An Unusual Case of Florid Osseous Dysplasia Complicated by an Oral Cutaneous Fistula

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ABSTRACT

Osseous Dysplasia is a benign disorder of the maxillary in which the cemento-osseous tissue replaces the normal architecture of the bone. It is encountered in African women from 40 to 50 years old. We report the case of a 53-year-old woman with florid osseous dysplasia complicated by cutaneous fistula. Florid Osseous Dysplasia is usually asymptomatic; the discovery is often fortuitous during a routine radiological examination. However, an infection may occur, and the lesion progresses to the symptomatic phase, which is manifested by pain, purulent exudate, fistulization, and sequestration. Various radiological aspects can be observed depending on the stage of development achieved. In the absence of infectious complications, the best course of action is abstention and surveillance.

KEYWORDS: Florid osseous dysplasia, cutaneous sinus tract, Complication

INTRODUCTION

Cemento-osseous dysplasia is an infection that is part of the maxillary benign fibro-bone lesions. Usually asymptomatic, discovery is most often incidentally done during a radiographic examination. More rarely, an infection can occur and the disease becomes symptomatic and manifests itself in pain or purulent exudate with fistulization and formation of sequestration. The radiographic aspect is initially osteolytic, and then there are radio-opaque inclusions which subsequently, converge to form a single mass surrounded by a radiolucent edging. The therapeutic attitude consists in monitoring the lesions, with surgery being justified only in cases of infectious complications. We report a case of florid osseous dysplasia that was complicated by a cutaneous sinus tract. Radiographic and clinical features of osseous dysplasia and its management will then be discussed on the basis of recent literature.

CASE REPORT

In 2018, a 53-year-old woman was sent to the Oral Surgery Service of the center of consultation and dental treatment (CCTD-Rabat) for a purulent discharge at the right submaxillary region. Symptoms began 2 years ago with episodes of swelling, suppuration, and regression, but always with the persistence of cutaneous fistula. Extraoral examination revealed a cutaneous fistula in the left submaxillary region with a bloody purulent exudate surrounded by silent sclerosis skin and the examination of the lymph nodes was negative (Fig. 1).

The intraoral examination showed widespread moderate chronic periodontitis. Upon the examination of the right mandibular molar region (Fig. 2), ulceration of the oral mucosa exposed a fistula at the summit of the alveolar crest (Fig. 2).
mucosa revealed a fistula at the top of the alveolar crest. The palpation made purulent serosities spring.

Dentascan showed multiple well-defined sclerotic masse with a hypodense border in the right the molar region of the mandible just below the summit of the alveolar crest. This sclerotic masse appeared to be attached to the root apices of the 46 and 45; the limits of the mandibular canal were closely related to the injury (Fig. 3).

The diagnosis obtained was then that of florid osseous dysplasia. The presence of purulent serosities indicated the over-infection of the lesion. The patient underwent antibiotics to limit the infection. Uninfected florid osseous dysplasia lesions didn’t require any treatment procedure; but due to septic complication and symptomatology, a decision was made to enucleate the lesion and extract the teeth (molar and pre-molar) that were related to the lesion (fig.4-6).

The flap was sutured with interrupted sutures. The patient was dismissed with routine postoperative instructions (antiseptic rinses) and antibiotic therapy: 1g amoxicillin/12h during 10 days.

The anatomopathological exam was compatible with the diagnosis of FOD and the examination revealed the presence of an amorphous and dense cement bone-like structures a fibrous stroma consisting of fibroblastic cells. A follow up was made only 4 months after surgical treatment (fig. 7).

Fig. 3: Dentascan. (Radial reconstruction) mandibular canal (arrow) away from the lesion.

Fig. 4: After incision and flap elevation, the hard mass is shown to be covered with a fibrous tissue.

Fig. 5: Bone surrounding the hard mass is removed.

Fig. 6: Enucleated surgical specimen.

Fig. 8: Presence of an amorphous, acellular eosinophilic matrix resembling cementum, within a fibrous stroma. (HE ×200)
DISCUSSION

As this observation points out, the complications are one of the modes of revelation of florid osseous dysplasia. The florid osseous dysplasia is a benign and rare fibro-osseous lesion. It was first reported by Bhaskar and Cutright in 1968 as multiple enostoses and then by Melrose in 1975. The new edition (WHO 2017 classification of odontogenic cysts and tumors) returns to cemento-osseous dysplasia (COD) terminology in order to recognize their odontogenic origin, with three variants1:

- Periapical bone dysplasia: the anterior region of the mandible
- Focal bone dysplasia: single injury in a region other than the anterior mandibular region
- Cement fluorde bone dysplasia: multifocal florid osseous dysplasia mainly affects women of middle age and African origin; our case belongs to this type.

Clinically, florid osseous dysplasia is mostly asymptomatic. In general, the teeth related to the lesions are alive. Symptoms arise from consecutive infectious complications resulting from exposure of lesions in the buccal area, by dental entry door (endodontic or periodontal) or surgical (avulsion, biopsy). There are then pains, a purulent discharge with fistulization and then the formation of sequestration imposing surgical treatment as is the case for our observation. In a few very rare cases of florid osseous dysplasia, a swelling corresponding to a cortical blast may exist. Radiologically, florid osseous dysplasia is characterized by several light-ray images with dented contours, sometimes with light-ray edging. These images are located at the level of the four oral quadrants, more willingly in the premolar region in an almost bilateral and symmetrical way. But earlier localizations are frequent.

Histological examination of florid osseous dysplasia lesions show a mixture of bone and pseudo-cemental material within a fibrous stroma with a large capillary network poor in cells. The masses of cement-bone tissue are irregular, lobulated, rather basophilic, relatively avascular and highly calcified. Within these calcified masses there are spaces resembling a spongy bone, poor in fibrous tissue, sometimes empty.

In the case of the sequestration of lesions after infectious process, these spaces contain necrotic tissue. There is sometimes a real ankylosis between cement-bone and dental roots. Differential diagnosis is made with diffuse sclerosing osteomyelitis (DSO), Paget’s disease, maxillofacial manifestations of Gardner’s syndrome, and family gigantiform cementoma. In the absence of clinical symptoms, FOD medical coverage is not required and the patient should be monitored for radiological changes in the behavior of the lesions. The biopsy is not indicated as it can trigger the transition to the symptomatic phase. In the presence of symptoms, two therapeutic attitudes can be considered. For some, antibiotic therapy would be sufficient to ensure the sequestration of sclerotic, avascular, cement-like lesions before their surgical elimination. For others, extensive surgical intervention is recommended at the outset because partial resection does not control the infection and may promote its spread to other OD zones. For our symptomatic case, the second attitude was adopted in a second time as there was no improvement after a long term antibiotic therapy prescribed by a fellow doctor; this is probably due to its low diffusion in sclerotic tissues.

In the absence of sequestration, the florid osseous dysplasia does not detach itself from the healthy bone into a block and it is necessary to carry out a significant curettage of the alveolar bone which is found in the periphery. In our case the surgical the procedure was very difficult because the florid osseous dysplasia was welded to the cellular bone and to the wall of the mandibular canal which was punctured after enucleation of the lesion and the nerve appeared by transparency. Before the procedure, it is necessary to inform the patient of the risk of sensitivity loss.

CONCLUSION

Florid Osseous Dysplasia is a lesion that evolves slowly and without clinical signs; the onset of symptomatology is related to the exposure of sclerotic lesions in the oral area. The discovery is often incidental following a routine radiological examination. The diagnosis is mostly clinical and radiological. The biopsy is counter-indicated due to the propensity for infection. In the absence of infectious complications, the conduct to be held is abstention and supervision. In symptomatic cases, slitting-up or sequestrectomy, if necessary, should be supervised by effective antibiotic therapy.

REFERENCES

Yousra Z et al.: Case of Florid Osseous Dysplasia Complicated by an Oral Cutaneous Fistula

CASE REPORT

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