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AMELOBLASTOMA OF MANDIBLE: A SERIES OF SIX CASES AND REVIEW

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ABSTRACT Ameloblastoma is a benign but locally invasive odontogenic tumor with unlimited growth potential. The most common site of occurrence is mandible. Histological presentation comprises of tall columnar ameloblast like cells with central stellate reticulum showing either cystic degeneration or squamous metaplasia or cells containing granules etc. radiologically they appear as unilocular or multilocular radiolucency sometimes associated with an unerupted tooth. Here we present six cases of ameloblastoma of different histopathological variant with a short review of literature.

KEYWORDS : ameloblastoma, mandible, follicular, plexiform

INTRODUCTION:-

Ameloblastoma was defined by Robinson as an unicentric, non-functional, intermittent in growth, anatomically benign and clinically persistent neoplasm.1 Cusack first described the tumor in 1827. French physician Malassez designated it as adamantinoma in 1885. Finally in 1930 Ivey and Churchill renamed it as ameloblastoma.2 It occurs mainly in mandible but can also occur in maxilla. It has a rare peripheral variant also.3while the odontogenic epithelial origin of ameloblastoma is well established even by the similar cytokeratin profiling of ameloblastoma and developing tooth germ (Brown and Betz, 2015) the exact etiopthogenesis is still nonconclusive. In this case series we describe six ameloblastoma cases of various histological variant all occurring in mandible.

Case no 1:

A female patient aged 19 years came to our department with the complaint of right sided painless facial swelling since last 8-9 months. The bony hard swelling caused expansion of the buccal cortex distal to 46. 47 was extracted from elsewhere. OPG showed an ill defined large multilocular radioucency with impacted 48 involving the ramus and the coronoid process of the right sided mandible. Incisional biopsy was taken from the representative site under LA. Histopathological evaluation revealed the presence of follicles of odontogenic epithelium with peripheral pallisading of tall columnar ameloblast like cells and central stellate reticulum like cells in a bland connective tissue stroma. The overall features were suggestive of follicular ameloblastoma.

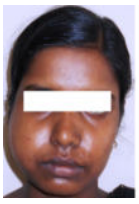
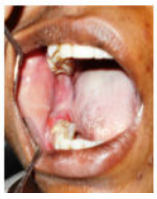
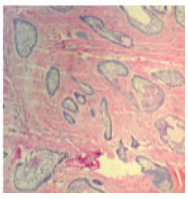

Fig1: Extraoral view
Fig2: Intraoral view
Fig3: Histopathological features

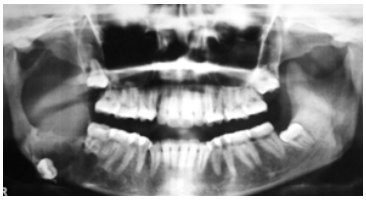

Fig4: Radiological features (OPG)

Case no 2:

A 61 years old female patient came to our department with the chief complaint of large swelling of the right side of the face with pus discharge since last 4 years. On intraoral inspection the swelling appeared to be a smooth surfaced lobulated one, firm to hard on palpation. Aspiration was negative. OPG revealed a multilocular radiolucency extending from 33 to 46 with root resorption of the regional teeth. Incisional biopsy was done under LA. Histopathological report revealed the presence of ameloblast like tumor cells as a network of interconnecting strands which were bounded by a layer of palisaded columnar cells with stellate reticulum like cells in between. The overall features were suggestive of plexiform ameloblastoma.


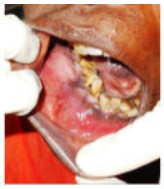
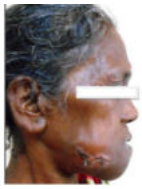

Fig5:Extraoral view
Fig6: Intraoral view
Fig7: Right profile view

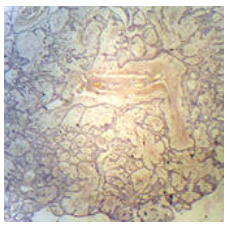
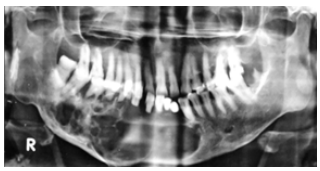

Fig8: Histopthological features
Fig9: Radiological features (OPG)

Case 3:

A female patient aged 40 years reported to our department with the chief complaint of swelling of the lower half of the face since last 3 years. On examination it was found that the buccal cortex in relation to 32 to 43 was expanded with some areas of fluctuation. OPG showed ill defined radiolucency with irregular margin regarding the same region. Histopathological report of the incisional biopsy taken under LA reveals the presence of follicles surrounded by palisading odontogenic cells with stellate reticulum like cells found central to them. The features suggestive of follicular ameloblastoma.


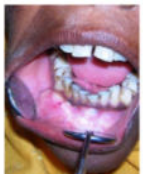
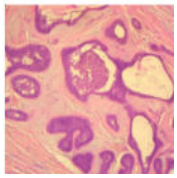

Fig10: Extraoral view
Fig 11: Intraoral view
Fig12: Histopathological features

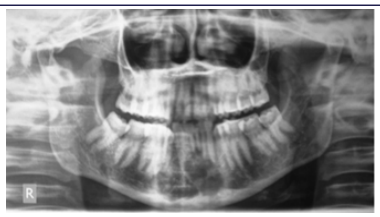

Fig13: Radiological features (OPG)

Case 4:

A 18 years old female patient reported to our department with the complaint of swelling of left lower jaw since last few years. On examination the bony hard mass showed buccal cortical plate expansion with egg shell crackling. Also an unhealed ulcer was noted distal to 36. OPG showed a multilocular radiolucency extending from 35 to the ascending ramus with dispkacement of 38 and root resorption of 35, 36, 37. Histopatholgy of the tissue taken by incisional biopsy under LA revealed the presence of nests of uniform basaliod cells with the peripheral columnar cells with reverse polarity. No stellate reticulum was seen in the central portion of the follicles. The lesional tissue was covered by fibrous capsule with fibrous septa intervening into it giving it a lobular pattern. The overall features were suggestive of basal cell ameloblastoma.

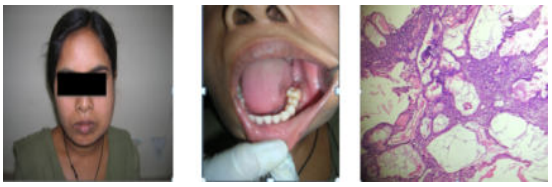

Fig14: Extraoral view
Fig15: Intraoral view
Fig16: Histological features

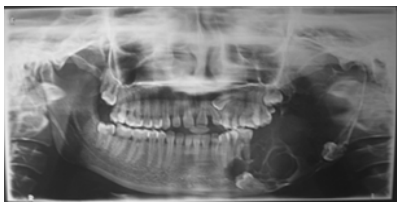

Fig17: Radiological features (OPG)

Case 5:

A 53 years old male patient came to our department with the chieft complaint of painless swelling in the front lower face since last few years. On inspection an exophytic mass was present in the lower alveolar ridge with expansion of buccal cortical plate. The lesion was firm to hard on palpation. OPG shows a unilocular radiolucency involving 43, 44 and 45 region. Histophathological section taken from incisional biopsy and stained with H & E revealed the presence of multiple follicles lined by tall columnar ameloblasts like cell with squamous metaplasia of central stellate retuiculum like cells along with keratin formation. The features were suggestive of acanthomatous amelobalstoma.

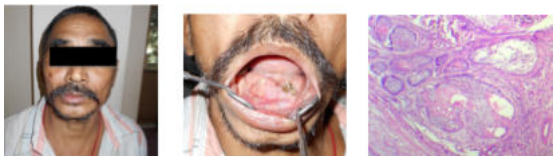

Fig18: Extraoral view
Fig19: Intraoral view
Fig20: Histological features

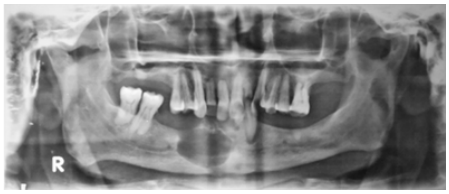

Fig21: Radiological features (OPG)

Case 6:

A female patient aged 12 years came to our department with a huge left sided facial swelling present since 6-7 years. Intraoral examination yielded the presence of a painless firm swelling with expansion of both buccal and lingual cortices distal to 34 upto the ramus of mandible. Contract CT scan of the left mandible revealed a lytic lesion with erosion of the bony outline and extension of soft tissue outside. Tissue section taken from incisional biopsy and stained with H & E revealed the presence the network of strands containing tall columnar ameloblasts like cells with stellate reticulum in between in the background of loose fibrous connective tissue. The features were suggestive of Plexiform Ameloblastoma.

Fig22: Extraoralo view
Fig23: Intraoral view
Fig24: Histological features

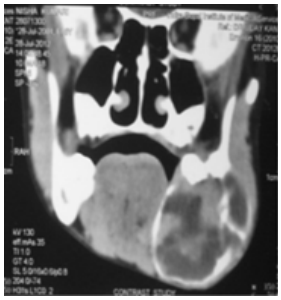

Fig24: Radiological fetaturs (CT Scan)

DISCUSSION:-

Ameloblastomas is a benign locally aggressive tumor of odontogenic origin. The word ameloblastoma derives from the early English word "amel," meaning enamel and the Greek word "blastos," meaning germ. It is thought to be originated from embryonic odontogenic rests (rests of Malassez and rests of Serres), the epithelial lining of odontogenic cysts (especially the dentigerous cyst) and basal cells of the oral mucosa.[4]

Ameloblastomas show a variable geographic prevalence with a global incidence of 0.92 cases per million person-years.[2] Among all odontogenic tumors approximately 11 to 18% are ameloblastomas, being the second most common just next to odontomas.[5] However the largest Indian study conducted in Maharashrta showed odontogenic keratocyst to be the most common odontogenic tumor followed by ameloblatomas.[2]

According to age ameloblastomas are mostly seen in third to sixth decade. In our case series three cases are seen in the mentioned age group and three cases are in the second decade of life with the mean age of 33.8 years. This finding presents similarity to mean age of 32.75 and 32.3 years in studies carried out by santanu patsa and Ritesh Jadav, Sriram and Shetty, respectively.[6,7] Only 10–15% of ameloblastoma cases occur in the pediatric population, but this can be as high as 25% in Africa and Asia (Bansal et al, 2015). However we have found only one such case in our series of six patients making it 16.6% of all cases.

Considering gender, some studies have described that there is no predilection,(Intapa 2017; LedesmaMontes et al. 2007) while others report predominance in men.(Reichart et al. 1995; Siar et al. 2012; 2007Hong et al). However a more recent study has found that Recurrent ameloblastoma are marginally female predominant with male to female ratio of 1:1.3.[8] interestingly in our case series five out of six patients (83.3%) were female.

Approximately, 80% of the tumors are found in the mandible. The molar/ramus area is the most preferred site in mandible. In blacks, ameloblastomas occur more frequently in the anterior region of the jaws.[9] All our presented cases happen to occur in mandible, three in the ramus- angle region, three in the body of the mandible with two of them crossing the midline.

According to Hamed M & Maryam B et al, around 15-20% of ameloblastomas are associated with impacted tooth specially in the posterior mandibular region around the third molar.[10] Another study by A T Vigneswaran and S Shilpa also found the similar incidence (15.7%) of ameloblastoma around impacted third molar.[11] In our case series 33.3% cases are associated with impacted molars. However in case 4 the 18 years old female patient may have 38 impaction due to age but also in this case 37 is impacted and both 38 and 37 were pushed to the lower border of mandible.

The most common symptom of ameloblastoma is a painless swelling as seen in our all six cases. Other than that loosening of teeth, malocclusion and occasional pain etc can take place. Facial deformity, if present, may range from very mild to severe in delayed presentations.[4] Due to progressive bone destruction eventually the tumor breaks through the cortical bone and extends in to adjacent soft tissues.[12]All our cases showed painless swelling with some of them showing facial deformity. All cases showed buccal cortical plate expansion whereas case 6 showed both buccal and lingual cortices expansion. Interestingly, in caso no 5 the tumor has extended to the soft tissue creating a soft tissue mass on the mandible.

Radiologically Ameloblastoma is usually detected by Orthopantomogram. However CT scan could be a better tool to determine the cortical destruction and soft tissue extension, if any. In this case series we have access to CTscan report only of one patient and other had OPG. Radiographically, ameloblastomas are observed as unilocular or multilocular radiolucent lesions with well-defined borders, often with buccal or lingual cortical plate expansion, root resorption and displacement of the teeth.(Milman et al. 2016). In this case series all lesions were multilocular except one which was unilocular lesion in the body of mandible. Root resorption and displacement of teeth are seen in case 3 & 4 whereas case 6 showed bony erosion and extension to soft tissue.

The classification of Ameloblastomas have been modified several times since the first edition of WHO classificstion in 1971. According to 2017 WHO clasification of benign epithelial odontogenic tumor, ameloblastomas are of the following four types: ameloblastoma, unicystic ameloblastoma, extraosseous/peripheral ameloblastoma and metastasizing ameloblastoma. The adjective 'solid/multicystic' for conventional ameloblastoma was removed since conventional ameloblastoams normally show cystic changes. On the other hand, Desmoplastic ameloblastoma, which had been sub-categorized under ameloblastoma in the previous 2005 WHO classification, was also removed, as it was considered merely a histological variant of conventional (meaning solid) ameloblastoma, though it bears some unique clinical and radiographic features. Hence the the histological variant which comes under conventional ameloblastoma are follicular plexiform, acanthomatous, basal cell ameloblastoma, granular cell ameloblastoma and desmoplastic. [2] The latest 2022 WHO classification remains same other than a new entity which has been introduced by the name of Adenoid ameloblastoma in addition to the previously mentioned four categories of ameloblastoma.[13] Out of these variants about 85% of the amelobalstomas are the solid/multicystic/conventional type making it the most common variant.[5] Among them one third is follicular, one third is plexiform and the other variants comprise the rest one third.[5] This finding completely comply with our case series. Here 2 cases are of follicular variety(33.33%) two cases are of plexiform variety(33.33%) and the rest two are acanthomatous and basal cell type.

Conventional ameloblastoma (solid/multicystic type) is treated by complete en bloc resection (radical surgery) with an 1-1.5 cm margin of safety.[14] Recurrence rate of conventional/ solid ameloblastomas ranges from 0.8 to 38% and higher recurrence could be attributed to conservative approach of treatment.[15] After initial diagnosis all our cases were sent to department of oral and maxiallofacial surgery for further treatment where en bloc resection of mandible were done followed by reconstructive surgery where needed. Till date no case of recurrence is known to the best of our knowledge.

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