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EXTENSIVE UNICYSTIC AMELOBLASTOMA: A CASE REPORT

Dr. Sonali Shrivastava	BDS, Post Graduate Student, Dept. Oral Medicine & Radiology, Hitkarini Dental College And Hospital, Jabalpur (M.P)- Corresponding Author
Dr. Sreedevi	BDS, MDS, Professor and HOD, Dept. Oral Medicine and Radiology, Hitkarini dental college and hospital, Jabalpur (M.P)
Dr. Anil Kumar Nagarajappa	BDS, MDS, Lecturer, Department of Maxillofacial Radiology, College of dentistry, Faissalyia, Sakaka, Aljouf University, Kingdom of Saudi Arabia, 42421
Dr. Nalin Saxena	BDS, Post Graduate Student, Dept. Oral Medicine & Radiology, Hitkarini Dental College And Hospital, Jabalpur (M.P)

ABSTRACT Ameloblastoma is a benign but locally aggressive epithelial odontogenic neoplasm which represents 1% of all tumours of the jaw bone. Mostly it is localized in the mandibular molar and ascending ramus area and associated with an unerupted tooth. It occurs over a wide range of ages having equal frequency in men and women. Usually it is discovered during a routine radiographic examination or as a clinically observed mass. It has a characteristic radiographic appearance. It can be treated by enucleation, bone curettage or wide resection.

SUMMARY: Ameloblastoma is the most commonly occurring odontogenic tumour in the mandibular body ramus region with differential diagnosis such as odontogenic keratocyst, odontogenic myxoma, central giant cell granuloma which is ruled out with advanced radiodiagnostic tools. The treatment of ameloblastoma is aggressive enbloc resection with simultaneous reconstruction.

KEYWORDS : Odontogenic tumor, unicystic ameloblastoma, mandible, radical resection.

INTRODUCTION

Ameloblastoma is a benign tumor which is rare, slow-growing but locally invasive neoplasm of odontogenic origin involving.[1] In 80% of cases mandible is the commonest site of involvement and in 20% of cases maxilla is the site of involvement. Third molar region of mandible is the commonest area of involvement.

The term ameloblastoma is derived from the French word "amel," which means enamel, and the Greek word "blastos," which means germ or bud. Other terminologies used are "cystosarcoma," "adamantine epithelioma," "adamantinoma," and finally "ameloblastoma".[1]

WHO defines it as a locally-invasive polymorphic neoplasia that often has a follicular or plexiform pattern in a fibrous stroma. Its behaviour has been described as being benign but locally aggressive.[2] Ameloblastoma is a true neoplasm of odontogenic epithelium and represents about 1% of all the oral ectodermal tumors and 9% of odontogenic tumors.[3]

It is usually diagnosed between the fourth and fifth decades of life, except in the case of the unicystic ameloblastoma, which is diagnosed between the ages of 20 and 30 years and has no gender predilection. 10-15% of the ameloblastomas are seen to be associated with an unerupted tooth. Usually asymptomatic except for the occurrence of swelling in most of the cases.[2]

CASE REPORT

A 24 years old female patient reported to the department of oral medicine and radiology of Hitkarini Dental College and Hospital with a chief complaint of swelling (Figure: 1) on left side of lower half of face since 1 year. Patient reported a small growth in the same region following the extraction of a tooth 3 years ago which was peanut size and progressed to present size in 1 year and was associated with parasthesia of lower lip and mild tenderness on mastication. There is no relevant medical history. All the vital signs were within physiologic limit.

Extra oral examination revealed facial asymmetry on left side of the face due to a solitary diffuse swelling of middle and lower 1/3rd of face measuring approx 5.5 × 6.5 cm in greatest dimension, dome shaped without any surface changes of overlying and surrounding skin (Fig: 1 and 2). Swelling was non tender, afebrile on palpation and was firm in consistency with no fixity to underlying structures with a pinchable overlying skin. A Solitary, mobile, non tender, bilateral submandibular lymph nodes were palpable with a soft consistency.

Figure 1: Extraoral photograph of patient, revealing a swelling over left side of the face

Intra oral examination revealed a solitary ill defined growth (Figure: 2) on left mandibular alveolar region extending from 33 to 38 measuring approx 4.4 × 5.5 cm, irregular in shape, smooth surface with normal color of overlying and surrounding mucosa and expansion of buccal and lingual cortical plates. The growth was non tender, fixed to underlying alveolar ridge with inferiorly displaced 35, firm in consistency, non fluctuant and non pulsatile.

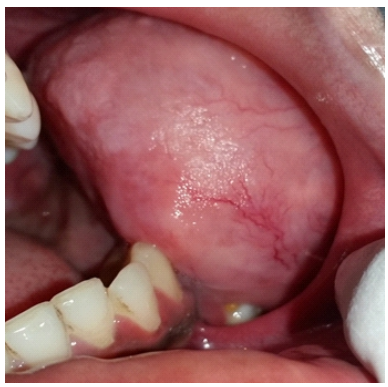

Figure 2: Intraoral photograph revealing a solitary extensive growth with displaced 35

Based on the history and clinical findings, provisional diagnosis of ameloblastoma was made. Ameloblastic fibroma, Calcifying Epithelial Odontogenic Tumor, odontogenic myxoma, Central Giant Cell Granuloma, keratocystic odontogenis tumor were considered in differential diagnosis.

Intra oral periapical radiograph showed flecks of radiopacities within the radiolucency and panoramic radiograph revealed an expansile ill defined non corticated radiolucency with scattered radiopaque flecks of remaining bone in left side of mandible involving body, ramus and angle with expansion of lower border of mandible and severe destruction of condyle, body, ramus and angle with complete destruction of coronoid process with no evidence of obvious internal loculations or septae.

CT imaging revealed an expansile enhancing radiolucent lesion showing cortical destruction in body and left mandible (Figure: 3 and 4). FNAC revealed a mixed epithelial and mesenchymal lesion with epithelial cells seen in mono-layered sheets with honey-comb pattern giving an impression of a benign complex odontogenic cystic lesion.

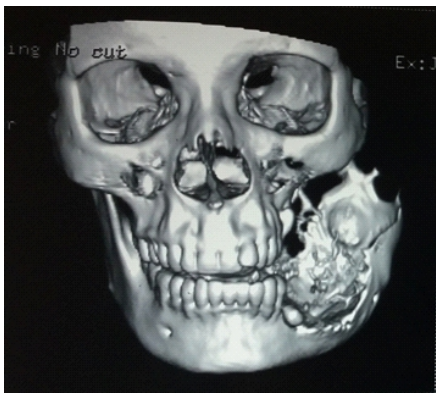

Figure 3: CT revealing 3D reconstructed image of ameloblatoma

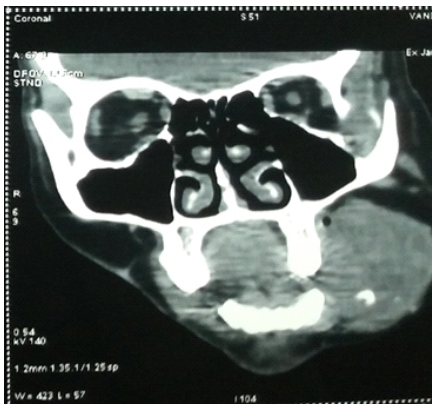

Figure 4: coronal CT revealing an expansile radiolucent lesion in the left side of mandible with displaced 35

Histopathologic sections showed parakeratinised stratified squamous epithelium with numerous follicles odontogenic epithelium of varying thickness surrounded by fibrous stroma with microcyst formation within follicles which confirmed the diagnosis of follicular ameloblastoma.

Hemi-mandibulectomy was done and reconstruction of left side of the mandible was done.

DISCUSSION

The ameloblastoma is an enamel tissue tumor, which does not differentiate to form the enamel.[4] Ameloblastomas are classified into various types based on clinical and radiographic characteristics, histopathology, and behavioral and prognostic aspects, three or four subtypes or variants of ameloblastomas can presently be distinguished:

1. The classic solid / multicystic ameloblastoma (SMA)
2. The unicystic ameloblastoma (UA)
3. The peripheral amelobalstoma (PA)
4. The desmoplastic ameloblastoma (DA), including so-called hybrid lesions.[5]

Ameloblastoma is thought to originate from residual epithelium from tooth germ; epithelium of odontogenic cysts; stratified squamous epithelium; and epithelium of the enamel organ.[6]

The most common presentation for ameloblastoma is an asymptomatic swelling of the mandible or maxilla[1] and identified as an incidental finding on radiograph or patient feels discomfort due to asymmetry of face because of the swelling.

Displacement of the tooth and root resorption are infrequent but have been reported in up to 25 % of desmoplastic ameloblastomas[1] whereas in the present case there was displaced premolar in unicystic varient. Paresthesias are uncommon, with rare reported cases of perineural invasion but in this case paresthesia was present. Ameloblastoma can be associated with unerupted third molar teeth, particularly in the unicystic type.[1]

Radiologically, the lesions are expansile, with thinning of the cortex in the buccal–lingual plane. The lesions are classically multilocular cystic with a “soap bubble” or “honeycomb” appearance.[6] Radiographs usually show a lytic lesion having scalloped margins, resorption of roots of the teeth, and impacted molars (unicystic). The classic "soap bubble" appearance is seen with the most common ameloblastoma, the multilocular/ solid type.[1]

Frequently a lingual and buccal cortical plate expansion is seen with the resorption of the roots of the teeth adjacent to the tumor. Unicystic ameloblastomas show a radiolucent image that surrounds the crown of an unerupted tooth or, they simply appear as a well defined radiolucent areas.[4]The radiographic appearance of ameloblastoma can vary according to the type of tumour.[6]

Computed tomography (CT) is one of the most useful diagnostic imaging modality, typically demonstrating well defined radiolucent unilocular or multilocular expansile lesions.[1]It is usually helpful in determining the contours of the lesion, its contents and its extension into soft tissues.[6]

Soft tissue extension and marrow extension beyond the lytic bone cavity is well appreciated in MRI when compared to CT.[1] MRI is essential for establishing the exact extent of an advanced maxillary ameloblastoma and thus determining the prognosis for surgery.[6]

Radiographic findings are characteristic but to achieve a final diagnosis histopathological examination is essential. Biopsy can be helpful prior to treatment to avoid unnecessary operations on lesions of alternative etiology which should be treated alternatively or simply observed, such as osteomyelitis, cystic fibrous dysplasia, giant cell tumor, ossifying fibroma, multiple myeloma, and rare sarcomas. Biopsy also allows for proper preoperative staging in malignant ameloblastomas.[1]

The solid/multicystic ameloblastoma can histopathologically be divided into a follicular and a plexiform type; the follicular type can be further subdivided into a spindle cell type, an acanthomatous type, a granular type and a basal cell type. The unicystic ameloblastoma represents an ameloblastoma variant that on gross examination, and not based on the appearance on the radiograph, presents as a cyst.[6]The unicystic type is the most benign and is further classified into intraluminal and intramural subtypes. The intraluminal unicystic subtype does not exhibit invasion of the supporting connective tissue, has the lower recurrence rate of the two subtypes, and may be the only histology amenable to conservative surgical treatment.[1]The plexiform type contains basal cells arranged in anastomosing strands with an inconspicuous stellate reticulum. The stroma is usually delicate, often with cystlike degeneration.[7]

Ameloblastic epithelium has been hypothesized to arise from the cells of rests of enamel organ or from cells of the sheet of Hertwig's or epithelial cell rest of Malassez or from epithelial boundary of an odontogenic cyst, particularly a dentigerous cyst or from the basal cells of the oral mucosa or from heterotopic epithelial from other parts of the body, perhaps pituitary. Cellular atypia and mitotic activity are rarely present in any histologic subtype of ameloblastoma.[1]

Ameloblastomas are treated by curettage, enucleation plus curettage,

or by radical surgery.[6] Surgery is the standard treatment for ameloblastomas. As the extent of resection has been controversial, comprising of two surgical options: "conservative" vs. "radical". The conservative treatment involves enucleation/curettage of the bony cavity, while the radical surgery involves a radical operation with appropriate margins.[1]

Unicystic ameloblastomas are usually treated by curettage with 10%-15% of recurrence; however, avoiding patient mutilation. In the solid or multicystic tumor, it is necessary to have radical surgical excision, with resection of the affected bone with at least 15 mm of healthy tissue as safety margin. The mucosa in contact with the tumor must be entirely removed, because it may contain ameloblastic cells that can contaminate the graft during reconstruction.[4]

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