

CERVICAL NECROTIZING FASCIITIS AFTER TOOTH EXTRACTION IN A PATIENT WITH CHRONIC EOSINOPHILIC LEUKEMIA: CASE REPORT

FASCITIS NECROTIZANTE CERVICAL POSTERIOR A EXODONCIA EN PACIENTE CON LEUCEMIA EOSINOFÍLICA CRÓNICA: REPORTE DE CASO

FASCITE NECROSANTE CERVICAL APÓS EXTRAÇÃO DENTÁRIA EM PACIENTE COM LEUCEMIA EOSINOFÍLICA CRÔNICA: RELATO DE CASO

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ABSTRACT

Cervical necrotizing fasciitis (CNF) is a rare and severe infection that affects subcutaneous tissue and muscle fascia, causing necrosis. Most cases arise from odontogenic or pharyngeal infections, evolving into extensive necrosis and gas formation in the subcutaneous tissue and underlying fascia, with a high mortality rate. CNF is more common among patients with chronic diseases or immune suppression, possibly being more severe in these cases. This work reports the case of a patient diagnosed with chronic eosinophilic leukemia (CEL) who developed a condition of extensive lesion in the right submandibular/cervical region after a tooth extraction, presenting necrosis, liquefaction, necrotized exudate, and compatible muscle tissue exposure – clinical aspect that, associated with data on computed tomography, enabled CNF diagnosis. The patient was hospitalized and followed-up by a multidisciplinary team. This article aims to report that CNF is a rapidly progressing, potentially fatal infection, requiring immediate diagnosis and intervention. The infection may arise from a complication following tooth extraction, especially in patients with chronic diseases or immunological suppression. Considering that, the oral surgeon must include this condition within post-extraction infections differential diagnosis, where a multidisciplinary approach is fundamental for diagnosing and establishing an appropriate treatment.

Keywords: Cervical Necrotizing Fasciitis; Diagnosis, Differential; Infections; Tooth Extraction.

RESUMEN

La fascitis necrosante cervical (FNC) es una infección rara y grave que afecta el tejido subcutáneo y la fascia muscular, causando necrosis. La mayoría de los casos surgen de infecciones odontogénicas o faríngeas, evolucionando hacia una extensa necrosis y formación de gas en el tejido subcutáneo y la fascia subyacente, con una alta tasa de mortalidad. La FNC es más frecuente en pacientes con enfermedades crónicas o inmunosupresión, pudiendo ser más grave en estos casos. Este trabajo reporta el caso de un paciente diagnosticado de leucemia eosinofílica crónica (LEC) que desarrolló una condición de lesión extensa en la región submandibular/cervical derecha después de una extracción dental, presentando necrosis, licuefacción, exudado necrotizado y exposición de tejido muscular compatible - aspecto clínico que, asociado a los datos de la tomografía computarizada, permitió la FNC diagnóstico. El paciente fue hospitalizado y seguido por un equipo multidisciplinario. Este artículo tiene como objetivo informar que la FNC es una infección potencialmente mortal que progresa rápidamente y que requiere un diagnóstico e intervención inmediatos. La infección puede surgir de una complicación posterior a la extracción dental, especialmente en pacientes con enfermedades crónicas o inmunológicas supresión. Por ello, el cirujano oral debe incluir esta condición dentro del diagnóstico diferencial de las infecciones post exodoncia, donde el abordaje multidisciplinario es fundamental para diagnosticar y establecer un tratamiento adecuado.

Palabras clave: Fascitis Necrosante Cervical; Diagnóstico Diferencial; Infecciones; Extracción Dental.

RESUMO

A fascite necrosante cervical (FNC) é uma infecção rara e grave, que acomete o tecido celular subcutâneo e a fásia muscular, resultando em necrose. A maioria dos casos tem origem odontogênica ou faríngea, evoluindo com extensa necrose e formação gasosa no tecido subcutâneo e fásia subjacente, com elevado índice de mortalidade. A FNC ocorre com mais frequência em pacientes com doenças crônicas ou supressão imunológica, podendo manifestar-se de forma mais grave nesses casos. No presente trabalho é relatado o caso de um paciente diagnosticado com Leucemia Eosinofílica Crônica (LEC), que após uma extração dentária, evoluiu com um quadro de extensa lesão em região submandibular/cervical direita com necrose, liquefação, exsudato necrotizado, e exposição de tecido muscular compatível – aspecto clínico que, estando associado aos dados da tomografia computadorizada, possibilitou o diagnóstico de FNC. O paciente foi hospitalizado e acompanhado por uma equipe multiprofissional. Este artigo pretende relatar que a FNC é uma infecção de progressão rápida, potencialmente fatal, que requer diagnóstico e intervenção imediatos, podendo ocorrer como complicação de uma extração dentária, principalmente em pacientes portadores de doenças crônicas ou supressão imunológica. Portanto, o cirurgião dentista deve incluir esta condição no diagnóstico diferencial de infecções pós-extração, sendo a abordagem multidisciplinar fundamental para o estabelecimento do diagnóstico e tratamento adequado.

Palavras-chave: Fascite Necrosante Cervical; Diagnóstico Diferencial; Infecções; Extração Dentária.

INTRODUCTION

Necrotizing fasciitis (NF) is a rare and severe infection that affects subcutaneous tissue and muscle fascia, rapidly progressing and causing extensive necrosis. Most cases occur in the abdominal region, extremities and peroneus, being less common in head and neck due to this region high vascular supply. However, cervical necrotizing fasciitis (CNF) can be fatal. Its most common causes are odontogenic and pharyngeal infections [1, 2, 3].

The infection is characterized by the presence of strict and facultative anaerobic bacteria (Type I) or by *Streptococcus* species isolated or combined with *Staphylococcus aureus* (Type II). When it occurs in the head and neck region, the patient may experience severe pain and trismus, as well as signs of edema, hyperemia, hyperthermia, tachypnea, and leukocytosis [2, 4, 5]. As CNF clinical signs and symptoms resemble those of other morbidities, such as cellulitis and erysipelas, and the literature lacks clinical guideline, arriving at a diagnosis is often complex. Although computed tomography (CT) fails in providing specific data, it offers an important tool for establishing diagnostic hypotheses and analyzing the extent of the disease, as well as clinical and surgical findings such as subcutaneous tissues low adherence, absence of bleeding, and subcutaneous fat liquefaction [1, 6]. CNF treatment involves broad-spectrum antibiotic therapy and aggressive surgical debridement, removing necrotic areas [4].

CNF is more common among patients with chronic diseases or immune suppression, possibly being more severe in these cases. This work reports the case of a patient diagnosed with chronic eosinophilic leukemia (CEL) – a subgroup of myeloproliferative diseases characterized by hypereosinophilia in bone marrow and peripheral blood, more common in men. In its original form, it is secondary to chromosomal abnormalities, among which the most frequent is caused by 4q12 rearrangement that leads to FIP1L1 / PDGFRA fusion. If left untreated, the disease damages several target-organs by infiltration, especially the heart (restrictive cardiomyopathy by endocardial infiltration), lung (pulmonary fibrosis), and spleen (splenomegaly). CEL typical hypereosinophilia is observed by blood count and defined as persistent eosinophil count of at least $1.5 \times 10^9/L$. Other tests, such as morphological evaluation of blood and bone marrow, standard cytogenetics, fluorescent in situ hybridization, flow immunocytometry, and t-cell clonality assessment are required to confirm LEC diagnosis and detect the specific rearrangement [7, 8]. The disease prognosis varies according to eosinophilia subtype, and the treatment aims to minimize organ damage and reach hematologic remission [9].

This article aims to report a rare case of CNF after tooth extraction in a patient diagnosed with CEL. The Free and Informed Consent Form was signed by the patient before the beginning of this work, according to the hospital's routine protocol.

CASE REPORT

A 40-year-old male patient referred to the Institute of Hematology Arthur de Siqueira Cavalcanti (HEMORIO) by the Irmandade da Santa Casa de Misericórdia de São Paulo – unit where he was diagnosed with LEC and treated the disease for two years. In a medical consultation for admission to HEMORIO, the patient denied history of smoking, alcohol consumption, and allergic diseases and reported persistent splenomegaly, occasional night sweat, anemia since childhood, and no use of medication for disease control (Hydroxyurea) for 3 months. A bone marrow biopsy was requested to confirm diagnosis and results showed hypercellularity, reduced erythroid series, hypercellular myeloid series with abundant myelocytes and eosinophils, and decreased hypolobulated megakaryocytes count and slightly increased reticulum, which lead to a myeloproliferative disorder diagnosis, favoring chronic eosinophilic leukemia (CEL).

The patient was submitted to outpatient care, where he was periodically followed by the hematologist and the nursing team. During a routine consultation, he complained of tooth mobility and pain, being referred to the outpatient Dental Service. The clinical evaluation confirmed the need for extracting the mandibular right first molar due to grade III mobility and marked bone loss verified by panoramic radiography (**Figure 1A**). On that date, laboratory data showed hemoglobin (8.77 g/dl), eosinophils (16,430,000/mm³), absolute

neutrophils (1,035,000/mm³), and platelets (57.3 thousand/mm³). Dental extraction was performed 2 weeks after the initial consultation, without complications and modification of the conventional protocol, under local anesthesia with Lidocaine 2% with Adrenaline 1:100,000 (0.01 mg/mL). Luxation was performed with wedge motions using an apexo elevator. Then, a no. 17 forceps was used to remove the element. Synthesis was performed by occlusive suture, using a 3.0 thread. At the end of surgery, Dipyron 500mg was prescribed every 6 hours or, in case of pain, for 3 days. The patient was instructed to bite a gauze placed over the surgical site for 30 minutes, apply an ice pack to the face for 20 minutes every hour during the first 24 hours, rest with the head supported on a higher plane than the rest of the body, perform no mouthwash, sanitize the mouth with 0.12% chlorhexidine twice a day (in the morning and before sleeping), and opt for a cold liquid to pasty diet in the first 48 hours.

The patient returned seven days after surgery complaining of trismus, which hampered a careful oral examination. The wooden toothpick method was used to stimulate muscle, Miosan 5mg was prescribed twice a day for 3 days to relieve muscle tension, and the patient was instructed to perform the method at home.

After 1 week, the patient returned to the Dental Service presenting right-sided hemifacial pain, edema, and erythema, so

amoxicillin 500mg every 8 hours for 7 days and Nimesulida 100mg every 12 hours for 5 days were prescribed.

The patient returned after 3 days without improvement, and surgeons determined hospital admission for airway monitoring and intravenous medication administration. Extraoral physical examination showed a swollen, hyperthermal, smooth, bright, reddish area with extraoral fluctuation point in the mandibular body region extending to cervical region. The patient underwent a surgical drain performed by the general surgeon in another hospital unit, respecting all biosafety and aseptic measures. On the second post-drain day, the patient presented extensive lesion in the right submandibular/cervical region with necrosis, liquefaction, necrotized exudate, and muscle tissue exposure (**Figure 1B and 1C**). The nursing team rinsed the wound with 0.9% saline solution (SS) and performed a hydrogel occlusive dressing. Then, intravenous antibiotic therapy with Ceftriaxone 1g, Metronidazole 500mg, and Vancomycin 1g was administered every 12hrs, and a CT scan of skull, face, and neck was requested. A new complete blood count found hematocrit 21.0%, hemoglobin 7.1g/dl, and absolute neutrophils 1,415,000/mm. Contrast-enhanced CT showed an extensive irregular set with permeating gas foci, walls, and some thick septa, extending

from the right anterolateral cervical region to the supraclavicular. The set was inseparable from the right parotid and matching musculature, closely related to the submandibular gland, internal carotid artery, and jugular vein, compressed by the set. The lesion measured approximately 13.8 x 2.2 x 7.0 cm and presented an aspect compatible with cervical necrotizing fasciitis (**Figure 2A**).

During hospitalization period, the nursing team rinsed the wound daily with 0.9% SS and performed hydrogel occlusion dressings, whereas the plastic surgeon performed serial debridement. After remission of the infectious condition, wound was closed in the operating room by skin incision with devitalized tissues removal and wound edges revival, subcutaneous dissection, revision of hemostasis, and synthesis with skin advancement flaps for primary wound closure (**Figure 2B**). The patient was monitored by the medical team and discharged after 58 days of hospitalization, being instructed to administer oral Clavulin every 8 hours for 7 days at home.

The patient remained in outpatient follow-up, and the team performed triamcinolone infiltrations in the wound to improve its aesthetics. He presented a surgical scar in the right anterolateral region, with mild hypertrophy, and no functional limitation (**Figure 2C**).

Figure 1- Panoramic radiography showing extensive bone loss associated with the mandibular right second molar (A). Lesion in right submandibular/cervical region with necrosis, liquefied, necrotized exudate, and muscle tissue exposure (B and C).



Source: Arquivo Hemorio

Figure 2- Computed tomography showing extensive irregular set with gas foci (A). Seven days post-surgery of wound closure in operating room (B). Final clinical aspect after one year (C).



Source: Arquivo Hemorio

DISCUSSION

Necrotizing fasciitis (NF) is an aggressive, rapidly progressive infection that

can quickly lead to patient death. Although uncommon in head and neck, occurrences within these regions are mostly due to odontogenic infections – involving dental

abscesses and chronic periodontal disease [4, 10]. The authors [3] evaluated 207 studies involving 1235 patients; among these, 47.04% reported that the source of infection was odontogenic. The mandibular second and third molars seem to be the main sources, as their roots are anatomically located below the mylohyoid line [3, 6].

Although the literature lacks reliable data on its real incidence, studies suggest a possible predilection for men. In the evaluation of 207 articles by the authors [3] on cases of NF, of these 64.23% occurred in men and mean age was 49.1 years. The authors [11] reviewed 125 cases of NF with age ranging between 12 and 82 years and found a mean age of 45.2 years and a prevalence of cases among men.

Chronic diseases and immune suppression may predispose the individual to NF and increase the risk of mortality [2, 4, 6, 10, 11]. In your study [12] reported that approximately 20% of diabetic patients were up to 9 times more likely to die from NF Complications than non-diabetic. The authors [13] reviewed 17 cases, of which 12 had immune suppression; anemia was a major finding in 10 of the 17 cases.

Our study patient had chronic immunosuppressed condition untreated for months. His last complete blood count before dental extraction found anemia, neutropenia, and thrombocytopenia, which may justify the quick evolution of the post-extraction condition and the advanced periodontal disease (PD)

compromising the extracted element support. The PD-associated microbiota would also pose a risk factor.

Initial symptoms such as swelling, redness, pain, and trismus are the most reported in the literature [2, 4, 6]. In this sense, the authors [11] evaluated 9 NF cases of odontogenic origin, and all of them presented swelling, trismus, or both. In our study, trismus was the first symptom presented by the patient, 7 days after tooth extraction. The history of recent surgery and the underlying immunosuppressive condition justify antibiotics prescription, stressing the relevance of the oral surgeon knowing and being aware of complications that may occur after extractions, as well as being prepared to treat them as soon as possible, avoiding further damage.

Computed tomography (CT) plays an important role in the diagnosis, surgical planning, and delimitation of NF extent [1, 6, 11]. The authors [3] found that 56.8% of the CTs found subcutaneous gas – which, although not a pathognomonic sign of NF, may indicate a diagnosis when combined with other clinical and laboratory data. CT employment is not unanimous in the literature; yet, it played a key role in diagnosing NF and supporting the therapeutic planning of the reported case.

Due to the polymicrobial nature of the infection and its rapid progression, the treatment of cervical necrotizing fasciitis (CNF) involves extensive debridement of necrotic tissue and broad-spectrum antibiotic therapy [4, 10, 11]. Patients often need to

remain hospitalized for long periods and, even after cured, they may still present extensive skin and soft tissues loss, which may require weeks or months of dressing changes or secondary reconstructive procedures, such as skin grafts [14].

NCF is a rare, rapidly evolving, and severe infection that requires a multidisciplinary approach for enabling an early diagnosis and appropriate treatment. In the reported case, the full care provided to the patient during hospitalization was essential for ensuring quality of life during hospital recovery and disease control.

CONCLUSION

NCF is a rapidly progressive, potentially fatal infection that requires immediate diagnosis and intervention. It may arise from tooth extraction complications, especially in patients with chronic diseases or immune suppression. Considering that, the oral surgeon must include this condition within post-extraction infections differential diagnosis, where a multidisciplinary approach is fundamental for diagnosing and establishing an appropriate treatment.

This study has some limitations, such as the absence of reported CNF cases in patients with immunosuppressive-associated myeloproliferative disorders. Cases of rare conditions must be reported to orient other

professionals regarding the appropriate diagnosis and management.

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