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Case Report
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Odontogenic Cervical Necrotizing Fasciitis and Descending Necrotizing Mediastinitis in a Diabetic Patient: Literature Review and Report of a Rare and Fatal Case

Henrik Holtmann[1], Julian Lommen[1], Karoline Sander[1], Norbert R Kübler[1] and Renan Langie[2]*

[1]Department of Oral and Maxillofacial Surgery, Heinrich-Heine-University Düsseldorf, Germany

[2]Centro de Odontologia/Santa Casa de Misericórdia de Porto Alegre (SCMPA), Brazil

Abstract

Cervical Necrotizing Fasciitis (CNF) is a rare yet critical wound infection with a tendency for rapid progression. Diabetic patients are particularly vulnerable due to their deprived wound-healing capabilities. We report the case of a black 18-year-old female patient of African descent with a type I diabetes mellitus and CNF. The patient presented in state after surgical removal of the lower right third molar. Despite a prescribed antibiotic therapy by the attending doctor the patient complained about persisting pain and a reduced overall well-being. Presenting at our clinic the patient showed no clinical signs of swelling, reddening, indurations or restricted mouth opening that would have indicated an abscess. Hence, we extended the antibiotic therapy and dismissed the patient. Two weeks later the patient returned in a considerably reduced general state of health including syncope and drowsiness. An immediately implemented CT-scan of the head and neck as well as the clinical appearance leads to the diagnosis CNF with profound mediastinitis. Despite surgical intervention, microbiological testing of wound swaps and an extended antibiotic therapy septic shock with acute hepatic failure caused the patient's death two weeks after her admission to our hospital. This case demonstrates the important role of odontogenic infections to the development of CNF and the difficulty of diagnosing early stage CNF especially in patients with dark complexion and diabetes mellitus. An early intervention is the deciding prognostic factor for the overall survival rate.

Keywords: Necrotizing fasciitis; Cervical necrotizing fasciitis; Mediastinitis; Descending necrotizing mediastinitis; Descending necrotizing fasciitis; Odontogenic infection; Dental infection; Diabetes mellitus

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Correspondence:

Henrik Holtmann, Department of Oral and Maxillofacial Surgery, Heinrich-Heine-University Düsseldorf, Zentrum für Operative Medizin II Klinik für Mund, Kiefer- und Plastische Gesichtschirurgie Moorenstr.5, D-40225 Düsseldorf, Germany, Tel: 49 211 81-07428; 49 211 81-18877;

E-mail: Henrik.Holtmann@med.uni-duesseldorf.de

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Introduction

Necrotizing Fasciitis (NF) is a rare life-threatening infection characterized by a rapid and progressive necrosis of subcutaneous tissues and fasciae with subsequent skin gangrene and further systemic toxicity leading to septic shock [1,2]. Areas of the body mostly affected by NF are extremities, trunk and perineum, while head and neck structures account for only 3% to 4% of all reported cases [2].

Due to the close proximity to the vessel and nerve cord Cervical Necrotizing Fasciitis (CNF) is an even more severe condition, with a high mortality rate, usually caused by dental infection, infection secondary to trauma, throat abscesses, osteoradionecrosis or salivary gland infection [2-4]. In patients developing mediastinitis an even higher mortality rate is reported. This condition is then called Descending Necrotizing Fasciitis (DNF) or Descending Necrotizing Mediastinitis (DNM) [5,6]. In cases where this complication occurs under immune suppression, such as diabetes, infection is often fatal [5]. The authors conducted a literature search about CNF and present a fatal case with DNF caused by a postoperative infection after extraction of a third molar, in an 18-years-old black female patient of African descent.

Case Report

Initially, the patient developed persisting pain after the surgical removal of the lower right third molar and showed a reduced overall well-being. The treating dental surgeon subscribed antibiotics (amoxicillin 1000 mg 3x/day without improvement of the clinical course. The patient's clinical history exposed a well-controlled type 1 diabetes mellitus (HbA1c 6, 5%). With health not improving the patient consulted our clinic for the first time. The observed swelling seemed adequate considering

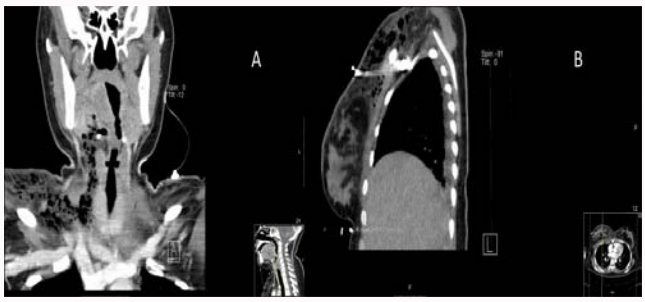

Figure 1: Parapharyngeal gas formation on the right side with extension to the right acromion. Above the parapharyngeal gas formation a hypodense mass is visible, that does not show significant rim enhancement on contrast injection as sign of liquefactive necrosis across multiple layers of the cervical fascia. The upper airway is almost completely obstructed (left picture: coronal slide). The gas formation infiltrates the breast tissue (right picture; sagittal slide of the right chest tissue).

the respective trauma set by the tooth extraction. Due to the dark skin complexion of our patient the buccal skin area did not present any signs of reddening or flush. Symptoms were un definable and there was no clinical evidence for an abscess with symptoms such as fever, trismus or hardened swelling. Hence we dismissed the patient with an extended oral antibiotic therapy (amoxicillin/clavulanic acid 875/125 mg 3x per day). Two weeks later the patient returned to the emergency room of our clinic with a considerably reduced general state of health, including syncope and drowsiness. An immediately conducted Computed-Tomography (CT) of the head and neck area showed signs of large gas cavities within the subcutaneous cervical and thoracic tissue compatible with the diagnosis CNF and profound mediastinitis (Figure 1). Initially the patient was stabilized in the Intensive Care Unit (ICU) followed by a radical surgical wound debridement. Our oral and maxillofacial surgeons drained the perimandibular abscess through an extra-oral incision while simultaneously the general surgeons performed a radical debridement of the necrotizing cervical, thoracic and axillary soft tissues. A mastectomy as well as a resection of the pectoral, sternocleidomastoid, trapezius and deltoid muscles was necessary.

On completion of the surgical procedures the patient was intubated and transported to the ICU. Here, the patient remained in a state of shock with an ongoing need for catecholamines. According to current clinical guidelines the antibiotic therapy was based on meropenem, teicoplanin and clindamycin but failed to improve the situation. In the following four days further surgical treatment with wound debridement was necessary. The results of intraoperatively conducted wound swaps proved diverse Candida species within the necrotic area leading to the initiation of a caspofungin-based treatment protocol. On day five the patient developed an acute abdomen with renal failure and an increasing hepatic dysfunction. Biopsies of the liver showed a general necrosis with rapid development of hepatic failure. Additionally, an abdominal sonography diagnosed a cholecystitis making a cholecystectomy necessary. For re-evaluation, new CT-scans of the head, neck, thorax and abdomen were executed. The radiological images showed bilateral infiltrate in both upper and lower pulmonary lobes as well as mediastinal fluids.

In the further course of events and after surgical debridement once again, the patient developed severe disseminated intravascular coagulation and was subjected to blood transfusion. On day twelve after introduced therapy leucocytes decreased to a level of 1100/µl and medication with growth factors and pentaglobin was initiated.

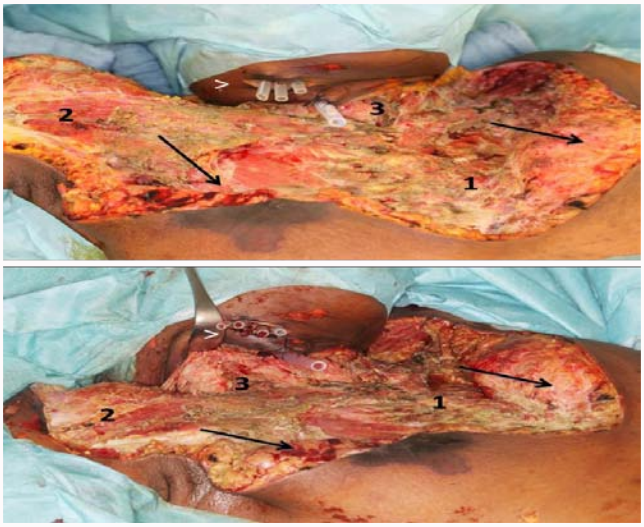

Figure 2: Advanced necrotizing fasciitis of subcutaneous and muscle tissue including fasciae of the anterior chest wall (1), right acromion (2) and neck (3). Condition after partial bilateral mastectomy (->). Submandibular drains (>).

Antiviral acyclovir was administered prophylactically. Nevertheless, leucocytes depletion to 200/µl and impaired wound healing could not be stopped.

Several days later radical necrectomy of the left thoracic and cervical soft tissues was unavoidable (Figure 2). Taken blood cultures showed increasing copies of Herpes Simplex Virus type 1 (HSV-1) justifying continuous antiviral therapy. Wound swaps as well as liver biopsies indicated an increasing fungal infestation leading to the beginning of an antimycotical therapy (ambisome 8 mg/kg per day combined with posaconazol 2 x 300 mg per day). To evaluate the aetiology of the leukocytopenia a bone marrow puncture was performed showing functionally aplastic bone marrow. Normally, chemotherapy would have been the first-line therapy, but the patient's poor general health made this impossible. Hence, a highly dosed glucocorticoid therapy was initiated which led to a fast increase of leukocyte numbers and decrease of inflammation parameters (i.e. CRP). Nevertheless, demand of catecholamine's increased steadily and stabilisation of vital parameters was no longer possible. Death occurred painless and symptom-controlled due to septic multi-organ failure.

Discussion

Necrotizing fasciitis is a severe soft tissue disease caused by a polymicrobial mixed aerobic-anaerobic infection [2]. NF is more often observed in skin of the extremities, abdomen and perineum, while the head and neck area presents a rarer site due to its higher vascularization supporting better wound healing [2]. Odontogenic infection is the main etiological factor of CNF [5,14]. Profound caries lesions as well as acute and chronic periodontitis, more often in molars, progressing to periapical abscesses, are reported to be the initial site of infection in most cases [2]. In the presented case report a lower third molar was the primary site of infection. Less frequently, additional causes such as tonsillar or pharyngeal infection, trauma, cervical adenitis, salivary gland and secondary tumor infection have been reported [2-5]. Infections in the head and neck area that are of dental origin are usually polymicrobial with a combination of aerobic, anaerobic and facultative anaerobic microorganisms

[3,6]. The synergy of these microbial species including their ability for quorum sensing makes such infections more aggressive than monoinfections [6]. Organisms most commonly isolated from wound swaps are oral streptococci, staphylococci and a variety of anaerobic bacteria. These pathogens also cause dental apical infection, acute localized cellulites and abscesses [5]. In our case the wound swaps revealed a polymicrobial fungal infection with *Candida albicans*, *Candida glabrata*, *Candida parapsilosis* and *Rhizomucor pusilus* being predominant. In addition, physiological bacteria of the mouth were found in wound swaps of the head and neck whereas physiological bacteria of the skin were found in swaps of the chest and abdomen.

NF frequently occurs in people with different forms of immunodeficiency, such as diabetes mellitus [8]. Insulin-dependent diabetes is the most common predisposing systemic factor for CNF, observed in 18% to 72.3% of the cases. Since chronic hyperglycemia impairs leucocyte functioning leading to a suppression of the immune system a higher susceptibility for exacerbating odontogenic infections is likely [13]. Predisposing factors for NF include alcoholism, tobacco abuse, immune suppression, neutropenia, malnutrition, higher age, peripheral vascular disease, renal failure, underlying malignancy, and obesity [3,5,14]. In the present case the patient's hyperglycemic condition, caused by a poorly controlled insulin-dependent diabetes mellitus, impaired leucocyte function suppressing the immune system. Hence, an adequate immune response to the infection was not possible.

There is no consensus regarding the mortality rate of NF, however early recognition and a prompt clinical and surgical intervention seem to positively influence the prognosis [11,12]. Banerjee et al. [15] reported that 44% of the CNF cases in their study had a mediastinal involvement, 38% of the patients died in the course of the disease. When an odontogenic CNF is complicated by Descending Necrotizing Mediastinitis (DNM) mortality rates can reach 41%, compared with 20% in cases with CNF alone [12]. According to Bucak et al. [6] the mortality rate in patients with CNF and DNF is 4 times higher compared with patients that suffer from CNF alone. The progression to sepsis was also shown to be an important prognostic factor for both CNF and CNF with DNM, and occurred more often when the thorax was involved [12]. A higher mortality rate is also related to preexisting systemic illnesses, especially diabetes mellitus, late surgical intervention, septicemia within the first 24 hours and higher age [4,5,2,11].

Classical NF usually begins 2 to 4 days after the initial injury [5]. NF presents as a rapid and progressive liquefactive necrosis of the subcutaneous connective tissue, while the overlying skin is spared [5]. Necrosis and liquefaction of fasciae and fatty tissues occur early, possibly mediated by collagenases and hyaluronidases produced by group a streptococci [5]. Liquefaction of fatty tissues results in a separation of the skin from the underlying tissues, producing edematous fluid and the path gnomonic 'dishwater pus' which has a strong odor in the presence of anaerobes [5]. Further necrosis and liquefaction of fat and fasciae lead to arterial thrombosis, wet gangrene, and finally ischemic death of the skin, while exposed underlying muscles may be unaffected [5]. Because gingiva, neck and mediastinum are in direct anatomical proximity odontogenic infections can rapidly spread to the mediastinum, facilitated by gravity, respiration, and a negative intrathoracic pressure [6]. There are two possible pathways for the infection to spread from the head and neck to the mediastinum: 1) by fascial spread along the carotid sheath or 2) by spread through the retropharyngeal or prevertebral space [10]. In our case the infection spread alongside the fasciae towards the mediastinum. Generally, spread of infection through the retropharyngeal space (danger space) accounts for 70% of DNM cases [6]. There is no specific sign that can reliably identify a patient with CNF and clinical features generally do not adequately correspond with the disease's rapid progression [4,9]. The onset of symptoms usually occurs only after an extensive infection has already been formed [9]. Early clinical presentation of CNF may be indistinguishable from a superficial soft tissue infection with mild symptoms [4]. However, cutaneous involvement may only occur in the later stages of the disease and is not directly correlated to the extensive underlying tissue necrosis [2,4]. Normally, a rapidly advancing odontogenic infection is the starting point for a developing CNF [3]. With the progression, CNF may be misdiagnosed as erysipelas or an acute neck abscess, presenting cervical tenderness, edema and erythema [4,8,9]. At this point, the skin may present an orange-peel appearance, something not visible in our presented case due to the patient's dark skin [8]. Furthermore, local ischemia and necrosis cause cutaneous anesthesia and emphysema due to anaerobic microbial metabolism [1,8]. NF may be accompanied by weakness, apathy, confusion, high fever, local pain, dehydration, tachycardia, hypotension, volume depletion, hypocalcaemia, increased glucose level and hypoproteinemia, with possible progression to septic shock and multiple organ failure [1,3,5,8]. Despite CNF's various clinical appearance rapid progression, systemic toxicity, and the presence of subdermal gas cavities are distinguishing features that should attract the attending physician's attention [1,5].

Computed Tomography (CT) examination is mandatory when NF is suspected [5]. The CT scan is able to identify the boundaries and severity of NF, since it is more sensitive than other imaging modalities in detecting inflammatory changes, such as fascial edema and thickening and enhancement of subcutaneous fat [5,10]. In contrast to abscesses, NF presents an area of fluid accumulation without significant peripheral rim enhancement [5]. Another characteristic CT feature is a hypodense area, often irrespective of the cervical fascial compartment, that does not show a significant rim enhancement on injection of a contrast agent. It has been suggested that the hypodense area is caused by the rapid spread of liquefactive necrosis across multiple layers of the cervical fascia [5]. Although accumulation of gas cavities is not a consistent finding it has been reported to be present in 64% to 66.7% of NF cases [3]. Abscess formation, mediastinal widening, gas tracking, empyema, pleural effusion and pericardial effusion in a CT scan are signs for a thoracic spread of the infection [10]. Specifically, gas formation was the predominant CT finding in our presented case (Figure 1 and 2). The main complications of NF are mediastinal involvement, septic shock, pleural effusion, lung empyema, airway obstruction, rupture of major vessels, brain abscess, cranial nerve palsy, disseminated intravascular coagulopathy, sepsis, acute renal failure and respiratory failure [5,6]. A higher mortality rate is found in patients where the infection has reached the mediastinum (descending necrotizing mediastinitis) [5,6,12]. When this complication is accompanied by a compromised immune system such as diabetes mellitus, the infection is often fatal [5]. Since mortality has been shown to increase with delayed intervention, once NF is diagnosed, treatment must immediately begin via different approaches [5]. Airway management is critical in CNF since edema and necrosis may complicate intubation [5]. After intubation a persistent throat irritation may be avoided if a tracheotomy is made [5,6,8,9].

Broad-spectrum intravenous antibiotics should provide effective coverage against gram-positive cocci, facultative anaerobes, gram-negative rods, and anaerobes [7]. Usually the regimen begins empirically with a triple therapy using a beta-lactame antibiotic, an amino glycoside and clindamycin or metronidazole [5,9]. In our case the antibiotic therapy began with meropenem, teicoplanin and clindamycin. Intra-operatively taken wound swaps should be used to adapt the antibiotic regimen to the predominant pathogens [7]. Our patient additionally received caspofungin because Candida species were detected during debridement. As mentioned before, therapy was extended to pentaglobin, Granulocyte-Colony Stimulating Factor (GCSF), prophylactic acyclovir and high-dose ambisome and posaconazol due to hypoimmunoglobulinemia and proven rhizomucor species. Aggressive surgical debridement including the removal of all nonviable tissues must be performed. Repeated debridement may be necessary until the infection has been adequately controlled [7]. Performing a second operative inspection of the wound after 24 hours to 36 hours is recommended [1]. The wounds should be rinsed and bandaged every four hours. Here hydrogen peroxide can be used as an ancillary antiseptic [1].

Recent literature has also suggested adjunctive therapy with fluid and electrolyte infusion, application of intravenous immunoglobulin G, and hyperbaric oxygen therapy, aiming to improve surgical outcomes and reduce mortality rates [2,4]. The hyperbaric oxygen therapy is capable of accelerating wound healing and to reduce the amount of microorganisms in the wound reducing hospitalization [2,4,8]. Reconstructive surgery immediately after the treatment of NF is contraindicated [8]. It should be performed only after the patient has been stabilized, the infection is fully eradicated and the wound presents a good vascular granulation [7,8]. Reconstruction is usually performed with skin grafts and flaps, analog to burn surgery [8]. However, in huge defects and limited donor-site availability artificial alternatives, like amniotic membrane, should be considered an alternative [8]. Regional or free flaps like anterolateral thigh, scapular, pectoralis major, latissimus dorsi and trapezius, are recommended for large defects as they allow good mobility of the skin [8].

Conclusion

The presented case highlights the important role dental infections play in the development of Cervical Necrotizing Fasciitis (CNF). Early recognition of the disease and prompt clinical and surgical intervention is essential for a successful outcome, especially in patients with systemic defects of the immune system.

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