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Case Report

A CASE OF AMELOBLASTOMA IN MANDIBLE – CLINICO RADIOLOGIC PERSPECTIVE

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ABSTRACT
Ameloblastoma is a benign, unicentric, non-functional and clinically persistent tumor. Ameloblastoma is the second most common odontogenic tumor in the oral and maxillofacial region. It occurs from second to sixth decades and the site of predilection is posterior mandibular region in 80% of the cases. Six histopathologic variants of ameloblastoma are recognized: follicular, plexiform, acanthomatous, basal cell, granular cell, and desmoplastic. Three clinico-radiologic groups namely solid or multicystic, unicystic and peripheral are recognized. Radiographically ameloblastoma represents as a unilocular or multilocular radiolucency. They are usually benign in growth pattern but frequently invade locally and rarely metastasize. This case report presents a case of plexiform ameloblastoma in the left mandibular posterior region diagnosed using radiographic examination and removed by surgical enucleation/curettage.

Keywords: *Ameloblastoma, Mandible, Plexiform, Unicystic*

INTRODUCTION
The ameloblastoma is true neoplasm of the enamel organ type tissue which does not undergo differentiation to the point of enamel formation (Rajendran and Sundharam, 2006). Regezi and Sciubba reported that ameloblastoma accounts for 11% of all odontogenic tumors in the jaw (Jayachandran and Singh, 2012); (Williams, 1993). Ameloblastoma is the second most common odontogenic tumor in oral and maxillofacial region. Ameloblastoma commonly appear in third through fifth decades and 70% cases develop in the posterior molar-ramus region of mandible, associated with unerupted/impacted mandibular third molar (Tozaki *et al.*, 2000). Six histopathologic variants of ameloblastoma are recognized: follicular, plexiform, acanthomatous, basal cell, granular cell, and desmoplastic (Rajendran and Sundharam, 2006). The plexiform pattern is common along with the follicular pattern. It is less aggressive and has a relatively lower recurrence rate (Çakur *et al.*, 2009). The main modalities of treatment that have been used include radical/conservative surgical enucleation, curettage, chemical/electrocautery, and less commonly radiation therapy or a combination of radiation and surgery (Rajendran and Sundharam, 2006). This case report presents a case of plexiform ameloblastoma in the left mandibular posterior region which was diagnosed using radiographic examination and removed by surgical enucleation/curettage.

CASE
A thirty five year old male reported to the department of Oral Medicine and Radiology with a chief complaint of swelling in the lower left back region of the jaw for the past three month associated with mild pain. History revealed that the swelling was insidious in onset, gradually increased and attained the present size. There was no history of fever, trauma, bleeding or purulent discharge in relation to the swelling. The past medical history, past dental history and family history was not contributory.
On general examination, the patient was conscious, normally built and moderately nourished. Extraorally, there was mild facial asymmetry and swelling in the left mandibular posterior region of the jaw. On palpation the swelling was firm to hard in consistency, fixed to the underlying bone, free from the overlying skin with mild tenderness on palpation. No secondary changes were evident in the extraoral skin over the swelling and no cervical lymphadenopathy was present. Intraorally a solitary swelling was

present in the left mandibular posterior buccal sulcular and gingival region measuring approximately 2.0*1.5cm, extending anteriorly from the gingival margin and sulcus in relation to 36, posteriorly extending upto the distal gingival margin in relation to 37, superiorly from the marginal gingiva and interdental gingiva in relation to 36 and 37, inferiorly it extended till the mandibular buccal sulcular/vestibular region [Fig 1a]. There was displacement of the posterior left mandibular molar (36) .The mandibular molar (36) appear submerged on clinical examination and the premolar (35) appeared impacted. The surface appeared smooth with diffuse borders and mild ulceration was present on the surface of the swelling. On palpation, swelling was non-tender, soft in consistency, partially compressible, not reducible or depressible. There was no bleeding or purulent discharge in relation to the swelling.

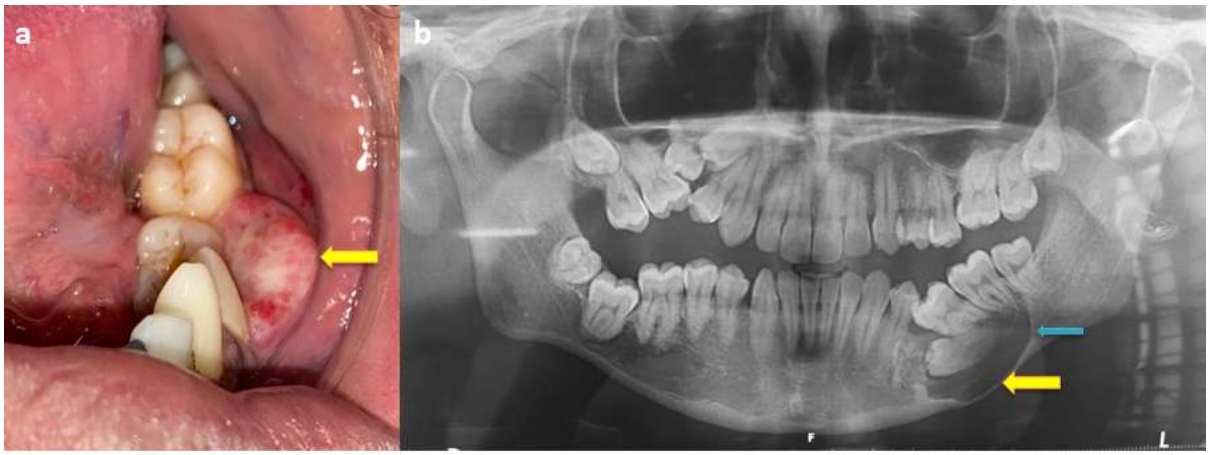

Fig1(a,b)Intraorally a solitary swelling was present in the left mandibular posterior buccal sulcular and gingival region(a) .The orthopantamogram revealed a well defined radiolucent area in the posterior left mandibular region with well delineated radiopaque corticated borders with blue arrow indicative of the radiolucent area and yellow arrow indicative of lower border of the mandible with intact cortical margin approximately 1mm in height (b).

The patient consent was obtained and radiological examination (orthopantamogram) was made. The aspiration was negative. The orthopantamogram (OPG) revealed a well defined radiolucent area in the posterior left mandibular region with well delineated radiopaque corticated borders. The lower border of the mandible had a thin intact cortical margin approximately 1mm in height [Fig 1b]. The inferior alveolar nerve and canal appeared displaced within the radiolucent area. The posterior mandibular premolar (35) was impacted horizontally and was completely enclosed within the lesion. The mandibular molars (36, 37) were displaced and appeared mesially inclined towards the lesion with resorption in the apical portion of the tooth roots. The orthopantamogram also revealed impacted maxillary posterior teeth (15, 18, 28), mandibular posterior (47, 48) and displaced teeth (16, 17, 14). Cone beam computed tomography (CBCT) was made to analyze and interpret the internal morphology, exact anatomical location, extent and density of the lesion. The axial, coronal, sagittal and three dimensional reconstruction of CBCT revealed a well defined hypodense area in the posterior left mandibular region in relation to 35, 36 and 37 with a well delineated corticated border measuring 19.3 mm mesiodistally, 16.7 mm superoinferiorly and 23.8 mm anteroposteriorly. The axial section revealed an expansion of the buccal bone and thinning of an adjacent buccal cortical plate leaving a thin “eggshell” of bone .The trabecular pattern of the mandible surrounding the lesion was altered. The mandibular premolar was horizontally impacted within the hypodense area. There was perforation in the lingual cortical bone [Fig 2]. The coronal and sagittal sections revealed the hypodense area in the posterior mandibular region with a defined corticated border. The buccal and the lingual cortical bone showed expansion. The buccal, lingual and the inferior border showed expansion. The mandibular premolar was impacted (35) and the molar teeth (36 and 37) were displaced. There was resorption of roots of the molar teeth (35, 36 and 37).The

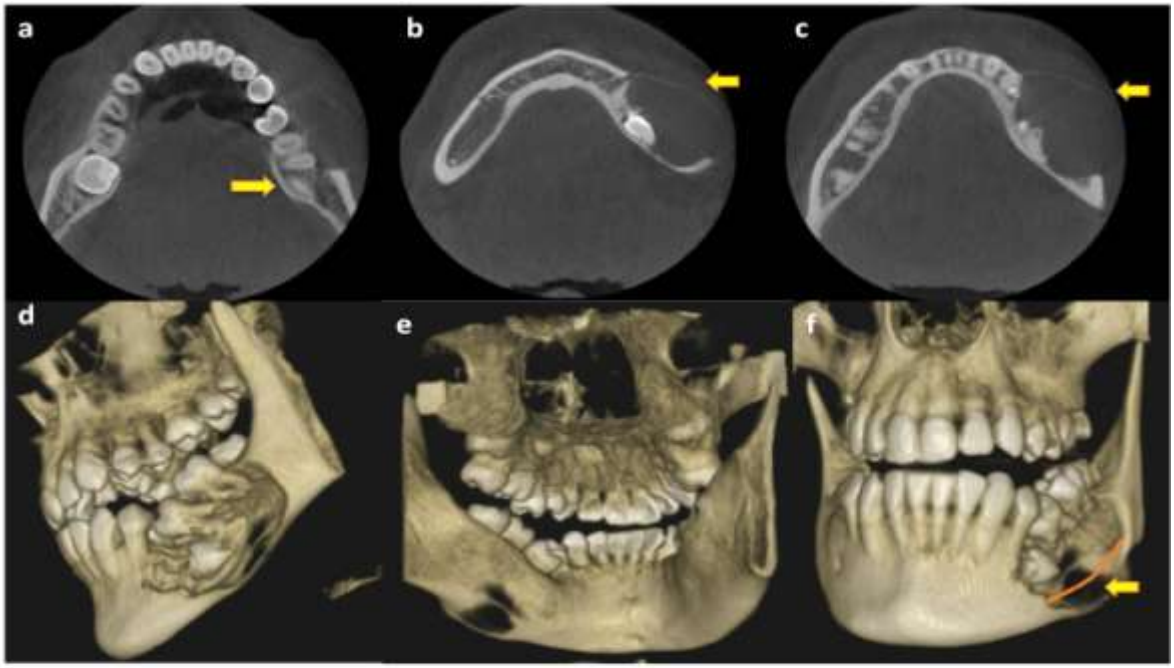

Fig2 (a,b,c,d,e,f) :Axial sectioning and three dimension of CBCT revealed a well defined hypodense area in the posterior left mandibular region in relation to 35, 36 and 37 with a well delineated corticated border. The axial section revealed an expansion of the buccal bone (b) and thinning of an adjacent buccal cortical plate leaving a thin "eggshell" of bone (C).The trabecular pattern of the mandible surrounding the lesion was altered (a).Three dimension with nerve traced model showed that the inferior alveolar canal and inferior alveolar nerve displaced inferiorly towards the lower border of mandible and approximated within the lesion (f)

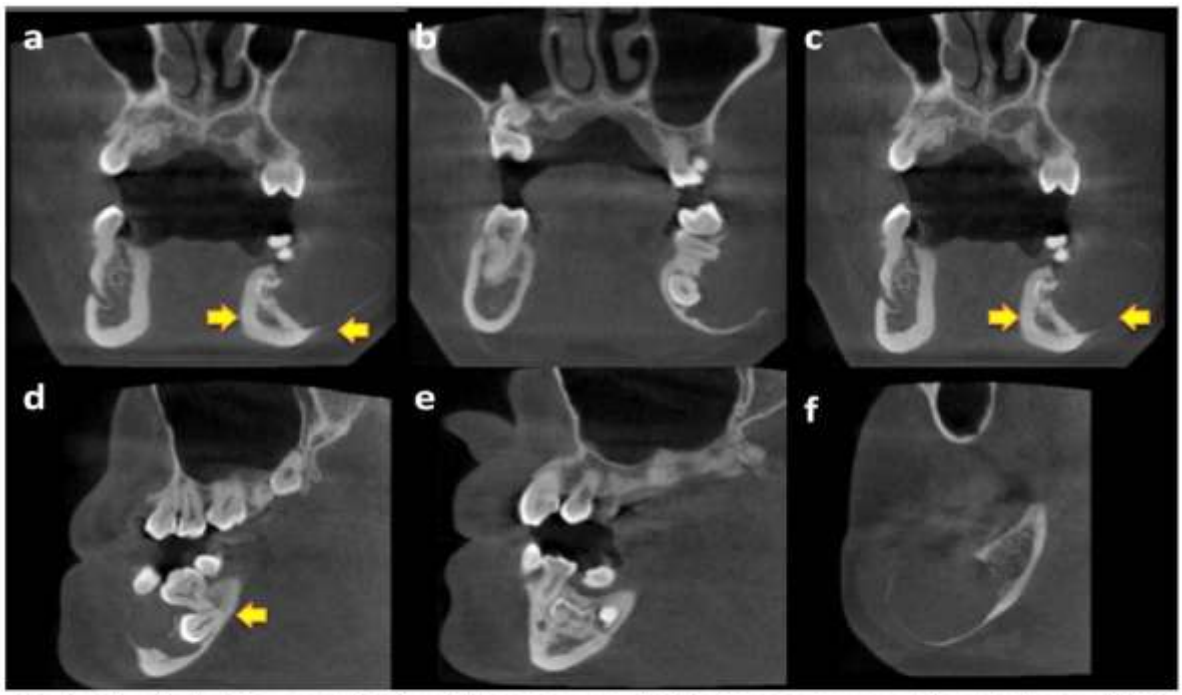

Fig 3 (a,b,c,d,e,f) : The coronal and sagittal sections revealed the hypodense area in the posterior mandibular region. The buccal and the lingual cortical bone showed expansion (a). The buccal, lingual and the inferior border showed expansion (b). The inferior alveolar canal appears normal in right side of the mandible (c)The mandibular premolar was impacted [35] and the molar teeth [36 and 37] There was resorption of roots of the teeth [35, 36 and 37](d.e)The lesion appears as a completely unicystic well defined lesion (f)

three dimension with nerve traced model showed that the inferior alveolar canal and inferior alveolar nerve displaced inferiorly towards the lower border of the mandible and approximated within the lesion [Fig 3]. The hypodense area was unicystic without any intervening septae. Atypical presentation of clinical swelling and unicystic radiologic presentation led to the provisional diagnosis of ameloblastoma. Surgical aspiration was done and proven to be negative. The haematological and biochemical investigations were within normal limits. Complete surgical enucleation/curettage of the lesion was done under local anaesthesia and teeth (35, 36, 37 and 38) were extracted. The mandibular posterior teeth roots were completely contained within the lesion on surgical exposure and hence were extracted. Histopathological examination revealed an odontogenic neoplasm characterized by proliferation of odontogenic epithelium within the stroma. The epithelial cells are arranged as a network of anastomosing strands and cords bound by tall columnar ameloblast like cells and intervening stellate reticulum like cells. Cystic degeneration within the stroma is noted in the same are suggestive of plexiform ameloblastoma [Fig 4].

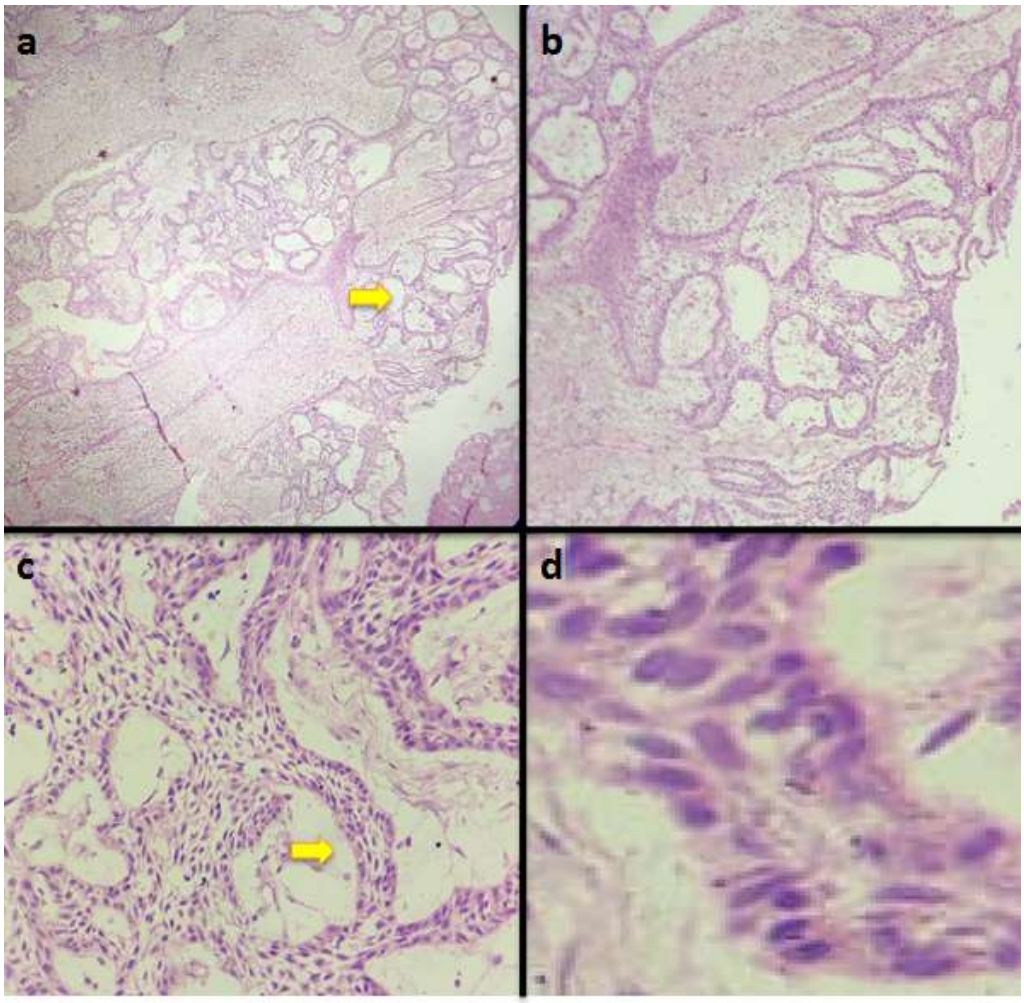

Fig 4(a,b,c,d): Photomicrograph of histopathological section at 10x (a,b) 40x (c,d) proliferation of odontogenic epithelium within the stroma. The epithelial cells are arranged as a network of anastomosing strands and cords bound by tall columnar ameloblast like cells and intervening stellate reticulum like cells. Cystic degeneration within the stroma is noted. Arrow indicative of anastomosing strands and cords.

The overall clinico-radiological and histopathological diagnosis confirmed the final diagnosis of plexiform ameloblastoma. The healing during the post operative period was normal. The patient is asymptomatic and under regular follow up for the past three months with no signs and symptoms of recurrence.

DISCUSSION

Ameloblastoma is unicentric, non-functional, intermittently growing, anatomically benign and clinically persistent. The first term coined was 'adamantinoma' which coined by Malassez and the term 'ameloblastoma' was suggested by Churchill in 1934 (Rajendran and Sundharam, 2006). The first ameloblastoma was reported by Broca in 1868 and the first thorough description of ameloblastoma was given by Falkson (Rajendran and Sundharam, 2006). The ameloblastoma represents about 1% of all oral odontogenic epithelial tumors and 11% of all odontogenic tumor (White and Pharoah, 2009). The predominant prevalence is in the range of 33–39 years and most cases cluster between second to sixth decades. The site of predilection is mandible in 80% of the cases. In the mandible 70% are located in the posterior molar region and the ascending ramus, 20% in the mandibular premolar region, and 10% in the mandibular anterior region (Adebiyi *et al.*,2006); (Varkhede *et al.*,2010).

The tumor is derived from the disturbances in the developing enamel organ and disturbances in epithelial lining of odontogenic cysts (dentigerous cyst and odontomas). They also originate from the cell rests of the enamel organ (remnants of dental lamina or remnants of Hertwig epithelial root sheath) and from epithelial cell rests of. The basal layer of oral epithelium and heterotopic epithelium in other parts of the body (pituitary gland) also gives rise to this tumor (Kovács *et al.*,1999); (Rajendran and Sundharam, 2006). Ameloblastomas are slow growing tumor, gradually increasing in size. The main clinical manifestations are swelling, facial asymmetry, displacement of the teeth, mobility of teeth involved, and bony expansion/erosion. Occasionally pain, paresthesia and secondary changes are evident.

Radiologically, this tumor shows multilocular or unilocular pattern. The multilocular pattern can have soap bubble (larger compartments of variable size), honeycomb (numerous small compartments or loculations) or spider-web appearances (Paramasivam *et al.*,2018). The periphery of the ameloblastoma is usually well defined and frequently delineated by a cortical border in mandible while the borders are ill defined in maxilla. The internal pattern varies from radiolucent to mixed with the presence of bony septa creating few to many internal compartments. The loculations are smaller in the anterior mandible and larger in the posterior mandible (White and Pharoah, 2009). Histologically, ameloblastoma is characterized by proliferation of epithelial cells arranged on a stroma of conjunctive vascular tissue within locally invading regions/structures that resemble the enamel organ at various stages of differentiation (Çakur *et al.,* 2009).

A plexiform pattern of epithelial proliferation which was associated with histologically characteristic ameloblastoma was first given by Vickers and Gorlin as plexiform ameloblastoma (Gardner,1981). In plexiform ameloblastoma the cells are arranged in irregular masses and as a network of interconnecting strands of cells. The masses or strands are bounded by a complete layer of tall columnar cells and stellate reticulum like cells are present between the layers. Double rows of the columnar cells are lined up occasionally. However, the stellate reticulum like tissue/cells is much less prominent in plexiform type. Areas of cystic degeneration of stroma are also common. The strong immunoreactivity of PAKT/PI3K (Phosphatidylinositol 3-kinase) in some variants of ameloblastoma, especially the plexiform histological pattern, suggests that AKT/PI3K pathway may be promoting proliferation of ameloblastic cells (Effiom *et al.*,2018). The p75-NTR (Neutrophin receptor- Tumor necrosis family receptor) immunoexpression was positive in peripheral cells of plexiform ameloblastoma which associates the differed biological behaviour of plexiform ameloblastoma (Ragunathan *et al.*,2016).

Advanced imaging and immunochemical diagnosis has the potential to provide improved diagnosis and superior surgical treatment planning (Singer *et al.*,2009). Surgical resection with intact margins of 1 cm has the least rate of recurrence in ameloblastoma and when treated inadequately, malignant

transformation and metastatic dissemination in ameloblastoma is rare but it does tend to occur. Long – term follow-up is necessary because ameloblastoma has been shown to recur 25 and 30 years following primary treatment (Çakur *et al.*,2009).

CONCLUSION

Plexiform pattern is less aggressive odontogenic tumor, has a relatively lower recurrence rate but it is locally invasive. Hence early diagnosis and optimal treatment planning with regular periodic clinical and radiographic follow up interval is mandatory to minimize the morbidity and to improve the survivorship of life.

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