

A Cemento-Ossifying Fibroma in the Maxilla

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Abstract: Cemento-ossifying fibroma is an uncommon, benign lesion, which can lead to swelling and deformity of the face. This report describes an uncommon presentation of this lesion with simple and effective measures to diagnose and treat it.

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Clinical Relevance: This case demonstrates the need for clinical, radiographic and histopathological information to be collated to reach a definitive diagnosis.

The cemento-ossifying fibroma is an uncommon condition which may occur at any age but is most often diagnosed in adulthood, typically during the third or fourth decades of life, with women more frequently affected than men.^{1,2} Seventy to eighty per cent occur in the mandible,³ mainly in the posterior tooth-bearing areas.^{1,2} The maxilla is a rare site of involvement but occurrences in ethmoid sinus and temporal bone have been reported.^{4,5}

Clinically, the lesion is usually asymptomatic but can lead to an asymmetric swelling with noticeable deformity. Displacement of teeth, with divergence of roots can be a presenting feature.⁶ The cemento-ossifying fibroma is a benign lesion and can be treated adequately by enucleation or curettage;⁴ unfortunately, extensive surgery is indicated for those lesions with rapid growth.⁷ Radiotherapy is contra-indicated owing to their radio-resistance and the potential risk of malignant

transformation.⁴ These tumours have also been known to recur,^{8,9} although this is uncommon.⁸

This report describes an uncommon presentation of this lesion with simple and effective measures to diagnose and treat it.

CASE REPORT

A 25-year-old white woman, otherwise well, presented with a swelling of the right cheek that had been gradually expanding over 3 years (Figure 1). No symptoms were present including visual disturbances. On examination, the swelling was firm and considered to be deep to the facial soft tissues as their mobility was not impeded. Intra-orally the swelling could be palpated in the upper right buccal sulcus, extending from the upper right canine to second molar (Figure 2).

A panoramic radiograph taken by the referring practitioner, four months prior to referral, showed an expanded posterior and inferior cortical margin of the right maxillary antrum compared to the left. The roots of the upper right canine and first premolar were also displaced (Figure 3).

An occipito-mental radiograph showed expansion of the right antral wall in the vertical and horizontal plane. A radio-opaque shadow was present in the antrum, with expansion into the right infra-orbital region. Soft tissue shadows could also be seen expanding into the right infra-orbital region (Figure 4).



Figure 1. Extra-oral view of swelling of right cheek.



Figure 2. Intra-oral view of swelling in right sulcus.

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Figure 3. Panoramic radiograph taken four months prior to referral.

An upper oblique occlusal radiograph of the upper right quadrant was also taken (Figure 5). This showed a radiolucent lesion over the apex of the upper right canine. It could also be seen that the antrum had expanded in a medial to lateral direction. The medial margin appeared scalloped and well corticated, indicating a slow growing, benign lesion. The lateral margin showed an irregular, poorly-defined margin of variable radio-density, indicating a rapidly growing aggressive lesion. However, in view of the clinical and radiographic findings, a provisional diagnosis of fibrous dysplasia was made.

The patient was admitted for biopsy of the lesion under general anaesthetic. Access to the lesion was made intra-

orally through the right buccal sulcus. The lesion enucleated easily, with no evidence of bone invasion. It had a white appearance, measured approximately 5 x 4 x 3 cm, and had a fibrous but gritty, 'rubbery' texture. It occupied the whole of the right maxillary antrum with expansion into the orbital floor and ethmoidal region.

The histopathological appearance showed a fibro-osseous lesion containing some bony trabeculae undergoing resorption. It also contained small islands of cementum-like material. One edge appeared to be well circumscribed. These findings, together with the radiographic appearance, were consistent with a cemento-ossifying fibroma (Figure 6a, b).

The patient recovered uneventfully with restoration of facial symmetry extra- and intra-orally (Figures 7 and 8). An upper oblique occlusal radiograph taken three months post-operatively showed adequate bony healing (Figure 9). There was no residual visual impairment or symptoms.

DISCUSSION

The cemento-ossifying fibroma belongs to a group of similar conditions classified as fibro-osseous lesions, which are usually benign. Such conditions include fibrous dysplasia, periapical cemental dysplasia and florid osseous dysplasia. However,

disagreement amongst authors does exist with regard to the classification of fibro-osseous lesions. The term cemento-ossifying fibroma was proposed for this lesion after Kramer *et al.*, on behalf of the World Health Organization (WHO),¹⁰ reported that the ossifying fibroma and the cementifying fibroma were variants of the same lesion owing to the histological similarity of bone and cementum. Voytek *et al.*¹¹ examined 56 cases of fibrous dysplasia and cemento-ossifying fibroma of bone (gnathic and extragnathic) morphologically, clinically and radiographically to determine the reliability of their distinction and whether their distinction had any bearing on clinical findings or prognosis. The finding suggested that, owing to histologic and radiographic overlap and similar (low) recurrence rates, the lesions were considered to be related. It was then proposed that these lesions were probably opposing ends of a morphologic spectrum. Koury *et al.*¹² and Waldron¹ proposed that fibro-osseous lesions can be categorized into three groups:

- fibrous dysplasia;
- reactive (dysplastic) lesions arising in the tooth-bearing parts of the



Figure 4. Pre-operative occipito-mental radiograph. Expansion into the infra-orbital region as arrowed.



Figure 5. Pre-operative upper oblique occlusal radiograph.

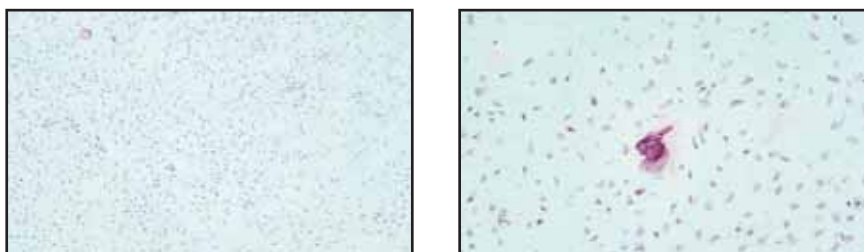


Figure 6. (a) Histological view showing the dense fibrous tissue. (H & E x100). (b) Histological view showing cementum-like material in the centre of the field of view. (H & E x250).

jaws, e.g. periapical cemental dysplasia; and

- fibro-osseous neoplasms, e.g. ossifying fibroma.

The aetiology of cemento-ossifying fibroma is still unclear; a variety of contributing factors have been proposed. Wenig *et al.*¹³ suggested that trauma can induce stimulation of progenitor cells. Hamner *et al.*¹⁴ looked at 249 cases of fibro-osseous lesions of the jaws and proposed that cemento-ossifying fibroma could be considered a tumour of multipotent mesenchymal blast cells, present in the periodontal membrane, that have the ability to produce cementum, alveolar bone and



Figure 7. Extra-oral view taken three months post-operatively showing good facial symmetry.

fibrous tissue.

Santos and Arianayagam⁴ described the histological picture of the cemento-ossifying fibroma as a fibro-osseous lesion, showing fibroblastic tissue containing rounded or lobulated masses of calcification with no relationship to the apex of a tooth. However, the histological picture of these lesions changes with time. During the early stages of development, these lesions are fibroblastic. As they progress and mature, cementum-like masses are deposited in the fibrous stroma, which enlarge and eventually coalesce with neighbouring masses. As with histological changes, the radiographic appearance of these lesions will also change over time. Raveh¹⁵ described the radiographic appearance of these lesions as being readily delineated from the surrounding bone, radio-lucent areas being present during their early stages of development, whilst later in their development they become more radio-opaque as the cementum-like masses fuse.

Owing to these progressive changes during development, and hence the varying different types of pictures these lesions can portray at the time when a patient presents, a variety of lesions must be considered in the differential diagnosis. Lesions of a similar appearance include:

- ossifying fibroma;
- cementoma, myxoma;
- calcifying cysts;
- adenomatoid odontogenic tumour;
- calcifying epithelial odontogenic tumour;
- osteoblastoma, osteogenic

sarcoma; and

- Paget's disease.¹⁶

To limit the differential diagnosis, other investigations can be undertaken, such as fine needle cytology¹⁶ and computed tomography.¹⁷ However, there are also disadvantages, such as increasing the expense of treatment and increased radiation exposure, as in the latter. In this case, it was thought unnecessary to perform any other investigations besides radiography prior to surgery, as the plain radiographic films allowed visualization of the lesion and its anatomical limitation as a consequence of its benign nature. This was then confirmed by its histopathological analysis.



Figure 8. Intra-oral view taken two weeks post-operatively.



Figure 9. An upper oblique occlusal radiograph taken three months post-operatively showing adequate bony healing.

CONCLUSION

This case demonstrates the need for clinical, radiographic and histopathological information to be collated together to reach a definitive diagnosis of a fibro-osseous lesion, whilst using simple, economical and non-invasive investigation techniques to diagnose and treat a lesion with a rare presentation.

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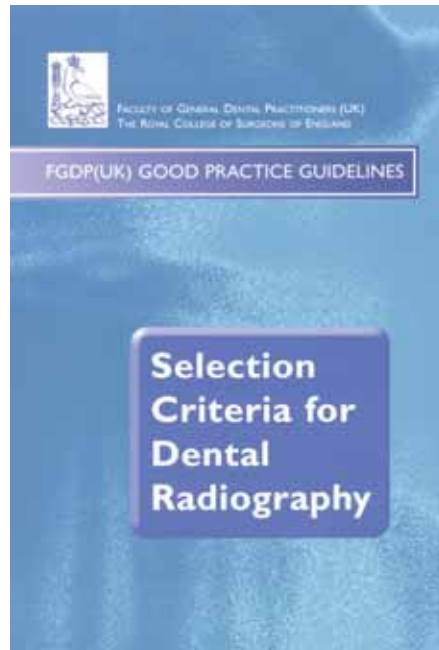
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BOOK REVIEW

Selection Criteria for Dental Radiography, 2nd ed (FGDP(UK) Good Practice Guidelines). M.E. Pendlebury, K. Horner and K.E. Eaton, eds. Faculty of General Dental Practitioners (UK), London, 2004 (108pp., £30.00). ISBN 0-9543451-1-8.

This is the second edition of a work originally published in 1998 by the Faculty of General Dental Practitioners (UK). The original guidelines were developed using scientific evidence where available. The second edition updates the guidelines in the light of new evidence. The levels of evidence supporting the recommendations are indicated by logos of small scales (rather like stars for hotels). The highest level has three scales and the evidence includes at least one randomized clinical trial. Two scales require well-conducted clinical studies, while one scale merely balances expert opinion/experience. Radiation doses and risks are discussed, as is panoramic radiography where three scales indicate that intra-oral radiographs should be considered first because of their better detail and lower radiation doses. There is a thorough discussion of



digital radiographs and their increasing use in practice. Subsequent sections cover radiographs in the management of the developing dentition, dental caries diagnosis, periodontal assessment, the heavily restored dentition, endodontics and implantology. A final section gives the dos and don'ts of good practice and gives ideas for audit. Finally, there are three charts with overviews of the recommendations.

This is an excellent publication. It is clearly written, succinct, easy to refer to and assimilate. The spiral binding allows it to rest open on a work surface where all can see it and the thick paper ensures it will not be easily destroyed by the constant thumbing it deserves. A copy should be in every practice in the land, preferably supplied by the Department of Health.

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NOVEMBER

CPD Answers:

- | | |
|---------------|----------------|
| 1. A, B, C | 6. B, C |
| 2. A, C | 7. A, B, D |
| 3. B, D | 8. A, D |
| 4. A, B, C, D | 9. A, B, C |
| 5. A, C, D | 10. A, B, C, D |