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Noma (Cancrum Oris): Report of a Mexican Case.

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Abstract:

"Cancrum oris" (noma) is a severe necrotizing infectious disease that typically begins as a localized ulcerative lesion of the oral mucosa and rapidly progresses to cause extensive destruction of the orofacial tissues. It predominantly affects individuals with compromised immunity and is strongly associated with malnutrition, poor oral hygiene, and systemic conditions such as HIV infection. We presented the case of a 44-year-old Mexican woman, living with untreated HIV infection, who presented with clinical features consistent with noma in Mexico City. Her management required a coordinated multidisciplinary approach. Although noma is a rare condition in the Americas, it remains a significant public health concern in sub-Saharan Africa. Identified risk factors include extreme poverty, severe malnutrition, inadequate sanitation, and underlying immunosuppressive states such as HIV/AIDS. Histopathological findings demonstrated sharply demarcated necrosis of the surface epithelium, accompanied by peripheral acanthosis and cytoplasmic ballooning of marginal epithelial cells. The underlying dermis showed marked edema with separation of collagen bundles, a dense polymorphonuclear neutrophilic infiltrate, and dilated blood vessels filled with inflammatory cells. Numerous phagocytosed microorganisms were identified, including Gram-negative cocco-bacilli and cocci of variable morphology. Is a polymicrobial infection, the most common pathogen identified is *Fusobacterium necrophorum*, the synergistic action of multiple anaerobic and facultative organisms, along with their virulence factors, contributes to the fulminant tissue destruction characteristic of the disease.

Treatment involves aggressive surgical debridement, antibiotic therapy, and addressing nutritional deficiencies.

Noma is a preventable but neglected disease that primarily affects impoverished populations. This case represents both a diagnostic and therapeutic challenge, and to our knowledge, constitutes the first documented instance of adult-onset noma in Mexico. A previous report from 1991 described a pediatric case, but no cases involving adults have been published in the Mexican medical literature to date.

Keywords: cancrum oris, necrotizing gingivitis, noma

Statement of Clinical Significance

I am pleased to submit an original case report entitled “Noma (Cancrum Oris): Report of a Mexican Case” for consideration for publication in the Journal of oral diagnosis.

We believe that this manuscript is particularly suitable for publication in the Journal of Oral Diagnosis, given the relevance of understanding this pathology for medical doctors and dental surgeons. Early identification, proper recognition, and timely intervention can significantly alter the patient's prognosis. Additionally, it is important to note that no similar cases from Mexico have been documented in the current literature. We are pleased to present this work as a case report, covering the entire process from the initial diagnosis to the final follow-up of the patient.

Introduction

“Cancrum oris” is a Latin term for a necrotizing infectious disease that starts as a sore in an infected individual’s mouth.[1] The term "noma" signifies 'to devour', illustrating the

disease's quickly progressing and aggressive 'tissue-eating' characteristics [1,2]. Most commonly reported in sub-Saharan Africa—particularly within the so-called “Noma Belt” (e.g., Niger, Nigeria, Senegal, Burkina Faso). Although cases have also been described in Asia and the Americas, its global incidence remains uncertain, with estimates ranging from 14,000 to 30,000 cases annually. Its clinical relevance lies in its rapid progression, high morbidity, and strong association with extreme poverty, malnutrition, and immunosuppression. We report a severe case of noma in an untreated HIV-positive woman in Mexico City.

Case report

A 44-year-old Mexican woman, malnourished and diagnosed with HIV nine years earlier, without current treatment (viral load: 141,371 and CD4 count: 47 cells/ml), presented to the emergency room with a 15-day history of severe lower gingival pain, a mass in the lower vestibular sulcus and sudden tooth loss, accompanied by edema and a red-violet coloration of the inferior lip. (Figure 1)

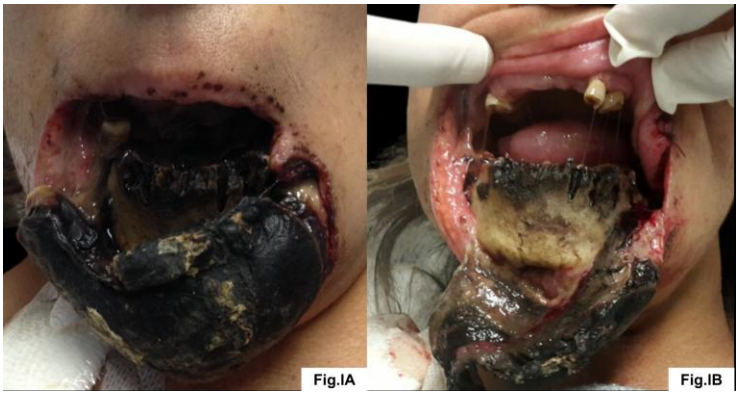

Figure I. Extensive tissue necrosis, ulceration, and denuded areas of the lower lip and gingiva with exposed jawbone.

During clinical evaluation, she showed necrosis of the lower lip, sloughing, drooling, purulent discharge, foul odor. And the presence of systemic symptoms such as fever and deterioration of the general condition.

Wound culture during hospitalization identified *E. coli* and *Fusobacterium necrophorum* resistant to ampicillin, cefazolin, ceftazidime, ceftriaxone, and trimethoprim/sulfamethoxazole. The lumbar puncture showed turbid, frothy cerebrospinal fluid with a glucose content of 24 mg/dL and a protein level of 77 mg/dL, the stains for India ink, Ziehl-Neelsen were negative.

A biopsy of the lesion was performed and revealed extensive coagulative necrosis of the epidermis, dermis, and subcutaneous cellular tissue (Figura 2a), In the superficial and mild corium there were dilated vascular capillaries, in addition to a perivascular inflammatory infiltrate composed of histiocytes, lymphocytes, neutrophils and plasma cells (Figure 2b).

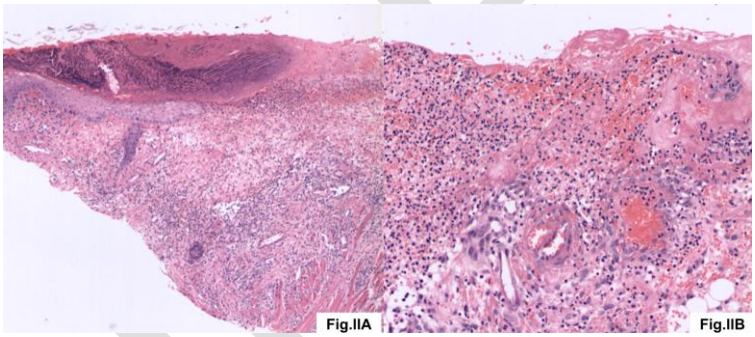

Figure II. IIA. HyE 10x. Extensive coagulative necrosis of epidermis, dermis and subcutaneous cellular tissue. IIB. HyE 40x. Superficial and mild corium exhibit dilated capillaries, in addition superficial and interstitial perivascular infiltrate composed histiocytes, lymphocytes, polymorphonuclear and plasma cells.

PAS and Grocott stains revealed the presence of hyphal structures consistent with *Candida* spp. Gram positive and negative cocci-bacilli were identified, and the Ziehl-Neelsen test yielded a negative result for acid-fast bacilli.

A multidisciplinary treatment approach was performed, with debridement, drainage of sublingual abscess, and partial anterior mandibulectomy. The patient progressed favorably with a good response, prompting consultation with plastic surgery for subtotal mandibulectomy of the entire necrotic segment from the right ascending ramus to the left condyle, followed by reconstruction with a microvascular fibular osteocutaneous flap, accompanied by multiple osteotomies. The fibular vessels were anastomosed to the external carotid artery and external jugular vein at the level of the neck on the left side (Figure 3).The patient was discharged after 114 days of hospitalization due to significant clinical improvement (Figure 4).

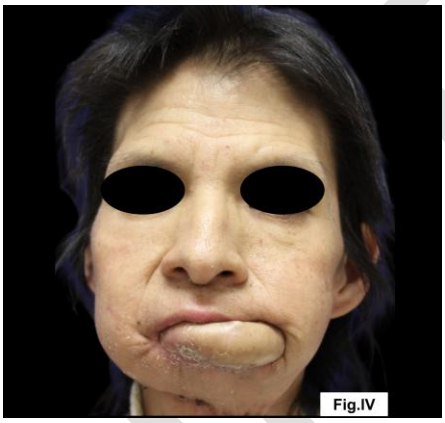

Figure IV. Photograph taken 14 days after mandibular reconstruction.

Although follow-up was planned, long-term monitoring beyond six months was not possible. Multiple attempts were made to contact the patient post-discharge in order to assess her long-term clinical and functional outcomes; however, these efforts were unsuccessful. This lack of extended follow-up represents a limitation of the present report.

Discussion

Although Noma is a very infrequent infectious disease, it is commonly seen in sub-Saharan Africa, with only some cases reported in Asia and the Americas. Reports from Latin America are particularly scarce, underscoring the relevance of the present case. Global incidence is unknown, but it is estimated to affect 14,000-30,000 individuals per year.[3] Most cases of Noma worldwide occur in countries such as Niger, Nigeria, Senegal, and Burkina Faso, which is known as the "Noma Belt" region. [4] To our knowledge, only isolated cases have been described in countries such as Brazil and Peru, primarily in the pediatric population and often associated with comorbidities. This case contributes to the limited body of literature on noma in adults outside of the African context, particularly within Mexico, highlights the need for increased awareness, documentation in non-endemic regions and the importance of clinical awareness and early recognition of this neglected disease, as timely diagnosis and intervention are critical to preventing severe complications and improving patient outcomes.

Risk factors include poverty, poor sanitation, deficient oral hygiene, limited access to health care services, immunosuppressive conditions (HIV infection, type 2 diabetes mellitus), chronic malnutrition, and is commonly associated with very low levels of most essential amino acids and significant reductions in plasma levels of vitamin A, vitamin C, zinc, albumin, and hemoglobin. [1,2,3,4,5, 6]

Noma may start in the mouth as a bacteria-induced necrotizing gingivitis that progresses to necrotizing periodontitis and then to necrotizing stomatitis. The exact causative agent is unknown, but it is thought to be multifactorial; frequently identified microorganisms include *Treponema vicentii*, other spirochetes or fusiform bacteria, *Staphylococcus*

aureus, α-hemolytic Streptococcus, and *Pseudomonas.* [5,7,8] *Fusobacterium necrophorum* has been observed to be the most frequent pathogen identified and the predominant microorganism for the development of Noma. Additionally, *Prevotella intermedia* has been recently reported to grow in several cultures[9].

Virulence factors *of F. necrophorum* include the capacity to synthetize proteolytic enzymes and toxins capable of tissue destruction; and to produce a growth stimulating factor for *P. intermedia*, which facilitates lipid degradation. [10]

Noma starts as a polybacterial infection, provoking necrotizing gingivitis, characterized by bleeding, marginal gingival necrosis, ulceration, and pain. Owing to local immune suppression, bacterial virulence factors, and HIV infection that induced immunosuppression targets T cells, inducing apoptosis through viral replication. This leads to colonization of lymphoid organs and destruction of their architecture, causing loss of TCD4 precursor cells (thymic invasion) and failure of the mononuclear phagocytic system.[11]

The necrotizing process may spread, extending from the marginal gingiva into the periodontal attachment apparatus to become necrotizing periodontitis and subsequently affect the adjacent mucosa, leading to necrotizing stomatitis, which is characterized by the destruction of oral soft tissues and bone. If left untreated, the necrosis may spread rapidly to the buccal mucosa and lip with subsequent full-thickness destruction of the epitheliums, muscles, and skin. The affected tissues become macerated, friable, swollen, and turn into black color. They rapidly slough leaving an extensive irregular defect, referred to as Noma. The course of the disease is very rapid, with progression from necrotizing stomatitis to full-thickness destruction in just a few days. [12]

The World Health Organization has classified Noma into five stages, the first stages (Stage 0 to 4) are the acute stages of Noma lasting only a few weeks. They include Stage 0: simple gingivitis; Stage 1: acute necrotizing gingivitis characterized by marginal gingival necrosis, bleeding, and pain; Stage 2: plus edema; Stage 3: gangrene that is an extension of necrotizing gingivitis into the periodontal attachment apparatus with progressive loss of affected teeth and necrotizing stomatitis is an extension of necrotizing periodontitis beyond the mucogingival junction with the necrotizing inflammatory process spreading into labial, buccal, lingual or palatal mucosa for example we can classify our patient in this stage. Stage 4: scarring, and Stage 5: sequelae. [7,13,14,15]

Symptoms and signs associated with the acute phase of Noma include mouth soreness, facial swelling, fetid breath, excessive salivation, difficulty eating, fever, and tachycardia. [16,17]

The diagnosis of Noma is primarily clinical and is characterized by a rapidly progressive gangrenous process affecting the orofacial tissues. Histopathological studies generally demonstrate necrosis of the surface epithelium with acanthosis around its borders. The necrotic area is well circumscribed; at the margins, the epithelial cells show cytoplasmic ballooning. The corium and dermis have a dense infiltrate of polymorphonuclear neutrophils and phagocytosed bacteria may be observed. Separation of collagen bundles due to edema is a common feature. Blood vessels are usually dilated and filled with inflammatory cells. Small and large cocci of various morphologies and Gram-negative cocco-bacilli may be seen. [17,18]

It is essential to consider a broad differential diagnosis that includes other ulcerative or necrotizing conditions such as ecthyma gangrenosum, mycobacterial infections (e.g.,

Mycobacterium tuberculosis, *Mycobacterium leprae*), mucocutaneous leishmaniasis, necrotizing fasciitis, oral and maxillofacial myiasis, osteonecrosis of the jaw, syphilis, and certain oral malignancies. In this case, mucocutaneous leishmaniasis was ruled out through negative parasitological smears and the absence of travel to endemic regions. Necrotizing fasciitis was excluded based on the lesion's confinement to the orofacial region without fascial plane involvement on imaging. [18,19]

Treatment focuses on halting the acute infection and addressing underlying nutritional deficiencies and hydration. Management includes tissue debridement, removal of necrotic bone, antibiotic therapy targeting anaerobic gram-negative microorganisms, wound care, and replacement of fluids, electrolytes, and good nutrition. [23]

The surgical treatment goal is to improve cosmetic appearance, as well as the recovery of oral functions like eating, drinking, swallowing, and speech. [18,19]

The patient underwent complete surgical reconstruction of the affected areas (Figure 3).

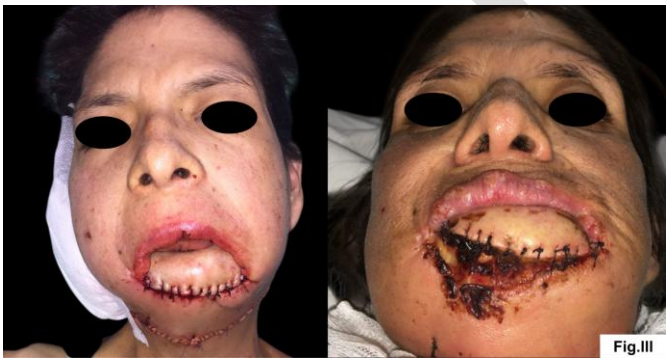

Figure III. Post surgical photograph.

VAC therapy was used following surgical intervention for preventing infection, with no evidence of wound dehiscence, complemented with piperacillin-tazobactam treatment post-operatively. The patient was monitored closely during her hospitalization and experienced no complications. She was discharged with clinical improvement and followed in outpatient care for six months, after which we lost contact for further follow-up.

Several factors contributed to the patient's survival, including prompt identification of the disease, effective management of underlying comorbidities, a multidisciplinary treatment approach combining broad-spectrum antibiotic therapy, and timely surgical debridement and reconstruction.

Conclusion

Noma is a preventable disease that primarily affects young children from vulnerable and impoverished communities, most of them from the African continent. In the literature, it is described as a neglected disease with significant gaps in knowledge, a devastating and fatal condition that requires urgent and intensive clinical and surgical management.

We consider this patient a case of diagnostic and treatment challenge and an opportunity to raise awareness of the disease in a population out of sub-Saharan Africa. To our knowledge, this is the first report of a Noma patient in Mexico. Understanding predisposing factors and the physiopathology of the disease will help to provide effective, targeted interventions to reduce the burden of this condition in the affected individuals.

Ethics Approval

Ethical approval is not required for this study in accordance with local or national guidelines. Written informed consent was obtained from the patients for publication of this case report and any accompanying images.

Disclosure Statement

All authors have no conflict of interest to disclose.

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Author Contribution Statement

Figueroa Hernández María Fernanda, Macedo Pérez Marysol and Mosqueda Taylor Adalberto wrote the original draft in support with Vega Memije María Elisa and Ramírez-Hinojosa Juan Pablo. Berumen Glinz Cristina and Santamaria Linares Eric supervised the manuscript. Toussaint Caire Sonia contributed to the histopathological photographs and diagnosis. All the authors were responsible for the design and critical revision of the manuscript. All authors approve the final version of the manuscript for submission.

Data Availability Statement

All data generated or analyzed during this study are included in this article. Further inquiries can be directed to the corresponding author.

Competing interests: None.

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