Case Report

DOI: 10.21276/ijchmr.2016.3.4.xx

Intricacies In Investigating Necrotising Ulcerative Stomatitis- A Case Report

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Abstract

The term Necrotizing Ulcerative Stomatitis (NUS) is derived from the Greek word stoma, meaning "mouth", and the suffix –itis, meaning "inflammation." Hence, it is an inflammatory disease of the oral cavity characterized by the destruction of epithelium, connective tissue and papillae leading to extensive bone loss. NUS usually has a sudden onset, and progresses from the mildest form on the spectrum, necrotizing ulcerative gingivitis (NUG), to the successively more severe condition of necrotizing ulcerative periodontitis (NUP), leading to necrotizing ulcerative stomatitis and finally cancrum oris (noma), which is frequently fatal. The etiology is somewhat unclear, but it may involve a complex of fusobacterium nucleatum along with spirochetes, borrelia or treponema. Well recognised host factors known to predispose to NUS include HIV infection, diabetes mellitus, immunosuppressant medications, malnutrition, psychological and physical stress, smoking and genetic prdisposition. NUS is frequently present, in combination with one of the other factors. The clinical characteristics of NUS include ulcerated and necrotic marginal and papillary gingiva covered by a yellowish-white or grayish slough or "pseudo membrane", blunting and cratering of papillae, spontaneous bleeding or bleeding on probing, pain and fetid breath. With this being said, the intense oral pain accompanying NUS is what usually causes the patient to seek dental treatment. We hereby present a case of an emaciated 70 year old diabetic male who was diagnosed with necrotizing ulcerative stomatitis.

Key words: Necrotising Ulcerative Gingivitis; Periodontitis; Stomatitis

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This Article May Be Cited As : Manchanda As, Kaur N,Kaur N And Kaur A: Intricacies In Investigating Necrotising Ulcerative Stomatitis- A Case Report. Int J Com Health and Med Res 2016;2(4):xxxx

Article Recieved: 01-10-16 **Accepted On:** 5-10-2016

INTRODUCTION

Necrotizing ulcerative stomatitis (NUS) is a uncommon, but potentially serious infection that can persist as a spectrum of clinical disease ranging from necrotizing ulcerative gingivitis (NUG) to noma (cancrum oris); and is characterized by the destruction of epithelium, connective tissue and papillae.[1,2] It is an uncommon disease caused most likely as a result of continuation of necrotizing ulcerative gingivitis or necrotizing ulcerative periodontitis (NUP); "NUG" is used when the disease involves only the gingiva, and "NUP" involves a loss of periodontal attachment.[3]

Necrotising stomatitis (NS) also called NUS is very difficult to differentiate from NUP. NS is generally localized, very rapidly destructive disease of the oral mucosa, alveolar bone and overlying gingivae.[1,4] Unlike other periodontal diseases, it presents substantial necrosis of gingival tissues, and loss of periodontal ligament and alveolar bone, in advanced stages it may lead to cancrum oris.[1,5] Significant predisposing factors include poor oral hygiene, unusual life stress, debilitated patients, recent illness (e.g. measles), malnutrition, smoking, immunosuppression and even inadequate sleep. It occurs commonly in developing nations with poor living condition.[6]NUS is characterized by a rapid acute onset which usually starts in the mouth, spreads

intra-orally destroying soft tissue and bone and progresses to perforate the facial skin, causing disfigurement (cancrum oris).[7.] Necrotizing ulcerative diseases are caused by infectious agents, although predisposing factors, such as compromised host immune response are the main factors facilitating bacterial pathogenecity.[8] This bacterial etiology was already demonstrated by Plaut in 1894 and by Vicent[9] in 1896, as microscopic examination of plaque samples retrieved from affected subjects clearly showed the presence of spirochetes and fusiform bacteria even within the tissues. The spirochetes and fusiform bacteria described in the necrotic lesions have the capacity to invade the epithelium[10] and the connective tissue[11], as well as to release endotoxins that may cause periodontal tissue destruction through the activation or modification of the host response.[8] Disease results when these bacteria proliferate, imbalancing the normal oral flora.[12] A multibacterial complex, in which Fusobacterium necrophorum, Treponema denticola, Porphyromonas gingivalis, Prevotella intermedia etc. are important components is implicated in causing this condition. [12,13]In order to prevent the progression of NUS to cancrum oris, affected patients should be vigorously treated and may require admission to hospital. Early diagnosis and interventions with local debridement, improved oral hygiene, systemic antibiotics and nutritional support are life saving and may reduce fibrosis and oral disfigurement that often follows wound repair.[13] As these diseases are often associated with systemic medical conditions, proper management of the systemic disorders is necessary in rehabilitating the patient.We report a case of an emaciated 70 year old diabetic male who was diagnosed with necrotizing ulcerative stomatitis.

CASE REPORT

A 70 year old male presented with the complaint of bleeding gums and pain in the oral cavity since 1 month. He also complained that his gums were brownish-white in colour and he had bad breath and altered taste. This was accompanied by a sore painful throat from the last two weeks. Patient had not brushed properly in over two weeks due to the pain. Eating was extremely painful for him resulting in concomitant malnutrition and at initial assessment, the patient appeared emaciated, unkempt and febrile. Upon asking, the patient revealed his poor living standards and economic background .On examination of his oral cavity, a whitish-erythematous, ulcerated gingival tissues

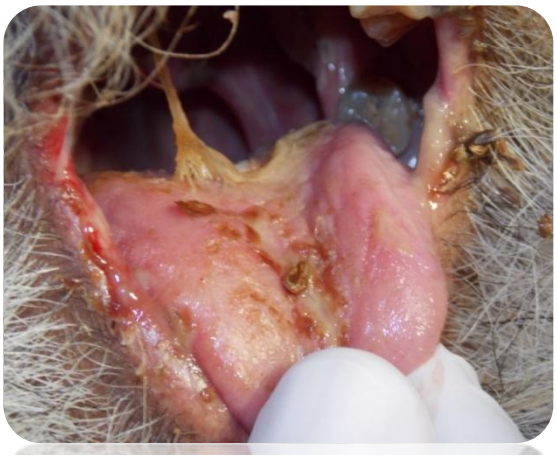

Figure 1 Ulcerations of the tongue and lip

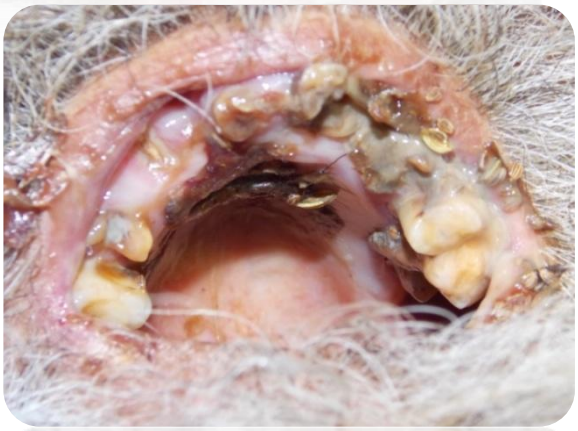

Figure 2- Necrotic ulcerations of palatal

was seen. A layer of grey slough like film formed from decomposed gum tissue was covering the labial mucosa and gingival region of the upper left oral mucosa. There was visible loss of papillary tissue with punched out ulcers in the same region. Lips were crustated, fissured, ulcerated and tender to touch, while the tongue presented with ulcerations and a depapillated surface **(Figure 2)**. Removal of slough, resulted in bleeding and the underlying tissue became exposed. Necrosis of the palate was evident **(Figure 2).** An excessive amount of saliva and extremely fetid odor perpetuated from his mouth. Multiple missing teeth, root stumps and mobility of teeth were seen

indicating periodontal involvement. The sockets from where the teeth were lost were necrotic.

The present condition was accompanied with bilateral submandibular lymphadenopathy, fever, malaise and an increased pulse rate. The patient also gave a history of loss of weight and appetite and general lassitude. Based on these signs and symptoms, a provisional diagnosis of necrotizing ulcerative stomatitis was arrived at. Histological examination was not carried out since the clinical presentation was adequate for diagnosis. Based on the above investigations a final diagnosis of necrotizing ulcerative stomatitis was made.Haematological examination revealed elevated TLC in the range of 14,300 mm^3 (normal range 4000-11000 mm^3) with a neutrophil count of 9×10^9/L (normal 2.0 - 7.5 $\times 10^9$/L), a C-reactive protein of 200 mg/L (normal range 0-5) and an ESR of 32 mm/hr (normal range 1-20 mm/hr). The patient was not anaemic and all other blood cell counts were within the normal range.Biochemical investigations found the patient to be diabetic with a random blood sugar (RBS) of 194 mg/dl (normal upto 160 mg/dl), SGOT level was 90 IU/L (normal range-5-34 IU/L) and SGPT was also elevated to 102 IU/L (normal range 0-40 IU/L). Screenings for HIV, HCV and Australian antigen were all negative. Oral swabs were taken for microbial culture examination and were reported as showing mixed oral microbial flora with no significant growth. Radiographic examination (Orthopantomogram; OPG) revealed severe bone loss, decayed teeth, multiple root stumps and multiple missing teeth in the maxillary and

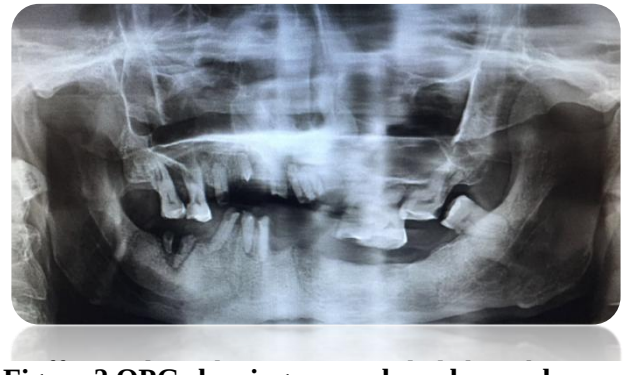

Figure 2 OPG showing severe bone loss and multiple missing teeth and root stumps

mandibular arches **(Figure 3)**. CT scan revealed extraoral marked lytic lesions in bilateral bones on either side of the midline with expansion and thinning of the overlying cortex and associated enhancing soft tissue density mass extending into the anterior portion of the oral cavity and premaxillary soft tissue with break in the cortex suggestive of destruction of palatal bone and its osteomyelitis. Rest of the visualized bones appeared normal **(Figure 4).**ulcerations. A normal chest radiograph and negative mycoplasma pneumoniae and hepes simplex serology excluded possible causes of erythema multiforme. The patient was counseled about the disease and admitted for treatment. He was reassured of the results of the treatment process. After written informed consent was obtained, 3% hydrogen peroxide was gently applied to the lesions using sterile swabs, for debridement of the necrotic areas. This was done and continued daily for a week. Topical lidocaine gel or benzydamine hydrochloride spray was given to control the severe pain. Patient was instructed not to use toothbrush as a method of cleaning for risk of interference with healing process. Chlorhexidine mouthrinses (0.12-0.2%) twice daily and saline mouthrinses throughout the day were advised. Metronidazole 500 mg every 8 hours to control the anaerobic bacteria, Vitamin C 300 mg tds, Tab folic acid 1 daily was included in the treatment protocol. Twice-daily regimen using isophane insulin (neutral protamine Hagedorn (NPH) insulin) or long-acting insulin analogues (insulin

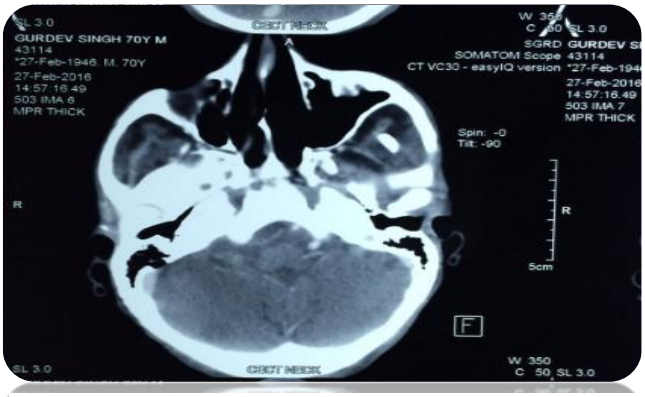

Figure 4 Expansion and thinning of the overlying cortex along with expansion of the soft tissue into the oral cavity, suggestive of osteomyelitis of the hard palate.

glargine) was also included. A follow up after one week revealed; better oral condition of the patient, resolutions in the necrotic areas and reduction in size of the oral lesions. Halitosis was reduced to a great extent. One week follow up revealed the patient to be much healthier and at ease with

normal blood glucose levels. Further blood investigations showed a decreased TLC level pointing to effect of treatment. Medication was continued for another week.

DISCUSSION

Necrotizing ulcerative gingivitis (NUG), necrotizing ulcerative periodontitis (NUP), and necrotizing ulcerative stomatitis (NUS), collectively termed necrotizing gingivostomatitis (NG), represent a dramatic oral infection associated with diminished systemic resistance, including HIV infection.[14] This form of gingival disease is relatively rare and always presents three typical clinical features which are: a) papillary necrosis, b) bleeding and c) pain which makes it different from other periodontal diseases.[8,15] If left unchecked the condition rapidly deteriorates to cancrum oris, which in turn is disfiguring and fatal.[16] Horning and Cohen[6] described seven stages of necrotizing periodontal disease according to the oral regions affected, ranging from necrosis of the tip of the papilla (stage 1) to necrosis that perforates the skin of the cheek (stage 7). This classification accords with the general view that this disease advances in a vestibular direction. The present case could be considered to be stage 5 of Horning and Cohen[6] (necrotizing stomatitis) because it shows a palatine progression and even approached the midline, where the palatal masticatory mucosa is affected instead of vestibular alveolar mucosa.[17]Knowledge of pathogenesis of the disease is limited because it is not clear whether spirochetes or fusiform bacteria observed in the lesion are the cause or the consequence, as secondary bacterial colonization may ensue after periodontal destruction since the necrotic tissues are the perfect environment for bacterial colonization and tissue invasion. [9,18] These anaerobic bacteria release virulent agents that are cytopathic to periodontal cells and to local immuno-inflammatory cells, that degrade extracellular matrix proteins and that disrupt the local vasculature, ultimately causing direct tissue damage with associated haemorrhage.[19]Localized ANUG is considered to be the key antecedent lesion which progresses to generalized necrotizing stomatitis in susceptible host, especially in immunocompromised patients, malnutrition and poor living conditions as well as in context of mental stress. [13,20] The treatment protocol includes irrigation and debridement of necrotic areas, oral hygiene instructions and use of mouthwashes, antibiotics and analgesics for pain.[21] In cooperative patients, gingivectomy or gingival grafting may be indicated after initial healing to resolve any residual effects.[1] All patients should be reviewed 1-2 weeks following treatment and require careful monitoring to ensure recurrence does not take place. [12] Also, we cannot underestimate the role of detailed history, proper clinical examination and correct diagnosis, which ultimately determine the treatment protocol.[1]

CONCLUSION

Necrotizing ulcerative stomatitis occurs when necrosis progress to deeper tissues beyond the mucogingival line, including the lip, cheek mucosa, tongue etc. The natural progression from an initial anaerobic bacterial infection of the marginal gingiva to full-blown case of NUS is the result of dynamic interactions between virulent bacteria on one hand and the host's general state of health, immune system, and local micro-environmental factors, on the other hand.The disease is not self limiting and will continue if untreated with periods of remission and exacerbation. Hence early diagnosis and intervention with local debridement, improved oral hygiene, systemic antibiotics and nutritional support are life saving and may reduce fibrosis and oral disfigurement that often follows wound repair.

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Source of support: Nil

Conflict of interest: None declared