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Clinicopathological and imaging variants of Ameloblastoma - Case series

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Abstract
Ameloblastoma is the second most common intraosseous benign odontogenic tumor usually affecting the posterior region of mandible. This tumor exhibits a locally aggressive behavior. It is seen in the third to fifth decades of life. In the recent histological classification of odontogenic tumors from the World Health Organization (WHO), Ameloblastoma is defined as a benign, locally invasive epithelial odontogenic neoplasm of putative enamel organ origin. Radiographically the lesion is variable in appearance and may be unilocular or multilocular, with well-defined cortical borders in the mandible and ill-defined margins in the maxilla. A number of morphological variants of Ameloblastoma have been documented in the literature and at times, may pose a diagnostic challenge to the pathologist. Recognition of the subtypes of Ameloblastoma is important, as it has been documented that some subtypes may exhibit a more aggressive biological behavior than the so-called "conventional" Ameloblastomas.

Keywords: Ameloblastoma, etiopathogenesis, case series, multilocular radiolucency, unicystic, plexiform, desmoplastic Ameloblastoma, tumor markers

1. Introduction
Ameloblastoma (from the English word, "amel," meaning Enamel, and the Greek word, "blastos", meaning Germ) is a rare, benign epithelial odontogenic tumor, representing 1% of all oral ectodermal tumors and 9% of odontogenic tumors. Ameloblastoma was recognized in 1827 by Cusack [1]. This neoplasm was designated as Adamantinoma in 1885 by the French physician Louis-Charles Malassez [2].
The term Ameloblastoma was coined by Ivey and Churchill in 1930, considered as a true neoplasm as it resembles the cells of the enamel-forming organ [1], [2]. This paper reports three patients who were diagnosed with Ameloblastoma of the mandible at our institute.

2. Case Reports
Case One
A forty eight year old female patient reported to our department with a painless, progressive swelling of lower left side of the jaw since two months. Extra orally, swelling was seen in left lower one third of face extending 2cm below lower border of mandible. Intraoral examination revealed a swelling, posterior to 34 obliterating the buccal and lingual vestibule (Figure 1a). Panoramic and occlusal radiographs showed multilocular lesion crossing midline, causing buccal cortical plate expansion (Figure 1b, 1c). Histopathological examination was suggestive of Desmoplastic Ameloblastoma (Figure 1d).

Case Two
A thirty five year old male patient reported with a painless swelling in lower right jaw since four months. Swelling was gradual in onset, slowly progressed to present day size. Extra orally, a solitary diffuse swelling measuring approximately 3.5x2.5cm over right parasymphysis region was evident. Intraorally, an expansile lesion was seen extending from 43 to 47 region obliterating buccal mucosa (Figure 2a, 2b) and panoramic radiograph showed unilocular lesion (Figure 2c). Histopathological features were suggestive of unicytic Ameloblastoma (Figure 2d).

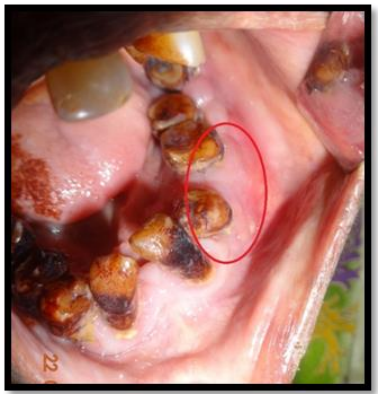

Fig 1 (a): Teeth in the region of swelling were non vital, mobile, missing w.r.t 31, 33, 35, 41, 42 and displacement w.r.t 32, 34, 43.

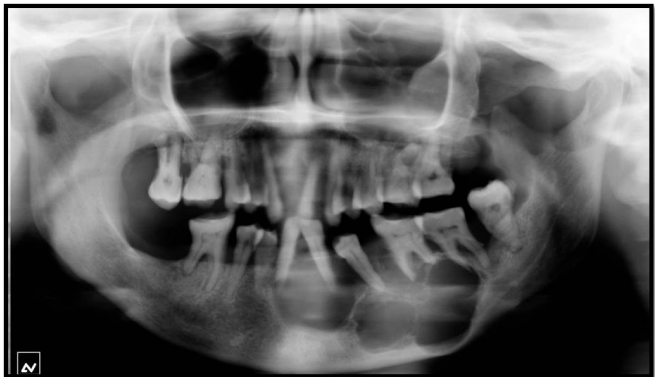

Fig 1 (b): Panoramic image shows a multilocular lesion with thinning of left inferior border of mandible with pathological teeth displacement w.r.t 32, 34, 43, and root resorption w.r.t 32, 34, 36, 37 and 43.

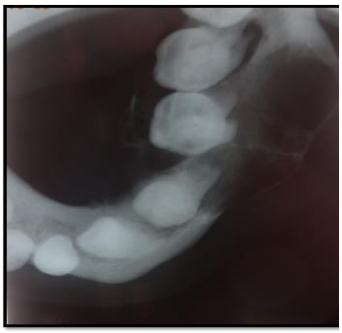

Fig 1 (c): Lateral Occlusal radiograph shows buccal expansion w.r.t 34, 36, 37 surrounded by corticated borders.

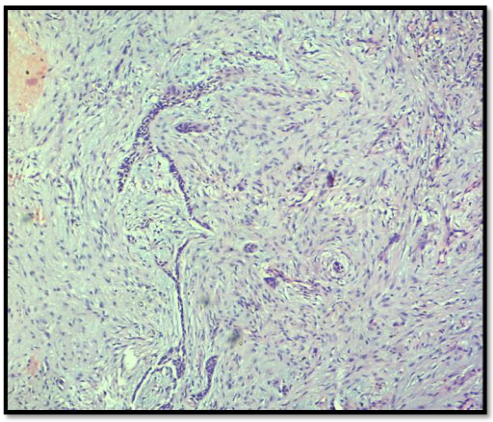

Fig 1 (d): On microscopic examination H & E stain showed dense collagen stroma, with epithelial proliferation compressed by dense collagen stroma suggestive of Desmoplastic Ameloblastoma

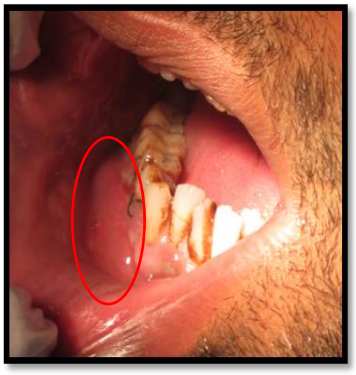

Fig 2 (a): Intraorally buccal expansion is seen extending from 43 to 47.

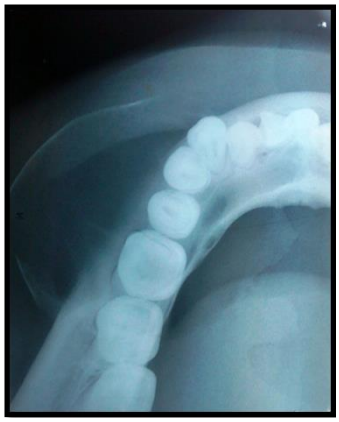

Fig 2 (b): Occlusal radiograph shows buccal cortical plate expansion extending from 43 to47 region surrounded by corticated borders, leaving thin 'egg shell of the bone'.

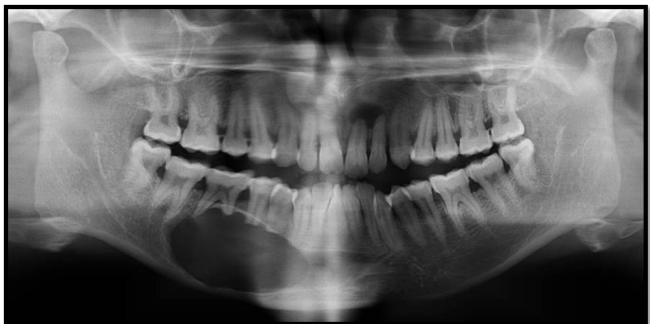

Fig 2 (c): Panoramic radiograph shows well defined unilocular radiolucency extending from 43 region to 46 region surrounded by corticated borders. Root resorption evident w.r.t 43, 44, 45, 46.

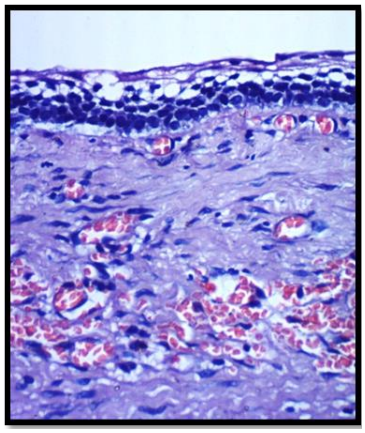

Fig 2 (d): H & E stain shows ameloblastomatous lining epithelium with hyperchromatic nuclei; connective tissue shows dense collagen fibers with chronic inflammatory cells predominantly lymphocytes.

Case Three

A forty seven year old male patient reported with painful swelling in lower right one third of face since five months. Swelling was sudden in onset, associated with dull pain, which aggravated on chewing food. Extra orally, a well-defined swelling was present in lower one third of face and intraorally the swelling extended from 43 to 46 (Figure 3a). Panoramic image showed multilocular lesion (Figure 3b). Histopathological examination confirmed plexiform Ameloblastoma (Figure 3c).

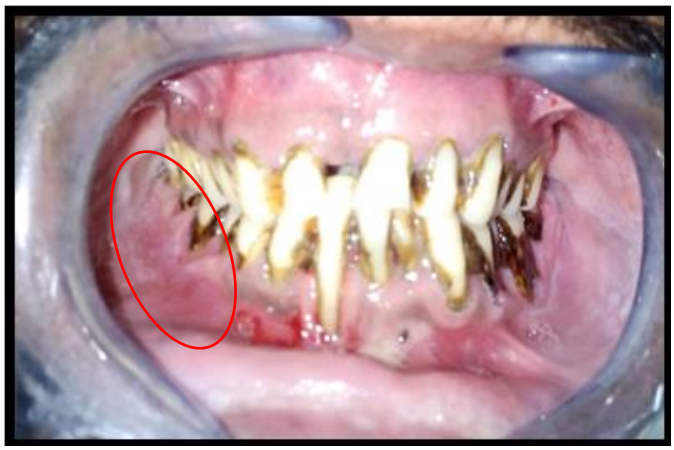

Fig 3 (a): Intraorally swelling is seen extending from 45 to 47, gingival recession w.r.t 31, 32, 33, 41, 42, 43.

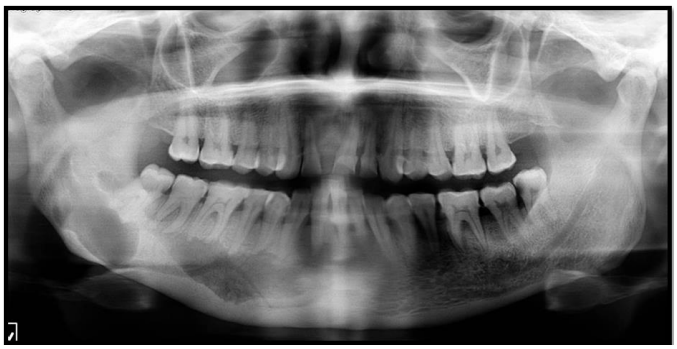

Fig 3 (b): Panoramic image shows multilocular lesion extending from 43 to 48 involving the inferior border of mandible, root resorption is evident w.r.t 43, 44, 45.

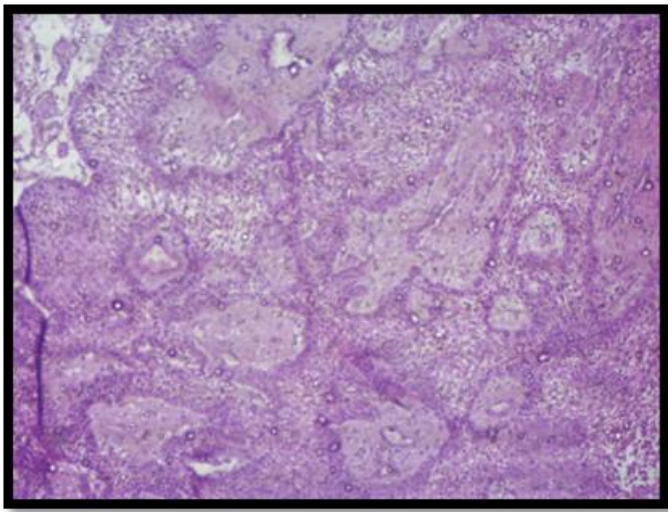

Fig 3 (c): On microscopic examination H & E stain shows strands of odontogenic epithelial cells and connective tissue stroma is seen enclosed between the networks of odontogenic epithelial cells suggestive of Plexiform Ameloblastoma.

3. Discussion

Ameloblastomas are an enigmatic group of oral tumors. Though they are usually benign in growth pattern, they not only frequently invade locally but occasionally metastasize. It may arise from rests of dental lamina, from a developing enamel organ or from basal cells of the oral mucosa [3].

Ameloblastoma is seen commonly in the third to fifth decades of life. In our series, Ameloblastoma was observed to occur in the 3rd to 4th decade of life, with the mean age being 45 years. This finding is not consistent with the studies conducted by Darshani *et al*., Fregnani *et al*., and Adeline *et al*. who found the average age of occurrence at 30years.

Various studies show inconsistent findings regarding gender predilection. The present report showed a slight male predilection, with a male: female ratio of 2:1. This is similar to the findings of Krishnapillai *et al*. and Potdar *et al.* [4]. In this present series, all the lesions were found in body-ramus region of mandible. Our observations are similar to the findings of Gunawardhana *et al*., Ogunsalu *et al*., Adeline *et al*., and Chidzonga *et al*. Ameloblastoma presents commonly as painless swelling (facial deformity). In the present series, 2 out of the 3 total cases were asymptomatic. Tooth displacement, root resorption and displacement or destruction of inferior alveolar canal, were the common findings in Ameloblastoma.

Multilocular radiolucency was the most commonly encountered radiographic presentation in the present series. Histologically, Ameloblastoma can be subdivided in four types: solid, unicystic, peripheral and desmoplastic. In relation to cellular pattern and organization, Ameloblastoma can be classified into five subtypes: follicular, plexiform, acanthomatous, granular Ameloblastomas cells and basal cells Ameloblastomas. These subtypes can occur in isolation or in combination [5].

Ameloblastoma is a tumor that frequently recurs after treatment. The rate of recurrence ranges from 17.7 % for en bloc resection to 34.7% for conservative therapy [4]. The size of the tumor, presence of ruptured basal cortical bone, and histological pattern seems to be an important factor for the management of Ameloblastoma. Treatment ranges from conservative surgery to radical procedures [6].

Latest Diagnostic Aids

Microgenomic procedure are done for identifying genes, these protein products are considered as diagnostic and prognostic markers as well as for potential therapeutic interventions [7].

Recent studies have established that, in benign tumors, a large number of cancer stem cells are present, which have great implications in tumor development [8].

Increasing evidence highlights the role of CD133 as a marker in various human tumors including Ameloblastoma. ABCG2 is a member of the ATP binding cassette transporter family, is widely expressed in stem cells, and is recognized as a universal marker of stem cells. Recent studies strongly suggest that ABCG2 expression in tumors may contribute to tumor growth initiation, invasiveness and relapse [9].

Etiopathogenesis

The cloning and characterization of expression of the Ameloblastin and Amelogenin genes in these tumors suggests that Ameloblastoma arise from the odontogenic apparatus or cells that are potentially capable of forming dental tissue.

The potential sources for this tumor are the cell rests of the enamel organ (cell rests of Malassez and cell rests of Serres), epithelial odontogenic cysts (Dentigerous cysts), basal cells of the surface epithelium of the jaws and heterotrophic

epithelium in other parts of the body. In the pathogenesis of Ameloblastoma different factors have been found to play a role and they potentiate their action through different mechanisms as shown in Table 1 [10].

Mechanism	Factors involved
Signaling Pathway	SHH
	WNT
	Notch
Growth factors	FGF
	BMP
Proteins	
	BMP
	Ameloblastin
	Enamel matrix protein
	Calretinin
	Syndeca-1
	TWIST
	FOS
Proteinases	MMP
Tumour suppressor genes	P53
	P63
	P73

4. Conclusion

Ameloblastoma is a benign odontogenic neoplasm that manifests with a wide range of varied clinical, radiographical and histopathological presentations. Though radiographs are an important aid for the diagnosis of intraosseous lesions, one should never rely on it alone. All such lesions should be biopsied and an accurate histological diagnosis should be obtained before definitive treatment is commenced.

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