Case Report

A parotid abscess out of control resulting in craniocervical necrotising fasciitis in the context of diabetes mellitus—a case report and review of the literature

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Background: Necrotising fasciitis is an aggressive life-threatening infective process rarely making an appearance in the head and neck region and its development secondary to parotid abscess is exceptionally rare and scarcely reported in the literature. This case report serves to guide otolaryngologists with respect to its recognition and offers an alternative approach to craniocervical necrotising fasciitis with multiple neck explorations, use of antimicrobial impregnated packing enabling delayed reconstruction with lower morbidity.

Case Description: A 76-year-old female with a body mass index of 36.2 kg/m^2 and a 30-year history of poorly controlled type 2 diabetes mellitus (HbA1c 91 mmol/moL), presented to the outpatient otolaryngology clinic with right sided parotid mass with minimal erythema, hyperglycaemia (19.2 mmol/L) and no cranial neuropathies. However, the aggressive nature of the parotid abscess triggered by *group A streptococcus* and *Staphylococcus epidermidis* led to sepsis and extensive non-odontogenic necrotising fasciitis involving the lateral neck mandating multiple surgical debridement and neck explorations, prolonged intravenous antibiotics with interval definitive reconstruction. A cervicofacial rotational sternocleidomastoid flap was utilised to conceal the defect with patient experiencing a remarkable recovery. The patient's immunosuppressive state from poorly controlled diabetes mellitus and multi-lineage cytopenia is likely to have contributed to a prolonged recovery.

Conclusions: This case report highlights the significance of repeat explorations and the need to give time for tissue healing as it unlocks options for reconstruction and reduce overall patient morbidity. Bismuth iodoform paraffin paste packing is a valuable tool with this case demonstrating its use an antiseptic and haemostatic agent in necrotising fasciitis and its ability to create an atmosphere to enable tissue healing minimising need for large-scale reconstructions. The absence of crepitus should not discourage the treating clinician from suspecting necrotising fasciitis of the neck. To limit successive cases, early prevention through aggressive control of predisposing systemic conditions including diabetes mellitus is needed. Moreover, when aggressive infections arise, the clinician should investigate for contributing systemic conditions.

Keywords: Parotid abscess; case report; necrotising fasciitis

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Introduction

Necrotising fasciitis of the parotid due to parotid abscess remains a rare clinical presentation in Otolaryngology and in the described literature. Necrotising fasciitis is an uncommon, life-threatening, single or polymicrobial infection of the soft tissue characterised by widespread necrosis of the skin, subcutaneous fat and fascia (1). Necrotising fasciitis rarely affects head and neck with an incidence of 4 per 100,000 (2) and necrotising fasciitis affecting the craniocervical region exists as a rare entity with only 38 cases reported in the literature (3). These usually originate from a dental source (50%), injury to skin/mucosa (25–74%), pharyngeal or tonsillar source (<15%), in addition to other triggers including radiotherapy and iatrogenic causes (3).

This case is unique with extensive necrotising fasciitis secondary to a parotid abscess in a poorly controlled diabetic requiring multiple surgical debridement and subsequent reconstruction. This is the second case of parotid abscess leading to craniocervical necrotising fasciitis but the first to employ a cervicofacial rotational sternocleidomastoid flap reconstruction in necrotising fasciitis secondary to a parotid abscess. Authors showcase an alternative approach to management of craniocervical necrotising fasciitis; one demanding utmost patience from the treating clinician with frequent neck explorations and use of antimicrobial impregnated packing to facilitate less morbific delayed reconstruction. We present this article in accordance with the CARE reporting checklist (available at https://gs.amegroups.com/article/view/10.21037/gs-23-365/rc).

Highlight box

Key findings

- Parotid abscess in a poorly controlled diabetic led to extensive lateral neck necrotising fasciitis secondary to *group A Streptococcus and Staphylococcus epidermidis* which is exceedingly rare.
- This case highlights absence of crepitus does not exclude necrotising fasciitis.

What is known and what is new?

- This unique and rare case of an aggressive parotid abscess resulting in non-odontogenic craniocervical necrotising fasciitis and the first case to describe use of a cervicofacial rotational sternocleidomastoid flap for its reconstruction.
- In our case, four neck explorations were performed with use of bismuth iodoform paraffin paste (BIPP) to enable good tissue healing.

What is the implication, and what should change now?

- Patients with necrotising fasciitis involving the head and neck region should have frequent reassessments and neck exploration to accurately determine appropriate reconstructive modality offering the lowest morbidity. This case highlights patience avoided need for pectoralis major flap reconstruction.
- BIPP in our case cultivated an environment to allow tissue healing thus minimising need for large-scale reconstructions.

Case presentation

This case features a 76-year-old lady who was seen in the outpatient otolaryngology clinic with a three-week history of a right sided parotid mass. She was clinically well with examination at the time revealing a large minimally tender and firm right parotid mass with some overlying erythema without crepitus and an intact facial nerve. She was instigated on oral co-amoxiclav 625 mg thrice daily (TDS) and was due to be followed up with an urgent outpatient magnetic resonance imaging (MRI) and ultrasound (US) parotid with fine needle aspiration. Her past medical history includes a 30-year history of poorly controlled type 2 diabetes mellitus, non-alcoholic steatohepatitis with portal hypertension, anaemia of chronic disease, hypertension, moderate aortic stenosis with moderately impaired cardiac systolic (ejection fraction 41%) and grade II diastolic function. Her regular medications included bisoprolol, doxazosin, amitriptyline, atorvastatin, ferrous sulphate, subcutaneous insulin (glargine) and metformin. She was a retired non-smoker, consumed normal intake of alcohol and mobilised with a stick, with no previous trauma or radiotherapy to the parotid. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the consent is available for review by the editorial office of this journal.

However, she presented to emergency department the day after at a district general hospital with new confusion, lethargy, swinging fevers, progressive right sided facial swelling with trismus and right sided otalgia. She had no native teeth and did not suffer from any dental problems and had no recent dental procedures. She denied urinary, gastrointestinal, musculoskeletal, cardiorespiratory, or dermatological symptoms. Her blood pressure was 149/70 mmHg and she was tachycardic. There was no weight loss

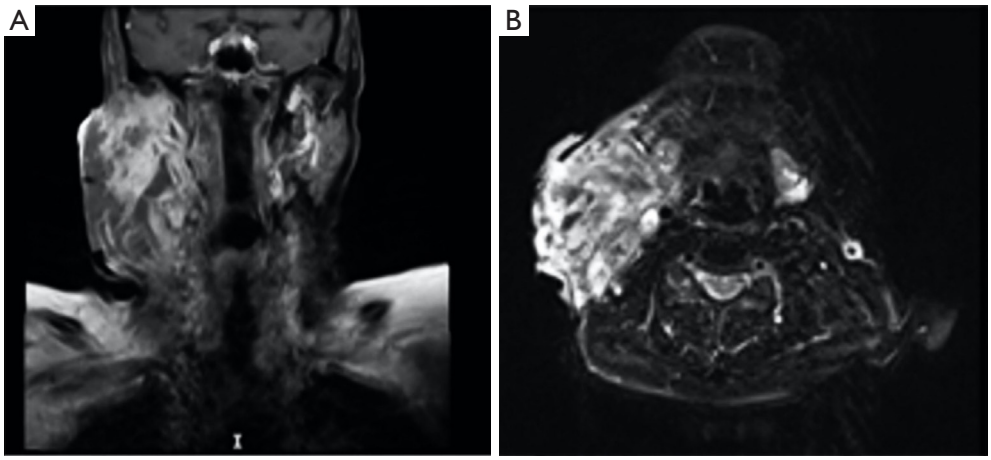

Figure 1 Coronal T2 weighted MRI (A) and axial T2 weight MRI (B) of head and neck. (A) It demonstrated large necrotising parotid abscess collection involving both superficial and deep lobes of the right parotid gland extending into the neck. MRI, magnetic resonance imaging.

reported. Initial blood analysis showed elevated C-reactive protein (CRP) 250 mg/L, elevated erythrocyte sedimentation rate (ESR) 105 mm/h, haemoglobin (Hb) 105 g/L, white cell count 9.4×10^9/L with mild neutrophilia 8.55×10^9/L and lymphopenia at 0.47×10^9/L, chronic thrombocytopenia at 114×10^9/L, impaired estimated glomerular filtration rate (eGFR) 44 mL/min, low albumin 21 g/L, elevated alkaline phosphatase 276 U/L, elevated alanine transaminase 85 U/L, elevated HbA1c 91 mmol/mol with hyperglycaemia (19.2 mmol/L). She was commenced on intravenous co-amoxiclav 1.2 g TDS and gentamicin 5 mg/kg OD, fluid resuscitation and variable rate insulin infusion for glucose control.

An urgent US characterised a heterogenous large mass below the angle of the right mandible with mixed echogenicity and several cystic spaces that is likely of parotid origin and likely infective aetiology. Fine needle aspiration cytology of the right parotid mass displayed acute inflammation with neutrophilia and presence of histiocytes in the absence of salivary gland acini. There was no sign of malignancy on the cytology. Blood cultures harboured no organisms.

The day after admission, the skin overlying her right parotid and neck broke down with necrosis and pus oozing from the aspiration site. She developed a right marginal mandibular nerve palsy within the first week of admission.

An MRI head and neck with gadolinium contrast (*Figure 1*) demonstrated a large right sided collection involving the right parotid gland but unable to exclude malignancy. Intraoperatively, exploration led to large quantities of pus and 'dishwater fluid' being expressed. The patient underwent urgent large-scale debridement of right superficial and deep parotid lobes, and level 2 neck region down to healthy tissue followed by washout with betadine-saline solution. The facial, hypoglossal, lingual, accessory nerves were not visualised intraoperatively due to the extensive inflammation and disease process. Radiological and intraoperative findings supported diagnosis of craniocervical necrotising fasciitis. One unit of packed red cell was transfused post-operatively due to low Hb at 77 g/L and 200 mL of blood loss. Intra-operative biopsies were taken that excluded malignancy but confirmed coagulase-negative staphylococcus epidermis and pus cultures yielded heavy growth of staphylococcus aureus with sensitivities to doxycycline, erythromycin, flucloxacillin and clindamycin. Microbiology advised intravenous clindamycin 1.2 g QDS and piperacillin-tazobactam 4.5 g TDS.

An autoimmune screen was undertaken which demonstrated mildly elevated rheumatoid factor 17 IU/mL (0–14 IU/mL) but negative for angiotensin converting enzyme, anti-myeloperoxidase and anti-proteinase 3 antibodies, Ro, La, Sm, ribonuclear protein (RNP), Jo-1, Scl-70, dsDNA, Centromere, Mi-2, Ku, Th/To, RNA polymerase III, PM-Scl, proliferating cell nuclear antigen (PCNA), and ribosomal P protein ruling out Heerfordt's syndrome in the absence of eye symptoms.

Computed tomography (CT) head, neck and chest (*Figure 2*) post-operatively revealed persistent soft tissue

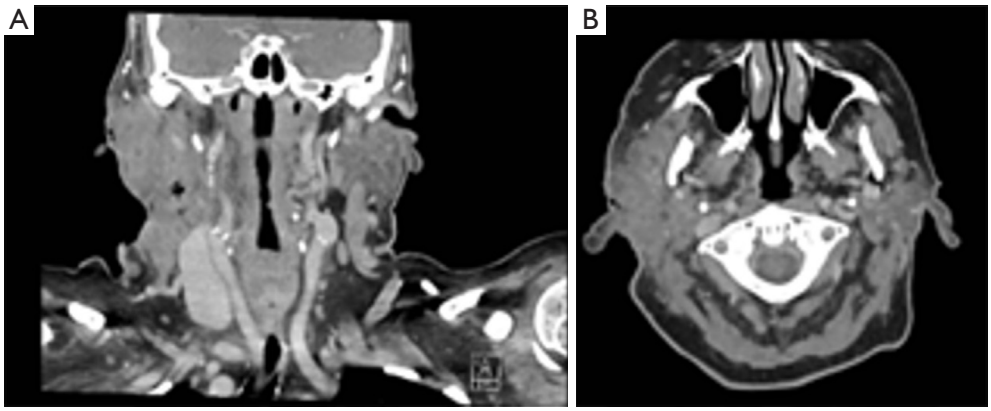

Figure 2 Coronal CT (A) and axial CT (B) head and neck. (A) It demonstrated persistent soft tissue with complex fluid gas containing collection within the parotid space and overlying fat with significant fat stranding of the right side of neck and around the right submandibular gland. There is extension of collection deep to the sternocleidomastoid with no extension into retropharyngeal or mediastinal spaces. The internal jugular veins were patent and there were no signs of mediastinitis. No pathological cervical or thoracic lymphadenopathy is noted. CT, computed tomography.

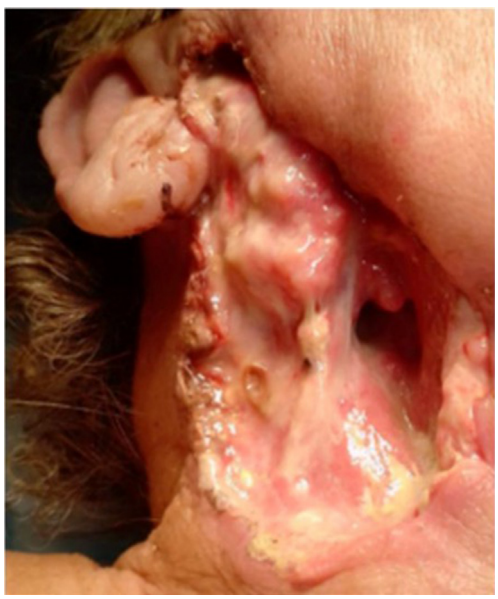

Figure 3 Medical illustration photograph of right neck demonstrated healing granulation tissue.

with complex fluid gas containing collection within the parotid space and overlying fat with extension deep to the sternocleidomastoid with no extension into retropharyngeal or mediastinal spaces. This reinforced the need for further surgical exploration and debridement.

A second surgical debridement was performed two days later until bleeding healthy tissue was visualised followed by betadine-saline washout. A second pus culture was sent. The cavity was packed with bismuth iodoform paraffin paste (BIPP) with jelonet dressing secured by prolene sutures to the skin. One unit blood transfusion of packed red cells was issued intra-operatively due to significant contact bleeding from tissues.

Following two debridement's and washout, her CRP improved to 22 mg/L. A third surgical exploration two days later showed no evidence of necrosis with granulating healthy tissue and no pus (*Figure 3*); the cavity was again cleansed with betadine-saline solution and re-packed with BIPP dressing. The absence of pus signified time to plan for definitive reconstruction of the wound defect.

Due to the suboptimal protection of the carotid sheath based on the third surgical exploration, initial plan was for a pectoralis major flap reconstruction via an inframammary approach with an overlying split thickness skin graft. However, fourth neck exploration exhibited significant improvement in appearance of neck defect with good tissue bulk of the remnant right sternocleidomastoid muscle. Hence, reconstruction consisted of coronal dissection of the right sternocleidomastoid muscle fibres utilising this as a flap to cover the right carotid sheath. A cervicofacial rotation flap from the right supraclavicular region was used to cover the right neck and face defect with 3'0 vicryl deep dermal suture closure with staples for skin closure. Due to considerable contact tissue bleeding, 1.2 litre blood loss was encountered warranting a unit of packed red cell transfusion and intravenous tranexamic acid intra-operatively and

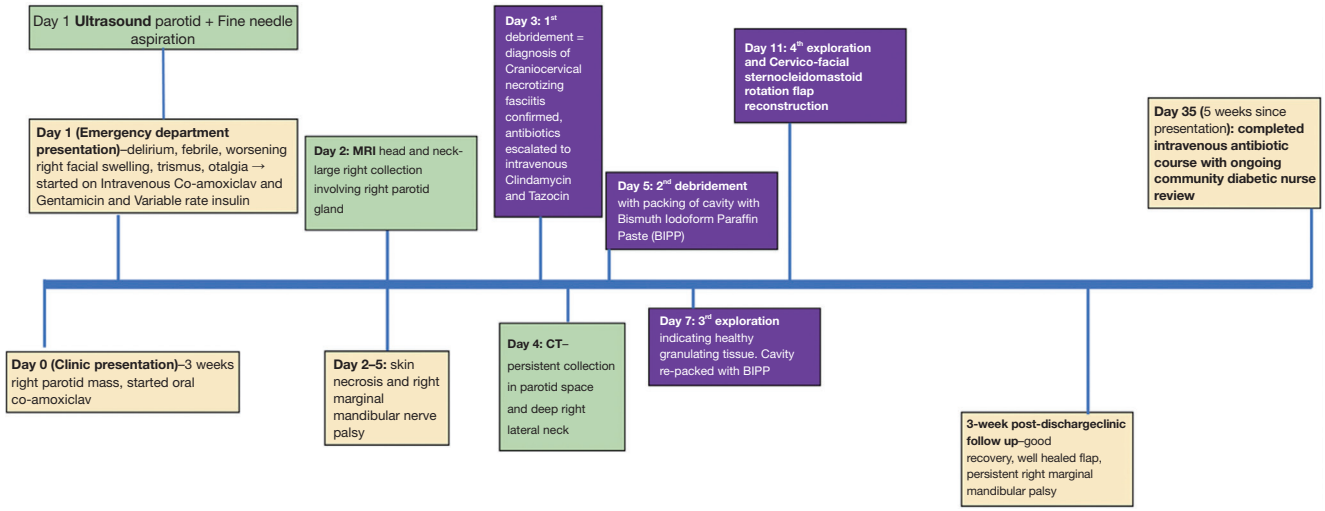

Figure 4 Timeline illustrating key events in the patient's journey. CT, computed tomography; MRI, magnetic resonance imaging.

a further unit of packed red cells post-operatively. A 10-french redivac drain was inserted due to the degree of blood loss which was removed 72 hours post-operatively. Post operative Hb was 73 mg/L increasing to 87 mg/L after 2 units of blood transfusion. Neck explorations, debridement and reconstruction were performed by two experienced head and neck surgeons.

The patient received a 5-week course of intravenous antibiotics, and electrolyte derangements namely hypomagnesemia and hypophosphatemia were corrected. Diabetic specialist nurses were requested to optimise glycaemic control and she was discharged on 44 units of long-acting insulin in addition to her oral hypoglycaemic medications with follow up in the community.

Topical chloramphenicol ointment was applied over the wound and staples removed after fourteen days. Overlying wound dressing were changed three times weekly with district nurse wound review.

She was subsequently followed up in a head and neck outpatient clinic 3 weeks later with a remarkably well healed reconstructed flap. Though the patient continued to manifest persistent right sided marginal mandibular palsy.

A timeline (*Figure 4*) has been included illustrating key events in the patient's journey.

Discussion

Parotid abscess leading to necrotising fasciitis remains a rare entity amongst craniocervical necrotising fasciitis, and this case described the second documented case in the literature. Our case originated from a bacterial parotid abscess where Staphylococcus aureus is typically the culprit organism (4). However, our cultures demonstrated polymicrobial involvement with *group A streptococcus* and *Staphylococcus epidermidis*. Immunodeficiency from poorly controlled diabetes mellitus and immunosenescence in our patient aided rapid evolution from a unilateral parotid abscess to life-threatening craniocervical necrotising fasciitis. Penetration of the parotid capsule and extension into the deep fascial planes of the neck led to the witnessed clinical picture (5).

Craniocervical necrotising fasciitis involving the parotid gland was described by Drake-Lee *et al.* (6) where an insulin dependent 24-year-old with preceding sore throat developed necrotising fasciitis overlying the parotid with pus being expressed from the parotid duct. However, the first case of a parotid abscess leading to necrotising fasciitis secondary to staphylococcus epidermidis was described by Marinoni (3) in a 67-year-old with a no history of diabetes or other co-morbidities; this case required one surgical debridement with subsequent adequate wound healing. Reported organisms of craniocervical necrotising fasciitis include *group A Streptococcus* (*Streptococcus pyogenes*, *Streptococcus viridans*), *Staphylococcus aureus*, *Peptostreptococcus species*, *Clostridia species*, *Enterobacteriaceae*, and *Haemophilus influenzae* (7,8) which is consistent with our case.

The mainstay treatment of craniocervical necrotising fasciitis involves aggressive surgical intervention with exploration of deep neck spaces and debridement of necrotic tissue. Prompt recognition and surgical debridement

is imperative in potential necrotising fasciitis especially in those with sepsis and features including erythema, fluctuance and crepitus. Case series of odontogenic necrotising fasciitis have demonstrated a 19.6% mortality if surgical debridement is performed within 24 hours compared to 50% mortality in subjects undergoing debridement after 24 hours (9).

However, in the case of parotid abscess necrotising fasciitis described by Marioni *et al.* (3), palpation revealed a parotid mass that was not fluctuant and displayed non-crepitant oedema similar to our described case. Choi *et al.* (10) and Yenigun *et al.* (11) described cases of submandibular sialadenitis causing necrotising fasciitis, initially signposted by an acute ill-defined erythematous swelling with documented crepitus highlighting that the absence of crepitus should not deter the clinician from suspecting potential necrotising fasciitis.

In this case report, we also present the use of BIPP as a good anti-septic and serves useful in packing large and small cavities in addition to providing haemostasis. BIPP has been exploited in ear, nose and throat (ENT) and maxillofacial surgery due to its versatility. However, attention should be made to documented risks of neurotoxicity due to bismuth being a heavy metal (12,13). Due to its absorption across damaged mucosa, bismuth can interfere with oxidative metabolism in the brain producing reduced cerebral blood flow and can potentially result in death (14). Bismuth subnitrate can also cause nitrite poisoning and iodoform toxicity (14). BIPP packing has not been commonly described in the literature for its management of necrotising fasciitis. Ross *et al.* (15) documented the use of BIPP packing in necrotising fasciitis affecting the posterior neck and a subsequent vacuum-assisted closure device. However, given the site of the necrotising fasciitis and the exposed vessels in our case, a vacuum-assisted closure (VAC) device could not be utilised and hence BIPP packing alone was used.

We advocate frequent reassessment of tissue defect and available tissue for reconstruction. The case avoided the need for pectoralis major flap which would have resulted in significant morbidity due to adequate time to promote wound healing with definitive reconstruction performed 2–3 weeks from first presentation. Multiple explorations and debridement are often needed to ensure there is no evidence of necrosis before deciding to close the wound defect following the reconstructive ladder (16).

Our case reinforces the importance of sending pre-operative group and save and ordering cross matched red blood cells in cases of necrotising fasciitis. This case had a pre-operative Hb of 105 g/L but still required 4 units of blood transfusion during her admission, largely due to the extensive contact bleeding during debridement.

Risk factors for necrotising fasciitis include diabetes mellitus, elderly, underlying malignancy, malnutrition, immunosuppression, cirrhosis, drug abuse, peripheral vascular disease, penetrating injuries and burns (17,18). Early prevention through strict control of predisposing systemic ailments including diabetes mellitus is needed. The treating clinician should investigate for contributing underlying medical conditions in cases of aggressive infections specifically disorders leading to an immunosuppressive state. A multidisciplinary approach was adopted with tissue viability nursing team, diabetic specialist nurses and microbiologists. Overall, a five-week course of intravenous antibiotics, regular wound monitoring and strict inpatient glucose control led to the patient being discharged.

With necrotising fasciitis involving the neck flaunting an overall mortality rate of 27% (11) urgent surgical debridement, appropriate antibiotic treatment, adequate treatment of systemic diseases, and early recognition of potential necrotising fasciitis are required to reduce patient morbidity and mortality. Moreover, other treatment options described in necrotising fasciitis include hyperbaric oxygen, intravenous immunoglobulin (IVIG) and sterile maggots (19). Hyperbaric oxygen has been utilised as a potential treatment due to its ability to inhibit anaerobic infection however there is no agreed consensus on its use (20-23); Tseros *et al.* performed a systematic review suggesting a significant reduction in mortality in those receiving hyperbaric oxygen as an adjunctive treatment in cervical necrotising fasciitis though higher quality randomised data is needed (23). IVIG and hydrocortisone immunosuppressive therapy were utilised by Koch *et al.* (24) due to its suppression of streptococcal superantigen induced T cell proliferation and cytokine production (25,26); though this has only been shown to be beneficial in necrotising fasciitis of staphylococcal or streptococcal origin (27).

Conclusions

We present a rare case of an erosive parotid abscess leading to necrotising fasciitis of the neck secondary to *group A Streptococcus* and *Staphylococcus epidermidis* mandating multiple surgical debridement. Cervicofacial rotation sternocleidomastoid flap reconstruction is a valid reconstructive modality in lateral neck defects following

necrotising fasciitis, producing aesthetically pleasing results. This is also the first case to use this method of reconstruction for necrotising fasciitis secondary to a parotid abscess.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at https://gs.amegroups.com/article/view/10.21037/gs-23-365/rc

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Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://gs.amegroups.com/article/view/10.21037/gs-23-365/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy and integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the consent is available for review by the editorial office of this journal.

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