Case Report**Fibrous epulis misdiagnosed for combined periodontic-endodontic lesion**Khzam N¹, Shah Mansouri R², Poli A³, Bakr MM⁴¹Dr Nabil Khzam

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ABSTRACT

Fibrous epulis or peripheral ossifying fibroma is a reactive non-neoplastic condition that affects the gingiva as a result of chronic irritation. A case of a 44 year old female is presented in this study with a gingival swelling related to the maxillary central incisors. The Patient reported a history of trauma ten years ago and a recent root canal treatment of tooth 21, followed by referral to a specialist with a misdiagnosis for a combined periodontic-endodontic lesion affecting the maxillary central incisors. Excisional biopsy of the lesion revealed a diagnosis of ulcerated fibrous epulis with osseous metaplasia also known as peripheral ossifying fibroma. Periodontal debridement was performed to eliminate supra and subgingival plaque and calculus as well as gingival inflammation that could have been the source of irritation. The clinical and histopathological pictures and the surgical procedures associated with management of the periodontal disease are described. The etiological factor behind the development of the gingival reactive lesion remains unknown and could be the history of trauma, the chronic irritation induced by the plaque and calculus associated with the periodontal disease or a combination of both. We endeavour to follow up the case in order to report any recurrence.

Key Words: Fibrous epulis, endo-perio lesion, trauma, misdiagnosis, gingival swelling

Introduction

Epulis is a very generic term referring to gingival masses of mixed cell origins and are mostly resides of periodontal cells. Histologically, they present as focal fibrous hyperplasia, ossifying fibroma, pyogenic or peripheral giant cell granulomas. [1] Three main types of epulis exist, and are grouped based on their tissue origin: granulomatous epulis (epulis haemangiomas), fibrous (fibroid) epulis and giant cell (myeloid) epulis. However, there are many other conditions and terms documented in the literature, such as congenital epulis, epulis fissuratum, pregnancy tumour, [2] pyogenic granuloma, fibrous hyperplasia, peripheral fibroma with calcification and lymphoplasma- cellular variety. [3]

Etiological factors determining their presentation, growth and recurrence rate are still unknown. However, several factors could be taken into consideration including history of injury, trauma or inflammatory process, oral hygiene status, nutrition, alcohol intake, smoking and nicotine consumption, pharmacotherapy,

hormonal imbalance, and immune efficiency. [4] Clinically, epulis usually presents as a round, oval or lobular gingival mass appearing between the teeth. It may occur at any age, but is most commonly found in patients in their twenties and sixties, and more frequently in women than men. The maxillary incisor region is the most frequent site for epulis development. The majority of epulis lesions are less than 2 cm, but may grow rapidly and exceed 4 cm depending on the degree of irritation. Sometimes they can grow into an irregular red fleshy mass with ulceration, bleeding or both. Occasionally an epulis may invade underlying bone resulting in mobility or displacement of the involved teeth. [5,6]

Fibrous epulides are reactive lesions that are usually related to some form of chronic irritation. [7] Microscopically, they are lined with a predominantly hyperplastic stratified squamous epithelium and consist of connective tissue characterized by variable collagen deposition and variable chronic inflammatory infiltration

depending on the stage of development. [8,9,10] Surgical excision is the treatment of choice for most epulides. A wider surgical excision is recommended, due to the high rate of recurrence (10.3%). [11] In some cases, scaling and root planning is needed, in limited and advanced cases, extraction of the affected teeth with the removal of the adjacent alveolar bone. [6] Recurrence is usually correlated with the failure to remove or stop the source of irritation, [12,13,14] inadequate excision, [13,14] or a lack of correction of the periodontal defect. [15]

In this article, a case of an ulcerated fibrous epulis with areas of osseous metaplasia (Peripheral Ossifying Fibroma) resulting from a history of trauma that was misdiagnosed for a combined periodontic-endodontic lesion is presented. The clinical and histopathological pictures of the gingival lesion are discussed in detail, together with the surgical procedures, as well as the supportive periodontal treatment provided.

Case Report

A 44 year old female initially presented to a General Dentist due to recurrence of swelling, tenderness and suppuration palatal to teeth 21 and 11. The patient reported a history of trauma on this area 10 year ago. Tooth 21 did not respond to cold vitality test (Endo frost). Tooth 11 had a delayed but positive response to the same vitality test. A decision was made by the General Dentist to perform root canal treatment (RCT) on tooth 21, patient was advised that tooth 11 may require RCT as well but was to be re-assessed in the future after completion and assessment of the outcomes of tooth 21 RCT.

One year later, patient was referred to a Specialist Periodontist for consultation regarding teeth 11 and 21 with possible combined periodontic-endodontic lesion. At this stage, the patient presented with a large swelling behind tooth 11 and in between teeth 11 and 21. There was a small diastema between the upper central incisors. Patient's chief complaint was: "I have a large swelling behind my front teeth, it does not hurt unless I bite on my front teeth. I had trauma 10 years ago to my teeth by my daughter's head

and have since developed this swelling, it was on and off in terms of size and was sometimes associated with bad taste in my mouth". A review of the patient's medical and social history revealed nothing significant other than heavy smoking for the last 29 years, on average 10 to 15 cigarettes/day. A review of oral hygiene measures revealed the use of manual toothbrush twice daily but no flossing or interdental cleaning. Patient's oral hygiene was poor.

Fig. 1 Initial presentation of the gingival enlargement associated with teeth 11 and 21

A full comprehensive examination was done and revealed a periodontal diagnosis of moderate to severe generalized chronic periodontitis modified by poor oral hygiene and heavy smoking. Gingival evaluation around teeth 11 and 21 showed inflammation, bleeding and tenderness. Gingival enlargement and swelling was evident around the palatal and mesial surfaces of tooth 11 and on the mesial aspect of tooth 21 as well. The swelling was oedematous with an ulcerated surface as a result of the trauma from the lower anterior teeth. The swollen area was full of supra and subgingival plaque and calculus. Teeth 11 and 21 showed grade I mobility. Pt was referred for a

cone beam CT scan to confirm the clinical finding radiographically and help in treatment planning. [Fig.1,2] The proposed treatment plan was patient education, in the form of information on periodontitis and gingival enlargement in terms of pathogenesis, treatment and prevention. Detailed oral hygiene instructions in the form of recommending the use of electric tooth brush, dental floss and interdental brushes. The patient was encouraged to quit smoking, maintain high level of oral hygiene, and attend regular periodontal review appointments in order to maintain her teeth.

Fig. 2 A cone beam CT scan images showing the bone loss associated with teeth 11 and 21

Surgical removal of the gingival enlargement affecting 11/21 teeth was performed, the excised tissue submitted for a biopsy. Non-surgical periodontal therapy, followed by re-evaluation of periodontal tissue conditions 12 weeks after completion of debridement was done. Direct access periodontal surgery if needed, followed by

supportive periodontal treatment (3 or 6 months interval – to be decided after active periodontal treatment is completed).

Excisional biopsy under 2.2 ml articaine local anaesthesia with 1:100,000 adrenaline concentration was performed at upper central incisors 11-21 area labially and palatally. A full muco-periosteal flap was elevated extending from tooth 11 distal to 21 distal, periodontal debridement was performed in the area and all subgingival calculus and plaque was removed using both hand and ultra-sonic instrument, two interrupted vicryl sutures were placed. Post-operative instructions as well as oral hygiene instructions were given. The excised tissue was sent for biopsy and histopathological examination.

Fig. 3 (a) Ulcerated stratified squamous epithelium covering the lesion (b) Ulcerated epithelium with evidence of acanthosis and thickening of the prickle cell layer

Biopsy results showed the diagnosis was Ulcerated Fibrous Epulis with Osseous Metaplasia. Microscopic examination revealed the presence of ulcerated stratified squamous epithelium showing acanthosis and thickening of the prickle cell layer, [Fig 3] with dense chronic inflammatory cell infiltrate mainly of plasma cells and mast cells present underneath. [Fig 4] The chronic inflammatory cell infiltrate was surrounding areas of newly formed irregular

collagen bundles [Fig 5] as well as irregular areas of osseous metaplasia, suggesting that peripheral ossifying fibroma could be a possible differential diagnosis, due to replacement of the newly formed irregular collagen bundles with bone-like tissue in some areas due to long standing chronic irritation [Fig 6].

Fig. 4 Dense chronic inflammatory cell infiltrate present under the ulcerated epithelium and consisting of plasma cells (green arrow) and mast cells (black arrow)

Fig. 5 Irregular newly formed hyperplastic collagen bundles surrounded by the chronic inflammatory cell infiltrate

Three weeks after the surgery, the site had healed well. [Fig 7] Patient was advised that the lesion could recur if the underlying plaque-related cause was not eliminated. Full mouth periodontal debridement was carried out under

local anaesthesia. Recommendations for smoking cessation as well oral hygiene instructions were reinforced. The patient will be reviewed again 3 monthly, followed by 6 monthly intervals once the periodontal condition becomes stable.

Fig. 6 Osseous metaplasia (a) Area of osseous metaplasia surrounded by chronic inflammatory cell infiltrate (b) Osseous metaplasia with bone-like tissue replacing the newly formed irregular collagen bundles in some areas due to chronic irritation (Peripheral Ossifying Fibroma)

Fig. 7 Healing of the surgical site 3 weeks after the surgery

Discussion

The gingiva is subjected to chronic local irritation from various sources including calculus, food impaction, restorations with poor margins, and low grade local trauma. Localized hyperplasia is the usual reaction to these irritants that could

form collagen fibers, cellular fibroblastic tissue, mineralized tissues in the form of bone-like or cementum-like material, endothelial cells, and multinucleated giant cells.^[16] The classification of localized reactive hyperactive gingival lesions apart from giant cell lesions can be confusing.^[17] The most recent and accepted classification consists of four types of lesions: Focal Fibrous Hyperplasia (FFH), pyogenic granuloma (PG), Peripheral Ossifying Fibroma (POF) and Peripheral Giant Cell Granuloma (PGCG).^[18]

Despite the clinical similarities between the fibrous epulis (peripheral ossifying fibroma) presented in this study and peripheral giant granulomas, both lesions could be easily differentiated through their histopathological picture. Peripheral giant cell granulomas consist of a proliferation of mesenchymal cells and multinucleated giant cells with an associated prominent vascularity. Mineralized tissue in the form of woven and / or lamellar bone can be identified in about one-third of these lesions.^[19] In the case presented in this study, multinucleated giant cells were not identified as part of the histopathological examination of the excised tissue. The histopathological picture described in our study resembles the diagnosis presented in a number of other cases in the literature.^[20] Fibrous epulides with osseous metaplasia (peripheral ossifying fibroma) could be both placed together with pyogenic granulomas and pregnancy tumors under the umbrella of focal fibrous hyperplasia as traumatic or irritational reactive fibrous lesions.^[21,22] The differential diagnosis in such lesions should include metastatic cancer, fibroma, hyperplastic gingival inflammation, hemangioma and angiosarcoma.^[23,24] Despite the nomenclature, fibrous epulides or peripheral ossifying fibromas are reactive, non-neoplastic lesions.^[25,14,26] The patient presented in this study was a 44 year old female, which confirms the female predilection in peripheral ossifying fibroma that was reported in previous studies.^[27,28,29] It is also consistent with the peak incidence of peripheral ossifying fibroma previously reported around the fourth and fifth decades.^[29,30]

Mast cells have been associated with secretion of a number of important mediators of fibrosis.^[31,32] It was shown that mast cells were present in higher numbers in inflammatory fibrous hyperplasia, as they were responsible for the adaptive and pathological responses in areas on chronic inflammation.^[33] This was attributed due to the presence of a number of chemical mediators including histamines, proteases, chymase and tryptase within the secretory granules of the mast cells.^[34,35,36] In addition to the above, mast cells were associated with increased migration and proliferation of fibroblasts^[37] as well as affecting the fibroblast's functional behaviour through releasing fibrogenic cytokines including platelet- derived growth factor (PDGF), tumor necrosis factor- α (TNF- α), and basic fibroblast growth factor (b-FGF).^[38,39] The last (b-FGF) was specifically associated with epulis formation.^[40] This is in agreement with the histopathological picture in the present case, where the chronic inflammatory cell infiltrate was densely populated with plasma cells and mast cells.

New treatment modalities were suggested for fibrous epulides including intra-lesional injections of pingyangmycin^[6] and propranolol.^[41] In the present case, a traditional surgical approach was used together with periodontal care to minimise the chances of recurrence. The case will be monitored in case of recurrence. Our case is unique as an ulcerated fibrous epulis was misdiagnosed for a combined periodontic-endodontic lesion. This confusion resulted from an endodontic problem related to the history of trauma, and was separate from the gingival swelling that was present due to the chronic periodontal disease. The bad taste experienced was due to suppuration from the periodontal lesion around teeth 11 and 21. The ulceration was the result of trauma from lower anterior teeth against the gingival swelling around teeth 11 and 21. All of the above factors played a role in the incorrect initial diagnosis. We endeavour to review the case and monitor for vitality tooth 11, progress and management of periodontal disease as well as recurrence of the fibrous epulis in case of failure to maintain good periodontal health.

Conclusion

A case of an ulcerated fibrous epulis also known as peripheral ossifying fibroma is reported. The presence of multiple etiological factors in this unique case led to an initial incorrect diagnosis of a combined periodontic-endodontic lesion. The correct diagnosis was confirmed through excisional biopsy. The surgical procedure, histopathological picture and suggested etiological mechanisms as well as alternative treatment approaches were discussed. Maintaining a healthy periodontium is the key for a successful management of this case in order to prevent recurrence.

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