

Necrotizing Fasciitis of Odontogenic Origin in a Patient with Unknown Diabetes Mellitus: A Case Report and Literature Overview

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Abstract

Case Report

Necrotizing fasciitis (NF) is a life-threatening emergency that requires prompt and aggressive surgery with broad-spectrum antibiotherapy and resuscitation. This report aims to raise the index of suspicion for NF allowing early diagnosis mainly in patients with diabetes mellitus and who received non-steroid-anti-inflammatory. A 52-year-old man, without noticeable illness, was admitted for progressive submandibular swelling, following wisdom tooth extraction, with extensive cervical redness centred by skin depression. The patient's chief complaint was a sore throat, trismus and intensive pain leading to self-administration of non-steroid-anti-inflammatory drugs. Laboratory tests revealed an unknown diabetes mellitus. Enhanced Computed Tomography revealed the disappearance of fascial planes and tissular swelling containing multi-space emphysema with an airway compromise. The patient underwent surgical debridement of the involved fascial spaces and necrotic tissues until normal bleeding was obtained combined with intensive resuscitation and 37 inpatient days allowed patient rescue without soft tissue sequelae. Thus, Odontogenic NF should be suspected in patients with diabetes mellitus who have taken non-steroid-anti-inflammatory medication especially when there are discrepancies between signs of toxicity and deceptive clinical symptoms of an odontogenic infection. Computed tomography is crucial for prompt diagnosis, therapeutic management and postoperative assessment. Removal of the odontogenic focus simultaneously combined with thorough surgical debridement with broad-spectrum antibiotherapy and resuscitation improves survival rates and reduces visceral morbidity and soft tissue sequelae. Otherwise, Oral health education and care mainly in high-risk patients remain a key to prevent this serious health problem.

Keywords: Necrotizing Fasciitis, Diabetes Mellitus, Computed Tomography, Biofilm, Diagnosis.

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1. INTRODUCTION

Necrotizing fasciitis (NF) of odontogenic origin is a rapidly spreading soft tissue infection that can result in death within 24 hours. It is associated with systemic toxicity and a higher risk of mortality mainly in patients with comorbidities and delayed therapy. Immunocompromised patients especially with poorly controlled diabetes mellitus are more likely to develop odontogenic infections that worsen more than in healthy individuals. It was reported that cervicofacial cellulitis was most likely to occur 2 years following the initial diagnosis of diabetes mellitus. The combination of host immunodeficiency and bacterial biofilm synergism favours the development of severe odontogenic infections usually resulting in death. NF appears unexpectedly, but can be predicted by experienced clinicians, in patients with debilitating conditions even

more aggravated by taking non-steroid-anti-inflammatory drugs leading to a feared life-threatening situation. The prognosis depends on prompt and aggressive medico-surgical treatment requiring an early diagnosis which is usually delayed because of the discrepancies between deceptive clinical features and the extent of the inflammatory necrosis well defined by computed tomography findings. On the other hand, the diagnosis should be suspected in front of pain out of proportion to clinical signs [Ko Hh *et al.*, 2017; Mehri Turki I, 2023]. In a matched case-control analysis of the United States Multiple Cause of Death Files database, diabetes mellitus, renal failure, and obesity were noted to be significantly associated with NF which has an overall mortality of about 13,36% [Arif N *et al.*, 2016].

Deep cervicofacial planes are priorly affected, and necrosis extends from the deep plane to the

superficial one reaching the cutaneous tissue. Thus, cutaneous necrosis indicates that NF is at an advanced stage where the respiratory tract is usually compromised and associated with an overwhelming sepsis syndrome that results in a rapid life-threatening situation [Leyva P *et al.*, 2013; Gunaratne DA *et al.*, 2018].

Enhanced Computed Tomography reveals pathognomonic signs of necrosis and delineates accurately the extent of the process to guide airway support and surgical strategy. This article reports the successful management of a patient with diabetes mellitus who had necrotizing fasciitis without skin necrosis following a tooth extraction, aggravated with self-drug administration.

2. CASE REPORT

A 52-year-old man underwent a wisdom decayed tooth extraction, at the right side of the mandible. A slight swelling of the right submandibular region appeared five days later associated with fever and intensive pain. Clinical examination revealed a patient with a right submandibular tender inflammatory swelling overlapping the submental area. The inflammation reached the anterior neck containing a noticeable excavation at its midline (Figure 1). However, there was no crepitant sensation on palpation or skin necrosis.

A thorough medical history revealed self-administration of non-steroid anti-inflammatory drugs due to intensive pain, a sore throat, trismus, difficulty speaking and moderate dyspnea. There was no history of comorbidity. Laboratory studies showed an elevated white blood cell count and elevated C-reactive protein levels of 35 mg/dL. Liver enzymes and blood coagulation system were normal. Urinalysis showed moderate ketonuria and his serum glucose level was 350 mg/dL. Enhanced Computed Tomography of the cervicofacial and thoracic region was performed upon his admission and showed enhancing swelling of soft tissue with a total loss of muscular and fascial plane involving a large amount of gas extending subcutaneously from the submandibular region,

anterolateral neck up to the hyoid bone with impingement on the oropharyngeal air column confirming NF complicating Ludwig's angina. However, there was no evidence of mediastinal involvement (Figure 2, Figure 3). Based on these findings, a diagnosis of NF was made. After receiving intravenously amoxicillin and clavulanic acid (2 g three times a day) associated with metronidazole (0,5 g three times a day) and ofloxacin (0,2 g twice a day), the patient was brought to the operating theatre six hours after his admission for a neck dissection and drainage, as well as the removal of necrotic tissues and revision of the alveolar socket. Nasotracheal intubation was conducted under fiberoptic guidance due to trismus. The submandibular, submental and retropharyngeal spaces were explored through a transcervical approach leading to a discharge of a putrid-smelling black pus with air bubbles. An extensive debridement of necrotic tissue was also performed until normal bleeding was obtained. The nasotracheal tube was removed, in the intensive care unit, three days postoperatively because of the airway concern due to postoperative oedema which decreased with adjuvant corticotherapy at high doses (dexamethasone 8 mg) administered over five days. Copious irrigation with an iodine solution in the tissue spaces was carefully performed, with low pressure, twice a day until the wound healed.

Pus culture produced growth of *Streptococcus viridans* and *Bacteroides Fragilis*, both of which were sensitive to empirical antibiotherapy (Amoxicillin – Ac clavulanic, Metronidazole, Ofloxacin). The patient recovered after two surgical procedures at two-day intervals and was discharged at 37 inpatient days, including 7 days in the intensive care unit, without soft tissue sequelae, and the serum glucose level steadily decreased to 167mg/dL as a result of insulin therapy. There was no evidence of residual infection up to three months of follow-up.

A written informed consent for participation and consent for publishing patient details in this study has been obtained.



Figure 1: Clinical picture showing submandibular swelling and extended cervical redness. A midline cervical excavation evocative of subcutaneous fat necrosis

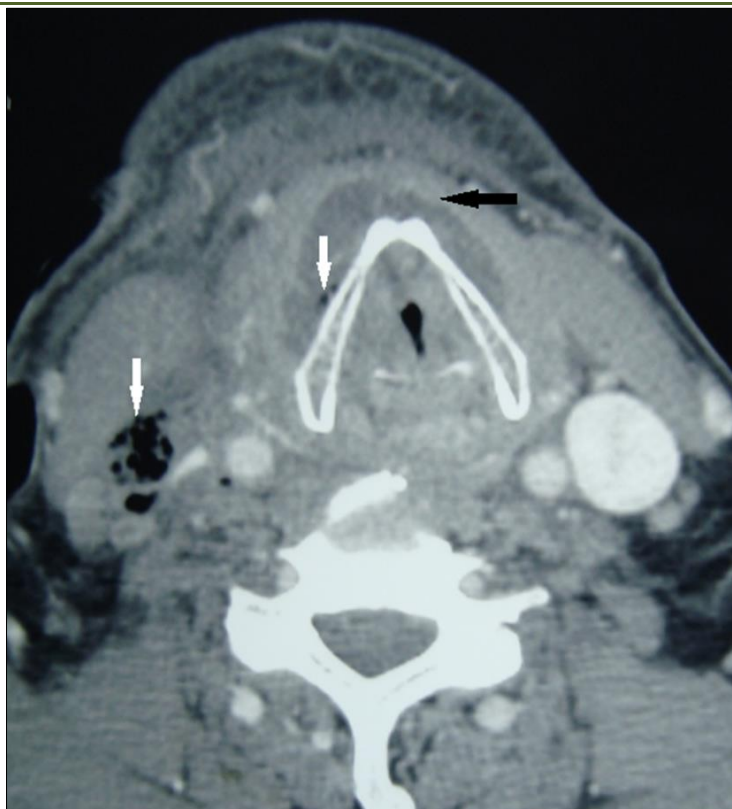


Figure 2: Enhanced computed tomography. Axial view at the level of the hyoid bone showing the compression of the internal jugular vein by gas formation and muscle swelling (arrow)

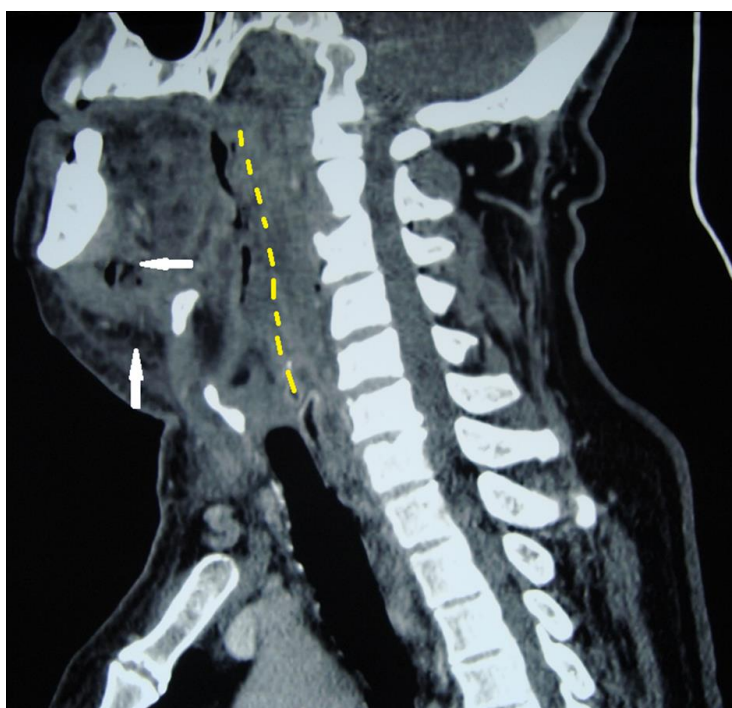


Figure 3: Computed tomography, Sagittal view. Narrowing of the airway tract from the retropharyngeal space up to the retrosternal level (Yellow discontinued line). Supra and infra-mylohyoid gas formation (arrow).

3. DISCUSSION

Necrotizing fasciitis is an uncommon fulminant soft tissue infection characterized by rapidly progressive necrosis beginning at tissue fascia and extending to

muscles, subcutaneous tissue and skin [Bosshardt TL *et al.*, 2001]. NF occurs rarely in the head and neck region accounting for only 1-10% of the whole body, because of the florid blood supply of this region. However, the

resurgence of this disease can be related to the widespread overuse of antibiotherapy increasing antibiotic resistance and the synergical work of polymicrobial species in an odontogenic focus that increases bacterial virulence. Additionally, host immunodeficiency such as in diabetes mellitus, as well as self-administration of inappropriate medications, notably non-steroid-anti-inflammatory drugs, aggravates the course of the infection [Mehri Turki I, 2023; Lin C *et al.*, 2001]. This fatal disease can be wrongly diagnosed as a common odontogenic infection at an initial stage causing delayed and improper treatment resulting in dramatic consequences. The occurrence of pain out of proportion to clinical signs with a rise of C-Reactive Protein should raise suspicion of NF mainly in immunocompromised patients. Additionally, tender oedema, frequently present in common odontogenic infections, can not predict skin necrosis which is more susceptible to occur when skin presents purplish discoloration, blisters and mottled aspect. However, a novel relevant cutaneous hallmark, not yet described in the literature, consists of excavation within the oedematous area. This clinical sign mirrors the subcutaneous fat loss due to necrosis. On the other hand, the occurrence of septic syndrome and vascular complications take a turn for a dangerous worse and death could occur within 12 to 24 hours despite customary surgical and medical treatment, particularly in immunocompromised patients. Regardless, one patient in four dies due to multiorgan septic failure, and those with debilitating conditions are more susceptible to this fatal prognosis [Böttger S *et al.*, 2022; Hohlweg-Majert B *et al.*, 2006]. In fact, diabetes mellitus is a well-established risk factor for head and neck NF, longer inpatient days, and higher morbidity and mortality [Taub D *et al.*, 2017; Verfaillie G *et al.*, 2002]. The present case illustrates the odontogenic infection complicated by unknown diabetes mellitus and aggravated with self-medication. Nevertheless, non-steroid anti-inflammatory drugs decrease pain and inflammatory reactions resulting in delaying diagnosis while necrotic infection spreads sufficiently to present fulminantly. Moreover, the harmfulness of these drugs is related to their immunosuppressive action by the inhibition of granulocyte adherence, activation of phagocytosis and cytokine production augmentation (Tumor Necrosis Factor) [Mehri Turki I, 2023; Taub D *et al.*, 2017; Verfaillie G *et al.*, 2002].

Contrast-enhanced computed tomography (ECT) is an excellent tool, readily available to confirm diagnosis while identifying odontogenic infection foci and delineating the involvement of adjacent soft tissue structures. It can also demonstrate vascular complications and airway compromise. However, predictive postoperative mechanical ventilation and tracheostomy should be planned with great care so as not to disseminate the infection in the respiratory tract. Furthermore, the lack of pathognomonic radiological features such as gas formation and fascia damage does

not rule out the diagnosis of NF [Gore MR *et al.*, 2018; Umeda M *et al.*, 2003; Liang J *et al.*, 2022; Rautaportas N *et al.*, 2023]. Despite the relevance of ECT for diagnosis and customizing treatment decisions, it should not delay the surgical treatment that helps the patient halt the spread of the necrotising process and thus stops the release of inflammatory mediators causing systemic complications. In addition, the removal of infarcted tissue improves antibiotic penetration and efficacy.

Multiple reviews have demonstrated that debridement within 24 hours is important for lowering mortality. However, necrosis may recur allowing further surgical procedures [Mehri Turki I, 2023; Malik V *et al.*, 2010]. Because of the impairment of the immune system caused by diabetes mellitus, serum glucose levels should be maintained within the normal range, as performed for the present case, to improve the survival of patients with diabetes mellitus.

As for common odontogenic infections, infection focus originates from the oral ecosystem. However, the triggering factor of NF is not yet established. Several researches suggest the rôle of synergy in the oral microbiome leading to this devastating infection, mainly a variety of bacteria species in which anaerobic bacteria of the oral cavity predominate such as *Prevotella*, *Porphyromonas*, *Fusobacterium* and *Peptostreptococcus*. *Actinomyces turicensis* and *Bacteroides thetaiotaomicron* whose virulence increases strongly in polymicrobial infection. Anaerobic bacteria produce volatile sulfur compounds giving more oxidative stress and proinflammatory activity. It also causes the putrid smell of pus and reflects gas formation often visualized as soft tissue emphysema on radiological explorations [Wu DD *et al.*, 2022]. The single “culprit bacterium” that triggers the necrotizing course could not be identified even by using molecular pathogen diagnostics. Therefore a broad-spectrum antibiotic therapy providing excellent empiric coverage should initially be performed and then adjusted specifically according to the antibiogram sensitivity. In some practices, authors suggest that a third-generation cephalosporin with clindamycin or metronidazole provides good initial antibiotic coverage [Sugata T *et al.*, 1997]. It has been shown that the combined use of antibiotics and corticosteroids has synergistic effects leading to decreased pain, post-operative oedema, and inflammation, and thus decreased hospital stay. Currently, there is no clear consensus, or evidence-based guidelines for clinical practice, in favour of corticosteroid administration because of their immunosuppressive action which could also worsen the infection [Chávez-Rimache L *et al.*, 2020]. Otherwise, removing the infected focus without delay has been widely recommended to avoid bacterial resistance due to the dental biofilm because of the inaccessibility of drugs to the deep layers of this biofilm where different bacterial species continue to interact [Mehri Turki I, 2023;

Treviño-Gonzalez JL *et al.*, 2023; Flanagan CE *et al.*, 2009].

Unfortunately, most patients with diabetes mellitus fail to show consistent improvement even when adequate prompt therapy is performed resulting in a mortality rate of about 19.2% [Umeda M *et al.*, 2003]. Some reports describe the interest in an associated hyperbaric treatment that improves tissue oxygenation and wound healing, but it is not always available and can never replace aggressive surgical treatment [Malik V *et al.*, 2010; Flanagan CE *et al.*, 2009]. The hospital stay is reported to be twice as long for patients with diabetes mellitus compared to patients with no comorbidity [Hohlweg-Majert B *et al.*, 2006].

4. CONCLUSION

NF of the head and neck is a rare but often fatal infectious disease hallmarked by initial intensive pain, out of proportion to deceptively clinical findings. Patients with diabetes mellitus are more susceptible to developing NF even more favored by non-steroid anti-inflammatory drug administration. A high index of suspicion for NF should be maintained for an early diagnosis enabling prompt and adequate treatment. Diagnosis of NF is obvious on Computed Tomography which is also critical in surgical decision, airway management and postoperative assessment. NF remains an unpredictable and life-threatening pathology. The author emphasizes the importance of checking periodically the oral health of patients with diabetes mellitus while ensuring the promotion of oral health education to prevent the occurrence of this serious odontogenic infection.

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Conflict Of Interest Statement: None

Patient Consent: Written informed consent for participation and consent for publishing patient details in the study has been obtained.

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