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RECURRENT UNICYSTIC AMELOBLASTOMA IN A 20 YEARS OLDFEMALE – CASE REPORT

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ABSTRACT:

Ameloblastoma is a slow growing, persistent and locally aggressive neoplasm of epithelial origin. According to the WHO ameloblastoma are classified into conventional, unicystic and peripheral. Unicysticameloblastoma refers to the cystic lesion that show clinical, radiological or gross features of a mandibular cyst but on histological examination show typical ameloblastomatous epithelium lining part of cyst cavity with or without luminal and or mural tumor growth. Moreover recurrence of unicysticameloblastoma may be long delayed and long term postoperative follow up is essential. Here we are presenting a case of recurrent unicysticameloblastoma in the right lower mandibular region in a 20 years old female patient.

Keywords: unicysticameloblastoma, recurrence, mandibular cyst

INTRODUCTION:

Ameloblastoma is reported to constitute about 1-3% of tumors and cysts of the jaw [1,2]. Although it is considered a benign tumor, its clinical behavior may be regarded as lying between benign and malignant [3]. It often presents as a slow growing painless swelling causing expansion of the cortical plates and infiltration of soft tissue. There is often delay in the diagnosis because of its slow growing nature [4]. Unicysticameloblastoma is a less encountered variant of the ameloblastoma, accounting for 10-15% of all intra osseous-

ameloblastomas [5,6]. Hong et al showed that the histopathology of an ameloblastoma is significantly associated with recurrence[7]. It was shown that the follicular, granular cell and acanthomatous types have a relatively high likelihood of recurrence, in contrast, the desmoplastic, plexiform and unicystic types show a relatively low potential for recurrence. Recurrence rate of 10 -20% has been reported after enucleation and curettage of unicysticameloblastoma. This is considerable less than 50 – 90% recurrence rate noted after curettage of conventional solid and

multicystic extraosseous ameloblastoma [8,9]. We present a case of large recurrent unicystic ameloblastoma in the mandible in a 20 years old female.

CASE REPORT:

A 20 years old female reported to our College with the complaint of swelling (figure 1) on the right side of the mandible of 2 weeks duration. There was no associated difficulty in opening the mouth, chewing or articulating. She had undergone surgical removal of unknown lesion under general anaesthesia 5 years back, which had presented as a swelling and began spontaneously without any history of trauma or infection. Root canal treatment was also done in relation to five teeth. Patient reported to our department with the complaint of swelling which increased gradually since two weeks in the same region (right lower mandibular region). On examination, extraorally there was solitary diffuse swelling seen over right lower mandibular region extending anteroposteriorly from about 2 cm lateral to midline and about 3 cm anterior to the angle of mandible. The skin over the swelling appeared to be normal. Intraorally there was mild obliteration of buccal vestibule on right mandibular region. There was

expansion of buccal and lingual cortex. The lesion was non-compressible and non-pulsatile. No neck nodes were palpable. Her medical history was unremarkable.

An orthopantomogram (OPG) showed (fig 2) solitary unilocular radiolucency extending posteroanteriorly from the distal aspect of 47 to distal aspect of 42. Superiorly it extended from the mid radicular level to the lower border of the mandible. Inferior border of mandible was intact. Radiopacity was seen in the pulp canal in relation to 41, 42, 43, 44, and 45 indicating obturation which was done 5 years back. Root resorption was seen in relation to 44, 45, 46. CT scan showed (figure 3 & 4) that cystic lesion was confined to the mandible and there was perforation of lingual and buccal cortex. The patient was treated surgically under General Anesthesia. Surgical enucleation (fig 5) of cystic lining along with extraction of 42, 43, 44, 45, 46, 47 was done followed by chemical cauterization with Carnoy's solution. Primary closure was done. The excisional specimen showed histological features of unicystic ameloblastoma which had luminal growth. Follow up was done after 6 months which showed signs of bony healing with reduction in the size of radiolucency.



Fig: 1. Swelling in the right side of mandible



Fig: 2. Panoramic radiograph showing unilocular radiolucency



Figs 3 & 4: 3D reformatted CT image of buccal and lingual view showing perforation of buccal and lingual cortex.



Fig 5: intraoperative picture showing intact lower border of mandible



Fig 6: post operative photograph

DISCUSSION:

Ameloblastoma is a slow growing, persistent and locally aggressive neoplasm of epithelial origin accounting for 10% out of 30% of odontogenic tumors [16]. According to WHO, ameloblastoma are classified into conventional, unicystic and peripheral. Unicysticameloblastoma was first described by Robinson and Martinez in 1977, referring to those cystic lesion that showed clinical, radiological or gross features of a mandibular cyst [16]. It is a rare type of ameloblastoma accounting for about 5-10% of all intra-osseous-ameloblastoma[5,10].

Unicysticameloblastoma is believed to be less aggressive and responds more favorably to conservative surgery than solid or multicysticameloblastoma [6]. Leider et al [11] proposed three pathogenic mechanisms for the evolution of unicystic-ameloblastoma. They are Reduced enamel epithelium, From dentigerous cyst and Due to cystic degeneration of solid ameloblastoma. Histologically the minimum criteria for diagnosing a lesion as unicysticameloblastoma is the demonstration of a single cystic sac lined by odontogenic (ameloblastomatous) epithelium often seen only in focal areas. Unicysticameloblastoma should be differentiated from odontogenic cysts because the former has higher rate of recurrence than the latter. In a clinicopathological study of 57 cases of

unicysticameloblastoma, Ackermann [12] classified them into three histological groups.

Group1: luminal unicysticameloblastoma(tumor confined to the luminal surface of the cyst); Group 2: intraluminal/ plexiformunicystic-ameloblastoma(nodular proliferation into the lumen without infiltration of tumor cells into the connective tissue wall) andGroup 3: mural unicystic-ameloblastoma (invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium). Another histological sub-grouping by Philipsen and Reichart [5] has also been described, they are: Subgroup 1: luminal unicystic-ameloblastoma; Subgroup 1.2: luminal and intraluminal; Subgroup 1.2.3: luminal, intraluminal and intramural and Subgroup 1.3: luminal and intramural. The unicysticameloblastoma diagnosed as subgroups 1 and 1.2 can be treated conservatively (careful enucleation) whereas sub group 1.2.3 and 1.3 showing intramural growth requires radical resection as for a solid or multicysticameloblastoma [13]. Following enucleation, vigorous curettage of the bone should be avoided as it may implant foci of ameloblastoma more deeply into bone. Chemical cauterization with carnoy's solution is also advocated for subgroup 1 and 1.2.Subgroups 1.2.3 and 1.3 have a high risk for recurrence requiring more aggressive surgical procedure. This is because the cystic

wall in these cases has islands of ameloblastoma tumor cells and there may be penetration into the surrounding cancellous bone [14,15,13]. In the present case since it was a recurrent unicysticameloblastoma in a young patient which was not involving lower border of mandible (fig 5), enucleation along with chemical cauterization was done. Histopathology showed unicysticameloblastoma with luminal growth. Follow up after 6 months showed no recurrence.

CONCLUSION:

Whatever surgical approach the surgeon decides to take, long term follow up is mandatory, as recurrence of unicysticameloblastoma may be long delayed.

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