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Case Report

CHRONIC NON-SPECIFIC ULCERS IN THE ORAL CAVITY CAN RESEMBLE DIABETIC FOOT ULCERS

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Abstract:

Oral ulcers are one of the most common mucosal lesions seen in different locations in the oral cavity. Most of these ulcers will resolve within two weeks. However, chronic ulcers remain problematic in the way they are managed and investigated for possible etiological factors. Viral infections, gastrointestinal disorders, blood diseases, cancer treatment, local trauma, medications or a combination of more than one factor contribute to oral ulcerations. The terms idiopathic or non-specific oral ulcers are still used when no possible explanation could be found regards the causative factors of the ulcerations. We present a unique case of a completely healthy 45 year old female, where a chronic fistula related to a failed root canal treatment developed into a chronic non-specific ulcer over the period of thirty years. The chronic ulcer resembled a diabetic foot ulcer in both clinical and histopathological pictures. Associated teeth with poor prognosis were extracted. Muco-gingival corrective surgery to close the associated gingival defect as well as a ridge preservation surgery to compensate for the associated bone loss produced excellent healing of both soft and hard tissues.

Key words: Oral ulcers – Root canal treatment – Oral Pathology – Oral medicine – Diabetic foot.

Introduction:

Oral ulceration is a very common problem encountered in clinical dental practice. It may involve both keratinized and non-keratinized epithelium. Diagnosing and identifying the etiological factor/s could be problematic, especially if it is not associated with other cutaneous and/or genital manifestations [1]. Oral mucosal manifestations including oral ulcers are inflammatory in nature and caused by multiple factors including genetic predisposition, viral and bacterial infections, food allergies, vitamin and micro-element deficiencies, systemic diseases (e.g., celiac disease, Crohn's disease, ulcerative colitis,

AIDS), increased oxidative stress, hormonal defects, mechanical injuries, anxiety, autoimmune disease and medications [2,3,4,5,6]. It was reported that oral ulceration was observed after misuse of Alendronate and Hydroxyurea as well [7, 8, 9].

One of the most common presentations of oral ulcers is Recurrent Aphthous Ulcers (RAUs) that usually resolve in one to two weeks. However, they indicate the presence of an associated illness, nutritional deficiency, autoimmune disease or an inflammatory disorder [10]. Clinically oral ulcers are classified according to the duration they remain in the oral cavity into acute and chronic ulcers [11, 12].Chronic ulcers that persist for longer than two weeks are often painful, stopping the patient from obtaining neither adequate nutrition through normal diet nor adequate fluid intake. They are resistant to conservative treatment such keeping good oral hygiene as well as therapeutic agents including topical corticosteroids, lidocaine and anti-fungal treatments [13]. Those cases are difficult to manage, can be misleading and confused with malignant ulcers. Terminology of non-specific oral ulcers is a bit controversial. The term non-specific implies that the causes and pathological mechanisms remain unknown [14, 15]. Non-specific oral ulcers can also be referred to as ulcers not otherwise specified (NOS) or necrotizing ulcerative stomatitis (NUS) [16].

Case presentation:

A 45 year old female was referred to a specialist periodontist for an initial periodontal assessment and treatment of gingival recession around her lower left central incisor. Patient's chief complaint was "I have a hole in my gums". Patient had no medical conditions and was not taking any medications. Patient was a smoker and her social history revealed no elements of stress. Patient was on a balanced diet and attended dental check-up appointments once every year.

Clinical examination revealed a large ulcer measuring 1cm X 2cm affecting lower central incisors and has been persistent for one year. There was a large bony defect around the lower anterior region. There was slight inflammation and swelling of the area buccally but no pus was evident. Both lower central incisors were tender to percussion. Patient's overall oral hygiene was poor with bleeding gums and generalized deposits of calculus [Figure 1].

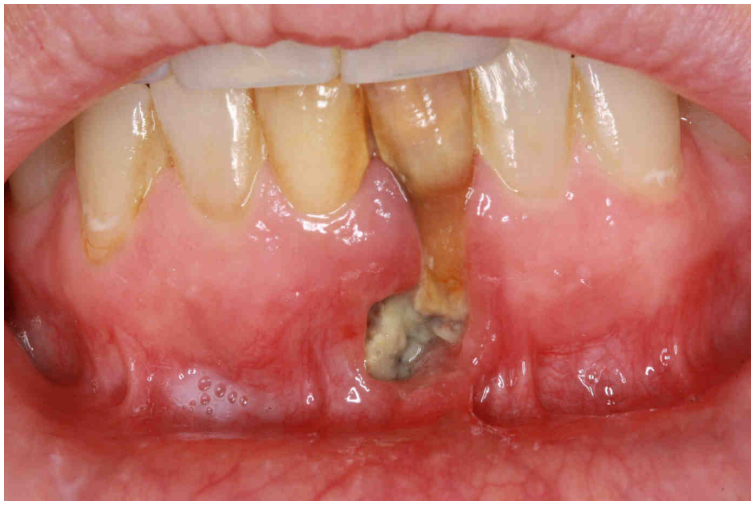

Figure 1: A photo showing the initial presentation of the ulcer-associated with lower central incisors.

A periapical radiograph of the lower anterior region was taken and showed a periapical radiolucency related to both lower central incisors. External inflammatory resorption and a failed root canal treatment were also observed on the radiograph [Figure 2]. Detailed dental history revealed that patient had severe trauma to her chin when she was 12 years old. As a result of that, her lower left central incisor was root canal treated. After three years patient had to have Endo-surgery (Apicectomy) on her lower left central incisor due to failure of the root canal treatment and the formation of multiple recurrent abscesses. Immediately after the surgery patient reported having a sinus that persisted until present days. One year ago, the sinus started enlarging and became a large hole of the present appearance.

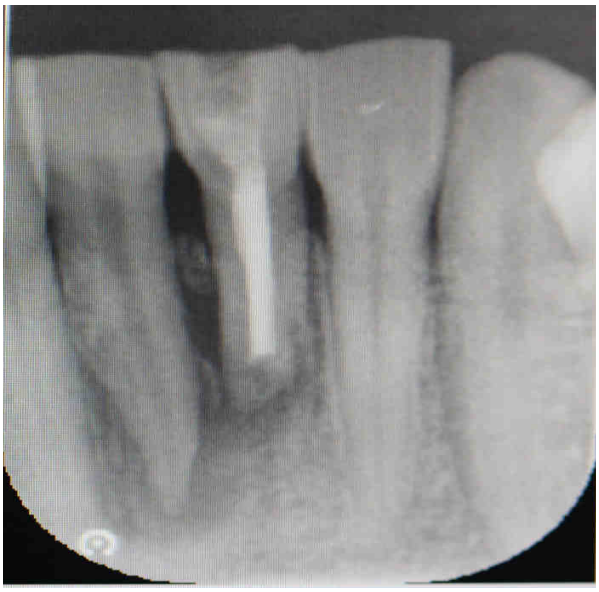

Figure 2: A periapical radiograph of the lower incisors showing the periapical radiolucency associated with teeth 31 and 41. Evidence of root canal treatment failure and external inflammatory resorption are also observed around tooth 31.

Due to financial limitations patient wanted to have both lower central incisors extracted and the defect in her gums to be treated. A full thickness mucoperiosteal flap was raised labially; a partial thickness flap was raised lingually to mobilize the keratinized gingival tissue to achieve a primary closure on the labial aspect under local anesthesia. Chronically inflamed tissue was excavated using surgical excavator to make sure all the ulcer lining was removed; all excavated tissues were submitted for histological analysis.

There was a large bony defect after all ulcer lining was removed. Peizo tips were used to smooth the sharp bony edges; a combination of BioOss granules (Bio-Oss; Geistlich Pharma AG, Wolhusen, Switzerland) and autogenous bone chips were harvested from the mental region using a surgical harvester and packed incrementally into the bony cavity. This was followed with resorbable Bio Gide membrane (Bio-Gide; Geistlich Pharma AG, Wolhusen, Switzerland) application to cover all bony chips. The lingual flap was approximated with the labial flap to achieve primary closure using

interrupted Prolene 05 and Vicryl 03 sutures [Figure 3]. A course of Augmentin 500mg antibiotic was prescribed tid for 5 days. Patient was referred to her treating medical practitioner for a complete blood picture.

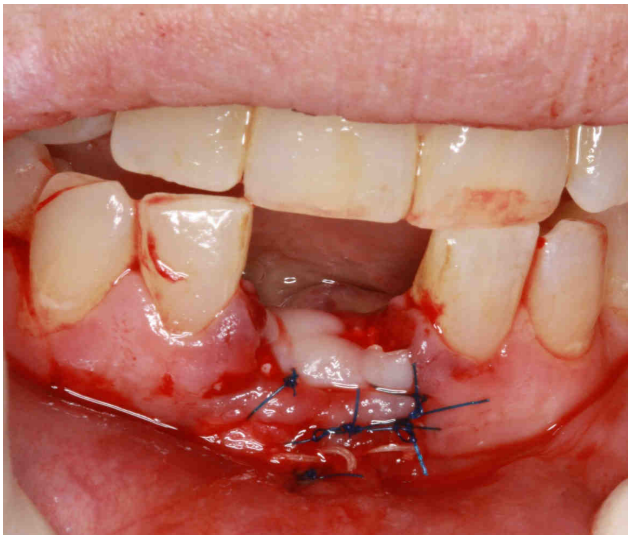

Figure 3: A photo showing the post-surgical picture after extraction of lower central incisors and closure of the associated bony defect (Muco-gingival and ridge preservation surgeries).

Patient was reviewed after four weeks of surgery. Overall, the healing was excellent with complete closure of the defect [Figure 4]. However, ridge augmentation might be needed labially at a later stage if implants are to be used to replace the missing lower central incisors. Results of the patient's blood picture were normal with no evidence of any blood disorder. The excisional results showed a diagnosis of chronic non-specific ulcer. Chronic inflammatory cell infiltrate, granulation tissue and areas of dystrophic calcification were evident in the lamina propria and submucosa [Figs 5, 6].This histopathological picture closely resembles that of diabetic foot ulcers. Finally, we endeavor to follow up this case to monitor for recurrence of ulcers and evaluate the success of prosthodontic treatment.

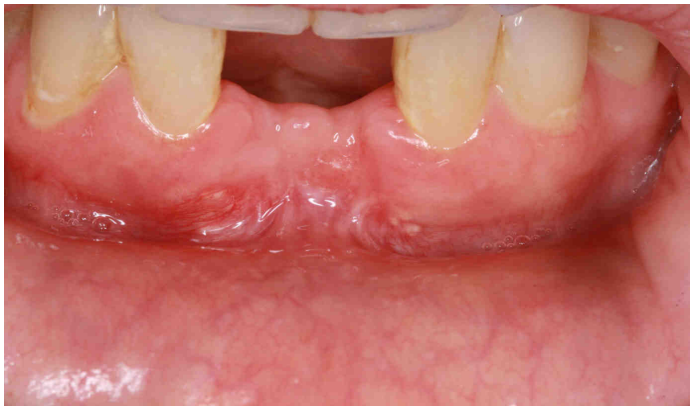

Figure 4: A photo showing the clinical picture of the lower anterior ridge four weeks after the surgery with complete closure of the defect.

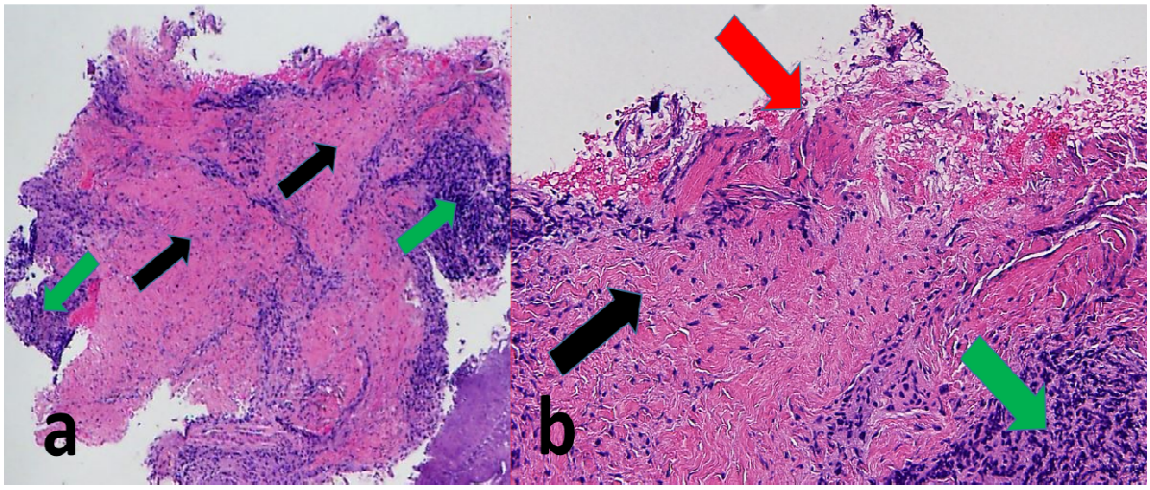

Figure 5: A histomicrograph showing an overview of the different zones of the chronic non-specific ulcer. (a): Black arrows are pointing to areas of granulation tissue and necrotic fibrous tissue, while the green arrows point to areas of dense chronic inflammatory cell infiltrate (H&E X 2). (b): Is a higher magnification of (a) showing a discontinued layer of stratified squamous epithelium (red arrow) in addition to the above mentioned layers.

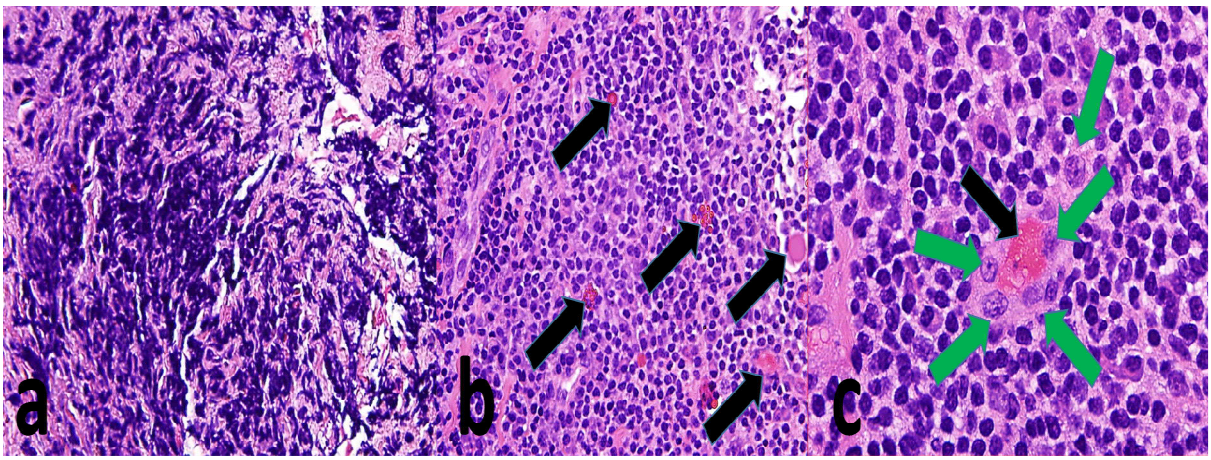

Figure 6: A histomicrograph showing higher magnifications of different zones of the chronic non-specific ulcer that demonstrates the similarity in heterogenous nature with diabetic foot ulcers. (a): Illustrates signs of tissue repair within the fibrous tissue layer within the ulcer including a major hallmark that is proliferation of fibroblasts (left side) and a dense layer of inflammatory cell infiltrate (right side) (H&E X 20). (b): Showing areas of dystrophic calcification (black arrows) that were formed due to long standing low grade infection over 30 years, embedded within the dense chronic inflammatory cell infiltrate (H&E X 20). (c): Higher magnification of the dystrophic calcification areas where it is surrounded by plasma cells with the characteristic eccentric cart wheel nuclues (H&E X 100).

Discussion:

Management of oral ulcerations has always been notorious for being problematic. In cases of RAUs for example, the treatment outcome remains a dilemma, as the etiology and pathological mechanisms of RAUs are still unclear. Despite the availability of different treatment protocols and modalities,

the aim remains to improve the quality of patients' lives until complete healing occurs. Recently, probiotics have been proven to be effective in gastric ulcers healing, which opens the horizons for future applications in healing ulcers in other peripheral parts of the body including the oral cavity [17]. However, no guarantee could be given regards prevention of recurrence [18].

Diabetic foot ulcers occur commonly due to external trauma, impairment of the peripheral blood circulation as well as peripheral neuropathy [19]. In our case the clinical and histopathological picture of the chronic non-specific ulcers resembled that of diabetic foot ulcers. This is justified by the history of trauma, and the fact the blood supply has been compromised in the lower anterior region for over 30 years, due to the persistence of a chronic infection as well as a fistula following a sub-optimal root canal treatment. The standard management of diabetic ulcers is monitoring, debridement and comprehensive wound care to avoid amputation of parts of the foot. However, in our case the decision was made to have the associated teeth extracted due to poor long term prognosis, as well as patient's choice due to financial constraints that did not allow for pursuing other treatment options.

There is extensive evidence in the literature showing the association between ulcerative lesions in the oral cavity and debilitating systemic conditions and/or nutritional deficiency [5]. The list of associated systemic diseases includes Primary Tuberculosis [20, 21], gastrointestinal disease, viral infections and hematological disorders [5, 22]. HIV positive patients and Epstein Barr virus positive patients are also frequent candidates for oral ulceration [15, 23, 24].In addition to the above, cancer treatment and immunosuppressive medications were also reported to be associated with oral ulceration [24, 25, 26]. In some cases oral ulcerations can be a manifestation of the combined effect of more than a single etiological factor [27]. Our present case is unique, as the patient is medically fit and there was no history of any relevant medical conditions, unlike other reported cases of gingival ulcerations, where patients suffered from an extensive history of medical conditions [28]. This shows that neglecting chronic oral infections or septic foci could lead to more serious consequences if left untreated even in healthy individuals. It also confirms that triggering factors and dynamics of oral ulceration is very complex. It is not yet fully understood and will remain a mystery until further notice.

A case of a similar chronic ulcer in the thorax region was reported to be a result of the presence of a chronic infection related to infected pacemaker leads [29]. In our case the unsuccessfully treated tooth, with a sub-optimal root canal treatment acted as a chronic septic focus that contributed to the etiology and pathogenesis of the chronic non-specific ulcer over the period of 30 years. This is also identical to diabetic foot ulcers; in addition the biopsy results and histopathological picture in our case supported the obvious similarities between chronic oral ulcers and diabetic foot ulcers, in terms of epithelial discontinuity, necrotic fibrous tissue, dense chronic inflammatory cell infiltrate and granulation. The only difference in diabetic foot ulcers is the difficulties and delays in healing due to the compromised blood flow to the extremities in case of diabetes [30].

The copious blood supply of the oral cavity and the absence of any underlying systemic condition allowed for excellent healing following the muco-gingival and ridge preservation surgeries, as opposed to diabetic foot ulcers which is known for its susceptibility to infection and delayed healing [15]. We endeavour to follow up the case further for any incidences of recurrence

of oral ulcerations and to explore different prosthetic options to replace the extracted lower central incisors.

Conflict of interest:

The authors declare that there is no conflict of interest regarding the publication of this article.

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