Hemangioameloblastoma of Mandible: A Case Report Regarding a Rare Form of Ameloblastoma

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ABSTRACT:

Hemangioameloblastoma is one of the rare entities of jaw pathologies. Though very few reported in literature, here we are presenting a case report which was treated in our department of maxillofacial surgery successfully. The present case report aims to add on the small literature regarding hemangioameloblastoma.

Key words: Hemangioameloblastoma, Resection, Swelling of Jaw

INTRODUCTION

Ameloblastoma is a benign epithelial odontogenic tumor that usually exhibits aggressive behaviour. It expands severely to the cortical bones and may have a high recurrence rate. It also may cause mobility and displacement of the teeth, as well as root resorption.¹ Hemangiomatous ameloblastoma (HA) was originally described as an ameloblastoma in which part of the tumor contained spaces filled with blood or large endothelial-lined capillaries.² The histological and radiographic

features of this tumor differed from those of a conventional ameloblastoma. Its' histopathological features were consistent with those of a hemangiomatous ameloblastoma, and its standard radiologic features and computed tomography mimicked that of cystic lesions. The behaviour and prognosis of the hemangiomatous ameloblastoma are uncertain because of the small number of documented cases and lack of long-term follow-up, but are thought to be similar to those of the conventional type.

CASE REPORT

A 45 year old female patient reported to the dental hospital with the complaint of swelling in the right jaw since 3 months. The swelling was gradually increasing in size (Figure 1). Extraorally, a diffuse swelling was seen in the right side of the body-ramus region of the mandible. Intra-oral examination revealed a diffuse swelling in relation to 45-47 region obliteration of the buccal sulcus. The case was provisionally diagnosed as a benign bony odontogenic neoplasm. A differential diagnosis of ameloblastoma or keratocystic odontogenic tumour was considered due to the multilocular/soapbubble appearance in the orthopantomogram involving the right side of the mandible (Figure 2).

Under local anaesthesia, an incisional biopsy was taken from the site and sent for histopathological examination. After proper tissue fixation, processing and haematoxylin and eosin staining, the section revealed cystic lining with changes like basal cell nuclei palisading with reverse polarity and sub nuclear vacuolisation. Microscopic examination of the enucleated tissue revealed a plexiform ameloblastoma with a prominent vascular component. The ameloblastoma consisted of anastomosing cords and sheets of odontogenic epithelium in a loosely arranged vascular connective tissue stroma. The epithelium was surrounded by columnar or cuboidal ameloblast-like cells and contained stellate reticulum-like areas.

Degenerative changes in the epithelium and the stroma resulted in the formation of cystic spaces and areas of necrosis. The vascular component consisted of numerous endothelial-lined channels, large blood-filled spaces in the stellate reticulum-like areas that were not lined by endothelial cells, and multiple thrombi with signs of organization. In sections of the lesion, ameloblastic elements and granulation tissue intermingled. In these areas a mixed inflammatory cell infiltrate was present, and reactive endothelial cells participated in the formation of numerous new capillaries (Figure 3). The lesion was diagnosed as an HA.

Since ameloblastoma is an invasive tumour requiring radical surgery, resection of pathology via

hemimandibulectomy of affected side under general anesthesia was planned. Under general anaesthesia, a lip split incision with lower submandibular extension till the post auricular region was given (Figure 4). Skin, fascia, platysma was dissected (Figure 5). Post ligation of the facial artery, the mylohyoid, masseter, medial pterygoid and temporalis muscle was dissected to swing the mandible extra orally through the incision area (Figure 6). The condyle was liberated from lateral pterygoid via myotomy and the capsule via dissection, the whole of the right side of affected mandible with the lesion was sent en toto for histopathological examination (Figure 7).

The area was closed layer by layer via sutures and a vacuum drain placed in the area. Post-operative amoxicillin clavulanic acid as antibiotic and diclofenac sodium as analgesic was provided to the patient. The post-operative stay of the patient was uneventful and the patient was discharged from the ward setup after 20 days and recalled after 1 month. The patient underwent a prosthetic mandibular guiding flange after 3 month of follow up (Figure 8a, b and 9).

DISCUSSION

Vascular lesions of the jaws are of particular interest to the dental community in that fatality following minor procedures is well documented and is found in the literature as early as 1933.³ Various theories have been formulated to explain the pathogenesis of the vascular component in ameloblastomas. During amelogenesis, many capillaries are associated with the outer enamel epithelium. They furnish the profuse blood supply necessary for enamel completion. It is probable that in the HA these blood vessels are abnormally induced and become part of the tumor.⁴ Alternatively, a traumatic incident such as a tooth extraction may provide the stimulus for proliferation of epithelial cell rests in the periodontal ligament and subsequent tumor development. Tissue damage is usually followed by repair, and this involves the formation of granulation tissue in which proliferating endothelial cells and new capillaries are prominent. A disturbance in the repair of

Figure 1: Patient with swelling in the right side of jaw

Figure 2: OPG showing multilocular radiolucency in the right side mandible

Figure 3: H and E section shows vascular component consisting of numerous endothelial-lined channels, large blood-filled spaces in the stellate reticulum like areas that were not lined by endothelial cells, and multiple thrombi with signs of organization.

Figure 4: Mandibular lip split incision on the Operating Table.

Figure 5: Sub platysmal dissection, the mandibular anterior osteotomy being placed in the symphysis region.

Figure 6: De articulation of resected hemimandible

Figure 7: Resected Hemimandible with visible proliferative ameloblastoma

Figure 8a: Post operative profile of patient

Figure 8b: Post Operative Intra oral View

Figure 9: Patient Provided with Guiding Flange

neoplastic odontogenic tissue may result in excessive granulation tissue formation or the development of an abnormal vascular component.⁴

Smith regarded the HA as histologically similar to one of the other recognized types of ameloblastoma and not as a distinct histologic entity. He thought the blood supply to these tumors was variable and that circumstances other than the number and size of the vessels influenced the blood supply.⁵

Ameloblastomas are benign asymptomatic intraosseous lesions that affect the bones of the maxilla-mandibular complex. They interfere with both function and facial esthetics. They originate from the epithelium involved with the formation of teeth: enamel, odontogenic rests of Malassez, reduced enamel epithelium, and odontogenic cyst lining; and are locally invasive with infiltrative growths and frequent recurrences even after radical surgical treatment.⁷

Ameloblastomas are the second most frequent benign odontogenic tumor.⁸⁻¹⁰ They occur in both the maxilla and the mandible, but mainly in the mandible, especially the molars, the mandibular angle, and the ramus. In the maxilla, the molar region is more commonly affected. There is no difference in distribution with regard to sex and race but age is a factor, with adults (mean age, 37 years) affected more often and children only rarely.^{8,11, 12}

Clinically, an ameloblastoma is a hard mass that can cause bone expansion. They are slow growing and progressive, and the adjacent mucosa generally has an aspect of normality without continuity of solution. They are painless, difficult for the patient to perceive in the initial stages, and, while they develop, cortical absorption occurs because of the compression produced by growth, making them palpable; in most cases, diagnosis is complemented by radiography.¹⁰

Ameloblastoma have 3 clinical forms that must be recognized and differentiated, because of different treatments and prognoses, and they are divided according to the histopathologic description into solid or multicystic, unicystic, and peripheral. The

solid or multicystic form is more aggressive and requires a more radical treatment than the unicystic and peripheral types, with a relatively higher rate of recurrence.¹³

Regarding its epidemiology, multicystic ameloblastomas affect patients between the third and seventh decades of life. Clinically, it is a more aggressive variant, because its capacity to infiltrate the bone trabeculae is more evident, and, therefore, there is greater risk of recurrence of these lesions when they are not efficiently removed. The radiographic aspect is a multilocular radiolucent lesion, described as 'soap bubble' or 'honeycomb'.^{10,14,15}

The unicystic ameloblastoma affects more young patients, generally in the second decade of life; its main site is the posterior region of the mandible. Radiographically, it is a multilocular radiolucent mass, which in most cases surrounds the crown of a tooth that has not yet erupted and is commonly mistaken for a dentigerous cyst. Its' low but relentless growth can cause movement of tooth roots and root resorption.⁸ The biologic behaviour of this variant tends to be less invasive than multicystic ameloblastomas.^{10,14-17} They respond more favourably to conservative surgery than do solid or multicystic ameloblastomas.⁶

The peripheral ameloblastoma accounts for only 1% of all cases and is found in the posterior alveolar and gingival mucosa. This lesion has a good prognosis if it is removed at an early stage when it is easily detected clinically, and, because the cortical bone is still preserved, it is a barrier to bone invasion by the peripheral ameloblastoma.¹³

With conventional radiographic examination and CT, it might be difficult to distinguish the HA from the desmoplastic ameloblastoma and other types of ameloblastomas or odontogenic tumors.¹⁸⁻²¹

Treatments can be varied, depending on the histologic type and the location site, as resection (marginal or segmental), enucleation, curettage, marsupialization, cryotherapy, or a combination of these techniques. In spite of these treatment modalities identified in the literature, there is still

controversy about the therapy, either its clinical presentation or its histopathologic characteristics.^{10,13,21} Furthermore, radiotherapy either with or without chemotherapy can also be recommended in specific situations: patients who have already been treated surgically more than once, patients with inoperable lesions, or elderly patients who could not withstand conventional surgery.^{13, 17}

The biologic behavior of HA is thought to be similar to that of the conventional ameloblastoma, but because few cases have been reported, the pathogenesis and clinical features are not yet fully understood and biologic behavior cannot be predicted.

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