Case Report

The Challenges of Extensive Face and Neck Necrotising Fasciitis Reconstruction: A Case Report

Hui Yuan Lam [1,2], Hamidah Binti Mohd Zainal[1], Khai Luen Koh[1*], Mohammad Ali Bin Mat Zain [1]

1. Department of Plastic Reconstructive Surgery, Hospital Kuala Lumpur, 50586, Kuala Lumpur, Malaysia
2. Science and Reconstructive Unit, School of Medical Science, Hospital Universiti Sains Malaysia, Kelantan, 16150, Malaysia

***Corresponding Author:**

Khai Luen Koh

Department of Plastic Reconstructive Surgery, Hospital Kuala Lumpur, Malaysia

Email: kkhailuen@moh.gov.my

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ABSTRACT

Necrotising fasciitis (NF) is a rare, severe, rapidly progressive, and life-threatening synergistic infection that primarily affects the superficial fascia. Although unusual in head and neck surgeries, necrotizing fasciitis may have drastic consequences. Dental problems, burns, trauma, and peritonsillar abscesses often cause the condition. We describe a rare case of extensive facial necrotising fasciitis secondary to carbimazole–induced agranulocytosis. A protective tracheostomy, prophylactic thyroidectomy, serial surgical debridement, and broad-spectrum antibiotics therapy were initiated. Our team referred the patient for subsequent soft tissue reconstruction. Reconstruction was challenging as it involved a full-thickness cheek defect extending to the oral commissure, lower lip, and anterior neck. In a single-stage procedure, we utilised a transverse upper gracilis free flap to reconstruct full-thickness tissue loss, provide tissue bulk, and seal the dead space. Reconstructing various parts of the face and lips while ensuring aesthetic and functional results can be difficult..

KEYWORDS
Carbimazole-induced agranulocytosis; Necrotising fasciitis; Lip reconstruction, Microsurgery

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INTRODUCTION

Necrotising fasciitis (NF) is characterised by fulminating, devastating, rapidly progressing, and generalized necrosis of the superficial fascial layer and cutaneous tissue[1]. NF primarily affects patients with weakened immune systems and predominantly occurs in the abdominal wall, perineum, and extremities, while the head and neck structures are rarely affected due to their abundant blood supply[1,2]. Managing necrotizing fasciitis on the face can be difficult due to its complex anatomy. Effective treatment requires a multidisciplinary approach and prompt medical and surgical interventions.

Here, we report a case of a young patient who developed necrotizing fasciitis due to carbimazole-induced agranulocytosis. This condition rapidly spreads across different anatomical subunits of the face and neck, including the cheeks, lips, and neck which presents a challenge in achieving both aesthetic and functional outcomes in these regions.

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CASE PRESENTATION

A man in his 30s was diagnosed with hyperthyroidism four months before the presentation. He was admitted to the Emergency Department with tonsillitis and swelling over his lips and neck (Figure 1 A, B, C, D). Further investigation revealed that he had been taking carbimazole 15 mg TDS for the past three months instead of carbimazole 15 mg OD. Physical examination revealed diffuse oedema over both cheeks, extending to the back of his neck. Full-thickness necrotic tissue covered the left angle and mandible of the mouth, resulting in eating and breathing difficulty for a patient due to limited mouth opening. Blood tests confirmed agranulocytosis and blood cultures revealed *Pseudomonas* infection.

Computed tomography (CT) indicated Ludwig's angina and small air pockets in both peritonsillar spaces. The surgical procedures were staged, with the initial procedure aimed at securing the airway using tracheostomy. A percutaneous feeding gastrostomy was performed in anticipation of saliva pooling to treat the patient's condition. Finally, extensive surgical debridement was done under similar conditions. The patient had unhealthy tissue and slough at the superior and inferior gingivobuccal sulci, extending to the pyriform aperture and left anterior wall of the maxilla. A necrotic patch was also found over the entire hard palate and tonsils. The thyroidectomy was carried out to prevent thyroid storm or thyrotoxicosis. After a series of debridement, the wound condition was deemed suitable for closure, and our team was consulted to restore oral cavity function, as this condition was caused by a full-thickness defect over the oral commissure and cheek (Figure 1 E, F, G). This patient had a composite tissue defect involving the left cheek and upper lip.

Moreover, the oral cutaneous fistula was connected to the neck, making reconstruction challenging. Therefore, we opted for a staged microsurgical procedure to replenish the dead space in the oral cavity using free-flap surgery. Once the bulk and volume of the face were restored, a second surgery was carried out to create a more aesthetically functional oral commissure region. We harvested a transverse upper gracilis myocutaneous (TUG) free flap supplied by the medial femoral circumflex artery and vena comitans. Identifying appropriate vessels for anastomosis was difficult because necrotizing fasciitis also affected the neck. It was a complex decision for the surgeon and the patient if



Figure 1: (A-D) Initial presentation prior to wound debridement. (E-G) Post wound debridement.

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we were to use double free flap to reconstruct the cheek, intraoral and neck defect. The initial arterial anastomosis was connected to the transverse cervical artery using a transposition vein graft. However, the flow rate was unsatisfactory. The decision was made to use the superficial temporal artery as the recipient vessel with a graft harvested from the great saphenous vein. The proximal part of the gracilis muscle was used to resurface the lower mandible, and the skin paddle lined the left cheek, upper lip, and oral commissure. A skin graft covered the gracilis muscle lining the intraoral region.

DISCUSSION

Antithyroid drugs, such as propylthiouracil, methimazole, and carbimazole, have harmful haematological effects, ranging from mild leukopenia to more severe agranulocytosis and aplastic anaemia. Agranulocytosis and neutropenia account for nearly half of all catastrophic responses to antithyroid medications[3] . Mild leukopenia is characterized by a total white blood cell (WBC) count of < 4,000/L[4] . This rare but potentially fatal condition can drastically reduce the number of granulocytes to an absolute neutrophil count of < 500/μL[5]. Agranulocytosis caused by carbimazole occurs in 0.3-0.6% of cases and has a fatality rate of 21.5%[6]. The most common symptoms of agranulocytosis include ulcerative changes in the upper respiratory mucous membranes, ulcerative-necrotic tonsillitis, gastrointestinal side effects, fever, regional lymphadenopathy, and fungal infections[6]. Administration of broad-spectrum antibiotics and haematopoietic growth factors, particularly granulocyte colony-stimulating factors, has improved the prognosis of this life-threatening condition

Pseudomonas is the primary cause of sepsis in patients with agranulocytosis caused by antithyroid drugs. The infection starts with colonisation and progresses to breaking down the host's defences, allowing for local invasion, eventually leading to dissemination and systemic illness. Only three instances of severe tissue infection caused by fulminant *Pseudomonas* secondary to drug-induced agranulocytosis have been reported: one case where the patient of ectyma gangrenosum and facial cellulitis temporarily resolved with medical intervention, another with necrotising fasciitis of the right forearm, necessitating emergency fasciotomy and skin grafting, and third case documented Fournier gangrene due to a *P. aeruginosa* infection[8,9,10].

Our case was the first documented instance of extensive necrotising fasciitis of the face requiring free-flap reconstruction to achieve optimal aesthetic and functional outcomes. The reconstruction process was particularly challenging, because it involved multiple aesthetic units, including the upper and lower lips, oral commissure, and cheek. Given the full-thickness loss in the orofacial region, tissue transfer utilizing microsurgical techniques was necessary. We opted for a transverse upper gracilis flap (TUG) free-flap because it eliminates dead space and inhibits infection in this complex, a *Pseudomonas*-infected wound. However, it was not easy in setting the flap due to this region's bulk and three-dimensional reconstruction. A substantial portion of the skin-grafted flap was inset at the cheek. Simultaneously, the distal end was directed towards the anterior lip and sutured to the contralateral oral layer and lip remnant to create a deep labial gingival sulcus to prevent saliva leakage and maintain oral competence (Figure 2 A, B, C, D). The patient got discharged ten days postoperatively, and the flap and skin graft survived successfully. In the future, flap debulking and commissuroplasty will be carried out to reshape the commissure and restore its symmetry (Figure 2 E, F, G).

For several reasons, in our case, the TUG flap was often preferred over the anterolateral thigh and the radial forearm flaps:

1. Vascularity: The gracilis muscle has a reliable blood supply, making it a safe and effective option for reconstruction. The skin paddle was also reliable for lip and cheek tissue.
2. Donor site morbidity: The gracilis flap was harvested near the groin crease, a hidden and cosmetically acceptable donor site. In contrast, the anterolateral thigh flap requires a larger incision, and the radial forearm flap requires the harvesting of the skin and fascia from the forearm, which may lead to scarring and a less cosmetically acceptable outcome.
3. Functionality: A gracilis flap could reconstruct the outer anatomy of the lip and the oral sphincter, essential for maintaining oral continence and speech. This makes it a more functional option than the anterolateral thigh or radial forearm flaps.

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Figure 2: (A-D) Intraoperative picture. (D-G) 2 months after complex wound reconstruction with transverse upper myocutaneous gracillis flap.

4. High affinity to infection control. Gracilis muscle was able to obliterate and separate intraoral and neck wounds. This combat infection from communicating in these regions.

The major disadvantage of this flap was the short pedicle which required vein graft. Thus, the composition of the flap was more challenging to contour accordingly without compromising the flap viability.

CONCLUSION

Optimal lip reconstruction outcomes depend heavily upon precise assessment of three-dimensional tissue loss and the availability of adequate volume for reconstruction. Frequently, lip defects may appear more severe because of the lateral displacement of the facial musculature and the amount of unhealthy tissue that requires debridement. The TUG flap reconstruction is a highly regarded technique for lip reconstruction that caters to the two key dimensions of functional and aesthetic outcomes.

CONFLICT OF INTEREST

Non-declared.

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