Florid Osseous Dysplasia: Report of Two Cases and a Review of the Literature

Displasia Òsea Florida: Reporte de Dos Casos y Revisión de la Literatura

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INTRODUCTION

Florid osseous dysplasia (FOD) is an osseous dysplasia rarely presenting in the jaws. It belongs to the spectrum of fibro-osseous lesions and represents a reactive process in which normal bone is replaced by poorly cellularized cementum-like materials and cellular fibrous connective tissues (Lin et al., 2010).

The latest World Health Organization’s classification (WHO) in 2005 of benign tumors recognized FOD as lesions associated with bone and designated them as florid osseous dysplasia that forms along with focal osseous dysplasia, peraipal osseous dysplasia and familial gigantiform cementoma, the group of osseous dysplasia (Barnes et al., 2005).

FOD is commonly seen in black women of middle age (40-50 years-old). It often occurs bilaterally in the mandible with symmetric involvements (Lin et al.), but it can also affect the maxilla. The process may be totally asymptomatic and in such cases, the lesion is detected when radiographs are taken for some other purposes.

But when it becomes symptomatic due to a secondary infection, the treatment is problematic because FOD lesions cannot be successfully treated only by the administration of antibiotics.

This paper describes the case of 2 patients who were diagnosed with FOD, revealed by secondary infections. The diagnosis was based on clinical and radiographic findings, as biopsy is contraindicated. Radiological and clinical features of FOD and its management will be also discussed on the basis of recent literature.

CASE 1

A 65-year-old white woman was referred for an acute pain associated with her complete denture and the apparition of a yellow tissue on the alveolar ridge of the left mandibular area. Symptoms started three months ago. The patient presented with hypertension and diabetes type 2.

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An extraoral examination did not reveal any particular signs and no swelling was detectable. The intraoral examination showed that the patient is edentulous and showed on the left alveolar ridge, in the premolar area of the mandible, an ulceration of the oral mucosa revealing a hard yellow tissue of 0.5 mm of diameter, surrounded by an inflammatory mucosa (Fig. 1). The right alveolar ridge of the mandible also presented the same tissue detected on the left side, but with a smaller diameter and no pain in this area (Fig. 2).

Panoramic radiograph showed in the right maxilla an impacted canine. In the mandible, lobular, irregularly shaped radiopacities were almost symmetrically observed in the premolar-molar area of the right and left region of the mandible. These opacities were surrounded by a radiolucent zone (Fig. 3). The lesion spared the inferior alveolar nerve. The lesion was supposed to be florid osseous dysplasia.
Conservative treatment using surgical debridement of the lesion and removal of sequestrum, was conducted under local anesthesia (Figs. 4 and 5). An anatomopathological examination of the specimen showed an irregular mass of cementum and osseous tissue within a fibrous stroma (Fig. 6). The diagnosis of FOD was then confirmed.

The case has been followed-up over the last 5 months with no symptoms in the left area of the mandible. In the right mandibular region, a purulent exudate was detected, and the same management was adopted.

CASE 2

A 70-year-old black woman was admitted in November 2010 with a painless swelling and the persistence of a sinus tract in the left submandibular area. Symptoms appeared 2 years ago with episodes of swelling and suppuration, followed by episodes of regression, but always with the persistence of the sinus tract. The patient never consulted for medical care and took a self-medication based on plants. The patient was systemically healthy.

The extraoral examination showed a facial asymmetry due to the swelling of the left mandibular area (Fig. 7). It also showed a sinus tract in the submandibular area with a purulent exudate that is surrounded by a sclerotic skin (Fig. 8). Palpation of the
swelling was hard and the examination of the ganglion areas was negative. The intraoral examination revealed some decayed teeth and missing teeth (Fig. 9). The vestibule of the region of /5 and /6 presented an expansion (Fig. 10). /5 was vital, /6 presented a radicular caries and 4/ was a retained root.

Orthopantomograph showed that the interradicular and the apical region of /6 presented throughout the alveolar process, irregular, amorphous radiopacities, extended to the left molar area but not to the inferior alveolar nerve, and surrounded by a radiolucent zone (Fig. 11). A mixed lesion near the periapical zone of /5, with small opacities surrounded

Fig. 8. Cutaneous sinus tract with purulent exudates surrounded by a sclerotic skin.

Fig. 9. Intraoral view showing missing teeth, decayed teeth and retained roots.

Fig. 11. Orthopantomogram showing irregular, amorphous radiopacities in both quadrants of the mandible. In the left region, radiopacities are surrounded by a radiolucent zone. In the maxilla, there was some radiopacities with regard to 5/, 6/ and 7/ (arrows).
by a radiolucent area, was also observed. In the right quadrant of the mandible, particularly in the molar area, there was lobular shaped radiopacities (Fig. 11). Additionally, there was some radiopacities with regard to the periapex of 5\textsubscript{i}, 6\textsubscript{i} and 7\textsubscript{i} (Fig. 11). The retained diagnosis was of florid osseous dysplasia.

The patient was then treated with antibiotics during 10 days. After that, a reduction of purulent exudates and the improvement of the skin aspect were noted, but a mild swelling with the sinus tract persisted (Fig. 12).

The excision of the lesion in the symptomatic area of the left mandible was therefore performed along with the extraction of /6 which apices were attached to a mass of the lesion (Figs. 13 and 14). The anathomopathological exam was compatible with the diagnosis of FOD. It revealed the presence of an amorphous, acellular eosinophilic matrix resembling cementum, within a fibrous stroma (Fig. 15). A follow-up was made only 4 months after surgical treatment. The patient was asymptomatic (Fig. 16). The extraction of all retained root and decayed teeth was also performed, to allow the patient to wear a complete denture.

Fig. 12. Aspect of the cutaneous fistula and the skin after antibiotic treatment.

Fig. 13. Surgical treatment: after incision and flap elevation, the hard mass is covered by a fibrous tissue.

Fig. 14. Lesion specimen with the extracted tooth /6 (arrow).

Fig. 15. Amorphous acellular eosinophilic matrix resembling cementum, within a fibrous stroma (hematoxylin and eosin stain, magnification x 100).
DISCUSSION

Osseous dysplasias (OD) are a variant of non-neoplastic fibro-osseous lesions (Barnes et al.; Heuberger et al., 2010; Kramer et al., 1992). These benign lesions are characterized by an alteration of bone structure. The normal architecture of bone is replaced by fibroblasts and collagen fibers, and also by a variable quantity of mineralized material (Su et al., 1997).

Florid osseous dysplasia (FOD), which is one group of osseous dysplasia, was first described by Melrose et al. (1976), and has been previously referred to as sclerosing osteitis, multiple enostoses, diffuse chronic osteomyelitis, gigantiform cementoma, and multiple cement-ossifying fibroma. The terminology and classification of these fibro-osseous lesions has long been a matter of discussion for pathologists and clinicians (Kramer et al.). The second edition of the WHO’s Histological Typing of Odontogenic tumours in 1992 recognized them as “florid cemento-osseous dysplasia”, that form with “periapical cemental dysplasia” and “other cemento-osseous dysplasia”, the group of cemento-osseous dysplasia (COD) (Kramer et al.) (Table I).

Because the discussions during these last decades about whether cementum-like tissue is present, it has been decided to give-up the term of “cement” in the latest WHO’s classification of odontogenic tumours in 2005, COD has been therefore called osseous dysplasias (Barnes et al.). The core of this latest classification is the concept of spectrum of clinicopathological entities in which the diagnosis can only be made on the basis of a full consideration of clinical, radiological and histological features (Barnes et al.; Speight & Carlos, 2006). This classification recognises in OD 4 groups (Barnes et al.): Periapical osseous dysplasia (POD) which involves a small number of teeth and it’s localised only in the anterior region of the mandible (Heuberger et al.; Kawai et al., 1999; Slootweg, 2010); focal osseous dysplasia (FocOD) which is the same lesion as POD but localised in the posterior region; florid osseous dysplasia (FOD) is a more extensive lesion that may be confined to two quadrants or more; and familial gigantiform cementoma which involves multiple quadrants while being expansible, and it shows an autosomal dominant inheritance (Slootweg) (Table I).

FOD is a very rare condition presenting in the jaw. The true incidence of the lesion is unknown and at present time, there is no satisfactory explanation for

Fig. 16. Orthopantomogram 4 months after surgery in the left region of the mandible.
the reported gender and racial predilection (Beylouni et al., 1998). These lesions are most commonly seen in middle aged (40-50) black women, unlike the first case reported where the woman was white. They also occur in Caucasians and Asians (Miyake & Nagahata, 1999). In a study of Kawai et al., OD lesions in Japanese men tend to occur at an older age than in women (Kawai et al.). This fact may support the hypothesis that OD lesions represent a dysplastic process related to hormonal imbalance, which influences bone remodeling (Kramer et al.). In a systematic review of MacDonald-Jankowski (2003), OD are most prevalent in women of middle to old age (92%). These would also suggest that sex-linked factors are implicated in the aetiology. However, the aetiology of this disorder is unknown (Beylouni et al.). Histopathology shows that it may be due to reactive or dysplastic changes of the periodontal ligament (Coleman et al., 1996), but when periodontal ligament spaces of related tooth are visible radiographically, it would seem that the lesions in the vicinity of the teeth generally have little possibility of originating from their periodontal ligament tissues (Kawai et al.). In the study of Kawai et al., almost all teeth related to the lesion had normal periodontal ligament spaces, suggesting these lesions are not of periodontal ligament origin but of medullary bone origin.

This disorder may also have a genetic component. The familial form is reported to be an autosomal dominant trait with variable expression (Young et al., 1989). It is also believed that the familial form is different from the non familial form, clinically and pathologically (Coleman et al.).

Clinically, the process of FOD may be totally asymptomatic and, in such cases, the lesion is detected when radiographs are taken for other purposes (Beylouni et al.). FOD lesions can easily become secondarily infected because they are located in the vicinity of apical areas of teeth which are frequently exposed to pulpal and periodontal infections (Kawai et al.; Marx & Stern, 2003). Symptoms such as dull pain or drainage, and mucosal ulceration, are almost always associated with exposure of sclerotic calcified masses in the oral cavity (Bencharit et al., 2003). This may occur as the result of progressive alveolar atrophy under a denture, as shown in the first case reported, or after extraction of teeth in the affected area (Beylouni et al.; Bencharit et al.; Waldron, 1985).

Some of these lesions will also form drainage tracts intraorally or extraorally, as reported in case 2 (Marx & Stern). These FOD lesions have generally a limited growth potential, whereas neoplastic lesions do

Table I. Classification of odontogenic tumors of the jaws (examples of bone related lesions) (Barnes et al., 2005; Heuberger et al., 2010; Kramer et al., 1992).

<table>
<thead>
<tr>
<th>WHO classification, 2nd Edition (Kramer et al., 1992)</th>
<th>WHO classification, the last Edition (Barnes et al., 2005)</th>
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<tr>
<td>Neoplasms and Other Lesions Related to Bone</td>
<td>Bone-related lesions</td>
</tr>
<tr>
<td>1. Osteogenic neoplasms</td>
<td>1. Ossifying fibroma (9262/0)</td>
</tr>
<tr>
<td>1.1. Cemento-ossifying fibroma (cementifying fibroma, ossifying fibroma)</td>
<td>2. Fibrous dysplasia (9262/0)</td>
</tr>
<tr>
<td>2. Non-neoplastic bone lesions</td>
<td>3. Osseous dysplasia (9262/0)</td>
</tr>
<tr>
<td>2.1. Fibrous dysplasia of the jaws (74910)</td>
<td>-Periapical osseous dysplasia</td>
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<tr>
<td>2.2. Cemento-osseous dysplasias</td>
<td>-Focal osseous dysplasia</td>
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<tr>
<td>-Periapical cemental dysplasia (periapical fibrous dysplasia) (927210)</td>
<td>-Familial gigantiform cementoma</td>
</tr>
<tr>
<td>-Florid cemento-osseous dysplasia (gigantiform cementoma, familial multiple cementomas) (927510)</td>
<td>4. Central giant cell lesion (granuloma)</td>
</tr>
<tr>
<td>-Other cemento-osseous dysplasias</td>
<td>5. Cherubism (9262/0)</td>
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<tr>
<td>2.3. Cherubism (familial multilocular cystic disease of the jaws) (70980)</td>
<td>6. Aneurysmal bone cyst (9262/0)</td>
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<tr>
<td>2.4. Central giant cell granuloma (44130)</td>
<td>7. Simple bone cyst (9262/0)</td>
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<tr>
<td>2.5. Aneurysmal bone cyst (33640)</td>
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<tr>
<td>2.6. Solitary bone cyst (traumatic, simple, haemorrhagic bone cyst) (33404)</td>
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not (Kawai et al.), but rare cases may show mild expansion (Marx & Stern).

The radiographic appearance of FOD lesions varies depending on the stage. The lesion is poorly circumscribed and initially radiolucent (Coleman et al.). The lesion opacifies progressively as it becomes more mature. The vascularity of the lesion decreases with the increased cementum-like deposition (Ackerman & Altini, 1992). The same lesion may appear in different stages. The classic appearance, as the first case reported, includes diffuse, lobular, irregular-shaped radiopacities throughout the alveolar process of maxilla and mandible (Beylouni et al.). The size of the lesion can vary from less than 1cm to 10cm (Ackerman & Altini). The location of the lesion is usually confined to the interadicular and periapical alveolar area, although an aggressive lesion may expand more inferiorly and superiorly (Kawai et al.; Marx & Stern). In the mandible, FOD lesions do not involve the inferior border, they do not occur in the rami and they are found superior to the inferior alveolar nerve (Marx & Stern; MacDonald-Jankowski, 2004). Finally, computed tomography scanning can be useful in the evaluation of the extension of the lesions and their relationship with neighboring structures, in particular in the maxilla (Beylouni et al.).

These clinical and radiographic features of FOD may suggest some other pathologies that are important to differentiate from FOD, such as diffuse sclerosing osteomyelitis (DSO) (Gonçalves et al., 2000). However, DSO of the jaws is characterised by symptoms of inflammation with varying degrees of sclerosis or lucency (Groot et al., 1996). It is not restricted to the mandible and is associated with clear-cut features of osteomyelitis that include pain, fistula formation, sequestration, and the presence of an infectious focus (Groot et al.). Another disease that may resemble FOD is Paget’s disease of the bone that may have a cotton wool appearance. However, this condition affects the entire mandible and shows loss of lamina dura, whereas FOD is centered above the inferior alveolar nerve (Langlais et al., 1995). Paget’s disease is often polyostotic, involving other bones such as spine, femur and skull (White & Pharoah, 2000), and produce elevated alkaline phosphate levels (Wolf et al., 1989). FOD can also suggest multiple endosteomas in Gardner syndrome, but in this syndrome, there is an involvement of other bones, dental abnormalities and benign skin tumors. The diagnosis is based on colonoscopy demonstrating polyps (Marx & Stern; Dghoughi et al., 2010).

The histopathological feature of FOD is similar to all the three other types of OD. It depends on the stage or the degree of calcification of the lesion. Early lesions are composed of cellular fibrous tissue containing variable amounts of new woven bone trabeculae with osteoblast rimming. Occasional spherical “cementicle” calcifications may also be seen. In more mature lesions, there may be woven and lamellar bone and cementum-like tissue coalesces into fused sclerotic masses of globular basophilic calcifications. There may be prominent reversal and resting lines giving a pagetoid appearance (Speight & Carlos; Marx & Stern).

An important association with FOD is the solitary bone cyst (SBC) (MacDonald-Jankowski, 2004). Non-epithelium-lined cyst formation associated with FOD was first reported by Melrose et al. This appears as a moderately defined radiolucency. Classically, it displays little or no bucco-lingual expansion and can displace the inferior dental canal downwards (MacDonald-Jankowski, 2004). Furthermore, Melrose (1997) noted that the classical SBC that affects teenage patients generally heals completely after surgery, whereas those associated with FOD may not do so. Instead, they are filled in by abnormal mineralized tissue similar to that of FOD (MacDonald-Jankowski, 2004). Hypothesis about cyst formation are conflicting (Wakasa et al., 2002). Speculation regarding the aetiology of SBC continues and there is no codified treatment of these cysts associated with FOD.

Regarding the treatment of FOD, in case of asymptomatic lesions, treatment and biopsy are not indicated. It is wise to keep the patient under observation (Melrose et al.; Beylouni et al.; Miyake & Nagahata; Dghoughi et al.; Waldron, 1993). In the absence of clinical signs, reevaluation with panoramic radiographs every 2 or 3 years is adequate (Beylouni et al.).

The treatment must be also preventive to avoid infectious complications provoked by untimely tooth extractions, biopsies and trauma of covering fibromucosa (Bayi et al., 2004). An antibioprophylaxis is also recommended prior to endodontic treatment or any other dental care in FOD affected patients (Dumas et al., 2000). Treatment is required when infection occurs. The secondary infection usually results from trauma to the area (Melrose et al.; Miyake & Nagahata). In edentulous area, minor irritation from a denture may cause serious consequences (Benchcharit et al.). Infection may not respond to
antibiotics because, when administered, they appear to have poor tissue diffusion (Beylouni et al.) due to the avascular nature of the lesion, requiring therefore surgical debridement or resection of the symptomatic area only, followed by an anatomicopathological examination of the specimen (Kawai et al.; Marx & Stern; Bencharit et al.). Asymptomatic areas should not be excised because the alveolar resection leaves a significant defect to the level of the inferior border (Marx & Stern).

Surgery is usually followed by healing in that area (Beylouni et al.). A more surgical approach is required when conservative treatment fails to control a recurrent lesion. Complete removal of necrotic tissue may result in a large discontinuity defect (Bencharit et al.), which may require bony reconstruction at a later date with a bone graft (Marx & Stern), it can be then followed by implant therapy to restore adequate form and function (Bencharit et al.).

In the two cases reported, FOD was symptomatic whether by an alveolar atrophy under a complete denture causing a mucosal ulceration (case 1) or a pulp infection (case 2). It was then clear that the patients needed surgery. The treatment consisted of conservative enucleation of the lesion in the symptomatic areas only.

CONCLUSION

FOD is a rare condition that can be encountered in clinical practice. Its diagnosis can be easy when it relies on adequate clinical and radiographic features, and the dentist should be able to evoke it easily, and therefore manage it adequately. Waldron wrote in 1993: “In absence of good clinical and radiologic information, a pathologist can only state that a given biopsy is consistent with fibro-osseous lesions, with adequate clinical and radiologic information most lesions can be assigned with reasonable certainty into one of several categories” (Waldron, 1993).