CASE REPORT

Necrotizing fasciitis in oro-maxillo-facial area after radiotherapy for squamous cell carcinoma of the soft palate

ALINA ORMENIŞAN¹, SILVIU HORIA MORARIU², OVIDIU SIMION COTOI³, MIHAI DORIN VARTOLOMEI⁴, RADU IONŢ GRIGORAŞ¹, SIMONA LILIANA MOCAN⁵, MIRCEA SUCKU⁶

¹Department of Oral and Maxillofacial Surgery, University of Medicine and Pharmacy of Tirgu Mures, Romania
²Department of Dermatology, University of Medicine and Pharmacy of Tirgu Mures, Romania
³Department of Pathophysiology, University of Medicine and Pharmacy of Tirgu Mures, Romania
⁴Department of Cell and Molecular Biology, University of Medicine and Pharmacy of Tirgu Mures, Romania
⁵Department of Pathology, Emergency County Hospital, Tirgu Mures, Romania
⁶Department of Oral Rehabilitation and Occlusology, University of Medicine and Pharmacy of Tirgu Mures, Romania

Abstract

The fascia’s and subcutaneous adipose tissue’s impairment by mono or polymicrobial infection, which also can involve the skin and the muscles, is rarely seen in oro-maxillo-facial area. The present case report is presenting a case of necrotizing fasciitis in a patient who had a history of an invasive squamous cell carcinoma of the soft palate, with surgical treatment and with radiotherapy. He was admitted in our Clinic with malaise and subsequently developed a toxico-septic shock. Clinical symptoms, serological and bacteriological analysis and histopathological examination confirmed the diagnosis of necrotizing fasciitis (NF). The patient subsequently underwent a series of surgical reconstruction and aesthetic treatments because of the complications that had arisen in the meantime. Postoperative evolution was favorable towards complete closure of the defect. The prognosis of this disease is generally reserved, the favorable evolution depending on the possibility of wound sterilization and the surgery is required despite its mutilating effect.

Keywords: cervical, facial, necrotizing fasciitis, infection.

Introduction

Necrotizing fasciitis (NF) is a rapid and progressive infection of the fascia and subcutaneous adipose tissue with secondary involvement of the skin and muscles, accompanied by severe general toxic manifestation [1]. It is produced by the combination of aerobic and anaerobic bacteria, predominantly Gram-negative. The factors that favor the occurrence of such pathology are the body immunodeficiency, diabetes, neoplasm, alcoholism, organ transplantation, radiotherapy and vascular insufficiency. NF type I, polymicrobial, is less aggressive compared to types II and III, monomicrobial and aggressive [2, 3]. NF type III is rapidly progressive, fulminate and can be produced by marine microorganisms that can supra-infect punctiform lesions.

NF frequently appears in the abdominal region or lower limbs, after trauma (31.4%) or surgery (4.3%) [4]. The incidence of NF in cervicofacial region is less than 3% [5–10]. Cervical necrotizing fasciitis (CNF) is more frequent in men in 4:1 ratio with average age between 40–55 years [1]. The cervical infection mortality rate is estimated up to 40% [11, 12]. NF due to radiotherapy is very rare in cervical facial area and often the cause is cervical facial neoplasia [13, 14].

In our particular case, the cervical NF developed after radiotherapy for squamous cell carcinoma poorly differentiated with oral cavity origins.

Case report

In this article, we present the case of a 52-year-old patient, Caucasian, which was transferred to the emergency room at the Oral and Maxillofacial (OMF) Clinic, with signs of right facial hemiparesis and toxico-septic malaise, associated with painful tumefaction and areas of necrosis in the right maseterin and latero-cervical regions.

The patient declared that, two weeks prior, he had noticed the appearance of a painful swelling in the right maseterin region subsequent of an odontogenic pain, which was associated with chewing disorders and progressive alteration of health, during the last four days. He had addressed an emergency surgical clinic where he was admitted and diagnosed with cellulitis, associated with chronic alcohol abuse. Seven days after the onset of the lesion, incision was performed, alongside drug treatment with Amoxicillin 4×250 mg/day, for seven days, Diclofenac sodium one tablet/day and Tramadol hydrochloride 3×1 tablets/day. The evolution was not favorable, showing fast alteration of the general health in the last three days. Locally, grey spots followed by necrosis had appeared and the decision was made to transfer the patient in the OMF Clinic. Physical examination of the cephalic extremity showed, on the right maseterin and right latero-cervical regions, an erythematous edematous plaque with poor delimitation, indurated, with hemorrhagic bullous lesions and grey spots, with a 7×8 cm wide necrotic area that produced foul smell. Intense pain, both spontaneous...
and palpatory, had been limiting movement in the temporomandibular joint. Profound palpatory examination revealed the presence of crepitus.

The physical examination determined the presence of icterus, cachexia, swollen lymph nodes in the submandibular, retro-auricular and latero-cervical areas, hepatomegaly, oliguria, fever of 38.6°C with chills, dyspnea and agitation.

The personal history revealed an invasive squamous cell carcinoma of the soft palate, poorly differentiated, operated and irradiated three years prior, with six weeks of daily radiotherapy (66 Gray). Histology findings showed in Figure 1.

![Figure 1](image1.jpg)

Figure 1 – Placards of malignant tumor cell, squamous-cell type, nuclear and cytoplasmic atypia and atypical mitosis, without keratinization. HE staining, ×100.

Laboratory data documented ALT (alanine aminotransferase) 57 U/L, AST (aspartate aminotransferase) 75 U/L, GGT (gamma-glutamyl transferase) 360 U/L, CRP (C-reactive protein) 15 mg/dL and CK (creatinine kinase) 750 U/L, glycemia 118 mg/dL. Bacteriologic examination was considered irrelevant because of the prior initial local and general antibacterial treatment.

The clinical diagnosis was established: NF, chronic alcoholic hepatitis, irradiated squamous cell carcinoma of soft palate. Subsequently, general treatment was induced: Ceftriaxone 2×1 g/day i.v., Gentamicin 2×80 mg/day i.v., Metronidazole 4×250 mg/day and hydroelectrolytic balancing.

In an immediate emergency, the necrotic tissues were widely debrided, all the damaged lodges were opened and the irrigations with H₂O₂, Metronidazole, Betadine and saline were done through the drainage tubes several times daily. Tissue fragments were sent for histology examination. Microscopic examination revealed a necrotic suppurative inflammatory process, extending to the subcutaneous fat and fascia underlying striated muscles. It was noted extensive soft tissue necrosis with imprecisely defined edges, swelling and numerous inflammatory cells: segmented neutrophils, macrophages, and lymphocytes. Among inflammatory cells were observed a number of cocci-type bacteria. Blood vessels showed an aspect of septic vasculitis, with thickening wall and the presence of inflammatory cells in the thickness. In some, thrombi were present (Figures 2 and 3).

After serial surgical interventions, at one month after admission, there was a secondary granulation with periosteum removal from ascending mandibular ramus. Postoperative, persistent trismus and facial nerve paralysis were installed. At the same time, the patient had received specific antibiotic therapy. Two months after the intervention, the area of the ascending mandibular ramus remained denuded, at histology was observed a foreign body granuloma formation around the suture threads, with granulation tissue in conjunctive organization phase (Figure 4).

The defect was closed by creating a pedicle submental flap, which was moved toward the ascending mandibular ramus. The cortical was trepanned to facilitate initiation of local vascularization. Submental flap is an axial flap based on submental artery, a constant facial artery branch, which follows a contact trajectory with the deep lobe of submandibular gland. The submental artery has an anastomosis with the contralateral submental artery. Veins drain into the facial vein and connect with the external jugular system by intraparotid communicating veins.

At six months after surgery, a good integration of the submental flap was observed, but with necrosis of the ascending mandibular ramus. Segmental resection of the mandible was used and the defect was closed with a rotated flap from the temporal region. The postsurgical evolution was favorable with entire closure of the defect. After a one-year follow-up, the patient is recovering very well with no recurrence of the squamous cell carcinoma (Figure 5).

![Figure 2](image2.jpg)

Figure 2 – Inflammatory infiltrate with neutrophils and necrosis. HE staining: (a) ×20; (b) ×40.
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Figure 3 – Inflammatory infiltrate with neutrophils and necrosis. HE staining: (a) ×40; (b) ×100.

Figure 4 – Clinical aspects before surgery.

Figure 5 – Clinical aspects after surgery.

**Discussion**

NF from the neck and head area are frequently related with primary odontogenic or post-extraction infections [1, 5]. Tonsillar infection, sialadenitis, those from ENT (ear, nose and throat) area or dermatological are rarely correlated with NF [15].

NF may appear following trauma of surgical interventions, sometimes even in the absence of trauma [16]. In the presented case, because of the particular evolution, with rapid aggravation of the general health, alongside the appearance post-op of the hemorrhagic bullae and grey stains followed by necrosis, it can be stated that the surgical intervention, especially the incision of cellulitis or of a dental abscess, associated with weakened immune system, can represent the determining factor for NF.

Complete blood count (CBC) shows specific changes. Changing the CRP values above 13 mg/dL and CK over 700 U/L can be explained by the presence of toxins and enzymes in the blood, this stage being correlated with NF's severity [4]. Microbial cultures used to identify the microorganism incriminated with the appearance of necrosis are sometimes hard to establish, because of prior general and local antibiotherapy, as in the presented case. The antibiotic adjuvant treatment is recommended using the antibiogram, but when it is not possible, the use of antibiotic associations with a large spectrum is recommended [5].

The NF diagnosis is often misdiagnosed because it is confused at onset with cellulitis or abscess – the situation in our case [17]. The severe pain in comparison with the initial cutaneous lesions, alongside the fast evolution towards necrosis and lack of response for systemic and local antibiotherapy would suggest the diagnosis of NF. During active state, NF should be differentiated by pyoderma gangrenosum, which is described by a deep ulcer, intense pain, with violet edges and areas of necrosis and calciphylaxis determined by vascular calcifications, which lead to painful cutaneous necrosis, surrounded by purpuric lesions with stellar shape that appear in patients with uremia [16].

NF is considered a particular form of cellulitis, rapidly progressive type that destroys the epidermis, dermis and subcutaneous soft tissues. It extends to the fascia or striated muscles, hence the name of necrotizing fasciitis.

In this category are included pyogenic infections lesions, with deep localization, called cellulitis. It affects both the skin and mucous membranes, especially those at ano-genital region such as Fournier’s gangrene [18]. Oral mucosa, cervical, facial and orbicular regions are rarely involved.

Histology diagnostic criteria include mandatory the presence of abundant polymorphic inflammatory infiltrate, predominantly neutrophils and granulocytes. Edema, necrosis and hemorrhage can complete this aspect. Blood and lymphatic vessels can present swelling or septic vasculitis lesions. Characteristic for necrotizing forms (NF) is the presence of intraluminal thrombus of fibrin.

The bacteria involved in these infections can be identified by microscopic examination of Hematoxylin–Eosin (HE) staining or in special staining: PAS, Giemsa or Gram. Bacterial cultures of these injuries can be stained with Gram or Giemsa. The term used to describe these microorganisms is “flesh-eating bacteria”, which denoted the rapidly progressive necrotizing disease often fatal in these patients [19].

The treatment is complex and serial, being susceptible to complications. Extensive surgical excision of the necrotizing fociars in association with antibiotic therapy is at first intent. An early diagnosis encourages a favorable prognosis and is influenced by predisposing factors that aggravate the disease [20]. In general, these patients have in their medical history a condition that predisposes them to infection, diabetes, hypertension, kidney and liver failure. Diabetes is the most common co-morbidity factor, affecting almost 50% of patients, because of hyperglycemia with predisposes to a low oxygen level environment, becoming

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favorable for the bacteria growth [21]. Another relevant co-morbidity factor is cirrhosis and chronic liver disease [17]. In this case, radiotherapy and ethanolic chronic hepatitis influenced the disease. There are reported associations between chemoradiation and NF, at seven months after treatment for T3 tonsil cancer and at nine months after treatment for T4 squamous cell carcinoma of oropharynx, but as in our case, severe infections complications may occur after years of remission [13, 14]. The symptoms presented by the patient are common to all cases; edema (80.8%), pain (79%), erythema (70.7%), tissue necrosis (24.1%) and crepitus (20.3%). The presence of crepitus in our case, lacking bacterial cultures, sustains the implications of anaerobic bacterial flora in the development of NF. Fever is present only in 40% of the cases. The septic shock is a late sign and has an incidence of 21.1%, with a close relationship with mortality [3]. Microbiological cultures reveal the prevailing presence of aerobic or facultative aerobic germs. In the cases of dental origin, Streptococcus oralis is most often involved. Among the anaerobic bacteria, Prevotella spp. is the most common. In 61% of cases, it can be found more frequently a mixed infection than strictly anaerobes (12%) or aerobes (28%) [22]. Sometimes lesions must be differentiated from ulcerated lipodic necrobiosis [23].

Conclusions
The case presented in our study had several particular features. First location at facial region was rarely reported in the literature. Second, the appearance at this level after a malignancy associated with important risk factors such as immunosuppressive status and chronic alcohol consumption. The prognosis of this disease is reserved and depends on making an early diagnosis. Initial surgical treatment is complex and depends on the possibility of wound sterilization, often mutilating, but it is required because of favoring factors and later complications. Subsequently, a series of surgical interventions are required to correct the defects.

Conflict of interests
The authors declare that they have no conflict of interests.

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Corresponding author
Silviu Horia Morariu. Associate Professor, MD, PhD, Department of Dermatology, University of Medicine and Pharmacy of Timișoara, 38 Gheorghe Marinescu Street, 540139 Timișoara, Mureș County, Romania; Phones +40265–215 551, +40744–757 246, e-mail: silviu_morariu@yahoo.com

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