Granular Cell Ameloblastoma of the Anterior Mandible: A Case Report

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Abstract:
Ameloblastoma, previously known as adamatinoma, a distinctive benign, locally invasive odontogenic tumor grows slowly and progressively. Among the six histopathological variants, the granular cell type is the rarest form but the most aggressive in terms of recurrence. A patient presented with a recurrent anterior mandibular swelling. The swelling was finally excised and was reported to be granular cell variant.

Introduction

The ameloblastoma is the second most common clinically significant and potentially lethal odontogenic tumour. Excluding odontomas, its incidence equals or exceeds the combined total of all other odontogenic tumours. 1 The amelobalstoma is a true neoplasm of enamel organ type tissue which does not undergo differentiation to the point of enamel formation. It has been very aptly described by Robinson as being a tumour that is "usually unicentric, non-functional, intermittent in growth, anatomically benign and clinically persistent". 2

The term amelobalstoma was suggested by Churchill in1934 to replace the term "adamantinoma" coined by Malassez in 1885. Its incidence, combined with its clinical behaviour, makes it the most significant odontogenic neoplasm of concern to the oral and maxillofacial surgeon.2

These tumors may arise from the epithelial lining of an odotogenic cyst or basal cell of the oral mucosa. The amelobalstoma occurs in three different variants, each with the specific implication of treatment and a unique prognosis: solid or multicystic, unicystic and peripheral. 1

We report a rare case of a granular cell variant of ameloblastoma in the anterior mandible with a brief review of literature.

Case report

A 46 year old male reported to the Department of Oral & Maxillofacial Surgery with a swelling in the anterior mandible since last 13 years. Patient gives history of a similar swelling 12 years ago for which he visited a local dental clinic where the lesion was excised. The patient had been asymptomatic for two years following which, the swelling recurred. It started as a painless, small swelling which gradually increased to its present size. He does not give any history of pain, paresthesia or hemorrhage from the area. He does not have any significant medical history. The patient is moderately built and nourished with no abnormalities detected on general examination. His mouth opening was found to be adequate with no deviation or deflection. The patient did not have any gross facial asymmetry. On intraoral examination, the swelling was found to be solitary measuring approximately 5 x 4 cm in the anterior mandible with the expansion of the buccal and lingual cortices. The overlying mucosa appeared normal. On palpation the inspection findings were confirmed. The swelling was found to be non tender, firm in consistency, no crepitation or fluctuation. The buccal cortex appeared to be perforated but the lingual cortex seemed to be intact. No occlusal derangement was noted except for increased mobility with respect to the lower anterior teeth.

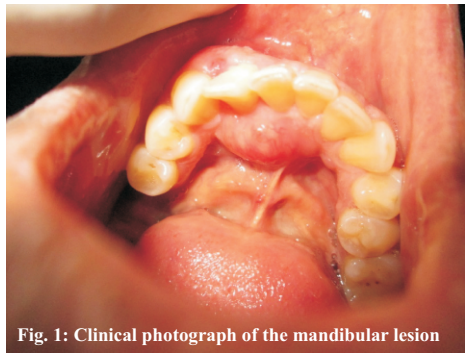

Fig. 1: Clinical photograph of the mandibular lesion

An orthopantamogram revealed a unilocular radiolucency on the anterior mandible with well defined, scalloped margins involving the roots of the left canine and extending to the right lateral incisor. A CBCT confirmed the perforation of the buccal cortex and significant expansion of the lingual cortex.

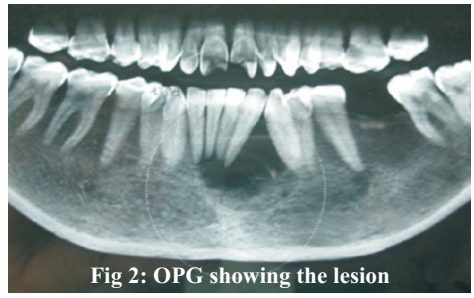

Fig 2: OPG showing the lesion

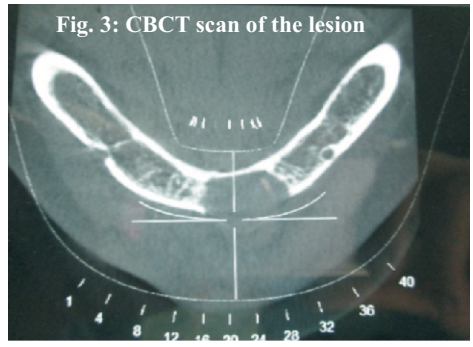

Fig. 3: CBCT scan of the lesion

An incisional biopsy of the area was done under local anesthesia and was reported as follicular type of ameloblastoma. Wide excision of the lesion and marginal mandibulectomy was planned. A degloving incision was placed in the anterior mandible extending from one the premolars of one side to the other side and subperiosteal dissection was performed. The mental nerve and vessels were identified bilaterally and preserved. The bone was osteotomized. En bloc resection was performed with 1 cm of safe margin of bone. The osteotomy cut was performed between the canine and the premolar on the left side and between the canine and lateral incisor on the right side. The geniglossus muscle was identified and detached. It was then sutured to the remaining mandible.

The bony margins were smoothened and the wound was closed primarily with 2-0 Vicryl. A pressure dressing was placed for two postoperative days. On histopathological examination, the stained sections showed odontogenic epithelium andconnective tissue. The epithelium was present as follicles which comprised of peripherally tall columnar cells with reverse architecture. The odontogenic cells were compose of granular cell cytoplasm. Connective tisuue was composed of collagen bundles, fibroblasts and a few extravasated RBCs. The final histopathological report of the specimen was suggestive of granular cell ameloblastoma.

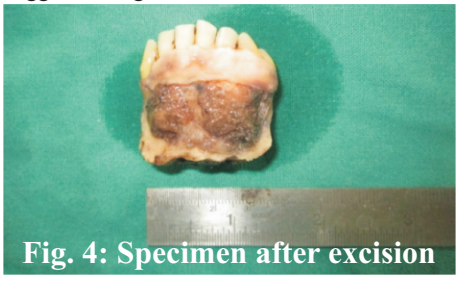

Fig. 4: Specimen after excision

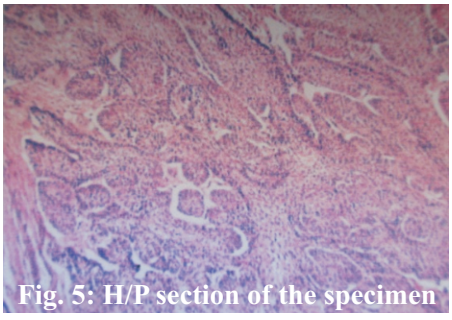

Fig. 5: H/P section of the specimen

Discussion:

Most authorities consider ameloblastoma of to be of varied origin, although the stimulus initiating the process is not known. Thus the tumor may be conceivably derived from:

- Cell rests of the enamel organ or that of Hertwig's root sheath
- Epithelium of odontogenic cysts, particularly dentigerous cyst and odontomas
- Disturbances in the developing enamel organ
- Basal cells of the surface epithelium of the jaws

Environmental factors and individual patient factors likely have a role in modulating the incidence of the disease.

A wide range of occurrence of the tumor from 10 years through 90 years is reported. The average age for diagnosis is in the range of 33- 39 years and most cases cluster between 20- 60 years. No significant sex predilection is noted. Ameloblastoma occurs in all areas of the jaws but the mandible is most common. Within the mandible, the molar- ramus area appears to be the most common site.2 In our case, the anterior mandible was affected which is not so common a site for ameloblastoma.

Ameloblastoma is described classically as a multilocular cyst like lesion of the jaws. The tumor exhibits a compartmented appearance with the septa extending into the radiolucent tumor. In many cases, however (as in our patient), the lesion is a unilocular one and presents no characteristics or pathognomonic features. The periphery is usually smooth although this regularity may not be borne out at the time of operation. In advanced cases, thinning of the cortical plate may be seen.2

Six histopathological subtypes of ameloblastoma are recognized: follicular, plexiform, acanthomatous, granular cell, basal cell and desmolastic. Most tumors show a predominance of one pattern but few lesions are found to be composed of purely one histopathologic type. The follicular variety has the highest rate of recurrence. 2

In granular cell ameloblastoma, there is marked transformation of the cytoplasm, usually of the stellate reticulum like cells so that it takes a on a very coarse, granular, eosinophilic appearance. Hartman reported that this type appears to be an aggressive lesion with marked proclivity for recurrence unless appropriate surgical measures are instituted. In addition several cases have been reported as metastasizing. 2

The ameloblastoma occurs in three different variants, each with its specific implications for treatment and a unique prognosis: solid or multicystic, unicystic and peripheral. The solid or multicystic variant tends to infiltrate between the intact cancellous bone trabeculae at the periphery of the tumor before bone resorption becomes radiographically evident. Therefore the actual margin of the tumor extends beyond the apparent radiographic or clinical margin. Therefore, the resection of the tumor with a 1.0 cm of linear bony margin is recommended. Soft tissue margins are best managed according to the anatomic barrier margin principles whereby one uninvolved surrounding anatomic barrier is sacrificed on the periphery of the specimen. Unfortunately, any less aggressive treatment modality may be fraught with inevitable persistence discovered at variable times postoperatively. Moreover, although persistent and unresectable ameloblastomas are radiosensitive, once this tumor defies curative surgical therapy, radiation is of questionable use.1

Unicystic amelobalastoma is most commonly seen in younger patients with 50% of the tumors being diagnose during the second decade of life. More than 90% of the tumors are in the mandible in the molar ramus area. Three histopathological variants of the tumor have been described that impact treatment and prognosis. The luminal unicystic ameloblastoma is confined to the luminal surface of the tumor. The intraluminal unicystic amelobalstoma contains one or more nodules of ameloblastoma that projects into the lumen of the cyst. In the third variant, known as mural unicystic amelobalstoma, the fibrous wall of the cyst is infiltrated by typical follicular or plexiform ameloblastoma. 1

When the ameloblastic elements are confined within the lumen with or without intraluminal extension, enucleation can be probably curative. When the cyst wall has been violated by tumor as in the mural variant the treatment is controversial. If the diagnosis is made postoperatively, the surgeon may wish to adopt a close, indefinite follow- up examination. If a preoperative biopsy reveals a mural type of ameloblastoma, the surgeon might recommend a resection of the tumor because this variant has the highest degree of persistence. The treatment of luminal and intraluminal variants is enucleation and curettage. 1

The peripheral ameloblastoma is the rare variant. Clinically these tumors are nonulcerated, sessile or pedunculated gingival lesions. Most examples are smaller than 1.5 cm with an average reported age of 52 years. They may be seen to “cup- out” the jaw bone. It is most appropriately treated by wide local excision. Malignant transformation is very rare.1

Elzay and Corio proposed that the term “ameloblastic carcinoma” be used to refer to any ameloblastoma with microscopic evidence of malignancy in either the primary or the recurrent tumor regardless of whether it has metastasized and the term “malignant ameloblastoma” be confined to the ameloblastomas that have metastasized and yet have maintained the classic well differentiated features of ameloblastoma. 6

Conclusion

To summarize, we have described a rare case report of granular cell variant of ameloblastoma in the mandibular anterior area. The treatment was planned considering the clinical, radiologic and histopathological features considering the aggressiveness and high chances of reccurrence of the granular cell variant. The patient has been on regular followup.

References:

References are available on request at editor@healtalkht.com