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Case Report

Ameloblastoma of the Anterior Mandible - A Case Report

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Abstract

Ameloblastoma is a neoplasm originating from the odontogenic epithelium. The tumor is considered benign despite its locally invasive nature. The pathogenesis of ameloblastoma involves the origin of the tumor from one of the following sources: remnants of dental lamina or remnants of the Hertwig's epithelial root sheath, basal cells of the oral mucosa, developing enamel organ, and epithelial lining of an odontogenic cyst. It can exhibit different clinico-radiographic presentations such as conventional, unicystic and peripheral. Ameloblastomas occur most predominantly in the posterior mandible, in the molar-ascending ramus area, and are quite uncommon in the anterior jaws. Third to seventh decade is the usual age range for the occurrence of ameloblastomas with no gender predilection. A case of ameloblastoma which occurred in the anterior mandible of a 24-year-old female patient is presented. The clinico-pathological features, diagnostic criteria, and management of the case are discussed. The case is important because of its unusual presentation in the anterior region of the mandible and the diagnostic challenge it posed during the initial investigation.

Keywords: Ameloblastoma; Mandible; Odontogenic Tumor; Follicular

Introduction

Ameloblastoma is a benign, locally invasive, polymorphic neoplasm arising from the odontogenic enamel organ which makes them unique to the jaws and tooth bearing areas. The most common presentation for ameloblastoma is a painless swelling of the mandible or maxilla [1]. Ameloblastomas can exhibit different clinic-radiographic presentations such as conventional, unicystic and peripheral. Among these, the conventional ameloblastoma is the most common to occur, followed by the unicystic ameloblastoma [2]. Approximately 90% of ameloblastomas represent the conventional solid or multicystic type. The average age at diagnosis is 36 years [3]. Ameloblastomas are tumors of odontogenic epithelium, potentially originating from dental lamina rests, the developing enamel organ, the epithelial lining of an odontogenic cyst, or the basilar epithelial cells of the gingival surface epithelium [3]. The tumor reveals a microscopic resemblance to the enamel organ of a developing tooth. The tumor cells within may mimic the inner enamel epithelium and central stellate reticulum of the enamel organ, which is considered a classical histopathological appearance

of these tumors. The tumor occurs most predominantly in the posterior mandible, in the molar-ascending ramus area. It is quite uncommon in the anterior jaws [3]. Studies have reported variation in the occurrence within the jaw in different races. Ameloblastoma of the posterior mandible were commonly seen in Asians whereas the anterior mandible is a common site of occurrence among blacks [4,5]. Surgery is the standard treatment for ameloblastomas. Treatment in the form of simple enucleation has not been advocated for many years owing to the aggressive and recurrent nature of the tumor, with a demonstration of recurrence rates of about 60 - 90%. The "radical" surgical option is the current standard of care for ameloblastoma and includes en bloc resection with 1 - 2 cm bone margins and immediate bone reconstruction to restore function and aesthetics. However, new studies have proposed two central molecular pathways, MAPK and SHH, that appear to play key roles in ameloblastic oncogenesis, and each of which offers potential avenues for personalized treatment modalities. Additionally, these discoveries open the doors to future research on odontogenic development, and the relationship of ameloblastoma to several other epithelial neoplasms [1]. A case of ameloblastoma in the anterior region of the mandible is presented in an Asian woman. The clinicopathologic features, diagnostic criteria, and management of the case are discussed.

Case Report

A 24-year-old female presented to the Department of Oral Medicine and Radiology with a complaint of a painful swelling in her anterior mandible. The onset of the swelling was approximately 2 - 3 months prior to her reporting to us. The swelling gradually grew since then, to the present size. There was no pain associated with the swelling initially, however she started to experience pain since the last two weeks. The pain was severe in intensity and continuous in nature, with no exacerbating or relieving factors. Patient was also concerned about the mobility of her lower anterior teeth, which she claimed only began a week ago. She gave no history of trauma and denied any change in sensations in the region. Her past medical, dental, and family histories were insignificant. Extraoral examination revealed an obvious facial asymmetry due to the swelling, which involved the symphysis region and seemed to extend to the right body of the mandible (Figure 1). The swelling was about 3 X 2 cm and tender on palpation. Skin over the swelling appeared stretched but otherwise normal. There was no associated lymphadenopathy.

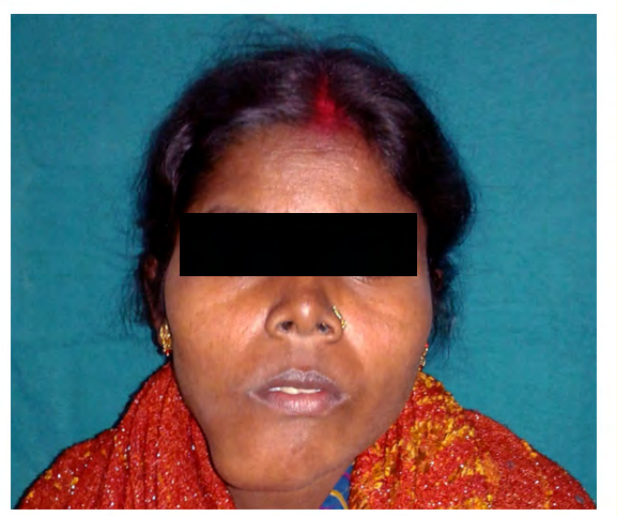

Figure 1: Extraoral presentation of the swelling showing facial asymmetry.

Intraoral examination revealed a swelling obliterating the buccal vestibule extending from the lower right first molar region to the left central incisor region (Figure 2). The overlying mucosa appeared normal. On palpation, the mass was soft, tender, fluctuant, fluid-filled, and non-pulsatile. There was no apparent discharge or bleeding in the region. Mild mobility was elicited from teeth #31, 41, 42, 43 and 46 and all the teeth in the region tested positive to electric pulp vitality testing. Additionally, teeth #31, 41, 42 and 43 appeared to be displaced labially, which according to the patient's history happened in the last one week.

An orthopantomograph (OPG) was ordered which showed a mostly unicystic radiolucency with a few areas of well-defined multilocularity (Figure 3). To our surprise, the radiolucency extended across the midline, from tooth #46 to tooth #35. Root resorption was noted in teeth #34, 33, 32, 42, 43, 46 and 47. The lesion seemed to have involved the lower border of the mandible almost completely from tooth #34 region up to tooth #45. At this stage, odontogenic keratocyst, central giant cell granuloma, ameloblastoma and odontogenic myxoma were considered in the differential diagnosis [3].

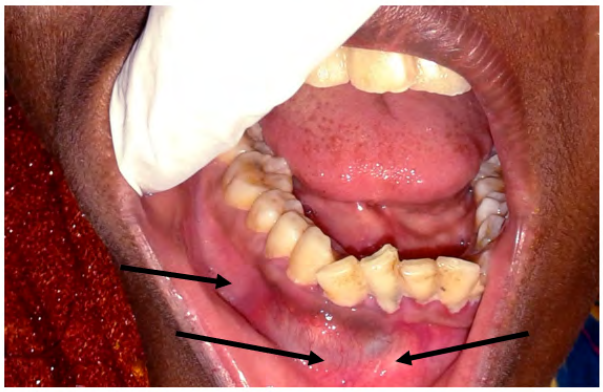

Figure 2: Intraoral presentation of the swelling obliterating the buccal vestibule (arrows).

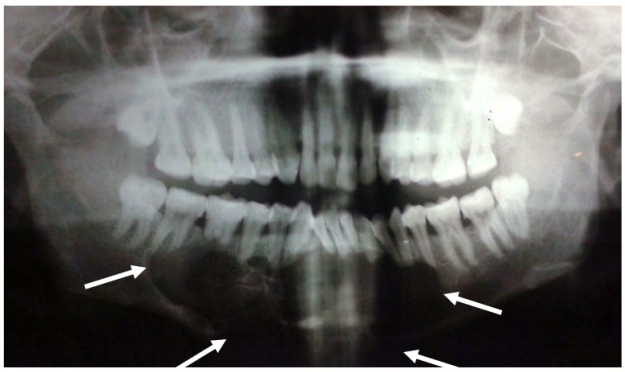

Figure 3: OPG showing unicystic radiolucency with a few areas of well-defined multilocularity. Arrows depicting the extent of the lesion.

Owing to the fluid filled nature of the swelling, a fine needle aspiration biopsy was performed, which produced a dark straw-coloured fluid tinged with blood. Cytological examination revealed the presence of keratin, lymphocytes, desquamated epithelial cells, and red blood cells. Based on the clinical, radiographic, and cytological findings a provisional diagnosis of odontogenic keratocyst was arrived at, and the patient was scheduled for incisional biopsy. The biopsied specimen was submitted for histopathological examination. The microscopic picture of the specimen revealed odontogenic epithelial islands strewn in a fibrous connective tissue stroma. The individual islands showed peripheral columnar cells fulfilling Vickers and Gorlin criteria such as hyperchromatism of basal cell nuclei, palisading with polarization of basal cells, and cytoplasmic vacuolization with intercellular spacing of the lining epithelium [6]. Few of the islands showed squamous metaplasia in the centre (Figure 4a and 4b). The histological features were suggestive of ameloblastoma, follicular type. Following the definitive diagnosis, the patient was scheduled for surgical resection with a 1 cm margin of normal bone and reconstruction procedure under general anaesthesia. Pre-operative blood workup of the patient showed no abnormal values. The mass was excised surgically and reconstruction with her iliac cancellous bone was performed. Post-operative healing was uneventful. The patient is being followed up routinely for the last 4 years and has shown no signs of recurrence so far.

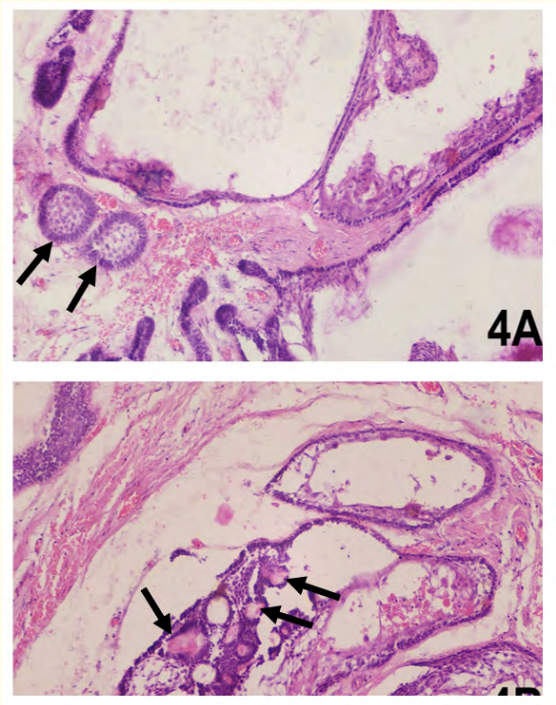

Figure 4A and 4B: Follicles of odontogenic epithelium (arrows 4A) strewn on a fibrous connective tissue background. Some of the islands showed squamous metaplasia in the centre (arrows 4B).

Discussion

Ameloblastoma usually takes a slow-growing course. Although these are considered locally invasive tumors, they are mostly benign [7]. Clinically they present as painless swellings or expansions of the jaws. Over time, if untreated, they may enlarge to enormous size [7]. They are not usually associated with pain. Third to seventh decade is the usual age range for the occurrence of ameloblastomas with no gender predilection [8]. Our case report describes a case of ameloblastoma in a 24-year-old woman, that was associated with pain. The case progressed at a relatively shorter period with a large area of bone destruction. The presence of a painful, fluid-filled lesion in the anterior mandibular region crossing the midline, rendered a diagnostic dilemma, initially, before the incisional biopsy.

The pathogenesis of ameloblastoma involves the origin of the tumor from one of the following sources: remnants of dental lamina or remnants of the Hertwig's epithelial root sheath, basal cells of the oral mucosa, developing enamel organ, and epithelial lining of an odontogenic cyst [9]. Apparently, 50% of ameloblastoma arise from the epithelial lining of dentigerous cysts [10]. At the molecular level, it is postulated to involve mutations or alterations in the genetic material of embryonic stem cells that would differentiate into future tooth forming cells [11].

Radiographically, majority of ameloblastoma appear multicystic. This is considered as the "classical" radiological appearance, popularly described as "soap-bubble" or "honey-comb". These multicystic ameloblastomas are more difficult to treat due to their recurrence potential [9,10]. Expansion of cortical plates and resorption of the roots involved are common features [12]. Unicystic radiographic appearance of solid ameloblastomas is not as common as the multilocular ones [11]. In our case, the radiographic presentation was mixed, mostly unicystic with a few areas showing multilocularity.

When it comes to the histopathological aspect of conventional ameloblastomas, they can paint a variety of pictures. They can be of the follicular and plexiform types, which are considered to be relatively common, when compared to the basal cell pattern, the least common type [13]. Other histopathological variants are acanthomatous, granular cell, and desmoplastic [1]. In our case, the histopathological picture was of the follicular type with areas of microcyst formation within the epithelial islands and large macrocystic areas. Reichart., *et al*. [13] have reported that follicular ameloblastomas recur more often than any other histopathological types, and hence require aggressive management.

Conclusion

Annual follow-up for a period of ten years is generally recommended after surgical treatment of ameloblastomas and in our case, we intend to do the same. The patient has shown no signs of recurrence until now, four years after the surgical treatment. This is probably due to the successful surgical resection of the tumour with wide margins and simultaneous reconstruction with grafts to restore function and aesthetics. Ameloblastoma must be considered in the differential diagnosis of any mandibular swelling, to be confirmed or ruled out, with further investigations. We have, herewith, reported a case of conventional ameloblastoma, of the follicular type, that occurred in an unusual site and presented an initial clinical diagnostic dilemma.

Conflict of Interests

The authors declare no conflict of interests.

Patient Consent

Obtained.

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