Hemangiomatous ameloblastoma: report of a case

***Adisa AO, **Lawal AO, **Olusanya AA, **Arotiba JT**

*Department of Oral Pathology, College of Medicine, University of Ibadan
**Department of Oral and Maxillofacial Surgery, College of Medicine, University of Ibadan, Nigeria

***Correspondence: Adisa AO**
Email: perakin80@hotmail.com

Abstract

Hemangiomatous ameloblastoma is a rare variant of ameloblastoma with only a few cases reported in the E nglish literature. This is a report of a 42 year old man with a recurrent mandibular neoplasm who had r epeated surgical interventions. Clinical and radiographic examinations were not significantly different from other regular variants of ameloblastoma. Microscopic examination however revealed islands of cuboidal and columnar cells arranged in a pallisaded pattern with a central area of squamous metaplasia and stellate reticulum-like cells. There were multiple vascular channels containing red blood cells within the stellate reticulum-like areas. Surgical resection of the mandibular mass was done with immediate reconstruction. It may be concluded that repeated surgical interventions may be a possible etiology for the development of hemangiomatous ameleblastoma.

Key words: Hemangiomatous ameloblastoma, Ibadan, Nigeria

Introduction

Ameloblastomas are benign, locally aggressive and slow-growing odontogenic neoplasms of the jaws which may be solid or cystic. Their polymorphic nature is reflected by the variety of recognized histologic patterns with which they may appear [1]. The follicular and plexiform patterns are the main histologic patterns[2] while the most common cellular variants are the acanthomatous and granular cell types [3]. Other histologic variants are basal cell ameloblastoma, desmoplastic ameloblastoma, hybrid ameloblastoma, keratoameloblastoma, papilliferous keratoameloblastoma, clear cell ameloblastoma and hemangiomatous ameloblastoma [2] . Clinical, radiological and histological reviews suggest that except for the unicystic ameloblastoma, no other histologic variant has significant prognostic value[4]. The 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[1]. The origin of the vascular component of the HA is however not completely resolved. HA was previously known as adamantinohemangioma, ameloblastic hemangioma or hemangioameloblastoma[5]. It was thought to occur due to rupture of enlarged vascular channels in the tumor which become unsupported after degeneration of loose myxoid tissue, leading to escape of blood into empty spaces [5]. Intraosseous purely v ascular lesions of the mandible are rare and mostly present radiographically as a multilocular radiolucent image with honeycomb or soap bubble appearance that mimics ameloblastoma[6]. HA is a rare variant of ameloblastoma with only few cases reported in the English literature. This case is reported to add to the knowledge base of existing literature on this type of tumour.

Case report

A 42-year-old Nigerian male presented at the Oral Diagnosis Clinic of the University College Hospital, Ibadan with a 15 months history of gradually enlarging asymptomatic left mandibular swelling. The patient reported that he had experienced two previous recurrences of a similar lesion in the same region with the present swelling being the third. The first occurrence was in 1992 and he had an enucleation done. The second swelling (first recurrence) occurred in 1996 and was associated with a yellowish odorless discharge. The swelling was enucleated and curetted under local anaesthesia at a private clinic. The second recurrence was in 2004 and enucleation of a residual cystic lesion wa s done under general anaesthesia. This present swelling was noticed about two years ago and had been asymptomatic but progressively increasing in size.
On extra-oral examination, there was a firm swelling of the left side of the mandible from the body to the left pre-auricular region measuring approximately 13 x 16 cm. An isolated area over the mandible appeared nodular (Figure 1).

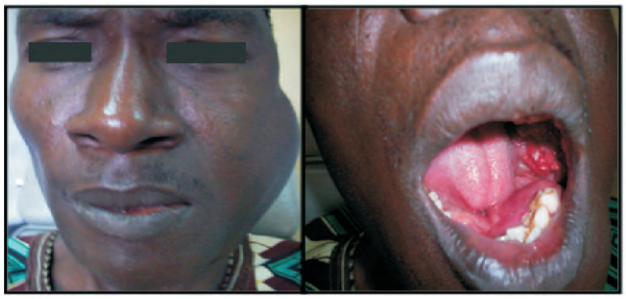

Figure 1. Diffuse extraoral swelling from the left body of the mandible extending to the pre-auricular area. Intra-oral lesion, with buccal/lingual plate expansion and associated exuberant hyperaemic soft tissue swelling.

Intraorally, there was no limitation to mouth opening but a bucco-lingual expansion was seen in the region of mobile teeth 31, 32, 33, 34 and 41. In this region the overlying mucosa was intact but hyperaemic. There was an extensive traumatic ulceration in the region of missing teeth 35, 36, 37 and 38. The lesion was fluctuant anteriorly and aspiration of the swelling yielded 5mls of purulent material stained with blood.
Orthopantomogram showed an osteolytic lesion with bone loss from the 44 extending to the left glenoid fossa involving the entire length and height of the mandible (Figure 2).

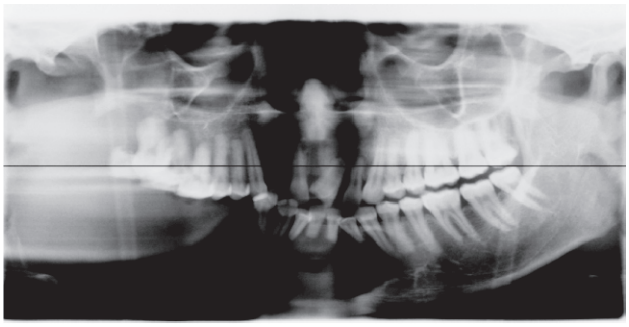

Figure 2. An orthopantomogram which shows a radioluscent left mandibular lesion, extending from the head of the condyle (arrow 1), to the apical region of 44 (arrow 2). The roots of 31, 32,33,34,41 and 42 are truncated.

An incisional biopsy done before treatment reported a hemangiomatous ameloblastoma. The patient had mandibular resection through the socket of 45 and a disarticulation of the left temporomandibular joint. The mandibular defect was reconstructed immediately with a Steinman's pin having an acrylic condylar bulb, whil e the residual soft tissue was closed in layers over it. The entire surgical specimen was then subjected to histopathological examination.
Microscopic examination revealed islands of cuboidal and columnar cells arranged in a pallisaded pattern with a central area of squamous metaplasia and stellate reticulum-like cells. There were multiple vascular channels containing red blood cells within the stellate reticulum-like areas (Figure 3). There was also an isolated area with a definite cystic lining containing ameloblast-like cells. The features were those of an acanthomatous ameloblastoma disposed in a follicular pattern with an associated hemangiomatous component.

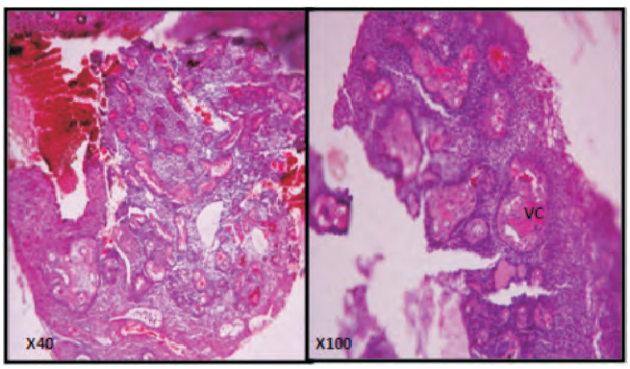

Figure 3. (Hematoxylin and Eosin X40 and X100): A photomicrograph showing islands of odontogenic epithelium consisting of peripherally palisaded columnar and cuboidal cells, and a central area of stellate reticulum-like cells. Some of these central areas have undergone squamous metaplasia with associated proliferation of thin-walled vascular channels (VC).

Discussion

Ameloblastoma is the most common benign tumor of odontogenic epithelial origin[7]. It is more common between 30 - 50 years of age[8] and about 80% involve the mandible[9]. Our patient had a mandibular lesion and was 42 years as at presentation but first noticed the lesion at about the age of 25 years. This connotes that other variants of ameloblastoma may have a similar age distribution with HA, apart from the ameloblastic fibroma and unicystic ameloblastoma that are frequently seen in the second decade[8].
Ameloblastoma with numerous vascular spaces have been described as hemangiomatous ameloblastoma but the biologic behavior and prognosis of these tumors are uncertain because of the small number of documented cases[10]. This paucity of literature on the hemangiomatous ameloblastoma may be a reflection of the rarity of the lesion. It is also possible that a low index of s uspicion for this neoplasm by the pathologist may lead to under-reporting, hence missing cases which may have added to knowledge about its epidemiology and biologic behavior. This is the first documented case from our centre, and to our knowledge the first from Nigeria.
The radiographic appearance in this case is suggestive of a cystic ameloblastoma with possible soft tissue extension, unlike in a study[2] which reported the radiographic picture of HA as being suggestive of a fibro-osseous lesion. The authors however noted that their finding may be non-specific since the radioluscent-opaque appearance was thought to be due to the dense vascular tissue and the thick endosteal reaction [11], and not necessarily due to the degree of calcification in the lesion.
The origin of the vascular channels in HA is still not clear. The controversy that the vascular component o f the HA is part of the neoplastic process, represent a separate neoplasm, or is a hamartomatous malformation has not been satisfactorily resolved[11]. They were thought to arise from vessels associated with the outer enamel epithelium which are abnormally induced during the neoplastic process. Also, the process of repair following tissue damage or surgery may be accompanied by the formation of granulation tissue in which proliferating endothelial cells and new capillaries are prominent[12]. The patient in this report had undergone three previous separate surgical interventions for the same lesion and formation of excessive granulation tissue may account for the vascular proliferation seen within the tumor mass.
It has also been suggested that the HA may represents a collision type of tumor in which two separate tumors grow in the same area and collide resulting in the intermingling of tumor elements [12] e.g a hemangioma and an ameloblastoma. The lack of significant bleeding before and during surgery in this case possibly suggests that this was not a collision type of tumor. Also the vascular channels seen at histology are not suggestive of any variant of a hemangioma or vascular malformation.
The clinical outcome of HA may not be different from that of a conventional ameloblastoma since it is generally accepted that the various histologic patterns of ameloblastoma do not influence the clinical behavior or prognosis [13]. Our patient volunteered a history of neoplastic involvement spanning almost twenty years and this is in keeping with the slowly progressive growth

pattern of other benign ameloblastomas. Report of more cases of HA may however reveal a different biologic behavior.

Conclusion

The etiopathogenesis, behavior and prognosis of the HA are uncertain as only a small number of cases have been documented and there is no report of a long-term follow-up. It may however not differ significantly from the conventional type of ameloblastoma as demonstrated by this report. It may be concluded that repeated surgical interventions may be a possible etiology for the development of HA. It is our consideration that as more cases of HA is reported, it's most probable etiology, biological and clinical behavior will become clearer.

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