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CASE REPORT

PLEXIFORM AMELOBLASTOMA- A CASE REPORT

Jaskaran Preet K. Gill, Savreen Kaur, Manroop Kaur, Kirtan Kaur

B.D.S. Intern, Sri Guru Ram Das Institute of Dental Science & Research, Sri Amritsar

ABSTRACT:
Odontogenic tumors are lesions that are derived from the tooth-producing tissues or their remnants that remain entrapped either within the jawbones or into the adjacent soft tissues. From a biological point of view, some of these lesions represent hamartomas with varying degrees of differentiation, while the rest are benign or malignant neoplasms with variable aggressiveness and potential to develop metastasis. Ameloblastoma a benign epithelial odontogenic tumor, is locally aggressive in nature and comprises about 1% of tumors arising in the jaws. It appears most commonly in the third to fifth decades and with equal frequency between sexes. Ameloblastoma prevalently occurs in the mandibular molar and the ramus areas. Recurrence frequently appears after inadequate treatment. Varying histological patterns of it are found, in which plexiform variant is a common type. Here we present a case of a 35 year old female diagnosed with ameloblastoma of a plexiform pattern.

Key words: odontogenic tumor, ameloblastoma, plexiform

Corresponding author: Jaskaran Preet K. Gill, B.D.S. Intern, Sri Guru Ram Das Institute of Dental Science & Research, Sri Amritsar, Punjab, India, E mail: gilljaskaran93@gmail.com

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INTRODUCTION

Odontogenic tumors (OTs) are lesions that are derived from the tooth-producing tissues or their remnants that remain entrapped either within the jawbones or into the adjacent soft tissues. From a biological point of view, some of these lesions represent hamartomas with varying degrees of differentiation, while the rest are benign or malignant neoplasms with variable aggressiveness and potential to develop metastasis.[1]

At present, it is known that the potential sources in developing of an odontogenic tumor are varied, and these include:

1. The pre-functional dental lamina (odontogenic epithelium with ability to produce a tooth), which is more abundant for obvious reasons distally to the lower third molars.
2. The post functional dental lamina, a concept that covers those epithelial remnants such as Serre´s epithelial rests, located within the fibrous gingival tissue; the epithelial cell rests of Malassez in the periodontal ligament and the reduced enamel organ epithelium, which covers the enamel surface until tooth eruption.
3. The basal cell layer of the gingival epithelium, which originally gives rise to the dental lamina.
4. The dental papilla, origin of the dental pulp, which has the potential to be induced to produce odontoblasts and synthesize dentin and/or dentinoid material.
5. The dental follicle.
6. The periodontal ligament, which has the potential to induce the production of fibrous and cemento osseous mineralized material.[1]

Odontogenic tumors are classified into three categories which are as follows: 1. Odontogenic epithelium with mature fibrous stroma, odontogenic ectomesenchyme not present 2. Odontogenic epithelium with odontogenic ectomesenchyme with or without dental hard tissue formation. 3.Odontogenic ectomesenchyme with or without including odontogenic epithelium.[2]

Ameloblastoma falls in the category of odontogenic epithelium with mature fibrous stroma, without odontogenic ectomesenchyme. Ameloblastomas are usually benign, locally aggressive neoplasms derived from the epithelial odontogenic tissues, which are part of the tooth-forming apparatus.[3] Ameloblastomas are divided into four categories based on radiological appearance, histological features, and anatomical location: unicystic, multicystic or solid, desmoplastic, and peripheral.[4]

Several predisposing factors have been proposed, including (1) nonspecific irritating factors such as extraction, caries, trauma, infection, inflammation, or tooth eruption; (2) nutritional deficit disorders; and (3) viral pathogenesis.[5,6]

Although ameloblastomas are histologically benign, they have a high rate of local recurrence, which ranges from 15 to 75% depending on the type of surgical treatment (conservative versus radical) and, exceptionally, may even metastasize despite their benign appearance. The biologic behavior cannot be predicted on the basis of morphology, although metastases usually follow multiple recurrences.[3]

Plexiform ameloblastoma, which is a histologic variant of solid multicystic ameloblastoma is a located centrally or intraosseously in both jaws. It may occur at any age, even though nearly half of the tumors do occur between the ages of 30- 50 years. Plexiform ameloblatoma has no gender predilection and no racial predilection. 80% of these ameloblastomas occur in the mandible –almost exclusively in the molar ramus region. Remaining 20 % occur in maxilla with maxillary tuberosity being the most common site. In the maxilla it can extend into the maxillary sinus and floor of the nose. These ameloblastomas are often associated with presence of unerupted teeth. Symptoms include painless swelling, facial deformity, teeth in the area may become loose, spontaneous fracture, and the tumor may cause the surrounding bone to thin up so that crepitation or eggshell crackling may be elicited, with perforation of the bone being a late feature.[2]

Radiographically, typical picture is a multilocular destruction of bone but unilocular appearances also occur. In the multilocular type the bone is replaced by a number of small, well defined radiolucent areas giving the whole lesion a honeycomb or soap bubble appearance of varying size. Resorption of the adjacent tooth roots is not uncommon. Histopathology it will show odontogenic epithelium arranged in a network which is bound by a layer of cuboidal or columnar cells and include cells resembling stellate reticulum.[2,5]

Here we present a case of a 35 year old female diagnosed with ameloblastoma of a plexiform pattern.

CASE REPORT

A 35 year old female reported to the department of oral diagnosis with the chief complaint of pain and swelling in left lower region of jaw since 7 months. The swelling was initially small in size and gradually increased to the present size of 3 cm x 2 cm. The patient also gave history of intermittent pain in the left lower alveolus region which aggravated on intake of meals. Pain was radiating in nature to left temple region and neck.

Extraoral examination revealed a solitary swelling, oval in shape, firm/ bony hard in consistency with diffuse margins and overlying skin being of normal colour in the left lower jaw. On palpation temperature of overlying skin was slightly elevated. Intraoral examination showed an edentulous third quadrant with obliteration of the buccal and lingual vestibule.

On radiographic examination (OPG), the lesion presented as a well defined multilocular radiolucency involving the left mandibular body region extending to mandibular ascending rami region, involving the superior-inferior border of angle of mandible, pushing the third molar upwards into the ramus of the mandible. (Figure 1) The borders of the radiolucency were well corticated, and fine bony trabaculae was seen throughout the radiolucency. Root resorption of the third molar in respect to the mesial root was also noticed. This picture resembled a typical soap bubble appearance.

Computerized tomography (CT) (Figure 2) revealed an evidence of expansile, multiloculated lesion involving the ramus and part of the body of the mandible on left side. It caused thinning of the cortex with breach at few places along the inner margins. High density fluid was seen within the locules with few air foci and air fluid levels, measuring 3.8 x 3.1 x 2.5 cm in size, with surrounding soft tissue appearing normal.

Haematological findings were not relevant. Keeping the clinical and radiographic picture in mind a differential diagnosis of ameloblastoma, aneurysmal bone cyst, multilocular cyst and cherubism was considered.

In Gross specimen multiple soft tissue bits of varying sizes were received which were creamish brownish in colour and firm in consistency. Histopathological examination showed an irregular network of anastomosing strands of tall columnar ameloblast like cells and stellate reticular like cells between the strands, the intervening connective tissue stroma was delicate too dense with fibroblast, blood vessel, extravasted RBCs, chronic inflammatory cells and areas of cystic degeneration (Figure 3). In few areas small discrete islands/ follicles of tumor were also seen in the connective tissue stroma. The above features were suggestive of plexiform ameloblastoma.

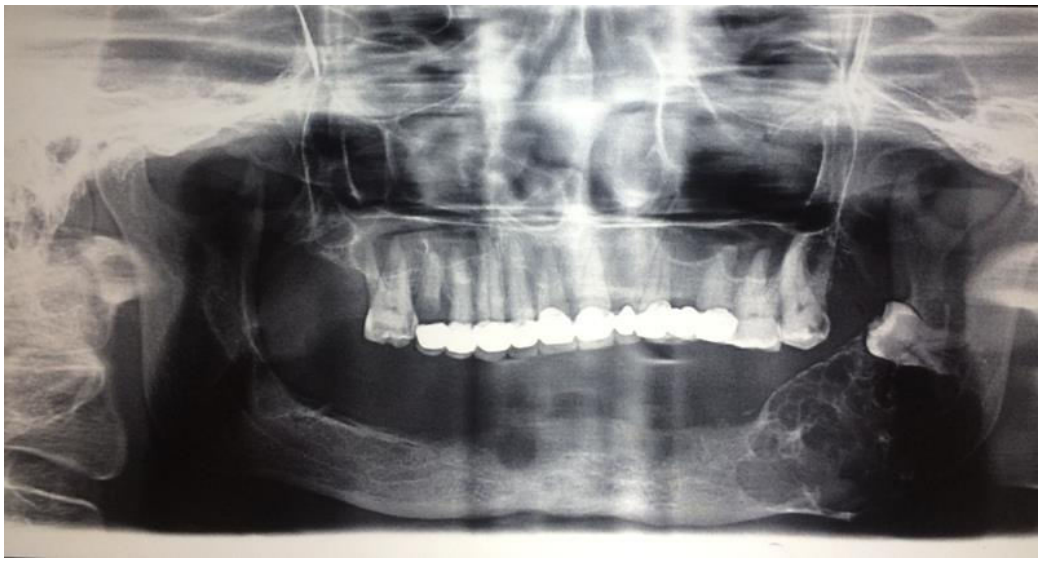

Figure 1: OPG showing a large multilocular radiolucency involving the left mandibular body region extending to mandibular ascending rami region, involving the superior-inferior border of angle of mandible, pushing the third molar upwards into the ramus of the mandible.

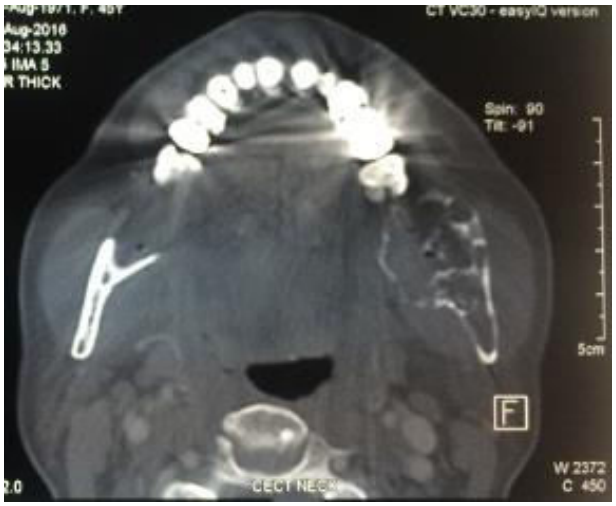

Figure 2: CT (coronal section) revealing expansion and thinning of buccal and lingual cortical plate with a breach in the latter.

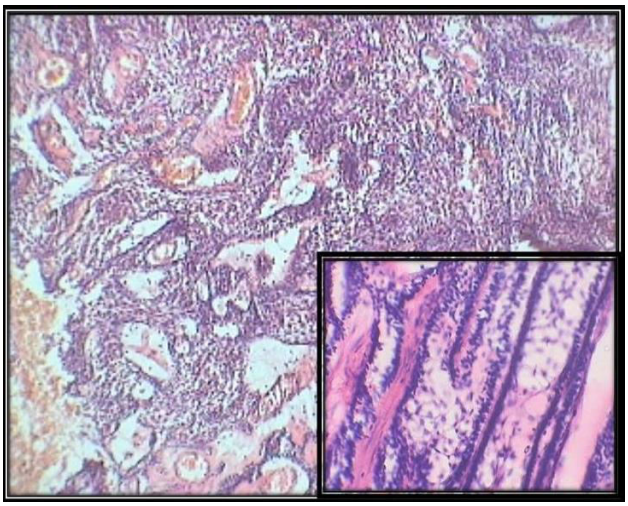

Figure 3: Anastamosing strands of odontogenic epithelium showing cuboidal to columnar cells in the periphery and central stellate reticulum like cells (H & E, X 10). Inset showing the same in H & E, X 40.

DISCUSSION

Odontogenic tumor represents the result of interruptions in or reactivation of tissues involved in the normal sequences of odontogenesis. The nature of the neoplasms is determined by the stage of development at which arrest occurs. The process of histodifferentiation and morphodifferentiation in odontogenesis require an elaborate interplay between epithelial and mesenchymal tissues viz;- ameloblastic epithelium followed by differentiation of fibroblast of dental papilla into odontoblast followed by dentin matrix differentiation into functionl ameloblasts and lastly enamel matrix, calcified enamel and dentin formed tooth.[7]

In ameloblastoma, a marked resemblance to phases of tooth development can occur and many studies have confirmed the similarities between tumor cells and dental lamina and enamel organ cells. Significant number of tumors arise in connection with unerupted teeth and in such cases point of origin is hardly in any

doubt, since tumor epithelium lining is found lining the dental follicle.[7]

Ameloblastoma is a benign epithelial odontogenic tumor but is often aggressive and destructive, with the capacity to attain great size, erode bone and invade adjacent structures.[8,9] Several features suggest that the ameloblastoma that arises allegedly in an antecedent odontogenic cyst exhibits a biological behavior different from that of the solid and multicystic forms of this neoplasm.[10] Hence a different entity called Unicystic ameloblastoma has also been accepted by WHO.

The location of the ameloblastoma in the jaw is important. First, the further the tumor is from vital structures, the less likely it is to infiltrate them. It follows that ameloblastomas of the body of the mandible and of the anterior maxilla are less dangerous than those of the ascending ramus and especially those of the posterior maxilla. This last region is the most dangerous, partly because of its close proximity to the vital structures of the orbit, pterygomaxillary fossa, and cranium. Ameloblastomas can kill by intracranial extension.[11] In the present case ameloblastoma was found involving the left mandibular region, without engaging any vital structure.

Ameloblastomas appear most commonly in the third to the fifth decades; however, the lesions can be found in any age group including children. When small in size, they are usually asymptomatic and are found accidentally on routine dental radiographs; however, larger lesions are associated with jaw expansion. Radiographically, ameloblastomas can either be uni or multilocular with well- circumscribed margins. Its slow but relentless growth may cause movement of tooth roots' or root resorption.[2] In the present case root resorption in relation to the mesial root of third molar was evident.

In multicystic or solid ameloblastomas, varying histological patterns can be seen, of which follicular and plexiform are the two most common ones. Some differences in the patterns of tumor involvement were recognized between follicular and plexiform patterns; infiltration of tumor cells around the nerve was primarily seen with the follicular pattern but not with the plexiform pattern. Follicular ameloblastoma has been postulated to have a more invasive tendency because of its multilocular or soap bubble configuration.[12] Such tumors may infiltrate the cancellous marrow spaces and proliferate around the mandibular canal, later destroying the bony canal, infiltrating the perineural tissue. On the other hand, plexiform tumors, push and deforming the mandibular canal, tend to grow expansively. Therefore, even in the multicystic or solid types, a more conservative approach and proper follow-up seems acceptable if the histologic pattern is plexiform and the canal wall is not destroyed.[13]

CT is usually helpful in determining the contours of the lesion, its contents and its extension into soft tissues for diagnosis.[14]Ameloblastoma typically shows expansive growth with an osseous shell. CT findings include cystic areas of low attenuation with iso attenuating solid regions and Contrast enhanced CT shows an enhancement effect in the solid components.[9]

Electronmicroscopically, the peripheral cells of the solid, follicular ameloblastoma were ultrastructurally similar to the inner enamel epithelium. Kim et al[15] found that, in addition to the strong resemblance of the columnar cells of the tumor to the cells of the inner enamel epithelium at an early stage of differentiation, the stellate cells of the tumor epithelium were similar in many respects to the stellate reticulum of the normal enamel organ. In areas of metaplastic squamous cell changes, the authors found that these cells had ultrastructural features similar to those observed in basal cells and lower prickle cells of the oral mucosa, especially in the epithelium of the palatal mucosa. In a transmission electro n microscopy (TEM) study of 12 plexiform and 9 follicular ameloblastomas, it was also found that the follicular variant consisted of two cell types, one resembling the stellate reticulum and the other resembling the inner enamel epithelium of the normal enamel organ.[2]

Plexiform variant did not show two cell type but resembled squamous epithelium. It should be mentioned that according to the clinical data given for plexiform variant, at least 10 tumors may qualify for the tumor currently known as the plexiform, unicystic ameloblastoma. Ultrastructure of a simple ameloblastoma, the occurrence of cells possessing single cilia which arose from a basal body and occasional cells containing langerhans granules has also been seen.[2] In addition the tumor stroma contains oxytalin fibers. Mucin producing cells were also reported to occur in a multicystic ameloblastoma. Occurrence of so called hyaline bodies, ultrastructurally similar to those found in odontogenic cyst epithelium and cyst walls have been

demonstrated in case of plexiform ameloblastoma by Takeda et al.[16]

Ameloblastomas are treated by curettage only, enucleation and curettage, or radical surgery. Ameloblastomas of the maxilla should be treated as radically as possible. Supraperiosteal resection of the bone is necessary when extensive thinning or perforation of the cortical plates is noted. Chemotherapy and radiation seem to be contraindicated. Postoperative follow-up is important in the management of ameloblastoma because more than 50% of all recurrences occur within 5 years of surgery.[2,5]

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