Noma in a Pacient whitout Sistemic Involvement

Noma em Paciente sem Envolvimento Sistêmico

Noma en un Paciente sin Participación Sistémica

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Abstract

Noma, also known as *Cancrum oris* or gangrenous stomatitis, is an infectious disease of bacterial origin, often in association with other microorganisms, affecting orofacial tissues and manifesting through a sequence of precursors. It begins as necrotizing gingivitis and progresses to necrotizing periodontitis and necrotizing stomatitis. In advanced stages, it can lead to facial mutilations, difficulty in eating, and psychological distress due to social stigma. Untreated, it can result in a mortality rate of up to 90%, mainly due to septicemia, dehydration, and malnutrition. When addressed early, the prognosis is favorable, and by treating its precursors, it is challenging to reach severe stages. The disease is often associated with children in impoverished social conditions, weakened immune systems, and malnutrition, especially in regions with low human development indices, such as sub-Saharan Africa, although some cases have been reported elsewhere. Treatment involves several phases, including debridement of necrotic areas, antibiotics, antimicrobial mouth rinses, and supportive periodontal therapy. Despite its rarity, infection can also occur in developed countries and affect adult patients, sometimes without underlying health conditions. The aim of this article is to report a clinical case of noma in a 31-year-old man, without systemic involvement, who sought treatment at the Maria Aparecida Pedrossian University Hospital in Campo Grande, Mato Grosso do Sul, Brazil, due to swelling in the submental region. After hospitalization, biofilm control and postoperative care, there was a satisfactory improvement in the clinical condition.

Keywords: Noma; Gingival necroses; Periodontitis.

Resumo

Noma, também conhecida como *Cancrum oris* ou estomatite gangrenosa, é uma doença infecciosa de origem bacteriana, frequentemente associada a outros microrganismos, afetando tecidos orofaciais e se manifestando por meio de uma sequência de precursores. Começa como gengivite necrosante e progride para periodontite necrosante e estomatite necrosante. Em estágios avançados, pode levar a mutilações faciais, dificuldade para comer e sofrimento psicológico devido ao estigma social. Não tratada, pode resultar em uma taxa de mortalidade de até 90%, principalmente devido à sepse, desidratação e desnutrição. Quando tratada precocemente, o prognóstico é favorável e, ao tratar seus precursores, dificilmente atinge estágios graves. A doença é frequentemente associada a crianças em condições sociais

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empobrecidas, sistemas imunológicos enfraquecidos e desnutrição, especialmente em regiões com baixos índices de desenvolvimento humano, como a África Sub-saariana, embora alguns casos tenham sido relatados em outros lugares. O tratamento envolve várias fases, incluindo desbridamento de áreas necróticas, antibióticos, enxaguatórios bucais antimicrobianos e terapia periodontal de suporte. Apesar da raridade, a infecção também pode ocorrer em países desenvolvidos e acometer pacientes adultos, às vezes sem con-dicões de saúde subjacentes. O objetivo deste artigo é relatar um caso clínico de noma em um homem de 31 anos, sem en-volvimento sistêmico, que procurou tratamento no Hospital Universitário Maria Aparecida Pedrossian, em Campo Grande, Mato Grosso do Sul, Brasil, devido a um aumento de volume na região submentoniana. Após internação, controle do bio-filme e cuidados pós-operatórios, houve melhora satisfatória do quadro clínico.

Palavras-chave: Noma; Necrose gengival; Periodontite.

Resumen

La noma, también conocida como Cancrum oris o estomatitis gangrenosa, es una enfermedad infecciosa de origen bacteri-ano, frecuentemente asociada a otros microorganismos, que afecta los tejidos orofaciales y se manifiesta a través de una secuencia de precursores. Comienza como gingivitis necrosante y progresa a periodontitis necrosante y estomatitis necrosante. En etapas avanzadas, puede provocar mutilaciones faciales, dificultad para comer y malestar psicológico debido al estigma social. Sin tratamiento, puede resultar en una tasa de mortalidad de hasta el 90%, principalmente por septicemia, deshid-ratación y desnutrición. Cuando se aborda de forma temprana, el pronóstico es favorable y, al tratar sus precursores, es difícil llegar a etapas graves. La enfermedad suele asociarse a niños en condiciones sociales empobrecidas, sistemas inmunitarios debilitados y desnutrición, especialmente en regiones con bajos índices de desarrollo humano, como el África subsahariana, aunque se han reportado algunos casos en otros lugares. El tratamiento implica varias fases, que incluyen el desbridamiento de las áreas necróticas, antibióticos, enjuagues bucales antimicrobianos y terapia periodontal de apoyo. A pesar de su rareza, la infección también puede ocurrir en países desarrollados y afectar a pacientes adultos, a veces sin condiciones de salud subyacentes. El objetivo de este artículo es informar un caso clínico de NOMA en un hombre de 31 años, sin compromiso sistémico, que buscó tratamiento en el Hospital Universitario Maria Aparecida Pedrossian en Campo Grande, Mato Grosso do Sul, Brasil, debido a hinchazón en la región submentoniana. Después de la hospitalización, el control del biofilm y el cuidado posoperatorio, hubo una mejoría satisfactoria en el estado clínico. Palabras clave: Noma; Necrosis gingival; Periodontitis.

1. Introduction

Noma, from the Greek "to devour", also known as oral cancer and/or gangrenous stomatitis, is an infectious necrotizing periodontal disease of bacterial origin, which affects orofacial tissues (Whiteson et al., 2014). Rapidly evolving, its manifestation occurs through bacterial interaction and is followed by necrotizing gingivitis, in which the affected region is restricted to the gums, progressing to necrotizing periodontitis, with involvement of the supporting periodontium, and necrotizing stomatitis, when it already affects muscle tissue, nervous and epithelial and there are systemic implications such as fever and anorexia (Ogunleye et al., 2022; Gasner & Schure, 2024). In addition to tissue injury, there is also the presence of a foul odor, pain and psychological impairment due to facial deformation. When the disease spreads and reaches more advanced stages, in the acute phase, with systemic involvement and tissue loss, with bone exposure and sequestration and gangrene, the mortality rate is high and, if left untreated, can reach 90% of cases, due to septicemia, dehydration and starvation (WHO, 2017).

The disease mostly affects children between 2 and 6 years of age on the margins of society, that is, exposed to precarious social conditions, often malnourished, living in extreme poverty, poor hygiene, with compromised immune systems and affected by other diseases such as measles, malaria and AIDS, being associated with countries with a low level of human development. There are reports of its manifestation in countries in Latin America and Asia, but they are rare (WHO, 2017; Khammissa et al., 2022; Feller et al., 2022). In sub-human conditions, the disease has been described in cases of war, in which poor nutrition, hypovitaminosis, psychological stress, infections with poor hygiene present, in concentration camp prisoners, usually children, and in soldiers of the First World War (Nolte, 2021; Tkacz et al., 2021).

According to the World Health Organization (WHO), the disease follows five stages of evolution, namely: 1- acute necrotizing gingivitis; 2- edema; 3- gangrene; 4- healing and 5- sequelae. The warning sign that precedes the first stage is simple gingivitis, when there is bleeding when touched and/or brushed and the gums have a reddish color with edema, denoting inflammation. During stages one (when there are painful ulcerated lesions, inversion of papillae and bad breath) and two (spread ulcerated lesions, edema, high fever and lymphadenopathy) the condition is reversible, with no physical sequelae. In stages three (tissue destruction, presence of fenestrations in necrotic areas, gangrene, difficulty feeding, anorexia and apathy), four (jaw constriction, tooth loss, bone exposure and beginning of healing) and five (disfigurement, fusion of structures jaws, tooth displacement, nasal regurgitation), there are physical consequences, such as facial mutilations and difficulty speaking and eating, in addition to psychological damage, such as difficulty in acceptance and social stigmatization (WHO, 2017; Dholam et al., 2022).

When the precursors of Noma are identified early, its prognosis is favorable, intervening as soon as possible (Masipa et al., 2013). To this end, the treatment is carried out in four phases. Immediately, debridement of the necrotic regions must be carried out with irrigation of 10% PVPI and saline solution, installation of drains, if necessary, and combination of medications (systemic antibiotics, corticosteroids, and analgesics). The second phase includes the use of antimicrobial mouthwashes, such as 0.12% chlorhexidine, due to its broad spectrum of action, in addition to control through gentle brushing to disorganize bacterial plaque. The third phase consists of monitoring the patient, which must be done after a period of 24 hours for new debridement, if necessary, monitoring drainage flow and carrying out scaling and root planning procedures to control infectious foci. The fourth phase covers supportive periodontal therapy, where control of the oral environment is carried out, validating the need for monthly monitoring and reaffirming hygiene instructions. When there are physical sequelae, such as mutilation and difficulty speaking and eating, there is also the possibility of using maxillofacial prostheses, specifically made to ensure greater comfort and dignity for the patient (Dholam et al., 2022).

Despite the rarity of cases, and their greater concentration in underserved children in underdeveloped countries, there are reports in adults and even in first world countries, such as the USA (Maley et al., 2014). Furthermore, the association of the disease with malnutrition and comorbidities, despite being a sufficient condition, in some situations is not a necessary condition (Feller et al., 2019). In this article, we will report a clinical case of Noma diagnosed in a 31-year-old male patient, without comorbidities, who attended the Hospital Universitario Maria Aparecida Pedrossian, in the city of Campo Grande- MS, Brazil, complaining of increased volume in the region submental.

2. Methodology and Results

This case is a report with descriptive, exploratory purposes and a qualitative approach (Pereira et al., 2018), show its clinical relevance and facilitate research and new reports with the same theme, always based on evidence. This case is a report with descriptive, exploratory purposes and a qualitative approach, in order to show its clinical relevance and facilitate research and new reports with the same theme, always based on evidence. A 31-year-old male patient was admitted to the hospital, reporting an increase in volume in the submental region followed by the formation of a cutaneous fistula. Patient denied allergies and/or controlled medications, claimed to be a smoker and social drinker. On extraoral physical examination, there was a significant increase in volume below the chin, redness and active fistulas with drainage of purulent exudate. On intraoral physical examination, poor hygiene was observed, large accumulation of dental calculus in the lower arch, drainage of purulent exudate in the periodontal region of element 43, accompanied by a foul odor (Figure 1).

Figure 1 - A- Initial extraoral appearance of the patient. B- Drainage of purulent exudate and presence of fistulas in the submental region.



Source: Authors.

After the physical examination was carried out and the patient's social conditions and history were assessed, the hypothesis of a diagnosis suggestive of cellulitis of odontogenic origin was raised. Based on this information, the patient underwent imaging and laboratory tests for systemic evaluation and extra and intraoral drainage procedures were proposed, under local anesthesia. To this end, submental access was performed, in the fistula region, and divulsion through planes, where bone exposure and communication of the region with the intraoral environment were perceived. In addition, a flap was created in the alveolar mucosa to visualize the subgingival region and scrape in an open field. There was drainage, followed by debridement of the wounds, washing with PVPI and saline solution and, finally, installation of Penrose drains, to continue the exudate elimination process until control of the condition, presented 48 hours after its installation (Figures 2 and 3).

Figure 2 - Panoramic radiography.



Source: Authors.

Figure 3 - A- Large amount of calculus on tooth surfaces. B- Tissue necrosis in the mental region.



Source: Authors.

After the procedure, the patient was hospitalized for a period of five days, where he was under observation by the maxillofacial surgery and traumatology team, and underwent intravenous administration of antibiotics (clindamycin 600mg every 8 hours and metronidazole 500mg every 8 hours), during every day of hospitalization, in addition to all dental assistance, supra and subgingival scrapings in the lower sextants, dressing changes and assessment of the progression of the clinical

condition. In relation to the tests carried out, there was no systemically important finding that would characterize impairment consistent with the presented condition.

On extraoral examination, on the third day after intervention, drainage of purulent exudate was scarce, edema and redness were moderate and the patient reported no pain to touch. During the intraoral examination, drains were in position, but there was no drainage, oral hygiene was still precarious, accompanied by spontaneous and generalized gingival bleeding and a foul odor. On the fourth day of hospitalization, the drains were removed and the patient was kept under observation until the following day, when he was discharged with no purulent drainage, no edema, redness or even pain to the touch. During the intraoral examination, poor hygiene, sutures in position and lack of drainage were observed.

The patient was notified and instructed about the necessary care to maintain hygiene and was discharged. To this end, antibiotic medication was prescribed for 7 days (metronidazole + clindamycin) and return scheduled for one week after discharge (Figure 4). The patient also had two meetings with the team to control his condition, assessing the need to maintain non-surgical periodontal treatment, as he continued to present stones in all sextants and this could lead to new inflammatory and infectious processes (Figure 4). Unfortunately, after the last two contacts, the patient abandoned treatment and did not carry out the proposed procedures to control his periodontal health in the long term.

Figure 4 - A- Improvement of periodontal condition after 12 days of intervention, requiring non-surgical periodontal control; B-Good scar appearance in the submental region. C- Intraoral appearance after three weeks of intervention was within normal limits, requiring non-surgical periodontal control; D- Clinical aspect of healing of the submental region within normal limits.



Source: Authors.

3. Discussion

Noma is known as a polymicrobial condition that affects the oral tissues, whose poor nutrition and factors such as lack of basic hygiene are readily recognized (Singh et al., 2024). With the advancement of antimicrobial therapy, there was a decrease in the mortality rate of noma cases, to 8% of cases (Feller et al., 2019). It is necessary to highlight the importance of identifying precursors while still having time to stabilize the infection and provide a much more favorable prognosis. When there is thorough monitoring, identifying any sign of changes in the oral cavity and treating not only the symptoms, but also the cause, the chance of developing a serious infectious condition, often with lifelong consequences, is low (Masipa et al., 2013). Therefore, early diagnosis and treatment are the only way to prevent the progression of the disease, reduce its morbidity and mortality, and

consequent severe facial disfigurement.

Thus, in order to identify the signs and symptoms related to each phase of the disease, it was necessary to standardize the stages of evolution (acute necrotizing gingivitis, edema, gangrene, scarring and sequelae), designed by the WHO, with the aim of guiding health professionals and population. This information helps professionals to define the stage in which each patient is and what measures are necessary to be taken based on this to improve their clinical condition. Despite this, there are articles that consider this current classification method to be incorrect, ending up facilitating the incidence of false positive diagnoses. These same studies suggest a more practical and more assertive classification, comprising only two stages of evolution: 1- acute noma; 2- arrested noma (Feller et al., 2019).

Based on the World Health Organization guidelines for noma in 2016, the ideal standard of care is to start antibiotic therapy as early as possible. Just as the patient in this report has already been treated with antibiotics to treat the lesion, progressing with reduction and control of the infectious condition. The microbiota present in periodontal disease is, for the most part, composed of obligatory and facultative anaerobic bacteria, such as the genera *Fusobacterium, Porphyromonas, Prevotella, Treponema, Aggregatibacter*, among others (Ahner et al., 2014). There is no single species responsible for the manifestation of the infection. There is an increased presence of some microorganisms such as *Fusobacterium necrophorum*, found in advanced stages of noma, and *Prevotella intermedia*, which worsens the clinical picture through the degradation of proteins and lipids, but they are not considered sufficient factors for the manifestation in isolation. There is a decrease in the proportion of *Capnocytophaga sp, Neisseria sp* and *Spirochaeta sp* (Baratti-Mayer et al., 2013). Furthermore, there is a relationship with colonization by fungi and viruses associated with the disease.

Due to the inherent characteristics of mixed infection and the unbalanced and complex composition of the altered microbiota, the administration of broad-spectrum antibiotics is necessary. Due to the predominance of gram negative anaerobic bacteria, the first choice antibiotic, as an adjunct to clinical procedures, is Metronidazole, a nitroimidazole that acts by interrupting the genetic replication of these microorganisms through the fragmentation of their deoxyribonucleic acid (DNA) (Ahern et al., 2014; Schneidereit et al., 2023) and can also be combined with another, which has a similar action and acts on other species, such as clindamycin, from the lincosamide class, administered in the case presented, since, like metronidazole, it promotes bactericidal action, depending on its concentration , and also has a broad spectrum of action, this time acting on positive and negative Grams (Luchian et al., 2021; Brandão et al., 2022). There is no standardization regarding the medication protocol and dose to be used, however, as it is a very serious infection, we opted for a higher dosage. discharge of medications administered. Therefore, in this clinical case, the approach was Metronidazole 500mg every 8 hours + Clindamycin 600mg every 6 hours during the 5 days of hospitalization and for a further 7 days after hospital discharge, under monitoring to assess the need to continue or interrupt the therapy.

Despite the aggressiveness of the acute stage of the infection and its high mortality, noma is preventable and there is a cure without sequelae, as long as it is diagnosed early and treated promptly. Affecting people, mainly children, in extreme poverty, malnourished, in unsanitary conditions and, often, without access to vaccines and information, the disease has not yet entered the category of Neglected Tropical Disease (NTD), which could change the course life of hundreds of thousands of affected patients (Feller et al., 2019). It has been demanded, several times over the last few years, that the WHO include it in the list of NTDs and, thus, public policies could be intensified with the aim of early diagnosis of both Noma and its precursors, providing effective treatment in a universal manner, and also widely disseminate knowledge about its severity in order to reduce its incidence and prevalence through prevention, by raising awareness among the hitherto vulnerable population (Engels & Zhou, 2020).

In line with this, it becomes evident, as demonstrated by Silva et al. (2020), the eminent need for knowledge about this

pathology, both in conditions of local and systemic aspects more commonly found, but especially in situations of an unusual nature as in the case presented.

4. Conclusion

The case presented culminated in an assertive treatment resolution, with a favorable prognosis, mainly due to the early approach. Although the characteristics are aggressive in the acute stage of the infection and have a high mortality rate, noma is curable and can be avoided without consequences, as long as it is diagnosed and treated early. The knowledge of dentists and other health professionals is an important factor that adds to the treatment of this disease. The development of actions that can promote the prevention and early diagnosis of the disease, through primary care health programs, minimizing the rapid development and reducing the lethality of noma, greatly favors oral health and its maintenance in the population.

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