CASE REPORT

A multilocular radiolucency with spindle cell proliferation in a case of ossifying fibroma: a potential pitfall

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Ossifying fibroma (OF) is a benign tumour characterized by cementum or bone formation accompanied by a fibrocellular stroma. The diagnosis of OF may be apparent when the histological features are supported by clinical and radiological information. In this report, an OF with predominantly spindle cell proliferation and purely radiolucent, multilocular lesion in a middle-aged woman is presented.

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Introduction

Ossifying fibroma (OF) belongs to the poorly defined group of fibro-osseous lesions (fibrous dysplasia, ossifying fibroma and osseous dysplasia) involving the jaws and craniofacial bones that result in the replacement of bone by fibrous tissue and subsequent mineralization.¹

Diagnosis of fibro-osseous lesions is troublesome, not only among themselves, but also with diagnosis of other jaw lesions, including those which are odontogenic and non-odontogenic in origin.^{1,2} In the jaws, the mandible is more frequently affected than the maxilla, with a preponderance of lesions diagnosed between the premolar–molar region in the mandible and the posterior region of the maxilla,^{2–4} with or without involving the maxillary sinus.²

We report an OF with predominantly spindle cell proliferation and purely radiolucent lesion with bilateral involvement of the mandible in a middle-aged woman.

Case report

A 45-year-old female presented with a complaint of mild pain and swelling over the lower jaw. The patient said she had been aware of the swelling for 5 years but

had not sought treatment owing to a lack of pain and because of her religious beliefs. The medical and dental history was not significant and there was no history of traumatic episodes preceding the development of the swelling. On examination, a smooth-surfaced swelling was noted in the mandibular anterior region. Introrally, the swelling was noted over the labial aspect of the anterior region. No pulsation was visible and no bruit was heard.

Panoramic radiographs showed a multilocular radiolucent lesion (Figure 1) extending from the mesial root of the right mandibular third molar to the ramus region on the left side of the mandible. Superiorly, it extended to the alveolar crest and the involved teeth lacked lamina dura. Inferiorly, the lesion extended to the lower border of the mandible and the margins were scalloped. Downward bowing of the lower border and circumscription were evident in the symphyseal region. The mandibular canal was displaced on the left side and tooth displacement was absent. An occlusal radiograph showed a multilocular radiolucent lesion imparting a soap bubble appearance (Figure 2a). CT showed an osteolytic, soft-tissue density lesion with thinning and erosion of the buccal cortex in the anterior region of the mandible (Figure 2b,c,d). The clinical diagnoses included ameloblastoma, keratocystic odontogenic tumour, central giant cell granuloma and simple bone cyst. Aspiration was negative and laboratory parameters were within the normal range.

Incisional biopsy showed a benign, monotonous proliferation of spindle cells arranged in short fascicular

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Figure 1 Panoramic radiograph (a) shows a well-defined, multilocular radiolucent lesion involving the right second mandibular molar to the left ramus of the mandible with invagination between the roots of the affected teeth, absent lamina dura and displacement of the inferior mandibular canal on the left side. Near the symphysis, the lower border of the mandible is well defined and scalloped margin is noted elsewhere—as shown in (b) (inverted with Photoshop). Arrows show the position of the mandibular canal

and storiform patterns (Figure 3). Neither basophilic calcification nor immature bone trabeculae were evident in the sections examined. A non-specific working diagnosis of benign non-odontogenic tumour was

suggested. A second opinion from a general pathologist favoured a diagnosis of non-ossifying fibroma.

At surgery, the lingual cortical plate was found to be intact while the buccal cortical plate appeared to be

Figure 2 (a) Occlusal radiograph shows a multilocular radiolucent lesion with a soap bubble appearance causing buccolingual expansion. (b) At the level of hyoid bone, a diffuse expansile osteolytic lesion involving the body of the mandible on the right side with thinning of the cortex and erosion near the parasymphyseal region is shown, as well as small lytic areas on the lingual aspect of the symphyseal region. (c) At the level of the body of mandible, an osteolytic lesion from the right mandibular body to the left side of the premolar region with thinning of the cortex in the symphyseal region is shown. (d) At the level of genioglossus muscle the maximum cortical expansion is shown

Figure 3 Monotonous spindle cell proliferation without cytological atypia set in a storiform growth pattern (haematoxylin and eosin, $\times 240$)

egg-shell thin. The lesion was removed in a single piece. Owing to root resorption, the incisors on the left side together with incisors and canine on the right side were extracted and the right premolars were endodontically restored. The surgical wound was closed without placing any graft or bone wax material. Histopathological examination of the surgical specimen showed cellular features similar to the incisional sample, but basophilic material with osteoid seam (Figure 4), bony trabeculae and fibrosis were evident in focal areas. There was no defined capsule and the lesion merged with the residual bone at the periphery. The final diagnosis was ossifying fibroma. The post-operative period was uneventful. At the 5-month follow-up, bone density in the left mandibular ramus region with flattening of the scalloped margins was evident on the panoramic radiograph (Figure 5).

Figure 4 Monotonous spindle cell proliferation and focal basophilic calcification with prominent osteoid seam [haematoxylin and eosin (H&E), $\times 100$]. Inset: basophilic calcification without osteoid seam within a cellular background (H&E, $\times 240$)

Figure 5 Follow-up panoramic radiograph shows bone formation on the left posterior region and flattening of the lower border of the mandible

Discussion

OF may present as an asymptomatic or symptomatic lesion with swelling or asymmetry of the jaws with mild tenderness.⁴ Liu *et al.*⁵ in an analysis of 20 Chinese patients with OF, found no gender significance compared with exclusive female involvement in 24 patients of Hong Kong Chinese descent.² The mean age also differed between these two Chinese groups, being 28 years (range 6–57 years) for Chinese and 38 years (range 10–80 years) for Hong Kong Chinese for both the maxillary and mandibular locations. However, both studies observed that patients with maxillary lesions were a decade younger at presentation than those with mandibular lesions.^{2,5}

Radiographically, OF may present as radiolucent, radio-opaque or both. The radiographic appearance is related to age at presentation, *i.e.* lesions found in older adults show a mixed pattern while a radiolucent pattern is frequently found with younger patients.² A similar observation was also espoused by Liu *et al.*⁵ in a recurrent case of OF, with the radio-graphic pattern changing from unilocular to multilocular and then to a radio-opaque lesion. Of the 20 cases examined separately by MacDonald-Janowski and Li² and Liu *et al.*,⁵ only a single case in each study showed a multilocular but radiolucent lesion: in a 26-year-old female and 12-year-old female, respectively. However, the current case occurred in an older patient (aged 45 years), who said that the lesion was present for more than 5 years without any discomfort and she had not sought treatment because of religious beliefs. According to Su *et al.*,⁴ the border characteristics of OF may be well defined with (21%) or without (40%) a sclerotic border or show an ill-defined border (15%). The current lesion, as evident from the conventional radiographs (Figures 1 and 2a), was a well-defined multilocular radiolucent lesion with scalloped margins but lacked a sclerotic border. Other radiographic features such as cortical expansion, absent lamina dura, involvement of both sides of the jaw, bowing of the lower border of the mandible and downward displacement of the mandibular canal are in accordance with the previous established reports.^{2,5}

To differentiate the present lesion from more common odontogenic lesions, such as keratocystic odontogenic tumour and ameloblastoma, based on conventional radiography may be difficult owing to the nature of multilocular radiolucency and scalloped margins, but central giant cell granuloma is more common in the paediatric population. Simple bone cyst, although unlikely to be extensive as in the present case, may simulate the invagination between the roots of the teeth and lack of displacement of the involved teeth.

Histologically, OF may show diverse features that vary from equal proportion of calcified material with prominent osteoblast rimming set in a stromal cellular fibroblastic spindle cell proliferation with focal areas of storiform growth pattern to predominantly cellular spindle cell population with focal areas of osteoid or bony trabeculae lacking osteoblast rimming.⁶ The present case falls at the latter end of the spectrum and was diagnostically difficult to categorize as OF based on the incisional sample from benign fibrous histiocytoma and non-OF (Figure 3). Although the former lesion is common in the mandible, the latter lesion shows a preference for the mandibular body and the ramus region but is uncommon in the maxillofacial skeleton.⁷ A reliable diagnosis of OF, however, in the present case, became evident after thorough examination of the surgical specimen and upon identification of basophilic material with prominent osteoid seam (Figure 4) and areas of bony trabeculae that constituted a minor

component of the entire specimen. The histological pattern, as noted in the current case, is considered to be a rare feature of OF and radiologically corresponds to purely radiolucent lesions.^{4,7} It would be prudent for a pathologist to exercise caution and corroborate pertinent imaging results before rendering a diagnosis when confronted with a similar situation to aid in the surgical planning.

The present case, although shelled out during surgery, lacked an identifiable capsule, a feature generally noted in only 44% of OFs.⁷ It should be emphasized that owing to the larger size and purely radiolytic and multilocular nature of the present lesion, the correct clinical diagnosis of OF was difficult to render. The literature also revealed that in only 39% of the cases was a correct clinical diagnosis of OF possible.³

The general consensus regarding treatment of OF is enucleation or curettage, although resection is performed in some cases.^{1,3} Although the follow-up is short, the current case showed a good deal of bone density 5 months post-operatively, indicating that conservative management can be more productive than resection,¹ which carries the risk of recurrence.³

In conclusion, although OFs with spindle cell components and storiform patterns are not rare, the case reported here could lead to potential misinterpretation owing to prominent cellularity, multilocular radiolucency and paucity of calcified material, especially from a limited biopsy.

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