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MANAGEMENT OF UNICYSTIC AMELOBLASTOMA CROSSING MIDLINE IN MANDIBLE IN GERIATRIC PATIENT.

Surgery

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ABSTRACT

BACKGROUND: unicystic Ameloblastoma's(UA's) refers to those cystic lesions that show clinical, radiographic or gross features of a jaw cyst but on histologic examination show a typical ameloblastomatous epithelium lining the cyst cavity, with or without luminal and/or mural tumor proliferation.It is a distinct entity of ameloblastomas, a less frequently occurring tumor .It accounts for 10 to 15% of all the intraosseous ameloblastomas, characterized by slow growth and relatively locally aggressive, with the main site of origin in the posterior portion of the mandible. (mandible to maxilla = 3 to 13:1) Approximately 50–80% of UAs are associated with the impacted tooth (dentigerous variant). Histologically, the minimum criterion for diagnosing a lesion as UA is the demonstration of a single cystic sac lined by odontogenic (ameloblastomatous) epithelium, often seen only in focal areas. The rate of recurrence for these lesions, as determined by long term follow-up observation in many patients, was distinctly lower than that associated with multicystic and solid ameloblastoma. This, coupled with the preservation of the unicystic character of the lesion throughout its course, is indicative of a much less aggressive variety of neoplasm. The adequacy of simple enucleation as a modality of treatment in the majority of patients with this type of lesion is suggested.

CASE PRESENTATION: We in this article report a case of an 80-year-old female patient presented with painless intraoral swelling increased in size with time since five years, located in the anterior mandible, radiographically a single unicystic radiolucency with a radiopaque rim at from 35 to 43, FNAC proved unicystic variant of ameloblastoma, treated successfully by enucleation under local anesthesia.

KEYWORDS

Unicystic Ameloblastoma, Anterior Mandible, FNAC, Enucleation

INTRODUCTION:

Benign lesions of odontogenic or non-odontogenic origin can cause swellings in the mandible. Ameloblastoma is the most common odontogenic tumor which develops from epithelial cellular elements and dental tissues in different developmental phases. It is usually a slow-growing, persistent, and locally aggressive neoplasm of epithelial origin, affecting the posterior area of the lower jaw in 80% of cases.[1] There are three different forms of ameloblastomas, namely, multicystic, peripheral, and unicystic tumors. [2] Robinson and Martinez first described a unicystic ameloblastoma (UA) in 1977.[3] It is although a variant of ameloblastomas, it has a relatively benign biologic behavior and better response to conservative treatment, and this makes it a distinguishable entity. It accounts for 15% of all intraosseous ameloblastomas and often affects the younger population, with half of the cases occurring in almost the second decade of life.[4]Unicystic ameloblastomas have a slight male predilection and frequently originate from the posterior mandible.[5] Approximately 50–80% of Unicystic ameloblastoma's are associated with an impacted tooth, the mandibular third molar being the most commonly involved.[6] The clinical and radiographic presentations and gross features of unicystic ameloblastoma, when it develops in relation to an unerupted tooth, are indistinguishable from those of dentigerous cysts.[3]Therefore, based on these parameters, it is impossible to distinguish between these two types of lesions. Unicystic ameloblastoma has become as a distinct clinicopathological entity on the general basis of its unicystic radiographic appearance, histologic findings, association with an unerupted tooth, occurrence in the mandible of younger patients, and a recurrence rate after conservative surgical treatment lower than that of its conventional counterpart.[7]The diagnosis of unicystic ameloblastoma can only be made when the presence of ameloblastic epithelium can be established unequivocally. [3]Unicystic ameloblastomas are believed to be less aggressive and respond more favorably to conservative management, including enucleation, curettage, and marsupialization. This article illustrates a case report of Unicystic Ameloblastoma in an atypical location in the mandibular anterior and premolar maxillary region without any association with impacted teeth in old aged women treated surgically by enucleation and no recurrence in periodic follow ups for one year.

CASE REPORT:

A 70-year-old female patient reported to the department of oral and maxillofacial surgery, GSL dental college and hospital, rajahmundry, to seek treatment for a progressive asymptomatic swelling in respect to the lower anterior mandible, which gradually increased in size in the past five years to attain the present size of concern, patients dental history revealed extraction of 32 by a local dental quack five years back,as there was lack of relief patient visited us for definitive treatment. Medical history data of the patient revealed no systemic disease or other health problems.

On extraoral examination there was no evident swelling, no palpable lymph nodes, there was no neurosensory deficit, Intraoral examination revealed a well a solitary oval swelling of 4x4 cm on the buccal aspect of mandibular lower anterior region, extending from the distal aspect of44tothemesialaspect of 34. The oral mucosa had a normal appearance, but a slight tenderness was found on palpation. Swelling was fluctuant, soft in consistency, compressible, and reducible. Expansion was noticed in the buccal and lingual cortical plate in relation to 34 to 44 with grade I mobility and 31,41,42.

A provisional diagnosis of the odontogenic cyst was made on clinical findings.

Panoramic radiograph revealed a well corticated unilocular radiolucent lesion approximately 5 × 5 cm in diameter, which was in contact with the roots of the teeth present from 35to 43, root resorption is evident in relation to 31,41.42 most probably due to the cystic pressure.

Differential diagnosis of a residual cyst, dentigerous cyst, keratocyst,or ameloblastoma was made based on clinical and radiographic findings, we planned and performed a fine needle aspiration cytology. The lesion was perforated with a wide bore needle to rule out vascular lesion and aspirate the content. The aspirate, which was straw-colored, was sent for pathological evaluation, and the report stated it to be a cystic fluid with inflammatory exudates, an infected cyst.

Enucleation of the lesion under local anesthesia was performed. Under local anesthesia,an incision was made from 44 to 34. A mucoperiosteal flap was raised, and it was possible to see a thin expanded cortical plate that was removed with a scalpel. The cavity contained a fibrous tissue wall full of liquid. The lesion, with all its fibrous tissue lining, was enucleated carefully to ensure complete removal. The remaining bone tissue showed normal contour and consistency without any clinical signs of the lesion. Collagen was used and mucoperiosteal flap sutured back after thorough curettage and lavage of the bony cavity.The entire specimen was then submitted for histopathologic examination.

Histopathology report revealed epithelial islands and nests of tumor tissue with cystic change. Epithelial nests show central stellate reticulum cells,surrounded by palisading ameloblastic columnar cells; cells were not showing off any atypia. Epithelium extended into stroma as plexiform cords-features suggestive of cystic ameloblastoma.

Based on the radiographic and post-operative histologic features. It confirmed as unicystic ameloblastoma.

Presently, the patient is under follow-up since one year without any signs of relapse and with bony restoration in the affected area.

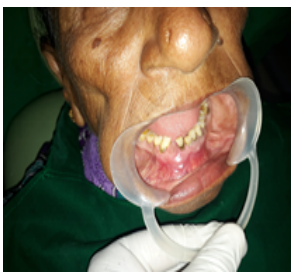

Fig.1. Intraoral Swelling At Anterior Mandibular Region.

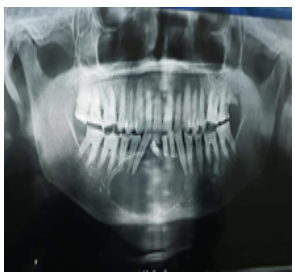

Fig.2. Orthopantomogram Of The Lesion.

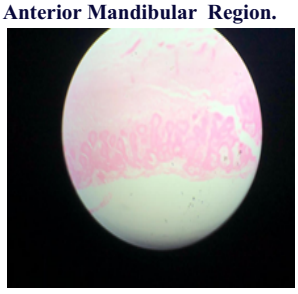

Fig.3. Histological Slide

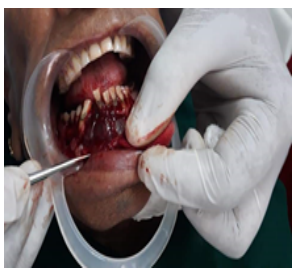

Fig.4. Exposure Of The Lesion.

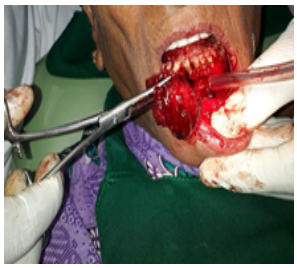

Fig.5. Enucleation Of The Unicystic Ameloblastoma.

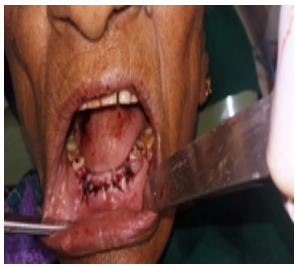

Fig.6. Suturing Done.

DISCUSSION:

The ameloblastoma is a neoplasm of true odontogenic epithelial origin. Second most common odontogenic neoplasm.Clinical behavior, combined with its incidence, makes ameloblastoma the most significant odontogenic neoplasm.[8] Ameloblastomas are classified into three types: multicystic intraosseous or solid conventional ameloblastomas,unicystic intraosseous ameloblastomas, and peripheral ameloblastomas.[9]Recognition and distinction of the different forms of ameloblastomas are fundamental because they are closely linked to the treatment plan and prognosis.
The term unicystic is derived from the macro and microscopic appearance, this lesion being, in essence, a well-defined, single cystic sac lined by odontogenic (ameloblastomatous) epithelium, whereas unilocular, on the other hand, is a term used in the radiographic interpretation of a radiolucency having only one loculus or compartment. Much confusion occurs from the fact that a Unicystic Ameloblastoma(UA) may appear not only as a unilocular but indeed also as a multilocular bone defect[5]

Unicystic ameloblastoma is characterized by the proliferation of an ameloblastomatous epithelium, which may be confined to the lining of the cystic cavity or may exhibit an intraluminal growth or mural proliferation. It has also been described as invasive ameloblastoma which presents as a single cystic cavity rather than multiple cystic spaces[1]Robinson and Martinez were the first to describe this lesion as a distinct entity. [3]According to Robert and Diane, unicystic ameloblastoma may originate from reduced enamel epithelium; or as a result of the transformation of the dentigerous cyst into unicystic ameloblastoma; or as a result of cystic degeneration of solid/multicystic ameloblastoma .[10]

Unicystic Ameloblastoma being a rare type of ameloblastoma, accounts for about 6% of ameloblastoma, 50% of these tumors diagnosed during the second decade of life (Roos*et al.*, 1994).The age is considerably lower and ranges from 19 to 27 years (Reichart&Philipsen).[11]When compared to multicystic ameloblastoma, UA occurs more commonly at younger, but contrary to literature in our case it occurred in an old adult patient. The most common site ofoccurance is in posterior mandible followed by parasymphysis region, anterior maxilla and posterior maxilla, again in contrary to literature it occurred in the anterior mandibular region crossing the midline. Approximately 50–80% of UAs are associated with impacted tooth mostly mandibular third molars termed as dentigerous variant, and the one which is not associated with impacted teeth is termed as nondentigerous variant. The “dentigerous” variant occurs approximately 8 years earlier than the “nondentigerous” variant. UAs are usually asymptomatic.[6] In this case according to literature as it is not associated with an impacted tooth is a non dentigerous variant of unicystic ameloblastoma. The chief presenting sign was enlargement without significant symptoms. However, many instances were detected following the procurement of oral radiographs.[12] Gender distribution shows a slight male predilection with a male to female ratio of 1.6: 1.However, when the tumor is not associated with an unerupted tooth, the gender ratio is reversed to a male to female ratio of 1: 1.8.5 as in our case it occurred in a female patient and it's a non dentigerous variant.

The unicystic ameloblastoma may reach considerable size with a multilocular or unilocular appearance . The border of the lesion is usually well-defined without any radiographic evidence of infiltrative growth into the surrounding bone. A diffuse border of the lesion may appear, perhaps representing a less favorable tissue response or faster growth . The cortical bone, however, is expanded, with numerous perforations but without any signs of clinical or histological neoplastic soft tissue in growth. In contrast to the primordial cyst, the unicystic ameloblastoma may cause root resorption,[11] as shown in the present case. Predominant radiographical patterns for UA according to Eversole et al. and Paikkatt et al.are : unilocular, scalloped macromultilocular, pericoronal, interradicular, or periapical expansile radiolucencies. Our case had a peculiar radiographic presentation of periapical unilocular radiolucency crossing the midline of the mandible.[9]

Histologically, the minimum criteria for diagnosing a lesion as UA is the demonstration of a single cystic sac lined by odontogenic (ameloblastomatous) epithelium often seen only in focal areas.

According to a clinicopathologic study of 57 cases of UA,

Ackermann[13] classified this entity in to three histologic groups:

I. luminal UA (tumor confined to the luminal surface of the cyst)
II. intraluminal/plexiform UA (nodular proliferation into lumen without infiltration of tumor cells intoconnective tissue wall); and
III. mural UA (invasive islands of ameloblastomatous epithelium in the connective tissue wall notinvolving the entire epithelium).

Another subgrouping by Philipsen and Reichart[2] has also been described as follows:
Subgroup1:luminalUA;
1.2:luminal and intraluminal;
1.2.3:luminal,intraluminal and intramural;and
1.3:luminal and intramural.

The UA diagnosed as subgroups 1 and 1.2 can be treated conservatively (careful evaluation), whereassubgroups 1.2.3 and 1.3 which has intramural growths require radical resection.

Our case according to histopathologic report falls under grade III mural unicystic ameloblastoma, and a sub group 1.3.

Perhaps the most crucial consideration regarding unicystic ameloblastoma is that of biologic behavior.It has been widely stated that these lesions are less aggressive than their solid or multicystic counterparts and should be treated by enucleation or curettage. However, Gardner has pointed out that there is a difference in biological behaviour between those lesions that are simply cystic or show intraluminal proliferation and those in which the epithelium penetrated and breaches the fibrous wall, therefore having the capacity to invade the cancellous bone.[14]

Conservative treatment is suggested, especially in younger populations, in light of the devastating impacts on the developing jaw,masticatory function, facial growth, and psychosocial aspects.Porgrel et aladvocated that enucleation followed by curettage and liquid nitrogen cryospray or Carnoy's solution cauterization would be appropriate for unicysticameloblastomas. In extensive lesions, marsupialization may be an alternative treatment, because it is easy to perform and safe, and can reduce the size of the lesion and surgical morbidity.Marsupialization is reported to be useful as preliminary management and as a more-effective treatment modality for cystic lesions in teenaged patients as well. Therefore, when planning treatment of unicystic ameloblastomas, patient's esthetic problems,masticatory function, facial growth, quality of life, and potential morbidity caused by the surgical intervention should be taken into account.

Hence, finally taking into account the age of the patient, clinical and radiologic features, the simple enucleation procedure was chosen as the treatment of choice, for it involves least patient morbidity and the effect on the quality of life are minimal.

The average interval of recurrence is seven years. Recurrence is also mostly related to histologic subtypes of UA, with those invading the fibrous wall having a rate of 35.7%, but others only 6.7%[12] in our case the patient was followed for 24 months now and there are no signs of recurrence.

CONCLUSION:

Neither the incisional biopsy nor the aspirational cytology may be able to reflect the true nature of the lesion. An enucleation biopsy best achieves the objectives of correct histological diagnosis, classification, and appropriate therapy. The histopathologic examination of the surgical tissue provided the final diagnosis of ameloblastoma because the lesion was in an atypical location. Long-term follow-up is mandatory for unicystic ameloblastomas since recurrence may take place years after removal. More than 50% of cases recurred within five years after the operation. Frequent postsurgical radiographic examinations favor early detection of recurrence. Unicystic variant of ameloblastoma with aggressive histologic behaviour also might be successfully treated with enucleation, and in this approach, the patient should be regularly followed-up.

DECLARATIONS

Patient Consent: we have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts Of Interest: There are no conflicts of interest :

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