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Cervicofacial necrotizing fasciitis: A rare disease with a high mortality requiring early debridement for survival☆

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ABSTRACT

Purpose: To review the clinical experience, management and outcome of cervicofacial necrotizing fasciitis (CNF) in patients treated in our institution.
Methods: A retrospective review of patients with CNF from two large health care institutions completed over a 10-year period.
Results: Five patients with complete data were identified. CNF was polymicrobial in 4 and monomicrobial in one patient and occurred as a result of odontogenic infection in 3, trauma in 1, and was idiopathic in one patient. All patients were treated with extensive debridement, broad spectrum antibiotics, and reconstruction with flaps. There was one death.
Conclusions: Early diagnosis and rapid aggressive debridement are key elements for reducing mortality and optimizing the cosmetic and functional outcome in patients with CNF.

Fasceítis necrosante cervicofacial: una infección severa que requiere tratamiento quirúrgico temprano

Palabras clave:
Fasceítis necrosante
Cervicofacial
Infección
Mortalidad
Desbridamiento

RESUMEN

Propósito: Revisar la experiencia clínica, el manejo quirúrgico y los resultados del tratamiento de pacientes con fasceítis necrosante cervicofacial (FNC) en nuestras instituciones.
Métodos: Un estudio retrospectivo de pacientes con FNC en un periodo de 10 años en 2 instituciones académicas.
Resultados: Cinco pacientes con datos completos (clínicos, imágenes, cultivos microbiológicos, tratamiento y seguimiento) fueron identificados. La FNC resultó de una infección

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polimicrobiana en 4 pacientes y monomicrobiana en un paciente. La etiología de FNC fue odontogénica en 3 pacientes, postraumatismo en un paciente e idiopática en un paciente. Todos los pacientes fueron tratados con tratamiento quirúrgico (desbridamiento) agresivo temprano, antibióticos de amplio espectro y reconstrucción con diferentes tipos de colgajos. Se registró una mortalidad.

Conclusiones: El diagnóstico temprano y un tratamiento quirúrgico agresivo son elementos clave en reducir la mortalidad y optimizar los resultados funcionales y cosméticos en los pacientes con FNC.

Introduction

Necrotizing fasciitis (NF) is an uncommon soft tissue infection that results in the rapid and progressive necrosis of the connective tissue and muscle fascia. In more advanced stages, it involves the skin and muscle, and the mortality rate is high. Cervicofacial NF (CNF) is a rare condition that constitutes between 2.6% and 5% of all the cases of FN.[1,2] It is not common for any one center to gather significant clinical experience with CNF.

There are many historical accounts of and references to NF that date back to Hippocrates (500 BC), who reported diffuse lesions that would not heal. Pouteau and Gillespie, in 1783 and 1785, respectively, described malignant, gangrenous ulcers.[3] In 1871, Joseph Jones, who had been a Confederate surgeon in the United States Civil War, was the first to offer a precise description of NF, which he referred to as "hospital gangrene".[4] Meleney reported 20 cases of hemolytic streptococcal gangrene in 1924[5] and, in 1952, Wilson used the term necrotizing fasciitis for the first time.[6] The information on the presentation, management and results of the treatment of patients with NF in the head and neck region is limited.

Necrotizing fasciitis is classified according to 3 different types, depending on the microbiological findings. Type 1 is a polymicrobial infection produced by a combination of anaerobic and aerobic bacteria, whereas type 2 is a monomicrobial infection due mainly to group A β-hemolytic streptococcus and, less frequently, to other streptococci and staphylococci; type 3 is a monomicrobial infection caused by a marine *Vibrio* species.[7] The clinical signs of NF include swelling, erythema, pain, skin blistering and crepitus.[8] The purpose of this article is to review our experience and report the demographic data, treatment microbiological findings and reconstruction carried out in 5 patients with CNF.

Materials and methods

We carried out a retrospective review of the cases of CNF treated between December 2002 and December 2012 in the oral and maxillofacial surgery units of 2 centers (MetroHealth Medical Center [MHC] in Cleveland, Ohio, United States, and the Instituto de Medicina Tropical Alexander von Humboldt [IMT] of the Universidad Peruana Cayetano Heredia in Lima, Peru). A total of 590 moderate and severe maxillofacial infections were identified, 7 of which (1.19%) were diagnosed as CNF. At the MHC, we identified 332 infections with 4 cases (1.2%) of CNF, and the IMC reported 258 cases, including 3 (1.16%) of CNF. Moderate maxillofacial infections were considered to be those that involved one or more of the following fascial spaces: submandibular, submental, sublingual, pterygomandibular, superficial temporal and deep temporal. Severe maxillofacial infections were defined as any infection that required in-hospital management and/or threatened to compromise the lateral pharyngeal, retropharyngeal, pretracheal and danger spaces, as well as mediastinal and intracranial infections.[9]

Results

Seven patients with CNF were identified on the basis of the data provided by the centers in which the present study was conducted. Two patients were excluded because of insufficient clinical data and a lack of follow-up. All of the patients were adults of the male sex, with ages between 30 and 61 years. Three patients were black, one was mestizo and another was white. All of them presented with severe pain, erythema, swelling, necrosis and subcutaneous gas. The clinical photographs can be seen in Figs. 1 and 2.

The demographic and bacteriological data, location and type of reconstruction carried out are summarized in Table 1. Two patients had systemic comorbidities: patient no. 3 had morbid obesity and patient no. 4 had type 1 diabetes mellitus and hypertension. The only death in our case series was that of a patient involved in a traffic accident as a pedestrian, who had multiple injuries (facial and rib fractures) and an in-hospital course complicated by prolonged ventilatory support and multiple respiratory tract infections.

Figs. 3 and 4 show the defects in the 5 reported patients.

Patient no. 1 underwent reconstruction with a skin graft. In patient no. 2, an advancement flap was created. Patient no. 3 underwent supraclavicular flap creation. The defect in patient no. 4 was closed by means of an advancement flap. Patient no. 5 underwent repair with a submental artery island flap which, unfortunately, failed; the defect subsequently healed by second intention, leaving a conspicuous scar; the patient refused to undergo revision of the scar.

Table 1 – Demographic and clinical characteristics of the patients and treatment of cervicofacial necrotizing fasciitis in each.

Case/age/G/R	Site	Bacteriology	Initial presentation	Initial white blood cell count	Antibiotic therapy	Reconstruction	Outcome
1/61/M/MR	Submandibular, right lateral neck	*Proteus mirabilis*	Swelling, pain, erythema and skin discoloration (black), skin blistering, subcutaneous gas, pus	24,600	Ceftazidime, ciprofloxacin, clindamycin	Partial-thickness skin graft	Survival
2/30/M/B	Submental	*Prevotella* spp., *Clostridium difficile*, *Peptostreptococcus micros*, *Streptococcus milleri*, *Streptococcus intermedius*, Coagulase-negative *Staphylococcus* spp., *Candida albicans*	Swelling, pain, erythema and skin discoloration (grayish), skin blistering, subcutaneous gas, pus	25,400	Vancomycin, piperacillin/tazobactam, ertapenem	Advancement flap	Survival
3/60/M/W	Sub mental, chin	*Streptococcus intermedius*, *Prevotella corporis*, *Bacteroides fragilis*, *Staphylococcus* spp., *Gemella morbillorum*, *Streptococcus constellatus*, *Prevotella melaninogenica*, *Prevotella oris*	Swelling, pain, erythema and skin discoloration (black), skin blistering, subcutaneous gas, pus	10,900	Ampicillin/sulbactam, vancomycin	Supraclavicular flap	Survival
4/56/M/B	Submandibular, right lateral neck	*Prevotella buccae*, *Prevotella intermedia*, *Peptostreptococcus asaccharolyticus*, *Bacillus* species, *Streptococcus constellatus*, Saprophytic *Neisseria* species, *Eikenella* species, α-hemolytic streptococcus, not enterococcus or pneumococcus, anaerobic Gram + cocci	Swelling, pain, fever, erythema, pus	13,700	Ampicillin/sulbactam, vancomycin, piperacillin/tazobactam	Advancement flap	Death
5/39/M/B	Right cheek	*Pseudomonas aeruginosa*, *Streptococcus gordonii*, *Klebsiella pneumoniae*, *Bacteroides capillosus*	Swelling, pain, pus, skin discoloration	25,400	Ampicillin/sulbactam, vancomycin	Submental artery island flap	Survival

B: black; G: gender; M: male; MR: mixed race; R: race; W: white.

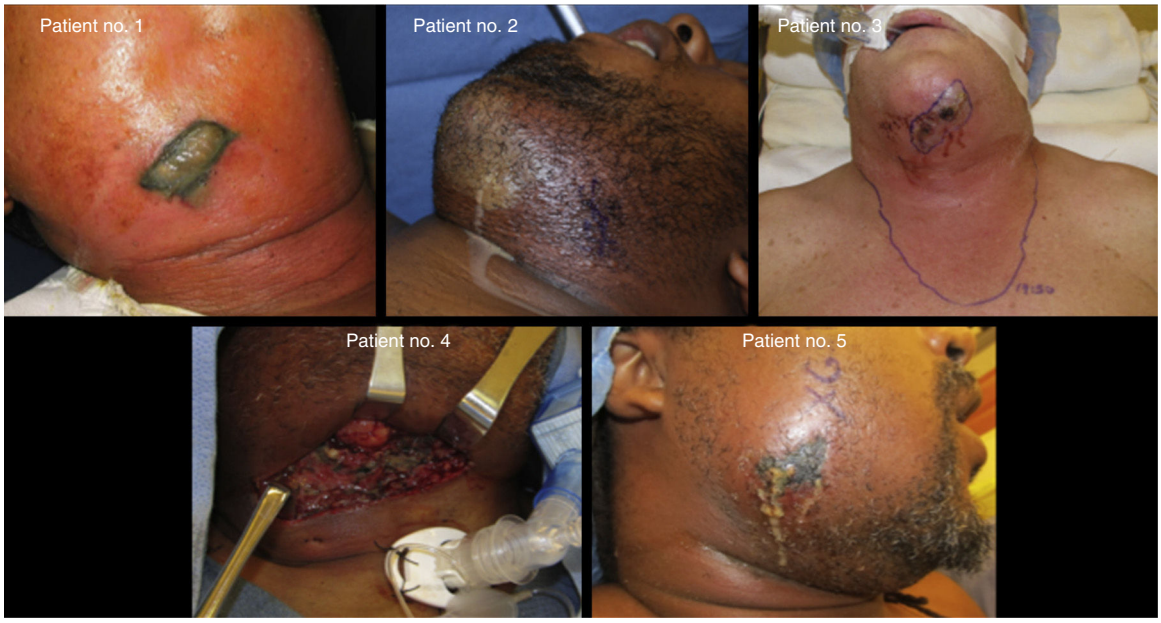


Fig. 1 – The initial clinical characteristics of our patients with cervicofacial necrotizing fasciitis.

Discussion

Necrotizing fasciitis is a serious infection associated with a mortality rate that can reach approximately 30%.[10] The most important factors in the reduction of the mortality are early diagnosis and early aggressive surgical treatment. The key findings that lead to a correct diagnosis are a combination of the clinical presentation (pain, which generally is severe and disproportionate with respect to the physical findings, erythema, swelling and induration, tenderness to touch beyond the region of erythema, subcutaneous crepitus, skin blistering and skin discoloration).[11,12] The radiographic signs include gas in soft tissues, very often associated with fluid collection in the connective tissue and cervical fascia. Reactive lymphadenopathy can also be observed.[13,14] The microbiological studies performed in our patients revealed polymicrobial infections in 4 and monomicrobial infection in 1 (patient no. 1), in whom only *Proteus mirabilis* was isolated. Streptococci and staphylococci were the predominant aerobic bacteria. The majority of the anaerobic bacteria isolated were peptostreptococci and *Prevotella* species.

McHenry et al. reported 65 patients with necrotizing soft tissue infections and only 2 (3%) were found in head and neck, whereas 37 (57%) were located in trunk and 26 (40%) in extremities. The mortality rate was 29%; the 2 patients with CNF survived. In their series, McHenry et al. found a number of factors that had a significant impact on the outcome, including time from hospital admission to operation, percentage of body surface area involved, acidosis, peripheral vascular disease, the number of systemic diseases and age. When the factors were correlated with mortality, the only statistically significant factor was a prolonged time between admission and surgical treatment.[13] This finding has also been reported

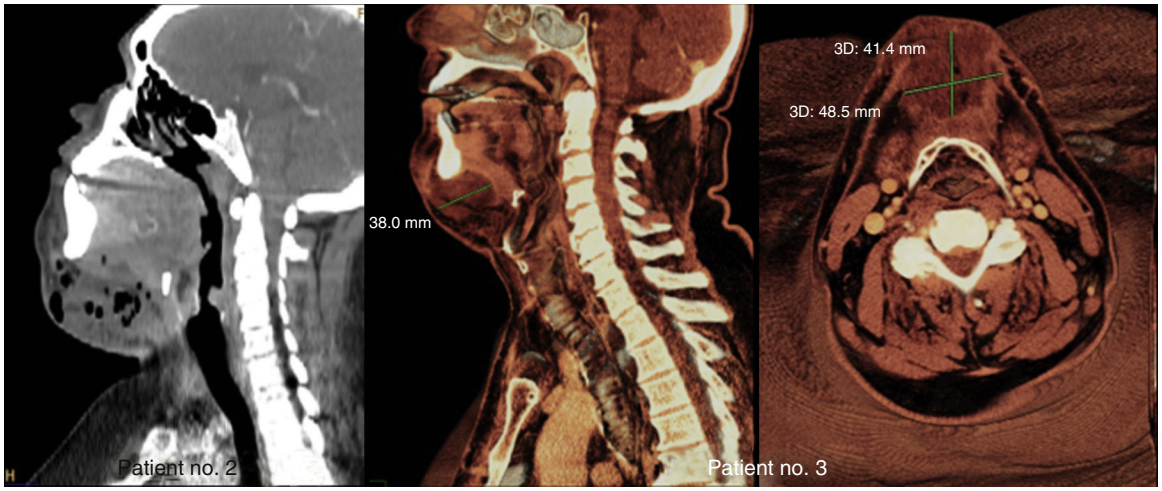


Fig. 2 – Examples of computed tomography slices and 3D reconstruction.

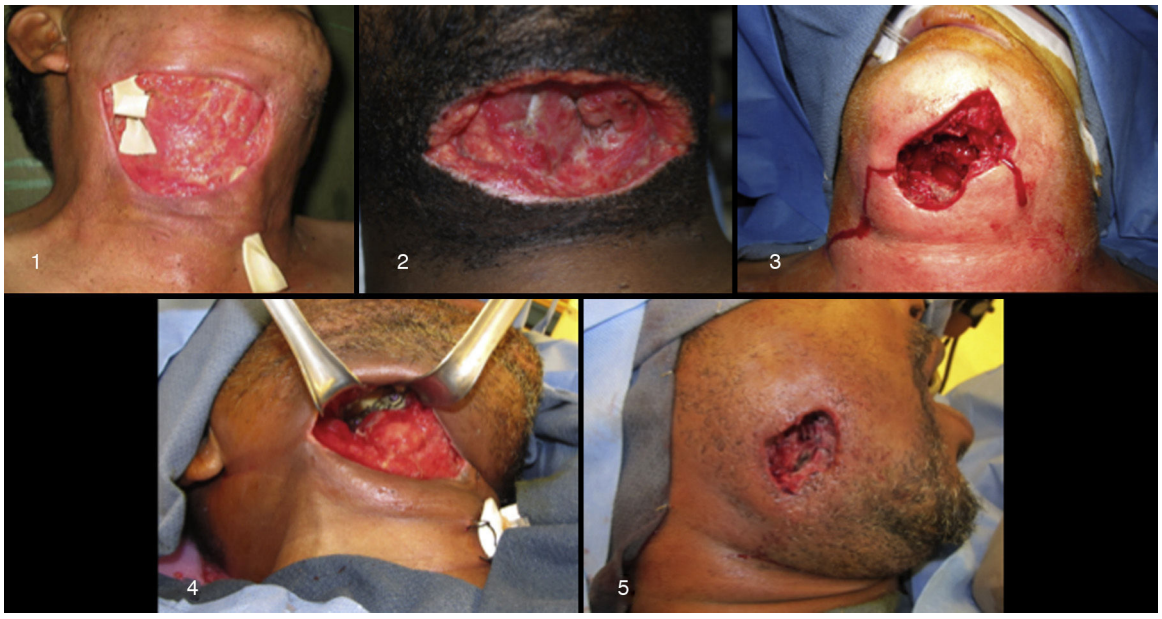
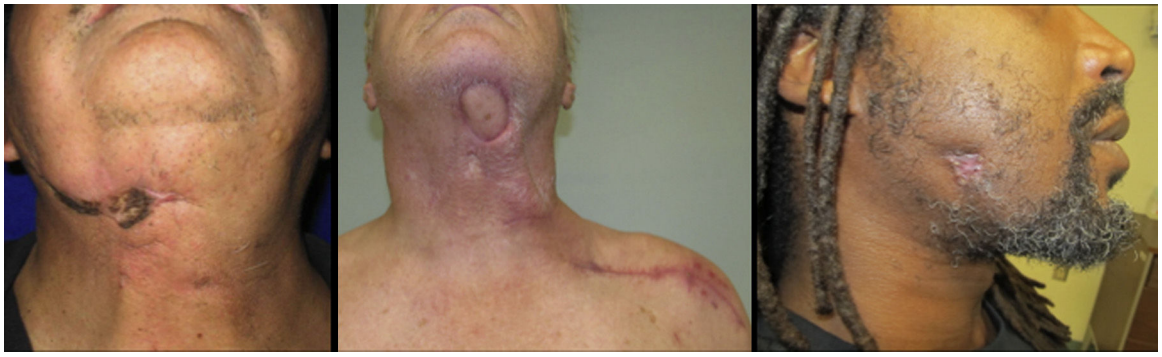


Fig. 3 – Different defects after aggressive debridement.



Patient no. 1. Twenty weeks after undergoing skin graft.

Patient no. 3. Six weeks after undergoing supraclavicular flap..

Patient no. 5. Scar left 8 weeks after healing by second intention.

Fig. 4 – Examples of the outcome of surgical treatment in patients 1, 3 and 5.

by other authors.[15–18] In our study, the rate of mortality was 20%, which is similar to or slightly lower than those observed by other authors.[10,19–22] In patient no. 4 of our series, there was a delay in the identification of a postoperative infection. The surgical wound was edematous, but showed no signs of skin necrosis. This delay in the identification of the infection may have contributed to his death, although it most probably was multifactorial, related to multiple comorbidities, injuries, hospital-acquired pneumonia and respiratory failure.

Owing to the fact that early aggressive surgical treatment is the key factor for survival, CNF should be suspected in patients with erythema and induration of the skin, severe pain that is disproportionate with respect to the physical findings, tenderness to the touch in areas free of erythema, rapid progression and hemodynamic instability, although the most important clinical signs of this disease are crepitus, blistering and skin necrosis.

Cervicofacial NF is commonly secondary to an odontogenic infection, and is less often found to be secondary to traumatism. On rare occasions, no etiological factor or obvious portal of entry for the bacteria is identified, as occurred in patient no. 1 of our series. This spontaneous presentation of NF has been reported in up to 20% of the patients.[18]

Conclusion

Cervicofacial NF is a rare infection associated with high rates of morbidity and mortality. Early diagnosis and rapid and aggressive surgical treatment can reduce these rates.

Ethical disclosures

Protection of human and animal subjects. The authors declare that the procedures followed were in accordance with the regulations of the relevant clinical research ethics committee and with those of the Code of Ethics of the World Medical Association (Declaration of Helsinki).

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors have obtained the written informed consent of the patients or subjects mentioned in the article. The corresponding author is in possession of this document.

Conflicts of interest

The authors declare they have no conflicts of interest.

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