

## CASE REPORT

# Management of hormonal induced recurrent ulcerative fibrous epulis with osseous metaplasia: A case report

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## Key Clinical Message

Understanding the role of hormones in periodontitis is important. Periodontal microscopic surgery approach in the treatment of fibrous epulis is not indicated. Wider flap access with root planning is indicated to control the lesion in one phase.

## Abstract

We present a case of a 40-year-old female who presented with a gingival hyperplastic lesion around the maxillary left permanent central and lateral incisors. Patient's medical history reveals a recent pregnancy, hypothyroidism, ulcerative colitis, and schizoaffective disorder. All medical conditions were controlled by medications. The lesion was excised using a minimally invasive periodontal surgical technique, and the biopsy results confirmed a diagnosis of ulcerative fibrous epulis with osseous metaplasia. No curettage or local debridement was done under the assumption that the patient's oral hygiene was satisfactory and due to aesthetic concerns of gingival recession and creation of black triangles. The lesion recurred after 3 months and was removed using a traditional more invasive surgical technique. The patient was followed up for 2 years, and there was no further recurrence due to the complete excision of the lesion in the second surgery and the disappearance of the hormonal-related factors post-pregnancy that could have contributed to the gingival hyperplasia. The unique component of the case we are presenting is the comparison between two different surgical techniques and the conduction versus absence of local periodontal debridement after surgical excision as well as the possible correlation between oral supplements and the calcific nature of the lesion(s) reported. The case we present demonstrates that a more invasive traditional surgical approach together with local periodontal therapy provide an optimum treatment outcome in conjunction with elimination of any associated etiological factors. We also propose that hormones are more important as an etiological factor in developing fibrous epulis lesions than medical conditions and medications.

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## KEYWORDS

fibrous epulis, hormonal balance, medications, osseous metaplasia, periodontal debridement

## 1 | INTRODUCTION

The term epulis refers to a group of mixed cells residing in the gingival or periodontal areas and can be subclassified into either fibrous epulis, ossifying fibroma, pyogenic granuloma, and peripheral giant cell granuloma.<sup>1,2</sup> Multiple other similar conditions are documented in the literature including congenital epulis, epulis fissuratum, pregnancy tumor,<sup>3</sup> pyogenic granuloma, fibrous hyperplasia, peripheral fibroma with calcification, and lymphoplasmic cellular variety.<sup>4</sup> Further classifications exist and are based on the presence of calcified tissue, the nature of the connective tissue, the type of keratinization, and the degree of epithelial thickness.<sup>5</sup> These lesions have also been grouped together under the umbrella of benign odontogenic tumors in human medicine and veterinary science.<sup>6</sup> Despite the controversy surrounding the classification of these lesions,<sup>7</sup> the etiological factors are similar between all different types of epulis, which are mainly reactive lesions to different sources of irritating agents.<sup>8</sup> The literature is divided on whether these lesions are actual tumors of an odontogenic origin or can be considered as a proliferation of a reactive nature.<sup>9</sup>

The clinical picture is usually a localized growth around the interdental papillae that is either sessile or pedunculated and usually small in size (less than 2 cm), with some larger lesions being documented in the literature.<sup>10</sup> The recurrence rate is usually between 8.9% and 20%, and the first recurrence usually occurs within 12 months of the first appearance of the lesion.<sup>11</sup> The recurrence is generally attributed to incomplete surgical removal and/or failure to identify and eliminate the local and/or systemic irritant/etiological factor.<sup>12</sup> Fibrous epulis can be misdiagnosed for other periodontal lesions or gingival growths and benign tumors.<sup>13–16</sup> Costa et al. in a recent systematic review described the diagnosis of gingival lesions as a dilemma and indicated that the patient's medical history is an important factor in determining the exact type of gingival lesions.<sup>17</sup> The management is complete surgical excision together with periodontal debridement and gingival recontouring as well as control of any local irritating factors to avoid recurrence.<sup>18–20</sup>

The current case report describes the significant factors for a recurrence, surgical technique, and treatment protocol for an ulcerative fibrous epulis with osseous metaplasia in a 40-year-old female with combined etiological factors and clinical features of fibroepithelial hyperplasia.<sup>21</sup>

## 2 | CASE PRESENTATION

Patient consent was obtained to use their deidentified records in research. A 40-year-old female was referred to a specialist periodontist for consultation regards gingival hyperplasia around the maxillary left permanent central and lateral incisors (Figure 1). Patient's medical history revealed she had a baby 4 weeks ago, and that the lesion appeared during pregnancy but increased in size after the delivery of the baby. The patient also reported a history of hypothyroidism that is currently managed by supplementary thyroxin (Eltroxin 50 mg once per day), ulcerative colitis that is currently managed by anti-inflammatory agent (Mesalazine) that is similar to aspirin (Pentasa Enema 1 g per day), and schizoaffective disorder that is currently managed by (Risperdal 1 mg once per day before bed). Furthermore, it should be noted that the patient has been on multivitamin supplements for a long period of time (self-prescribed) as well as calcium/vitamin D3 (Ostelin—Calcium Carbonate 600 mg and Vitamin D 500IU Colecalciferol 12.5 mcg) and folic acid (Blackmores Folate 500 mg Folic Acid) supplements during her pregnancy as prescribed by her medical practitioner.

Dental, periodontal examination, orthopantomography (Figure 2) and bitewing radiographs (Figure 3) revealed good oral hygiene, absence of sinus-related problems and the absence of any active caries or periodontal disease, apart from localized periodontitis stage 1/grade A around the lesion. Mandibular third molars were horizontally impacted, and patient was aware of the impacted teeth but did not wish to proceed with any surgical intervention at



FIGURE 1 Clinical photograph showing the initial presentation of the hyperplastic lesion.

this stage. There was evidence of white spot lesions on multiple teeth in the oral cavity and pulp stones (denticles) in the pulp chambers of some posterior teeth on the radiographs. Periapical radiographs were taken and showed no periapical pathology around the maxillary left permanent central and lateral incisors nor any other anterior teeth on the left side (Figure 4). Extra-oral examination revealed no temporomandibular related issues nor any lymphadenopathy.

Upon the examination of the hyperplastic lesion, the area was 3×1.5cm in size, enlarged with small ulcerations. There was bleeding upon gentle probing. The lesion was pedunculated with a hard consistency. The overlying mucosa was normal in color with the exception of the central area which was reddish in color. There was pseudo pocketing due to the swelling rather than actual periodontal attachment loss. The patient was booked for an excisional biopsy after 2 weeks.

The excisional biopsy was undertaken under local anesthetic using C15 scalpel with no complication. A minimally invasive periodontal surgical technique that preserved the soft tissue to minimize future cosmetic defects was used. Therefore, a partial thickness papilla preservation flap design was used to try and preserve the interdental papillae and the marginal gingiva. The excised tissue was sent for biopsy. No periodontal debridement of the area was performed due to the absence of any visible local

etiological factors such as subgingival plaque and calculus and the excellent status of oral hygiene. The risks of gingival recession and the creation of black triangles were very high. A conservative versus a full surgical approach were presented to the patient during the informed consent procedures. The patient for aesthetic reasons chose to have minimally invasive surgery and requested to start with the conservative approach.

The biopsy report indicated the diagnosis of ulcerative fibrous epulis with osseous metaplasia. The biopsy specimen showed hyperplastic epithelium with areas of ulceration and an underlying dense fibrous connective tissue showing an increased number of chronic inflammatory cells. There were areas of calcification in the form of trabeculae and drop-like metaplastic newly formed bone (bone-like material) with an irregular pattern of incremental lines and a smaller number of osteocytes as well as multiple blood vessels present in the center of the lesion (Figure 5). The patient was scheduled for a review appointment after 4 weeks. The healing was complete with some fibrous scarring and no signs of swelling, inflammation, or infection. The base of the lesion could still be identified after the surgical excision (Figure 6).

Three months later, the patient presented with a similar lesion that was smaller in size (2.5×1 cm) compared to the baseline size (3×1.5 cm) (Figure 7). Another excisional biopsy under local anesthetic was performed using C15 scalpel. A large full thickness muco-periosteal flap was used to completely elevate the interdental papilla and the marginal gingiva. The flap extended from the mesial of tooth 21 to the distal of tooth 22. The biopsy result was identical to the initial lesion (ulcerative fibrous epulis with osseous metaplasia). The second excisional biopsy was performed using a traditional surgical technique which was less conservative than the first surgical technique. Local periodontal debridement of the area was conducted. The patient was reviewed again in 6 months, and the healing showed no fibrous scarring in opposition to the post-surgical review of the original lesion (Figure 8). Up to date, 2 years post the initial lesion was detected, no further recurrent lesions were reported by the patient.



**FIGURE 2** Orthopantomography showing no significant findings apart from the impacted mandibular third molars and pulp stones (denticles) in the pulp chamber of some posterior teeth.



**FIGURE 3** Bitewing radiographs showing no interproximal decay or interdental bone loss.



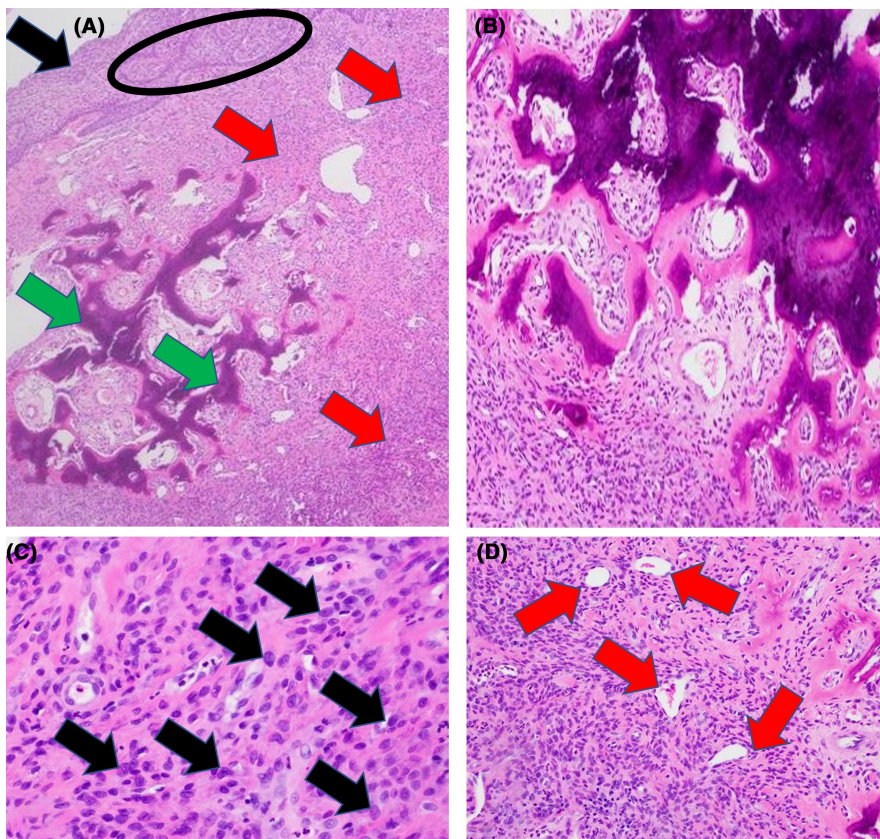


**FIGURE 4** Periapical radiograph showing no periapical pathology around the roots of anterior teeth in the upper left quadrant.

### 3 | DISCUSSION

The diagnosis of gingival enlargements has proven to be challenging to the extent that a decision tree was deemed necessary to systematically reach a conclusive diagnosis.<sup>22,23</sup> Etiological factors also remain unclear. Radanović et al. demonstrated a fibrous epulis with no obvious clear etiological factors apart from poor oral hygiene and sub-standard restorative work.<sup>24</sup> On the contrary, the present case has a clear etiological factor, the history of a recent pregnancy, and the postpartum hormonal imbalance.

The three medications that the patient is on are linked with dry mouth. Khzam et al. described a similar case of generalized gingival enlargement linked with Felodipine; an antihypertensive medication with a known adverse effect of xerostomia revealed exact same diagnosis (ulcerative fibrous epulis with osseous metaplasia).<sup>25</sup> The medication-induced xerostomia could have been a contributing risk factor to the development of the fibrous epulis since dry oral mucosa is more susceptible to damage from local irritants (hormonal imbalance and localized periodontitis in this case) when compared to a moist and hydrated surface. However, in the abovementioned case, felodipine could have also contributed to the gingival overgrowth by acting on long-lasting type (L-type) calcium channels that are characterized by a slow rate of



**FIGURE 5** (A) Showing the overall histological picture of the lesion with hyperplastic epithelium (black arrow) with areas of ulceration (black circle), dense fibrous connective tissue (red arrows), and areas of calcification in the form of metaplastic bone-like material (green arrows) (H&E  $\times 10$ ). (B) Higher magnification of the calcified areas showing irregular trabeculation and drop-like areas of bone-like material with features supporting a rapid rate of formation (H&E  $\times 20$ ). (C) Higher magnification of the dense fibrous connective tissue that is heavily infiltrated with inflammatory cells (black arrows) (H&E  $\times 40$ ). (D) Showing the central part of the lesion with signs of an inflammatory reaction (left side) and multiple enlarged blood vessels (red arrows) (H&E  $\times 20$ ).



**FIGURE 6** Clinical photograph showing complete healing with some fibrous scarring after 4 weeks of excising the lesion.



**FIGURE 8** Clinical photograph showing the clinical picture 6 months after the excision of the recurrent ulcerative fibrous epulis with osseous metaplasia.



**FIGURE 7** Clinical photograph showing the recurrent hyperplastic lesion that developed 3 months after the presentation of the initial ulcerative fibrous epulis with osseous metaplasia.

inactivation.<sup>26</sup> It is important to note that calcium channel blockers which act only on L-type calcium channels, such as felodipine, tend to induce gingival enlargements at a higher rate compared to other calcium channel blockers that act on L-/T-/N-type calcium channels.<sup>27</sup>

In the present case, due to the absence of visible signs and symptoms of periodontal disease and the acceptable status of oral hygiene, the risk of gingival recession and the aesthetics concerns of creation of black triangles was very high; therefore, a decision was made not to perform periodontal debridement. It is a standard procedure that area of excision should be debrided to minimize the chances of recurrence.<sup>28,29</sup> The current case highlights the importance of periodontal debridement irrespective of the oral hygiene status. In the present case, despite the observation and the concerns listed above, at least a closed debridement should have been performed. The recurrence of the lesion could have been attributed to the incomplete

excision of the lesion in the first instance which was evident by the fibrous tissue observed in the review appointment. It could have been also due to not performing local periodontal debridement of the area after the first surgical excision. However, the most significant factor contributing to the recurrence of the lesion could have been the post-pregnancy changes in hormones which has been linked to higher recurrence rates in previous reported cases.<sup>30,31</sup>

Adequate and complete excision of the lesion is very important to avoid the recurrence of the fibrous epulis.<sup>32</sup> Previous research has shown that minimally invasive plastic periodontal surgery could produce more superior results than traditional surgery in selected cases.<sup>33</sup> In the current reported case, a more invasive traditional surgical technique was more effective in stopping the recurrence of the lesion than the initial minimally invasive surgical technique used in the first instance of excisional biopsy. Laser has been used successfully in surgical removal of fibrous epulis and has the advantages of a better bleeding control, short operation time, elimination of the requirement of sutures, no antibiotic, no pain killers, and superior healing.<sup>19,34–36</sup> Paglioni et al. concluded in a systematic review that laser excision might reduce the recurrence of some oral lesions like leukoplakia.<sup>37</sup> On the contrary, Rodriguez-Lujan et al. reported that further assessment of different parameters that affect recurrence should be evaluated after laser removal of oral lesions.<sup>38</sup> In the present case report, it is questionable whether laser excision would have prevented the recurrence of the lesion in the presence of other risk factors including the hormonal changes post-pregnancy, the medications, and the medical history of ulcerative colitis.

The patient in the current case report was 40 years of age, which is slightly older than common demographics of similar lesions that usually occur in females between the age of 20 and 30 years of age.<sup>39</sup> Osseous metaplasia



starts as an inflammatory process of the periodontal cells and the periosteum that leads to metaplasia of the surrounding fibrous connective tissue and consequently dystrophic calcification and bone formation.<sup>40</sup> Katanec et al. demonstrated that high parathyroid hormone blood levels could be related to the development of calcified lesions such as peripheral ossifying fibroma that are very similar to the current reported lesion.<sup>39</sup> The current reported cases suffered from hypothyroidism and were taking Eltroxin 50 mg once per day. A follow-up blood test would have been beneficial in determining the blood thyroid and parathyroid hormone levels. The presence of pulp stones that were evident in the patient's radiographs could be correlated to the ossifying nature of the lesion, especially that the patient was on multivitamin supplements as well as calcium and folic acid supplements during her pregnancy.

In the present case, the most significant factor contributing to the development and recurrence of the lesion was the pregnancy-related and postpartum hormonal imbalance. Most cases of pregnancy-related gingival overgrowth would resolve fully or reduce in size after 6–8 weeks postpartum.<sup>29,41</sup> Zhao et al. reviewed 2917 cases of fibrous epulis and reported an average recurrence rate of 11.28% with pregnancy-related lesions having the highest recurrence rates.<sup>42</sup> Furthermore, Zhao et al. concluded that regular periodontal supportive therapy and management of the periodontal inflammation had a significant impact in reduction the recurrence rates.<sup>42</sup> This emphasizes the point raised in the current case report where local periodontal debridement should have been performed after the first excisional biopsy in order to reduce the chances of recurrence. After the recurrence of the lesion in 3 months, no further recurrence was reported after following up the case for 2 years. This is explained by the fact that postpartum hormonal changes in estrogen and progesterone reset to pre-pregnancy levels.<sup>22,43,44</sup> The patient was advised to follow-up with her medical practitioner for advice regards the risks versus benefit of taking daily self-prescribed multivitamin supplements, monitoring thyroid/parathyroid hormones.

In summary, the following treatment protocol is recommended for ulcerative fibrous epulis cases:

1. Surgical removal of the entire lesion using a traditional invasive surgical technique with a full thickness mucoperiosteal flap to completely elevate the interdental papilla and the marginal gingiva.
2. Scaling and root planning (periodontal debridement) of the surgical area.
3. Consultation with medical practitioners to ensure the elimination of systemic etiological factors, in particular hormonal imbalances.

4. Review appointments after 4 weeks, 3 and 6 months to monitor healing, oral hygiene status and confirm the absence of recurrent lesions.

## 4 | CONCLUSIONS

1. Hormonal imbalance is the most significant risk/etiological factor in developing fibrous epulis.
2. Local periodontal debridement of the area after surgical excision is highly important in minimizing the chances of recurrence of fibrous epulis lesions irrespective of the oral hygiene status or etiological factors.
3. Traditional invasive surgical techniques could contribute to less recurrence of fibrous epulis when compared to conservative minimally invasive periodontal surgery techniques.
4. Vitamin and mineral supplements could have contributed to the calcified nature of the lesion(s) presented in this report.

## AUTHOR CONTRIBUTIONS

**Mahmoud Bakr:** Conceptualization; data curation; formal analysis; investigation; methodology; project administration; supervision; validation; writing – original draft; writing – review and editing. **Mahmoud Al-Ankily:** Formal analysis; project administration; validation; writing – original draft; writing – review and editing. **Nabil Khzam:** Conceptualization; data curation; investigation; methodology; project administration; resources; validation; writing – original draft; writing – review and editing.

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## CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

## DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

## CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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