

Case Report

Fungal necrotizing fasciitis of face- a reconstructive challenge

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ABSTRACT

Fungal necrotizing fasciitis (NF), particularly in the face, is an unusual infection. It is mostly seen in immunocompromised individuals and can be gravely destructive if diagnosis and treatment are delayed. We report this rare case of a 27-year-old, immunocompetent male who presented with gangrenous patch of skin on the cheek following blunt trauma to the right side of the face. Till now no case has been reported in literature with such an extensive soft tissue loss of face and involvement of facial skeleton due to fungal etiology. Clinical suspicion of necrotizing fasciitis led to early surgical intervention. Histopathological examination of the debrided tissue identified the infective organism as *Apophysomyces elegans*. Intravenous antifungal therapy with liposomal amphotericin B was initiated. Despite the prompt commencement of the treatment, the infection continued to spread, and the patient had to undergo serial debridement which resulted in orbital exenteration, partial maxillectomy and mandibulectomy on the right side. This resulted in a huge soft tissue defect requiring flap cover. A free anterolateral thigh flap was harvested to cover the soft tissue defect, but the blood flow could not be established. For salvage, scalp and pectoralis major muscle flaps were raised and used to cover the large hemifacial defect. The oral lining was created with a folded deltopectoral flap in a second stage. However, there was persistence of the fungal elements in wound bed even after prolonged systemic liposomal amphotericin B therapy and it invaded the flap margins due to which there was partial necrosis of the flap. In this article, we aim to describe the difficulties faced by us in the management of such devastating infection and the reconstructive challenge that it posed.

Keywords: Necrotizing fasciitis, Fungal infection, Flap cover, Liposomal amphotericin B

INTRODUCTION

Necrotizing fasciitis is a potentially life-threatening soft-tissue infection; that has a sudden onset and is characterized by rapid spread of infection along the fascial planes. It has been classified into four type: type I is polymicrobial, including aerobic and anaerobic organisms; type II is caused by group A streptococci, with or without staphylococci; type III is monomicrobial due to clostridium species or gram-negative bacteria; type IV is of fungal etiology seen mostly in immune compromised patients.¹ Extremities, perineum and genitals are the common sites involved in NF. Face is rarely affected, with a predilection for peri-orbital region if involved.²

The fungus *Apophysomyces elegans* is a thermotolerant species, reported mainly in areas with warm climates. It mostly causes cutaneous infections after injury to skin; however, rhino-orbito-cerebral and renal infections have also been described.³

The diagnosis of NF in a patient is usually made clinically in presence of symptoms such as pain, swelling, fever, weakness, dizziness and diarrhea. The affected part will be swollen with black discoloration. The presence of white, fluffy growth is an indicator of fungal etiology. Surgical exploration with radical debridement aids in the definitive diagnosis of NF. Early, aggressive debridement and timely initiation of antimicrobials form the keystone of management in NF.

Fungal necrotizing fasciitis is a rare entity and, cranio-facial involvement is even more unusual, 74 cases are reported from India and abroad, with a very high mortality. Our patient had fungal NF affecting the face and, survived this potentially fatal infection with prompt medical and surgical intervention.⁹ The radical debridement necessary for eradication of the infection, resulted in a huge soft tissue defect that needed cover for aesthetic and functional rehabilitation. In our report, we aim to describe the challenges faced by us in managing the infection and in reconstruction of the complex soft tissue defect.

CASE REPORT

A 27 years old man reported to us with painful swelling on the right side of the face, progressive blackish discoloration over cheek, diminished vision in ipsilateral eye, and reduced mouth opening for three days. He gave a history of physical assault two weeks back, during which he sustained blunt trauma to the face with a wound below the right eye. The wound was sutured at a local hospital. There was no history of any past illness or surgery. On examination, a gangrenous patch of skin measuring 8x6 cm, was present over malar region. A friable, white fluffy growth was observed in the infra-orbital region. Edema, erythema, severe tenderness and induration was seen in surrounding areas (Figure 1).



Figure 1: Case at presentation with gangrenous patch of skin, edema, erythema and serosanguinous discharge. Note white fluffy cotton like fungal growth infraorbital region.

Mouth opening was 24 mm. Patient was febrile, tachycardic and hypotensive. Ophthalmic examination revealed a visual acuity of 6/60 in the right eye, with chemosis, and corneal opacification. The laboratory parameters were normal except for leukocytosis (24,000/cu mm) and thrombocytopenia (40,000/cu mm). NCCT face revealed diffuse soft tissue enhancement over right malar region and infratemporal fossa, without any intra cranial pathology.



Figure 2: Full thickness cheek defect after removal of necrosed skin, muscle and fat.



Figure 3: Composite soft tissue defect with exposure of facial and skull bones after second debridement.



Figure 4: Defect after removal of right zygoma, inferior turbinate, outer table of mandible, partial maxilla with alveolus has been removed. Note incision in neck for attempted free flap.

A provisional diagnosis of fungal NF was made, and the patient was started on intravenous liposomal amphotericin B along with broad spectrum antibiotics. Patient was stabilized and shifted to the operating room. Tracheostomy was performed in view of anticipated difficult airway and need for further surgical interventions. A radical debridement was performed under general anesthesia. The cheek skin, muscles, fat and nerves were found to be necrosed and removed (Figure 2). Thin, murky, foul-smelling fluid was oozing from the dead tissues. Tissue was sent for bacterial and fungal culture along with KOH mount which showed broad, aseptate and pauci-septate hyphae.



Figure 5: Scalp transposition flap to cover upper part of defect and pectoralis major muscle flap with split skin graft over outer surface, to cover lower half of the defect.



Figure 6: Extended deltopectoral flap folded over itself to create oral cavity lining and outer surface.

A second look debridement was performed after 48 hours, due to extension of the blackish discoloration along with development of fluffy, white growth beyond the debrided wound borders to involve half of the face. Right orbital exenteration was performed in view of intra-orbital spread of the infection with corneal perforation, eyeball collapse and, extra-ocular muscle and fat necrosis. Aggressive removal of dead and necrotic tissue was done including devitalized facial muscles and parotid gland, which left the facial bones (temporal, zygomatic, nasal, maxilla and mandible) exposed (Figure 3).

The patient improved clinically with reduction of facial swelling and erythema. Ryle's tube feeding was started to maintain adequate nutrition. Wound was managed with regular dressings. Results of fungal culture from the first debridement identified the organism as *Apophysomyces elegans*. Bacterial culture yielded klebsiella and providencia species sensitive to colistin. Liposomal intravenous amphotericin B was continued and colistin was added to the treatment. Serum potassium levels were monitored and maintained within normal limits. A repeat KOH mount after two weeks revealed the persistence of fungal elements in the wound bed, despite treatment.

Four weeks later, patient was taken under general anesthesia with a plan of definitive debridement and flap cover. Right zygomatic bone, right inferior turbinate, outer table of mandible with condyle and coronoid, right part of maxilla with alveolus was excised (Figure 4). To cover the defect free thin ALT flap was attempted but flow across the anastomotic site of vessels could not be achieved. Scalp transposition and pectoralis major muscle flap were done to cover the upper and lower part of defect, respectively. The muscle flap was covered with a split thickness skin graft and the flaps were sutured to each other (Figure 6). The flaps settled and liposomal amphotericin B was continued for three more weeks till the KOH mount showed fungal element of doubtful significance (broken hyphae).

Two months later, definitive reconstruction to create inner nasal and oral lining was planned. Right deltopectoral flap was delayed and raised to inset into the margins of pharyngeal wall, palate and folded on itself to suture to pectoralis major muscle flap. Post-operatively, the flap looked healthy for four days. On the 5th day, distal necrosis was observed. On exploration, the cavity was found to be filled with soft, friable, pale white fungal mass which has been removed and sent for fungal culture. The necrosed part was debrided and the flap re-insetted (Figure 6). Culture from both necrosed flap and mass, yielded fungal growth. intravenous liposomal amphotericin B was restarted and continued for another three weeks. Patient was gradually started on oral feeds and the Ryles's tube was removed. Tracheostomy tube was removed after a laryngoscopic examination. Patient refused for further surgical intervention and was discharged. He was prescribed itraconazole to be taken orally for eight weeks. Patient is now under follow-up,

gaining weight, feeling comfortable and on regular oral diet.

DISCUSSION

Apophysomyces elegans was earlier considered a rare pathogen with a propensity to infect immunosuppressed individuals. The most common systemic conditions that predispose to such an infection are uncontrolled diabetes mellitus, malignancy, leucopenia, immunosuppressive therapy, organ transplantation and alcoholic cirrhosis. However, mucormycosis cases caused by this fungal species have increased over the past few decades, especially in immunocompetent hosts, like our patient.⁴ In one of the largest series, Jain et al reported 18 patients with zygomycotic necrotizing fasciitis of which 15 were immunocompetent. Common regions involved were the abdomen, chest wall, lower extremities and gluteal region.⁵ Fungal necrotizing fasciitis of the cranio-facial region is rare and can be devastating with a high morbidity and mortality of about 30%. Our patient survived and underwent flaps coverage for large composite defect.

Percutaneous inoculation of pathogen after trauma is the most common mode of infection. Intramuscular injections have also been identified as other possible route of infection.⁷ Our patient had a sutured wound over the right infra-orbital region that may have been the portal of entry.

Clinical signs such as skin discoloration, necrosis, swelling and erythema with presence of white, fluffy growth aid in early diagnosis of fungal NF, prompting early treatment. Patient may also exhibit systemic features such as hypotension, tachycardia, and fever. A scoring system has been developed for early detection of NF. A score of more than or equal to 6 on LRINEC scoring system should raise suspicion of NF.⁸ Direct demonstrations of hyaline aseptate or pauci septate hyphae by KOH mount in the sample, culture on SDA with antibiotics and induction of sporulation by special methods provides a definitive diagnosis.⁹ A fungal culture usually takes around two weeks and is needed for species identification. KOH mount takes considerable lesser amount of time and if positive, is helpful in starting antifungal therapy early.

The quick progression or rapid deterioration is explained by growth of fungus in vascular lumen (angioinvasion) leading to thrombus formation and tissue ischemia.¹⁰ No cases with such significant soft tissue loss of face and facial skeleton involvement due to fungal etiology have been documented so far in the literature. Early, aggressive debridement is the key to gain control of the infection and prevent its spread. Despite repeated debridement in our patient, there was progressive tissue damage with persistence of fungal elements and a prolonged anti-fungal therapy was required.

The standard antifungals for the management of *Apophysomyces mucormycosis* are amphotericin B and itraconazole. Alternatives such as Posaconazole in Amphotericin B-resistant cases and Isavuconazole are used in susceptible strains.⁶ There is no standard duration of treatment for mucormycosis. Decisions are made on an individual basis, and as a principle, antifungal therapy of mucormycosis is continued until resolution of all clinical, laboratory and imaging signs of infection and, reversal of immunosuppression.¹¹

NF of the face poses a unique challenge of secondary reconstruction due to complexity of soft tissue defects. The choice of reconstruction depends on the site and extent of involvement. Extensive soft tissue defects require skin and subcutaneous tissue for bulk. The resultant fibrosis and loss of subcutaneous tissue under adjacent skin makes it difficult to advance and rotate local skin flaps. In such cases, fasciocutaneous free flaps can be used to cover the skin defect as well as to provide sufficient bulk for restoration of facial contour. Scapula, lateral thigh, and radial forearm are potential donor sites for fasciocutaneous free flaps.¹² Our patient had a large right hemifacial defect on with bone loss.

Our unsuccessful attempt at free flap cover was attributable to compromised recipient vessels. The persistence of fungal elements despite prolonged intravenous therapy made the reconstruction of the defect even more demanding. A single flap may not be able to cover such huge, complex defects. So, regional flaps in the form of scalp, pectoralis major muscle and deltopectoral flaps were ultimately utilized to cover the defect and achieve some functional rehabilitation for the patient.

CONCLUSION

Fungal necrotizing fasciitis of the face is a distinct clinical entity with high morbidity and mortality. Clinical suspicion with the aid of histopathological examination is paramount for early diagnosis. Repeated, aggressive debridement and early initiation of parenteral antifungal improve the survival chances of the patient. The resultant soft tissue and bone defect is, however, difficult to reconstruct and needs the whole surgical armamentarium. As of now, the focus while providing soft tissue cover for such large and complex defects is mainly on functional rehabilitation of the patient.

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