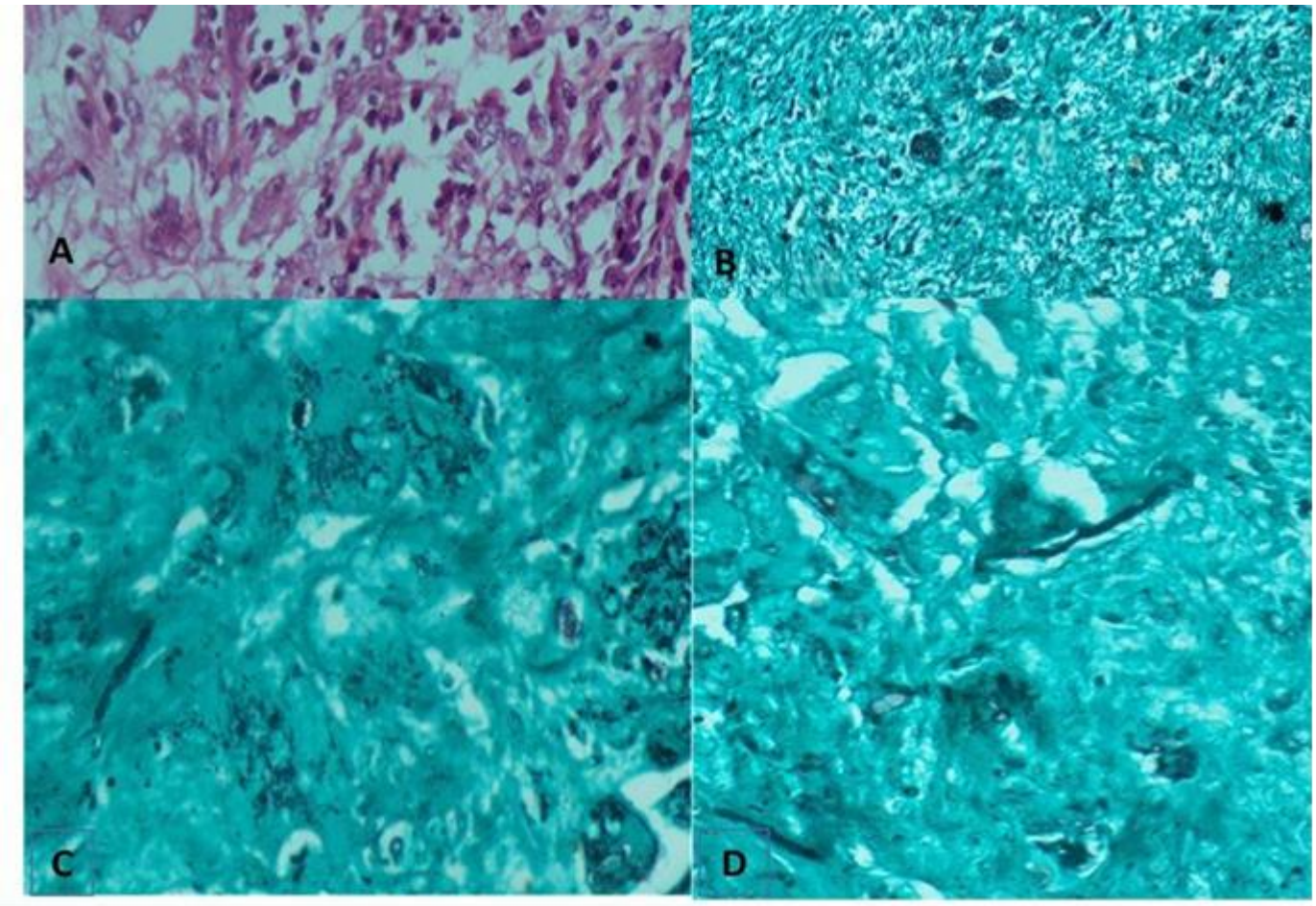


Figure 4
Photomicrograph showing:
a. Fungal hyphae in the excised mass, PAS, X400
b. Branching and non-branching hyphae, PAS, X400
c. and d. Hyphae and spore within multinucleated giant cells, GMS, X100



Based on the clinical, histopathological, and microbiological findings, a diagnosis of zygomycosis, probably entomophthoramycosis was made. She was treated with oral Fluconazole for eight weeks and fared well with treatment. However, severe ptosis with poor levator palpebral superioris action persisted (Figure 5a). She subsequently had ptosis correction with tarsofrontal sling 18 months after completing the antifungal medication with satisfactory outcome (Figure 5b). She has remained free of disease after 24 months of follow-up.

Figure 5a

Clinical picture of the patient after second surgical excision with left severe ptosis

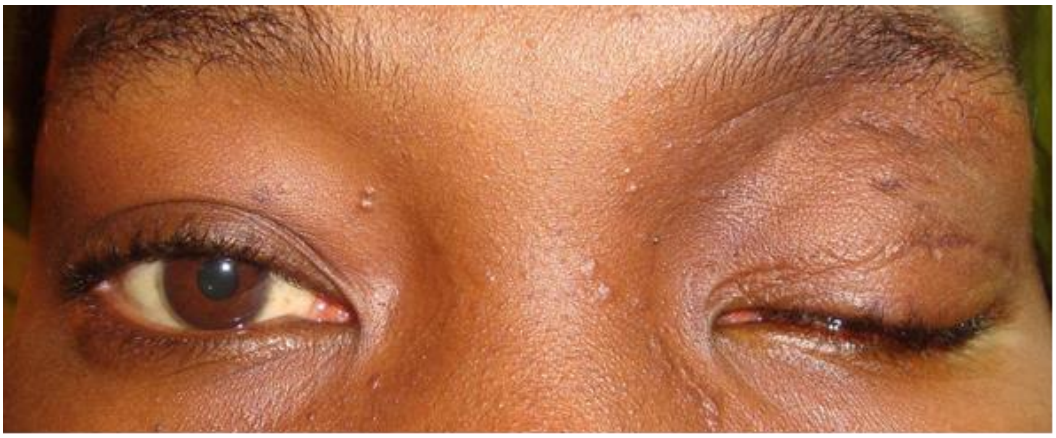
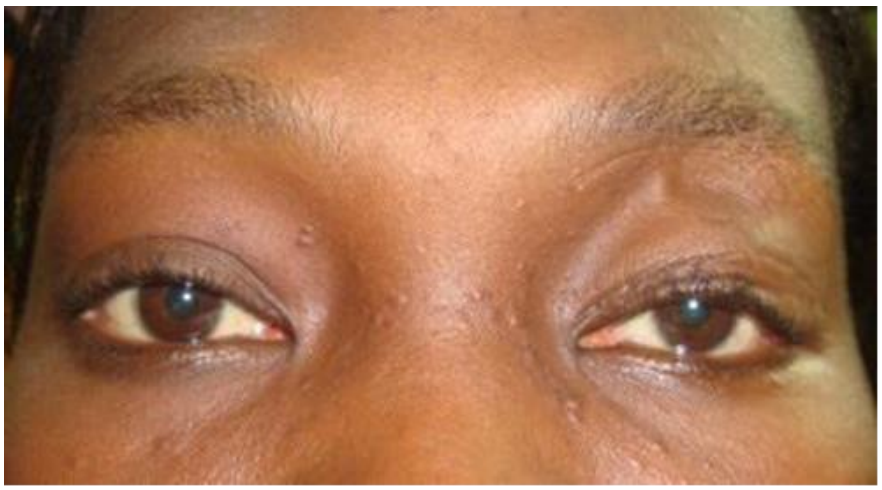


Figure 5b
Clinical picture of the patient after ptosis surgical correction by tarsofrontal sling



DISCUSSION

Cutaneous mucormycosis occasionally affects immunocompetent individuals and trauma is the commonest predisposing factor (2). Inoculation through surgical wound sites and sites of insertion of intravenous catheters as nosocomial infections had also been documented (6). Although the predisposing etiologic factor of the infection in our patient is unknown, the presumed mode of infection could have been direct inoculation through a minor skin abrasion, the organism being ubiquitously present in the soil and environment (7), which serve as its natural habitat and reservoir (7,8).

Cutaneous mucormycosis commonly presents as non-healing ulcers and necrotizing fasciitis (4) and, the risk of developing uncontrolled necrosis is said to be related to the size of the injury, degree of contamination, and the length of contact period (7).

Presentation as firm, well circumscribed, subcutaneous mass as it occurred in our patient is unusual, but might be due to her being immunocompetent with no obvious predisposing risk factors such as trauma, thus, suggesting limited contamination with the organism.

The organisms in the two orders causing zygomycosis have dissimilar ecologic, morphologic, epidemiologic and clinicopathologic characteristics, with Mucorales having a worldwide distribution while Entomophthorales are distributed mostly in the tropical and subtropical regions (9,10). Morphologically, pathogenic Mucorales fungi generally produce numerous asexual spores within the sporangium, while pathogenic Entomophthorales produce a single conidium on each conidiophore (9). Also, pathogenic species of Mucorales primarily affect immunocompromised individuals causing acute angio-invasive infections whereas Entomophthorales cause chronic subcutaneous infections in immunocompetent individuals (9). However, there is a great overlap in the clinicopathologic manifestations of atypical infections by organisms in these orders, hence, the inclusive name "zygomycosis" for cases with indistinct clinicopathologic manifestations, and unavailable culture studies (1). Specific organism identification in our patient was difficult as we could not carry out molecular sequencing and Lactophenol Blue stain of the excised tissue. Another challenge encountered in her management was socio-economic, leading to abandonment of treatment after the first surgical excision with subsequent recurrence. A second surgical excision was thus necessitated.

In conclusion, we have reported an unusual presentation of a localized subcutaneous fungal lesion involving the upper eyelid of an immunocompetent Nigerian female, successfully managed by surgical excision, medical treatment, and ptosis surgery. Cutaneous zygomycosis is uncommon, and presentation as a well circumscribed, painless, subcutaneous eyelid mass is unusual, but this should be considered a differential in immunocompetent individuals.

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