

Fungal Necrotizing Fasciitis of the Left Hemiface with Subsequent Temporomandibular Ankylosis and Eyelid Lymphedema: A Case Report

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Abstract

A 35-year-old female presented with progressive swelling and necrosis of the left cheek after sustaining maxillofacial trauma due to a motorcycle accident. Intravenous antibiotic was given, and emergency debridement was performed. Tissue culture showed *Aspergillus flavus complex*, *Acinetobacter baumannii*, and *coagulase-negative Staphylococci*, so her antibiotics were adjusted to amphotericin B, cefoperazone-sulbactam, and fosfomycin. After the infection was improved, her left hemifacial defect was covered with a skin graft. One month later, she developed a gradually diminishing mouth opening and progressive swelling of her left eyelid that completely obscured her vision. Left temporomandibular joint (TMJ) ankylosis and eyelid lymphedema were diagnosed. Preoperative investigations were performed, and the staged surgical reconstruction strategy was developed. After removing the scar tissues and releasing ankylosis, reconstruction was performed using an anterolateral thigh (ALT) free flap, which directly contacted the left eyelid to facilitate lymphatic drainage. Early postsurgical mouth-opening rehabilitation was introduced. Her swollen left eyelids significantly improved thereafter, and the patient could open her mouth freely with an interincisal gap of 3.5 cm. The second operation involved contouring of the flap, smile reconstruction with a Tensor Fascia Lata sling, and partial resection of the left eyelids to reestablish her vision. The patient can now eat without difficulty, and she has favorably reintegrated into society with good mental health.

Keywords: Fungal infection, Necrotizing fasciitis, Facial reconstruction, Ankylosis, Lymphedema

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INTRODUCTION

Necrotizing fasciitis (NF) presents a rapidly progressing infection that affects the fascial layer and subcutaneous tissues. This condition may arise from idiopathic or secondary causes, such as trauma and skin eruptions, potentially leading to life-threatening conditions.¹ In addition to the rare NF of the face compared to other parts of the body, treatment of NF of the face poses a surgical challenge.² Only 29 cases of NF of the face have been reported.³ Due to the thin nature of the skin and muscles, deeper structures, such as facial nerves, may become involved, and debridement may be unavoidable. The surgeon balances the necessity of thorough surgical debridement with the preservation of functional and aesthetic considerations.^{4,5}

CASE REPORT

A 35-year-old Thai female sustained a right zygomatic complex fracture and multiple facial wounds in a motorcycle accident. The zygoma was treated with internal fixation using miniplates. All wounds were decontaminated and dressed in standard fashion. The patient was discharged one day after the operation and was scheduled for daily wound dressing.

One week after discharge, she developed cellulitis on her left face. The infection progressed to necrotizing fasciitis, which required prompt surgical debridement (Figure 1). She was admitted, intravenous amoxicillin-clavulanate was given, and emergency debridement was performed. Tissue culture showed *Aspergillus flavus complex*, *coagulase-negative Staphylococci*, and *Acinetobacter baumannii*. The antibiotics were then switched to amphotericin B, sulbactam/cefoperazone, and fosfomycin to cover the identified organisms.



Figure 1 (A) Abrasion wound and contusion at left face at 1-week post-accident. (B, C) Progressive swelling and the development of necrotic tissue with fluffy white cotton wool-like growths at the left side of the face two weeks post-accident. (D) Rapidly progressing necrotizing fasciitis.

She underwent a tracheostomy and 14 serial debridement procedures to remove infection and necrotic tissue. The debridement process resulted in a large soft tissue defect on her left face that involved her left forehead, temporal area, cheek, upper neck, and posterior auricular area. The left parotid gland, muscle of facial expression, and left facial nerve were removed during debridement. After gaining control of the infection, the defect on her left face was temporarily closed using the skin graft, and the patient was referred to our center.

At the first visit, she had thin skin coverage with scar tissue and loss of facial contouring over her left face. She had lymphedema of the left eyelid that completely obstructed the vision of her left eye. She had malocclusion and limited mouth opening with an interincisal distance of 1 cm. She also had left facial palsy with total paralysis (House-Brackmann grade VI). Her body mass index (BMI) was 33.7 kg/m^2 without other underlying disease (Figure 2).



Figure 2 (A-C) Patient photos on the first visit, left eyelid was marked swelling due to lymphatic obstruction. (D) Intraoral examination shows limited mouth opening with an interincisor distance of less than 1 cm and malocclusion.

The two times of stage reconstruction were planned. The first operation included the total excision of the scar tissues and skin grafts, then the release of soft tissue ankylosis around the temporomandibular joint (no bony ankylosis was found) and reconstructing the entire defect with anterolateral thigh (ALT) free flap (The ALT free flap

size 21×13 cm). The intraoperative results demonstrated an interincisal distance of 3.5 cm, and the patient's malocclusion was improved. The edge of the flap was connected with the raw surface of both the upper and lower eyelids to facilitate lymphatic connection and drainage (Figure 3).

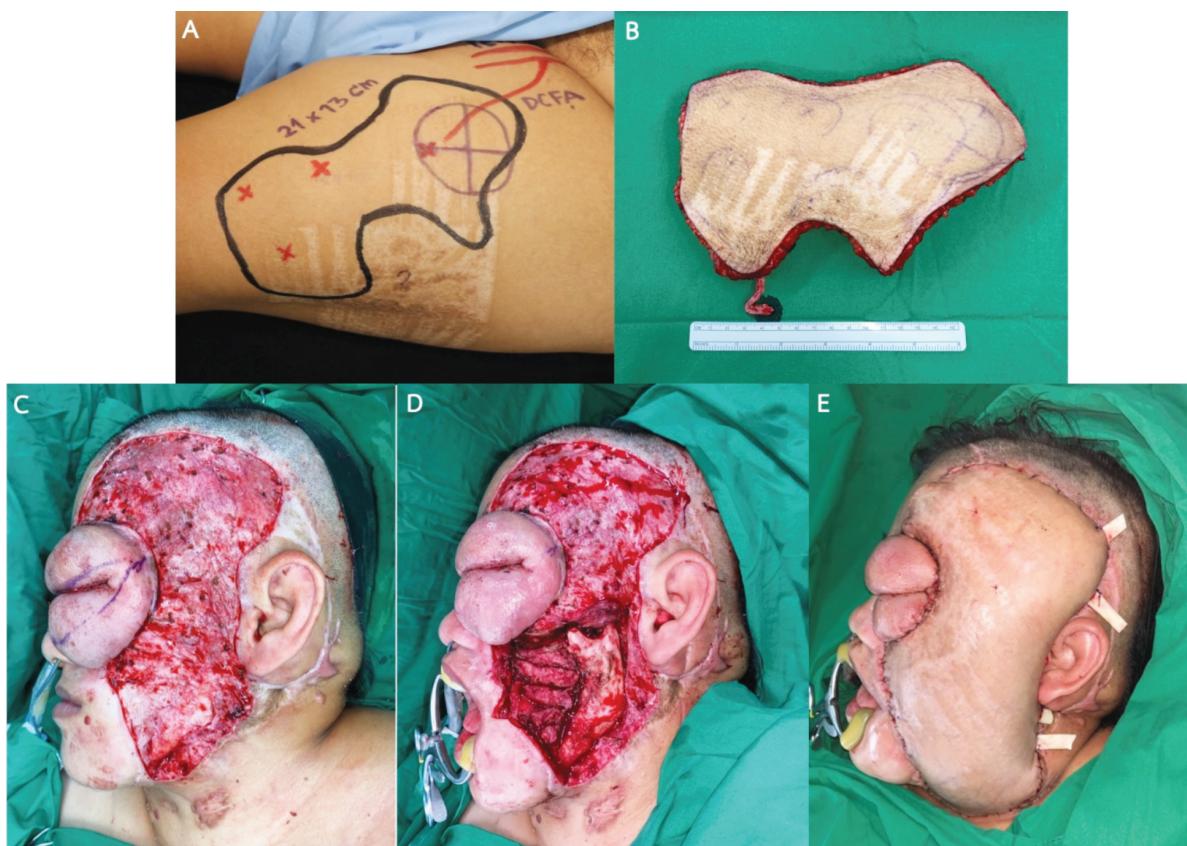


Figure 3 (A) Anterolateral thigh fasciocutaneous flap was designed according to the recipient defect. (B), Flap harvested with the dimension of 20×13 cm. (C) Defect after debridement. (D) Temporomandibular joint after exploration and removal of surrounding scar tissue. (E) Immediate postoperative results.

The second operation was performed 6 months later and focused on improving the function and aesthetics. Indocyanine green (ICG) lymphography was performed at the left eyelid, and the results revealed the spontaneous drainage of lymph fluid through the neo-lymphatic connections between the left eyelids and the ALT free flap

(Figure 4). The debulking procedure with liposuction and partial resection of excess skin and subcutaneous tissue were performed. Finally, the static smile reconstruction by re-positioning of the left oral commissure was performed by Tensor Fascia Lata (TFL) sling and suture hanging to the periosteum of the left zygoma (Figure 5).



Figure 4 Intraoperative indocyanine green (ICG) lymphography. (A) Immediately after injection. (B) After 10 minutes. (C) After 20 minutes, ICG was spontaneously drained into the reconstructed anterolateral thigh-free flap.

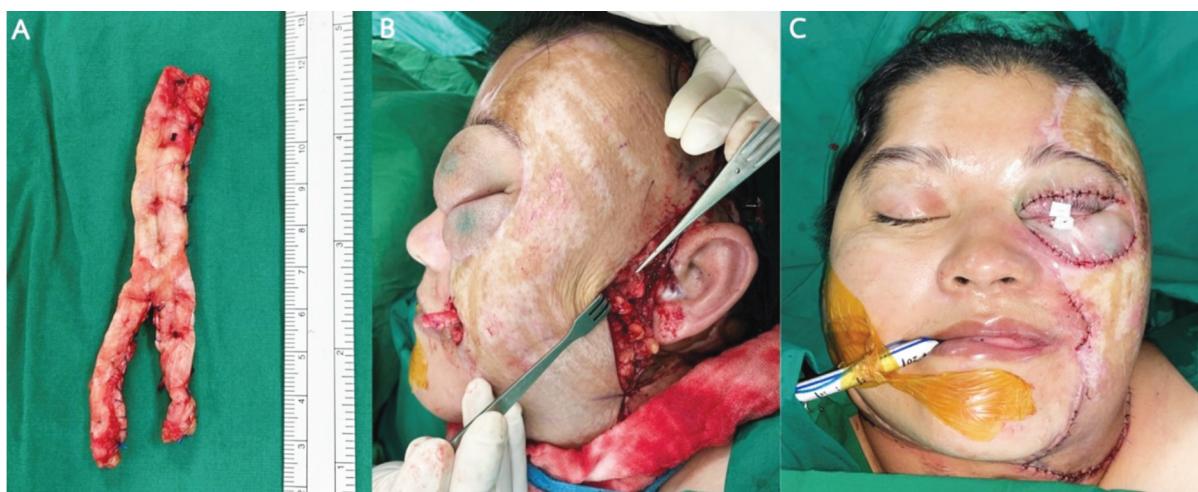


Figure 5 (A) Tensor Fascia Lata (TFL) from the right thigh was prepared. (B) The fascial graft sling was inserted beneath the flap at the left cheek, and the suture hanging for static smile reconstruction. (C) The immediate postoperative outcomes after fascial sling, flap debulking, and partial resection of lymphedema tissue from the left upper and lower eyelids.

At the 1-month postoperative follow-up, the visual field of the patient's left eye was restored, and we plan

to correct some ectropion via lateral canthoplasty under local anesthesia in a future procedure (Figure 6).

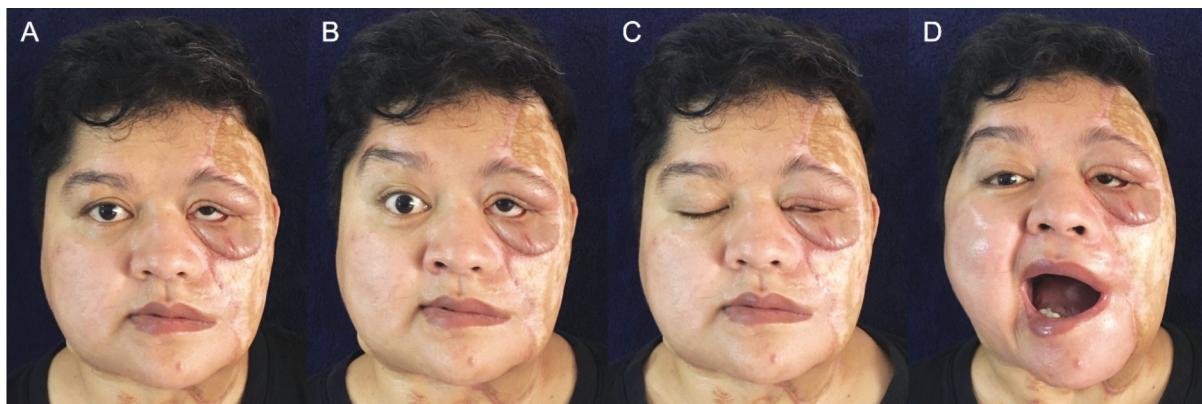


Figure 6 One month after the second operation, (A) Frontal view showed improvement in facial contour. (B, C) The patient can open and close her left eye without difficulty, but she still had some ectropion and scleral show during eye closure. (D) Mouth opening was improved with an interincisal distance of 3.5 cm and good occlusion.

DISCUSSION

Necrotizing fasciitis (NF) of the face is a rare and life-threatening disease.¹ Our patient presented with only an abrasion wound, which later progressed to NF. We found no identifiable predisposing factors, including diabetes, hypertension, or chronic alcohol abuse.^{2,6}

The most commonly isolated microorganisms are *Group A Streptococcus*, *Candida spp.*, and *Enterobacter cloacae*. Polymicrobial infection is more common than monomicrobial infection.³ Broad-spectrum antibiotics should be given immediately after diagnosis, and adequate surgical debridement should be performed.⁷ In this case, tissue culture showed *Aspergillus flavus* complex, *Acinetobacter baumannii*, and *coagulase-negative staphylococci*.

Fungal NF is relatively rare; it may be caused by direct infection by a fungus or as a secondary superimposed infection. A higher prevalence of fungal infection is observed in patients with diabetic mellitus due to decreased neutrophil chemotaxis and phagocytosis.^{1,8} Fungal necrotizing fasciitis is characterized by specific signs, including the rapid progression of black tissue necrosis, the formation of cotton wool-like material over the wound, and the absence of clinical response following treatment with broad-spectrum antibiotics. These indicators are crucial in diagnosing and treating this serious condition. A definite diagnosis can be made from tissue culture for fungus and tissue pathology.

Aspergillus flavus, found in soil and outdoor air, is a common cause of fungal infection after trauma.⁹ There is a report of NF caused by *A. flavus* in an immunocompromised patient, but this infection in an immunocompetent host has not been previously reported.¹⁰ Amphotericin B is the drug of choice for treating this fungus.¹

Regarding the outcome of treatment, severe functional deficit and disfigurement of her left hemiface were inevitable. The split-thickness skin graft was employed to cover the entire wound prior to referral temporarily, but the patient developed malocclusion, left TMJ ankylosis, and left eyelid lymphedema. The first stage of the procedure aims to correct the malocclusion and the TMJ ankylosis, to downsize the upper and lower lymphedematous eyelids, and to cover the entire defect with well-vascularized skin and soft tissue.

Concerning facial function and aesthetics, our patient exhibited a diminished left facial expression resulting from multiple aggressive debridements aimed at eradicating the infection and necrotic tissues. Static smile reconstruction with a TFL sling was selected in this case after discussing the pros and cons of each reconstruction technique with the patient.

For left eyelid lymphedema, we managed by creating new lymphatic circulation via a well-vascularized flap. We also partially resected redundant skin and soft tissue. After surgery, our patient can now open her left eye spontaneously, take an oral diet without difficulty, and reintegrate into society with a good mental health status.

CONCLUSION

Necrotizing fasciitis of the face is a rare and devastating infection. Polymicrobial infection, including fungus, should be suspected. Appropriate surgical debridement should be performed, and proper intravenous antibiotics and antifungals should be given as soon as possible. Wound coverage with subsequent defect reconstruction should be considered after achieving infection control. The well-planned staged reconstruction can yield favorable aesthetic and functional outcomes.

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CONFLICT OF INTEREST

All authors declare no personal or professional conflicts of interest relating to any aspect of this study.

FUNDING DISCLOSURE

This was an unfunded study.

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