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Up-to date review and case report

Active surveillance for intraosseous mandibular haemangioma. Case report and literature review

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Abstract – Introduction: Intraosseous mandibular haemangiomas are rare benign tumors.To this date there is no general consensus regarding their treatment. **Observation:** The authors present a case of a 56-year-old patient with swelling of the basilar left edge, without any other associated symptom. Radiolucent images with a well-defined honeycomb structure supports the vascular injury found using dental orthopantomography and cone beam computed tomography. Dynamic magnetic resonance angiography (MRA) showed mild intense intralesional vascularity and a biopsy confirmed the diagnosis of mandibular cavernous haemangioma. Clinical and radiological active surveillance were carried out. The size of the lesion had regressed by more than half in one year and there was no more palpable lump. **Discussion:** The most commonly approved treatment approach of haemangioma is selective embolization followed by complete surgical resection, which may be complicated by bleeding. From the follow-up of this case it seems that an active clinical and radiological surveillance can be proposed in the management of intraosseous mandibular haemangioma with few persistent symptoms and a low risk of complication, these may even regress with time.

Introduction

Intraosseous mandibular haemangiomas are rare benign vascular tumors and represent less than 1% of all intraosseous tumors. They are more predominant with female adults (2:1) with a higher occurrence between the second and fifth decade [1]. The pathophysiology of hemangioma is explained by an early disruption of the vascular system during embryogenesis and should be differentiated from the arteriovenous malformation. The latter is the consequence of late disturbance in embryogenesis with persistence of vascular anastomoses [2]. Haemangioma tends tog row quickly during childhood, then stabilizes during adulthood and may spontaneously regress. It is not usually associated with massive bleeding but remains at high risk of bleeding during its diagnostic investigation [2-4].

Case report

A 56-year-old patient visited with a slight mandibular asymmetry discovered by self-examination two months prior to his visit with no other associated symptoms

This asymmetry was due to a swelling at the left basilar edge of the mandible a without any cutaneous change over the swelling and no major esthetic impact. The intra oral examination showed a palpable lesion extending from the canine's apex to the second premolar tooth at the left mandibular body. The concerned teeth had a positive vitality test and they were motionless. Dental orthopantomography (DOP) with cone beam computed tomography (CBCT) revealed a well-defined lesion of 2 cm, in honeycomb with cortico-medullary dedifferentiation at the lower border of the mandible (Fig. 1-2). The lesion is away from the inferior alveolar nerve and the dental apex. There was a communication between the cubicles

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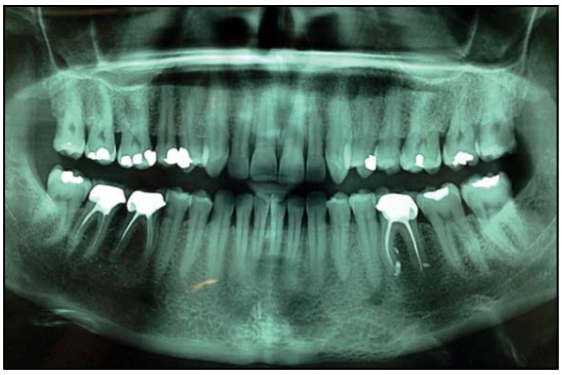

Fig. 1. The orthopanthomogram shows a well-defined lesion 2 cm long axis, honeycombing at the left mandibular basilar edge.

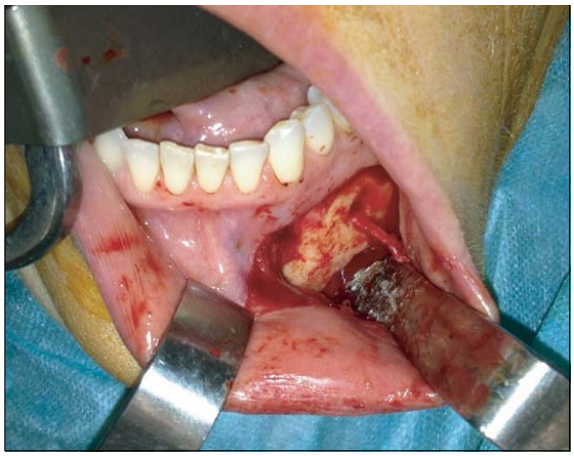

Fig. 3. Intraoperative photograph after biopsy. The limits of the lesion are labeled with felt pen.

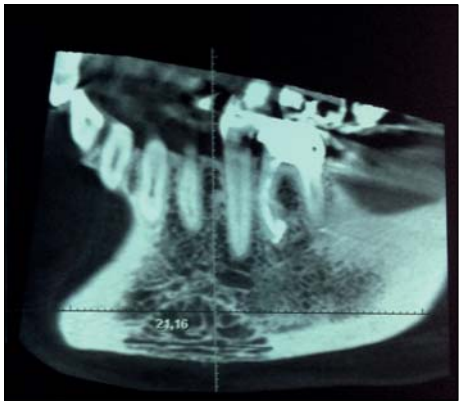

Fig. 2. Cone beam computed tomography shows the cortico-medullary dedifferentiation of the left basilar edge.

evoking vascular injury and more accurately intraosseous haemangioma.

The differential diagnoses of osteolytic multilocular lesion are ameloblastoma with its related aspect of "soap bubbles", and more rarely the central giant cell granuloma or odontogenic myxoma [5].

Because of this radiological evocative aspect of vascular lesion, dynamic MRA used and found a mildly intense intralesional vascularity.

After a multidisciplinary discussion, a biopsy was taken under general anesthesia knowing the risk of potential bleeding and the possible differential diagnosis.

During surgery, the bone appears bluish, slightly rounded and brittle. The bleeding was moderate, controlled by a haemostatic dressing (Fig. 3).

Histological analysis found the presence of vascular capillary structures characteristic of a cavernous haemangioma.

This case describes an asymptomatic mandibular lesion (apart from a discreet bone deformity), with low neurological risk, fracture or bleeding because of its size and location.

Moreover, according to the natural history of this lesion, it is often stable and can regress.

An active clinical and radiological surveillance with DOP is proposed to be carried out annually. At the follow-up consultation one year later, the patient remained asymptomatic and there was no palpable swelling opposite the lesion.

The DOP showed the lesion poorly limited but identifiable through the cortico-medullary dedifferentiation at the left basilar edge (Fig. 4). Comparing the first DOP with the one performed one-year later, the mass was reduced by half with remineralization of a large part of the cortical bone. It was measuring approximately 1 cm at the long axis (Fig. 5).

There was therefore a regression of the lesion with bone remodeling that might have been accelerated by the performed biopsy. This clinical situation was in favour of an active surveillance of intraosseous mandibular haemangioma.

Discussion

The diagnosis of mandibular intraosseous haemangioma is made difficult because of the great clinical and radiological variability and because the patients are usually asymptomatic [1].

Symptoms often result in bones welling. Other symptoms may be present such as pressure sensations, significant gingival bleeding, bluish coloration of the gums, tooth mobility, or paresthesias in the mandibular nerve territory [5].

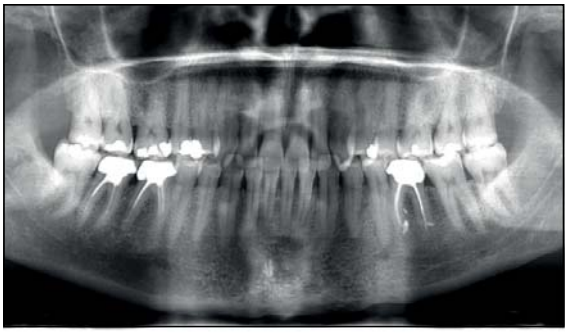

Fig. 4. Orthopantomogram one year of evolution.

Radiologically, the intraosseous haemangioma appears most often as a nonspecific multilocular radiolucent area of poorly defined "soap bubble" or "honeycomb" shapes. It may also appear as a unilocular lesion, sometimes associated with root resorption at the adjacent teeth [1, 5-7]. A cortico-medullaryde differentiation, as in the case presented, can also be a suggestive sign.

CBCT or MRA are useful tools for the etiological assessment and staging of the lesion [1].

Angiography, which is the reference examination, can confirm and quantify the intralesional vascularity [5, 8]. Today this radiological examination is increasingly replaced by dynamic MRA, which is much less invasive [7, 9].

Histologically, hemangiomas are divided into three types: cavernous, capillary, and mixed [1, 10, 11].

They are vascular spaces of variable sizes interposed by fibrous connective tissue. The capillary hemangioma is composed of small vessels with a thick endothelium, while the cavernous hemangioma consists of large vessels and lined with a thin endothelium. The mixed hemangioma hasc haracteristics of both types.

The therapeutic management and the prognosis are similar for all types of hemangiomas [12, 13]. The histological analysis only allows a definitive diagnosis of haemangioma and defines the type. However this biopsy may lead to extensive bleeding [5].

The decision must be made in a multidisciplinary way with advice from radiologists and anesthetists. The biopsy should be performed under general anesthesia with controlled hypotension in order to anticipate possible complications [12].

Some authors advocate a needle aspiration biopsy prior to the biopsy to check its feasibility. In case of major bleeding during the aspiration, this makes the biopsy inappropriate [5].

However, when significant radiological presumption of haemangioma, treatment may be initiated without prior histology [5].

The differential diagnosis includes multiple myeloma, exostosis, osteoid osteoma, eosinophilic granuloma, dermoid cyst and fibrous dysplasia [8].

Currently, there is no consensus on the treatment management of mandibular haemangioma. The treatment depends on various conditions, such as the age of the patient, the size of the lesion, the degree of deformation, and the predictable complications [5, 14].

The most frequently performed procedure is a selective embolization, followed by surgical resection [5, 12, 15, 16]. However, this surgery may cause large osseous defect with immediate reconstruction issues [12].

In the literature other medical and surgical alternatives are available. One of them is a partial resection when the mandible presents a deformity without any other associated symptoms.

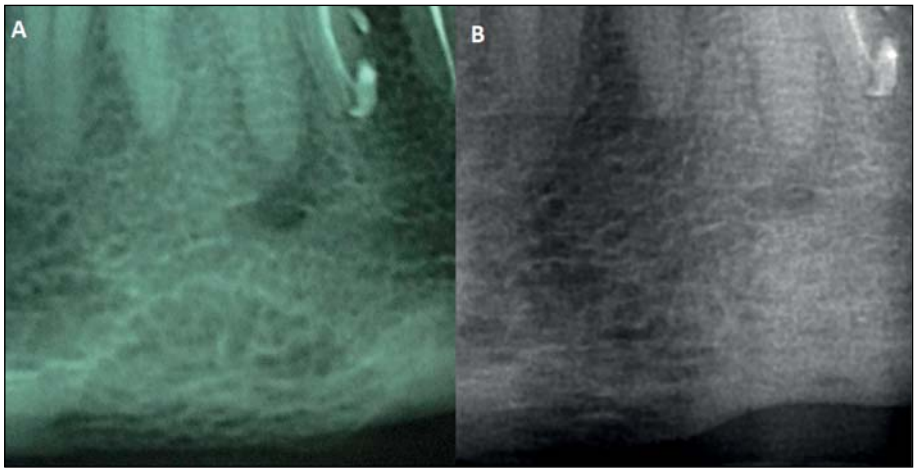

Fig. 5. Comparative orthopantomograms one year of evolution.

Embolization prior to the resection is then not systematic, and control of hemostasisis generally done through bone waxing and electrocautery [12].

Also cryotherapy has been proposed and is adapted to small size lesions but with some side effects such as loss of innervation of adjacent tissues [5].

Injection of intralesionals clerosing agent induces an inflammatory response of the endothelium and causes a localized vascular obliteration while the healthy bone is spared [5, 17].

The sole selective embolization is often associated with the development of collateral vessels and has potentially serious complications such as pulmonary or cerebral embolizations [5]

Finally, external beam radiation therapy, is quite effective, but should performed due to significant side effects such as radiation-induced cancers [1, 17].

The therapeutic decision must be multidisciplinary and most of the seprotocols require specific technical platforms [14].

The active surveillance is rarely mentioned, it is indicated in two conditions: asymptomatic patients or those with minimal facial deformity and without active bleeding [12]. This does not take into account the risks of specific complications of mandibular location: the nervous risk with the lower alveolar nerve, the pathological fracture risk and above all the haemorrhagic risk related to the proximity of the dental structures.

The relationship between hemangioma and teeth can be manifested by the daily hemorrhagic episodes when brushing, feeding or during dental care such as avulsions. Also dental mobility can occur due to involvement of the alveolar bone.

Given the steady natural history for potentially regressive haemangioma, annual monitoring protocol may be proposed and seems suitable. We suggest a quarterly clinical monitoring with a radiological control orthopantomogram. And the patient should be educated to see his physician whenever a new symptom appears

As can be seen in the presented case, active surveillance is a good alternative to the usual managements because after one year of the diagnosis the lesion regressed by more than half its original size, but that might have been accelerated by the performed biopsy.

Conclusion

Active surveillance can therefore be considered for some mandibular haemangioma with few symptoms and low risk of complications in the medium term. Depending on the progression of the lesion, current treatment can obviously be considered secondarily.

Conflicts of interests: Authors' disclosures of potential conflicts of interest.

Competing Interests: None

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Ethical approval

This study was carried out after institutional approval obtained from the Ethics Committee of Pierre Paul Riquet Hospital (n° 523-756-02/15).

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All authors have viewed and agreed to the submission

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