Case Report

Necrotizing Stomatitis in a Non-HIV Patient: A Case Presentation

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Abstract

Necrotizing stomatitis (NS) is an uncommon oral ulceration associated with reduced immunity, recent illness, and unusual life stress. It can be seen in malnourished children and HIV-infected adults, but it is very uncommon in non-HIV adults. Objective. To present a case of NS in a non-HIV adult. Methodology. We present a previously misdiagnosed case of NS in a non-HIV 27-year-old female whose immunity was compromised by several factors including malnutrition, chronic anemia, drug treatment of rheumatoid arthritis, and fibrous TMJ ankylosis. Several factors mitigating the diagnosis and successful treatment of this patient are discussed. Conclusion. This presentation highlights a challenging case of NS complicated by several unusual factors. It serves to educate the dental practitioners on the interplay of factors which if not addressed will mitigate successful management of the patient.

Keywords necrotizing stomatitis; malnutrition; anemia

1. Introduction

Necrotizing stomatitis (NS) is an inflammatory disease of the oral cavity characterized by rapid destruction of epithelium, connective tissue, and dental papillae [3].

It can be defined as a painful ulceronecrotic lesion of the oral mucosa which may expose underlying bone [17] and extend into contiguous tissues [7].

It may be a sequelae of necrotising gingivitis or necrotising periodontitis or may arise on the oral mucosa separately from the periodontium [10,13]. It involves mainly the soft tissues but may extend into the underlying bones, causing massive tissue destruction and oro-antral fistula. If the necrotic process spreads through the oral mucosa to the facial skin, it will result in cancrum oris (NOMA) [4,5,10].

The predisposing factors include anemia, malnutrition, physical and emotional stress, anemia, poor nutrition, smoking, recent illness (example, measles), agranulocytosis or neutropenia, chemotherapy, and immunosuppression [3,15].

The pathogenesis of NS is multifactorial. It includes specific micro-organisms which are normal commensals that multiplied and become virulent when there is impairment of the host immune system. Fusiform bacteria (example, Fusobacterium necrophorum), Spirochettes (example, Borrelia species) [2,14,17], and Pseudomonas aeruginosa have been implicated; likewise herpes viruses which result in local immune suppression leading to overgrowth of microbacteria [9,16] and Candida which produces eicosanoids and these induce host release of arachidonic acid, and other pro-inflammatory mediators that promote fungal colonization and invasiveness, by shifting T-helper 1/T-helper 2 (TH1/TH2) balance of cellular immunity to humoral immunity that is less effective in combating fungus. Such interference in the local immune and inflammatory response in the oral soft tissues has the potential to contribute to the development of necrotic oral lesions [11].

The clinical presentation includes fever, cervical lymphadenopathy, foul/fetid breath, painful ulcer with necrotic base, and dysphagia. In advanced state, there could be sequestratum formation.

The treatment is very effective and is similar to the treatment of necrotising ulcerative gingivitis/periodontitis. The treatment modality include counseling and reassurance; oral toileting—debridement, irrigation, and oral hygiene instructions; antibiotics—metronidazoles; analgesics—paracetamol; mouthwashes—chlorhexidine/hexedene; and supportive therapy—folic acid, B-complex (Bco), vitamin (Vit) C. Recall visits are important to monitor progress of treatment and institute rehabilitation of any lost tissue where necessary.

2. Case report

A 27-year-old female patient presented at the Oral Medicine Clinic with a chief complaint of pain and sores in her mouth.

Patient noticed an ulcer with associated pain on the left cheek about four years ago. She presented at the Dental clinic of a General Hospital (GH) 2 years ago after visiting several clinics and hospitals with no improvement. She was treated with antibiotics and incisional biopsy taken at the GH. A diagnosis of chronic non-specific ulcer was made. There was an initial period of remission, but there was a recurrence of the lesion 6 months later which involved the soft palate.
She then presented at the Oral Medicine Clinic, Department of Preventive Dentistry, Lagos University Teaching Hospital (LUTH) with large oral ulcers involving the soft palate and left buccal mucosa. There was an associated severe pain, fever, malaise, dysphagia, and difficulty in speech.

She is a known rheumatoid arthritis, gastric, and duodenal ulcer patient. Her father and two of her sisters also had rheumatoid arthritis. She was single and unemployed; she neither took alcohol nor smoked cigarette.

On examination, an ill-looking young lady in distress, very pale and febrile, was seen. There was no facial asymmetry. Submandibular lymph nodes were slightly enlarged and tender. On palpating the temporomandibular joint, there was no tenderness, crepitus, nor clicking sound. The knee joints were swollen bilaterally and were tender.

Intra-orally, the mouth opening was 15 mm (inter-incisal distance). Oral hygiene was poor with marked halitosis. Large ulcers with overlying white sloughs were seen on the soft palate (Figure 1) and left buccal mucosa. Fibrous band was also noticed on the buccal mucosa. The actual size of the lesion could not be ascertained due to limited mouth opening.

Patient had been on these medications at various times—Oruvail, Naproxen, Cerebrex, Tramadol, Spacibact, hydrogen peroxide mouthrinses, and Augmentin. She had been on the non-steroidal anti-inflammatory drugs (NSAIDs) for about 6 years for the treatment of rheumatoid arthritis.

Clinical impressions of chronic ulcer with overlying candidiasis, fibrous ankylosis of the Temporomandibular joint (TMJ) secondary to chronic ulcer, and anemia were made. Full blood count with differentials, erythrocyte sedimentation rate (ESR), exfoliative cytology for Candida, TMJ views (open and close), and Human immunodeficiency virus (HIV) test were requested. The patient was counseled and re-assured and was placed on preliminary treatment of chlorhexidine alternated with warm saline mouth wash, miconazole self-adhesive tablet, ferrous sulfate one tablet, 3 times daily (i tab tds), Vit C 300 mg tds, Bco i tab tds, folic acid i tab daily. She was given a week (1/52) for review.

A week after, the patient came with the result of hemoglobin (Hb) of 6.7 g/dl, packed cell volume (PCV)—21.3%, platelets—176,000, ESR—60 mm/h, Polymorphs—34%, lymphocyte—60%, monocyte—4%, basophil—0%, eosinophil—2%, HIV (Elisa) test—negative for HIV I and II, exfoliative cytology—negative of Candida, and TMJ views (open and close) which revealed no pathology of the joint. She informed us that the investigations were done before commencing the therapy and also complained of heart burn.

On examination, palatal and left buccal lesions have cleared considerably, and a necrotic ulcer on the right buccal mucosa was seen with a characteristic oral malodour (Figure 2). A diagnosis of NS secondary to severe anemia and malnutrition was made.

She was advised on proper/balanced diet—protein-rich food, examples, fish, meat (mash/chop/blend for easy swallowing), fruits, and vegetables—and was referred to oral surgery for excision of the fibrous band on the buccal mucosa. Hb and PCV were to be repeated in about 2 weeks.

She was placed on hydrogen peroxide (1:3 dilution) alternated with 0.2% chlorhexidine mouth wash tds × 1/52, tabs metronidazole 400 mg tds × 1/52, syrup Asfyfer 10 mL tds × 3/52, tabs Vit C 300 mg tds for 3 weeks (3/52), tabs Folic acid i daily × 3/52, Tab Vit Bco i twice (bd) × 3/52, and Gelusil i 4 times daily (qds) × 1/52.

The second hematology result was—Hb—8 g/dL and PCV—24%. She was treated again in the same manner, and a referral letter was written to her rheumatologist for possible change of the NSAIDs.

On her next appointment in 2 weeks, she was better, and the lesion had healed considerably. The NSAIDs had been changed to low dose prednisolone, 5 mg daily (Figures 3 and 4).

However, the lesion re-occurred 3 weeks later (Figure 5). Emphasis was made again on proper diet, and she was placed on the supportive therapy, antibiotics; and oral...
Figure 3: The lesion has healed considerably on the palate.

Figure 4: The lesion has healed considerably on the right buccal mucosa.

Figure 5: The lesion re-occurred on the palate.

toileting was done. She was now scheduled for a week appointment. She was advised on the importance of the excision of the fibrous band.

Patient had not really been keeping to her appointments afterwards for various reasons, chiefly because of financial constraints. She could not adhere to the dietary instructions given, which was key in her management. This had led to the deterioration in her condition.

Presently, the mouth opening has worsened and is now about 12 mm. The intraoral ulcers have spread to the throat, making swallowing very difficult.

3. Discussion

NS is a localized, acute, rapidly destructive, ulcerative, and necrotizing lesion of the oral mucosa that exposes underlying bone and extends into contiguous tissues. Usually, the lesion extends from the periodontium into adjacent mucosa [6,12,17]. Progression of necrotizing periodontitis to NS may subsequently result in progressive osseous destruction with the development of oro-antral fistula and osteitis [8].

NS sometimes arises without apparent origin from necrotizing periodontal diseases, necrotizing gingivitis and necrotizing periodontitis [2,14] which is similar to this case where the lesion was seen on the palate and buccal mucosa.

A study by Buchanan et al. revealed NS in an HIV-negative malnourished 68-year-old man with occult hypothyroidism [3]. Malnutrition was the major underlying factor which was also seen in our patient because she was not feeding well.

Another case was a malnourished old patient who developed NS following chemotherapy for leukemia [15]. This patient had more than one factor contributing to the development of NS. Our case was not different because she was also malnourished, had fibrous ankylosis, and had with underlying rheumatoid arthritis where she was on NSAIDs.

NS has been reported in HIV seropositive as demonstrated by Feller et al. [6] and Agbelusi et al. [1]. In our own case, the patient was HIV negative with underlying medical conditions which would have lowered her immunity, predisposing her to NS. This is similar to the finding of Buchanan et al. [3] and Santos et al. [15].

A young thalassemic major patient placed on deferoxiprone to reduce iron overload developed agranulocytosis which resulted in NS [18]. This also demonstrated reduced immunity.

The diagnosis of the lesion was based on clinical presentation of necrotic foul-smelling ulcer in a malnourished and severely anaemic patient. This is in line with the findings of other authors like Buchanan et al. [3] and Agbelusi et al. [1]. A biopsy of NS was not needed because its usually not indicated since the histology is not pathognomonic [7]. The histological features of NS are usually non-specific. It usually includes surface ulceration covered by a fibropurulent membrane with acute or mixed inflammatory cell infiltrate and hyperemia of underlying lamina propria [10].

The patient was treated based on the clinical presentation was placed on antibiotics and mouth wash. She was encouraged to eat balanced diet. During follow-ups, the lesion was seen to be waning and waxing. Whenever she adhered to instructions, especially feeding well, the lesion disappeared.
Efforts were made through phone calls for recall visits, but patient always complained of financial constraints. Challenges were faced in her management as she was not gainfully employed and had financial difficulties. She could not maintain a balanced diet, which was very important in her management. The little money is channeled to her drugs and the chronicity of the underlying medical condition (crippling rheumatoid arthritis), mega dose of NSAIDS, limited mouth opening, making the lesion inaccessible; lesion spreading to the throat resulting in dysphagia and compromised food intake, leading to further malnutrition and anemia, thus creating a vicious circle.

4. Conclusion
We present NS in a non-HIV patient complicated by several factors. NS is an uncommon condition with limited literature about it. This led to the interest in this case and also its contribution to the body of knowledge. This case is unusual because it is chronic and slowly progressive unlike the acute rapidly progressive type seen in patients who are severely immunocompromised. There is no social security. Patient had to pay hospital bills, buy drugs and pay for transportation. The only support was from her family. She was not feeding well which was very important in her management. This case also serves to educate the dental practitioners on the interplay of factors which if not addressed will mitigate successful management of the patient.

References