

Severe Nasofacial Necrosis From *Capnocytophaga canimorsus*: A Case Report of Disease and Reconstruction

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1 | Introduction

Capnocytophaga canimorsus is a Gram-negative bacilli bacterial pathogen found on the gingival flora of various canine and feline species [1]. The most common clinical presentation of patients who are infected with *Capnocytophaga canimorsus* is sepsis [2]. A subset of these patients can develop purpura fulminans (PF), a syndrome that leads to rapidly evolving necrosis of tissue secondary to microvascular thrombosis [3]. The management of these patients is complex, and often requires surgical debridement or amputation of necrotic tissue and limbs. A review from 2019 found that an average of 4 surgical procedures is used to treat each PF patient [4]. For those who suffer necrosis of the face, nose, and ears their care is further complicated by the extensive scarring and tissue loss that is caused by disseminated intravascular coagulation and PF [5]. Reconstruction of these areas can be difficult due to the complex aesthetic and functional requirements of these facial subunits.

Here we present a case of a 60-year-old immunocompetent man who presented with an insidious *Capnocytophaga canimorsus* infection later deduced to be due to a cat lick on a lower limb wound. He subsequently developed PF and was left with a significant facial defect of his nose, septum, and upper lip. We undertook the debridement and subsequent reconstruction of this defect to improve his aesthetic and functional outcomes.

2 | Case Report

A 60-year-old immunocompetent man presented to the local emergency room with 2 weeks of mottled legs and cold feet with

weak pulses. The patient's medical history was significant for hypertension and dyslipidemia, he had no history of hyposplenemia. He consumed 5–6 units of alcohol consumed per week and had a pack per day history of smoking. His bloodwork showed marked thrombocytopenia and anemia raising concern for thrombotic thrombocytopenic purpura. He was transferred urgently to a tertiary care hospital for plasma exchange. He was also started on piperacillin-tazobactam and vancomycin for broad spectrum antibiotic coverage. After multiple treatments with plasma exchange without improvement, in addition to an elevated prothrombin time (PT) and partial thromboplastin time (PTT), it was felt that disseminated intravascular coagulopathy and PF were more likely. On day 9 of his admission, a source of infection was identified when *Capnocytophaga canimorsus* was isolated in blood cultures after being incubated in chocolate agar for several days with 5% CO₂. The patient developed significant necrosis of the lower limbs, upper limb digits, and face secondary to PF. He remained intubated and in the intensive care unit for a total of 13 days until he was medically stabilized. The patient underwent bilateral below-the-knee amputations and subsequently, facial debridement by the Otolaryngology team. The entire nose distal to the nasal bones was necrosed (Figure 1). The cartilaginous septum was also necrotic and devascularized. The right side of the upper lip was also involved. A large defect in the center of the face was left behind after the initial debridement (Figure 2).

Reconstruction of the defect began approximately 2 months after his initial debridement. To begin the reconstruction, septal hinge flaps were raised bilaterally to form the lining. The structure of the nasal dorsum and columella was recreated with a large strip of posterior septum as well as bone from the

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FIGURE 1 | Patient's necrotic nasal tip prior to debridement.



FIGURE 2 | Large facial defect following debridement.

maxillary crest. These were fixed with sutures superiorly to the nasal bones, inferiorly to the maxillary spine, and anteriorly, together to re-create the nasal tip support (Figure 3). Subsequently, cartilage from the conchal bowl of both ears was used to construct the new alar. Finally, coverage was obtained using a right-sided paramedian forehead flap.

A second stage was performed 5 weeks later wherein the vascular pedicle was divided and the forehead flap further refined to provide a more natural contour to the nose. The upper lip scarring was then also addressed with a crescent alar advancement flap. Following the second stage of the procedure, there was satisfactory nasal tip projection, and the patient reported good functionality (Figures 4 and 5).

3 | Discussion

Capnocytophaga canimorsus was initially identified in 1976 [6] and has become an important organism on the differential for sepsis, especially when history identifies recent animal exposure. It has been recognized as the second most common organism involved in infections after dog bites, second only to *Pasteurella multocida* [2]. Since it was first identified, the total estimated number of infections by 2014 was 484 [2]. It possesses several traits that provide protection from the innate immune system. It is a catalase-positive organism, allowing it to survive phagocytosis [2, 7]. It may also elicit lower quantities of proinflammatory cytokines, which allow it to evade surveillance by the innate immune system [2]. Common risk factors for

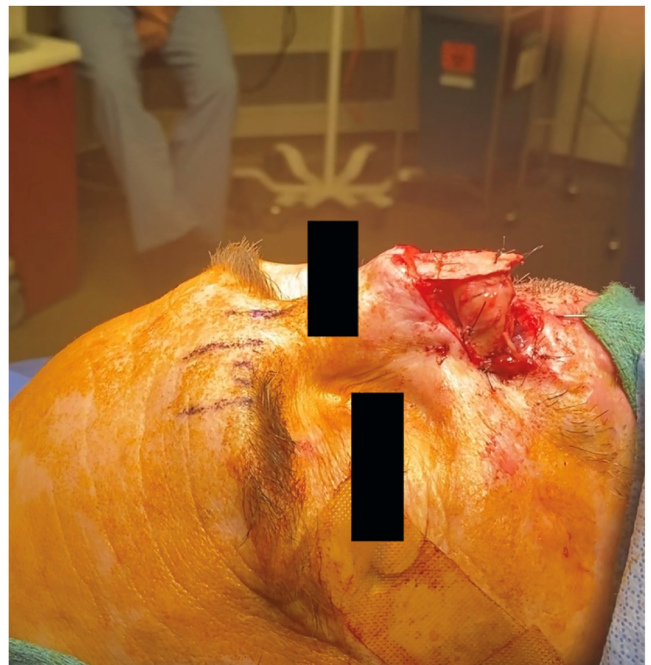


FIGURE 3 | Bony reconstruction of nasal dorsum.

infection include male sex, age greater than 50, previous splenectomy and alcoholism [2]. It is thought that because splenic macrophages can provide some protection against *Capnocytophaga canimorsus*, patients with co-morbidities that result in

hyposplenias/asplenia such as alcoholism, previous splenectomy or sickle cell disease are at higher risk for infection [2, 8].

While PF is a rare clinical entity, it has been described secondary to more common pathogens in the literature. Djurdjevic et al. described a case of an immunocompetent male developing PF as a result of *Streptococcus pneumoniae* infection [9]. Of note, their patient had not received the recommended pneumococcal vaccinations. Their case was not marked by severe ischemia requiring debridement, the patient ultimately improved with intensive medical therapy.

Urushidate et al. described a nose and upper lip reconstruction for a patient with PF secondary to *Klebsiella pneumoniae* infection [5]. Their patient had a significant defect of the external nose and central upper lip. They employed bilateral nasolabial orbicularis oris myocutaneous flaps to reconstruct the upper lip. The columella, ala and septum were reconstructed using a forearm free flap and the external coverage of the nose was achieved with a paramedian forehead flap. The use of a forearm free flap versus the reconstruction in our case is likely due to surgeon comfort and patient specific anatomical differences. Their patient, like ours has a satisfactory degree of nasal aeration.

A systematic review performed in 2019 [4] showed that *Capnocytophaga canimorsus* was the third most common organism causing PF, behind *Neisseria meningitidis* and *Streptococcus pneumoniae*. It is thought that inflammatory cytokines result in the consumption of key anticoagulants such as protein C, protein S and antithrombin III during the pathogenesis of PF [3]. Given its known pro-inflammatory properties and capacity for high grade bacteremia [2], this appears consistent with the pathophysiology of *Capnocytophaga canimorsus*. The mortality rate of

PF in adults has been reported as 18% but is likely even greater [4]. The most common procedure performed is below-the-knee amputation which is a significant source of morbidity for



FIGURE 5 | More than 1 year post reconstruction.



FIGURE 4 | After stage 2 of the reconstruction. All incisions had healed well, and the graft had taken well.

patients [4]. Given its devastating sequelae, PF requires urgent recognition and in the specific case of *Capnocytophaga canimorsus*, special attention should be paid to older male patients with hyposplenia.

4 | Conclusion

In this case, our patient presented with a 2-week history of mottled legs, cold feet and weak pulses. The initial diagnosis was thrombotic thrombocytopenic purpura, later found to be PF secondary to bacterial infection. It was not until day 9 of the patient's ICU admission that the culprit organism was identified as *Capnocytophaga canimorsus* from a cat licking a wound. This highlights the importance of a thorough social history, especially from collateral sources when the patient presents with an altered level of consciousness.

The resulting purpura fulminans caused massive nasofacial necrosis. Here we describe the nasal reconstruction with a two staged paramedian forehead flap, bilateral septal hinge flaps, bilateral conchal grafts, and bony reconstruction. To our knowledge this is the first described nasal reconstruction in a patient with PF secondary to a *Capnocytophaga canimorsus* infection. The aesthetic and functional outcomes were satisfactory to the patient.

Author Contributions

Armon Hadian: conceptualization (supporting), data curation (lead), investigation (lead), writing – original draft (lead), writing – review and editing (equal). **Jason Azzi:** writing – review and editing (equal), supervision (supporting). **Benjamin Taylor:** conceptualization (lead), methodology (lead), supervision (lead), writing – review and editing (equal).

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Ethics Statement

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. Institutional review board approval was not required in accordance with institutional policy for single case reports.

Conflicts of Interest

The authors declare no conflicts of interest.

Data Availability Statement

Data sharing is not applicable to this article as no datasets were generated or analyzed during the current study.

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