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Ameloblastoma of retromolar region extending to ramus: A case report

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Abstract
Ameloblastoma is the most common benign odontogenic tumour of the jaws constituting about 1% of all cysts and tumours of the jaws. It is more commonly found in mandibular posterior region. Conventional ameloblastoma are common than other counterparts which appear radiologically as multilocular radiolucency exhibiting soap bubble pattern. Here we present a case report of conventional ameloblastoma which appeared as desmoplastic variant histopathologically.

Keywords: Ameloblastoma, odontogenic tumour, cysts

Introduction
Ameloblastoma, derived from an English word "Amel" which means enamel and Greek word "blastos" which means germ, is the most common benign odontogenic tumour of the jaws constituting about 1% of all cysts and tumours of the jaws [1]. It can arise from the multiple sources of odontogenic epithelium that remain with the alveolar soft tissue and bone. It is painless, slow-growing, locally aggressive tumour and can cause expansion of the cortical bone, perforation of the lingual or the buccal cortical plates and infiltration of the soft tissues. The peak incidence of ameloblastoma is in third and fourth decades of life but it can also occur in any age group with equal gender predilection (1:1) [2]. It is more commonly found in the mandibular posterior region [3]. According to WHO 2017 Clinical classification of ameloblastoma, it is of three types: conventional, unicystic or peripheral ameloblastoma [4]. In a conventional radiograph such as an orthopantomogram, conventional ameloblastoma can present as multilocular radiolucency surrounded by a corticated border; the bony septae contribute to make honeycomb or soap bubble appearance, or tennis racket pattern. In Unicystic variety, it presents as unilocular radiolucency associated with impacted tooth. Cortical plates may be spared in some areas and expanded as they may be destroyed in other regions along with root resorption [5]. Conventional radiographs are sufficient for smaller mandibular lesions but CT and MRI are required in maxillary lesions and extensive mandibular lesions to establish the extent of the lesion [6]. The challenge in the management of ameloblastoma is the complete excision and reconstruction of the defect when the lesion is quite large. Treatment options include enucleation, curettage or surgical excision (segmental resection/enblock resection or excision with peripheral ostectomy) depending on the size and type of the lesion [1].

Case report
A 52 year old female reported to the Dept. of Oral Medicine and Radiology, H.P Government Dental College, Shimla with chief complaint of swelling on right side of face for 3-4 months which was accompanied with pain particularly on biting and chewing. Patient was apparently well 4 months back when she first noticed a swelling which was insidious and painless initially and later it gradually increased in size and became painful. Pain was moderate and intermittent in nature and aggravated on chewing and swallowing.Though there was pain on biting but no dysesthesia in the lip, chin or overlying skin, and no discharge was noticed. Patient had no history of trauma or toothache and there was no decrease in the size of the swelling. After consultation from a nearby primary health center she was referred to this institute for evaluation.

Patient was a known case of hypothyroidism and was on medication for last 4-5 years. Extraorally, on inspection, face was apparently asymmetric with diffuse, smooth–surfaced swelling on right middle and lower 3rd of the face, extending superior-inferiorly (Measuring 7 cm approximately) from right portrayal region to inferior border of mandible and beyond. Anteroposteriorly (measuring 5.5 cm approximately) extending 1cm behind right commissure region to posterior border of the ramus of mandible. Skin overlying the swelling was stretched and exhibits no change in color. Swelling was non-tender on palpation and hard to palpating fingers with no evidence of crackling sound. Overlying skin was smooth in texture and no local raise of temperature is seen. Right submandibular lymphadenopathy evident with tenderness on palpation. (Fig. 1 and 2) Intraorally, a diffuse, smooth surfaced swelling evident at right retromolar pad region and beyond with slight lingual protuberance and obliteration of buccal vestibule. Overlying mucosa was blanched but similar to the colour of adjacent mucosa. On palpation it was non-tender, hard in consistency along with buccal and lingual cortical plate expansion. Along with clinically missing teeth with respect to (w.r.t.) 12, 14, 15, 16, 21, 23, 25, 26, 36, 37, 42, 46, 47. Root stumps w.r.t 24, 31, 34, 41, dental caries w.r.t. 11,22,43,44 and generalized attrition. (Fig 3) Based on the clinical findings, a provisional diagnosis of benign tumour of the right mandible was made. Dentigerous cyst, odontogenic myxoma, Keratocystic odontogenic tumor, Pindborg tumor and ameloblastic fibroma can be the differential diagnosis. Patient was advised routine haematological examination-which was within normal limits, Intra oral Periapical Radiograph (IOPAR) w.r.t.47 and 48 region, Orthopantomogram (OPG), CT of concerned region and incisional biopsy.

IOPAR W.R.T. 47 and 48 revealed multilocular radiolucency with scalloped border (producing soap bubble appearance), extent not clear. (Fig 4) OPG displayed a well-defined multilocular radiolucency with scalloped border (soap bubble appearance) extending from the right second mandibular premolar to the mandibular ramus. Extensive thinning, bulging of the cortical plate and downward projection of inferior border of mandible. (Fig 5) Plain axial/coronal CT revealing an expansile radiolucent lesion in the right body-ramus region of the mandible causing expansion of the ramus & body. Reformatted 3D IMAGE showing multiple small perforations in buccal cortical plate. (Fig 6, 7 and 8) Histopathological section revealed multiple fragments lined by acantholytic stratified squamous epithelium. Subepithelium revealed round to irregular angular odontogenic islands in desmoplastic stroma. Segments are lined by peripherally palisading cuboidal to flat cells with central spindle cells. Occasional islands revealed central stellate reticulum implying desmoplastic ameloblastoma. (Fig 9) Based on clinical, radiographic and histopathologic examination a final diagnosis of Ameloblastoma of right mandibular ramus and body was made. As the lesion was very extensive, a hemi-mandibulectomy along with reconstruction was planned. After hemi-mandibulectomy defect was reconstructed using reconstruction plate with condylar prosthesis. Figure 10 showing fragment of the mandible removed after surgery and submitted to confirm the initial biopsy. Figure 11 showing Post-operative extraoral view of patient. Figure 12 and 13 showing OPG and Posteroanterior view post operatively depicting reconstruction of mandible.

Discussions

Ameloblastoma, being a benign epithelial odontogenic tumour is usually aggressive and destructive with the capacity to cause erosion of bone and invade adjacent structures.[7] Ameloblastoma in mandibular region can progress to variable sizes (1–16 cm) and may cause facial asymmetry, displacement of teeth, malocclusion and pathological fractures [1]. In the present case also the patient's clinical examination revealed a large hard swelling in the retromolar region of the mandible which caused the facial asymmetry and expansion of the buccal as well as the lingual cortical plate. Ameloblastoma commonly presents as a painless, slow growing hard mass [1] but in this case, it was painless and presented a hard swelling. Other clinical presentations of this lesion are pain or anaesthesia of the affected area but that was not seen in our case. Based on WHO 2017 clinical classification, our case was conventional Ameloblastoma. There are six histopathological variants of Ameloblastoma: follicular, plexiform, acanthomatous, granular cell, basaloid and desmoplastic [4]. Our case presented a desmoplastic variant. The incidence of Desmoplastic Ameloblastoma (DA) ranges from 0.9% to 12.1% among other types [8]. Treatment of Ameloblastoma involves curettage, segmental resection, enbloc resection, excision with peripheral ostectomy. Concerning the biological behavior of Desmoplastic variant, WHO classification of odontogenic tumors suggests that DA, like unicystic ameloblastoma and peripheral ameloblastoma, can possibly have a lower recurrence rate than another ameloblastoma [9]. In contrast to that statement of the WHO, literature review provided the information that DA also has a similar recurrence rate (15.9%) with the other types of ameloblastoma. Keszler *et al*. even reported a higher recurrence rate (21.4%) of desmoplastic variant than the other type (10.1%) of ameloblastoma. The patient should be on periodic follow up after the treatment.

Fig 1: Right submandibular lymphadenopathy evident with tenderness on palpation

Fig 2: Right submandibular lymphadenopathy evident with tenderness on palpation

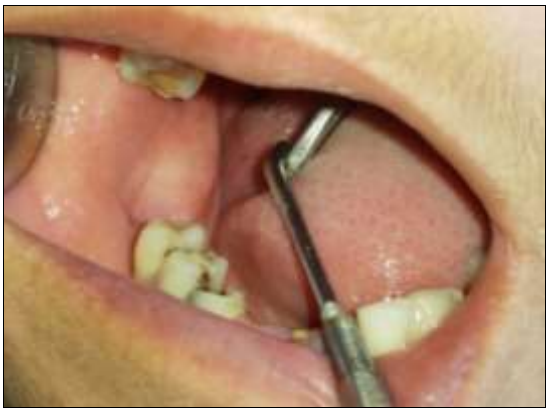

Fig 3: Clinically Missing Teeth

Fig 4: IOPAR W.R.T. 47 and 48 revealed multilocular radiolucency with scalloped border (Producing soap bubble appearance), extent not clear

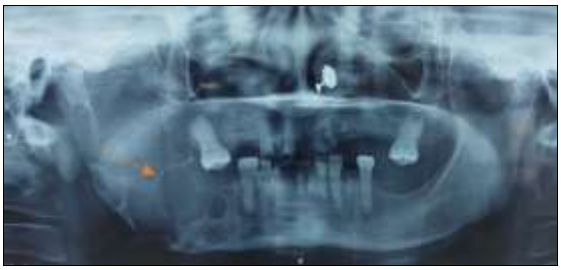

Fig 5: Extensive thinning, bulging of the cortical plate and downward projection of inferior border of mandible

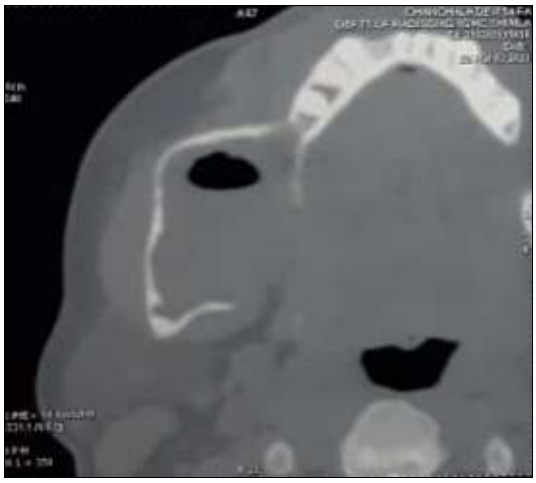

Fig 6: Reformatted 3D IMAGE showing multiple small perforations in buccal cortical plate

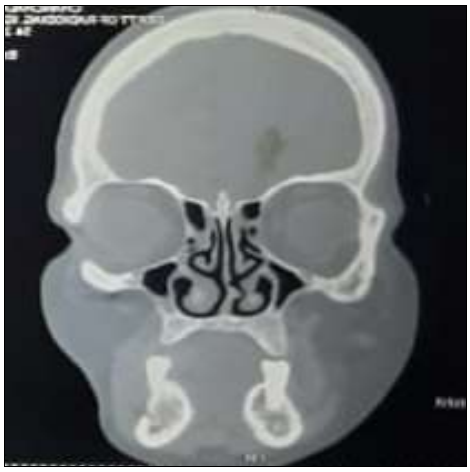

Fig 7: Reformatted 3D IMAGE showing multiple small perforations in buccal cortical plate

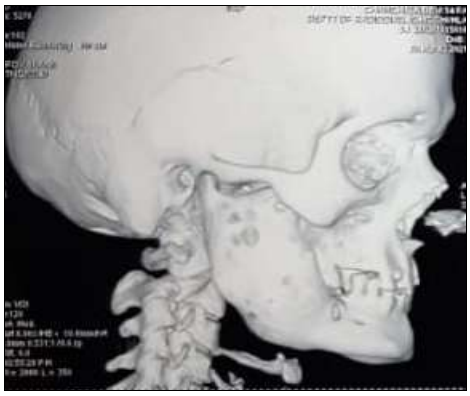

Fig 8: Reformatted 3D IMAGE showing multiple small perforations in buccal cortical plate

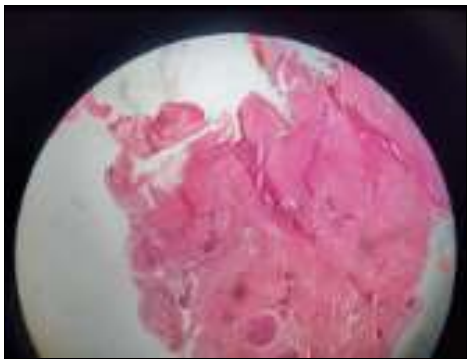

Fig 9: Occasional islands revealed central stellate reticulum implying desmoplastic ameloblastoma

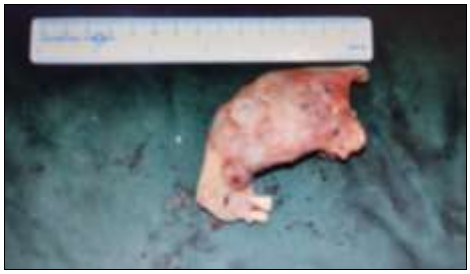

Fig 10: Fragment of the mandible removed after surgery and submitted to confirm the initial biopsy

Fig 11: Post-operative extraoral view of patient

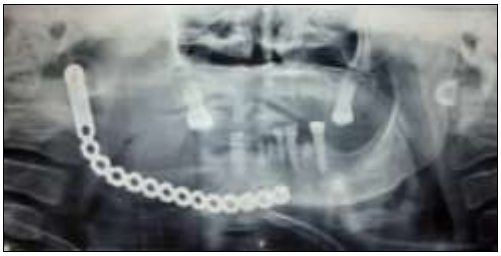

Fig 12: OPG and Posteroanterior view post operatively depicting reconstruction of mandible

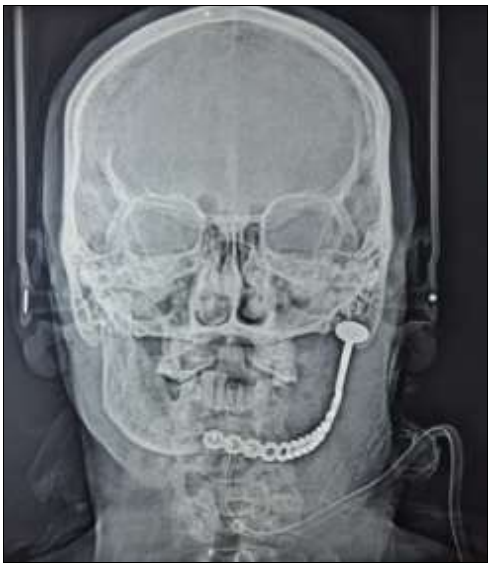

Fig 13: OPG and Posteroanterior view post-operatively depicting reconstruction of mandible

Conclusion

Ameloblastoma should always be considered as a differential diagnosis for the swelling in the mandibular posterior region, especially in the middle age group. Radiographs alone are sufficient for diagnosing smaller lesions but larger lesions need advanced imaging such as CT to see better extent and surgical management of the lesion. Wide resection of the lesion along with a safety margin of healthy bone will prevent recurrence as multilocular ameloblastoma has the highest recurrence rate among all other types of ameloblastoma. Resection followed by reconstruction using grafting procedure will restore aesthetics of the patient as well as function of the mandible.

Conflicts of interest

There are no conflicts of interest.

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