Mural Ameloblastoma of the Mandible: Radiological-Pathological Correlation

Yadavalli Guruprasad, Dinesh Singh Chauhan, Amit Byatnal[1], Shrinivas C Koppal[1], Umashankar Kura[2], Pradeep Mattighatta Rudraiah[3]

Departments of Oral and Maxillofacial Surgery, [1]Oral Medicine and Radiology, AME'S Dental College Hospital and Research Center, Raichur, [2]Department of Orthodontics, Government Dental College and Research Institute, Bellary, [3]Department of Oral and Maxillofacial Pathology, AME'S Dental College Hospital and Research Centre, Raichur, Karnataka, India

Correspondence: Dr. Yadavalli Guruprasad, Department of Oral and Maxillofacial Surgery, AME'S Dental College Hospital and Research Center, Raichur - 584 103, Karnataka, India. E-mail: guru_omfs@yahoo.com

ABSTRACT

Ameloblastoma is a benign odontogenic neoplasm, which frequently affects the mandible. The term 'ameloblastoma' includes several clinicoradiological and histological types. The radiological appearance is most commonly of a multilocular radiolucent type, with a high incidence of poorly defined borders. Definitive diagnosis of mural ameloblastoma can only be done by histopathological examination and cannot be predicted preoperatively on clinical or radiographical grounds. We are presenting a case of mural ameloblastoma in the body and ramus of the mandible in a 65-year-old female patient, with emphasis on its radiographical and histopathological appearances.

Key words: Mandible; multilocular; mural ameloblastoma; odontogenic tumors

Introduction

Ameloblastomas are the most common benign neoplasms of an odontogenic nature that directly affect the jaws. According to the World Health Organization (WHO) it is defined as a locally invasive polymorphic neoplasia that often has a follicular or plexiform pattern in a fibrous stroma.[1] In 20% of the cases, it is located in the maxilla, which can give rise to potential complications because of the close proximity to the vital structures and difficult surgical access, to obtain clean margins. However, 80% of the ameloblastomas occur in the mandible and, among these, 70% are detected in the ascending ramus or molar region, while 20% are found in the premolar area, and only 10% in the anterior mandible.[2] It appears most commonly in the third to fifth decades, but the lesion can be found in any age group, including children. Men are affected slightly more often than women, with a 1.4:1 ratio. The most common is the multilocular form, with multiple cysts that occur in groups or are separated by osseous septa, giving a soap bubble appearance, with scalloping and sclerotic margins. The teeth adjacent to the tumor may be displaced or may show root resorption.[2,3] From a histopathological perspective, the literature reports different types with prognostic significance. Multicystic ameloblastoma is the most frequent subtype and it generally causes gross facial deformities with both functional and aesthetic consequences. As the tumor tends to infiltrate cancellous bone trabeculae, the incidence of recurrence is very high, reaching up to 90%, despite extensive curettage. Histopathologically, a solid multicystic ameloblastoma is characterized by the occurrence of islands, strands, and irregular configurations of the tumor epithelium, consisting of a central mass of polyhedral cells resembling a stellate reticulum surrounded by a layer of cuboidal or columnar cells, similar to preameloblasts. When degeneration of centrally placed cells occurs in several tumor islands, the term 'multicystic' is often used.[3,4] We report a case of mural ameloblastoma affecting the right mandible in a 65-year-old female patient and the correlated radiological and histological findings.

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Case Report

A 65-year-old female patient was referred to the Department of Oral and Maxillofacial Surgery for evaluation of a right facial swelling. The lesion had been slowly increasing in size since it was first noticed about one year prior. There was no history of associated trauma, pain, palsy, paresthesia or

lymphadenopathy. There was significant facial asymmetry caused by an approximately 5.5 × 4.5 cm mass involving the right mandible. The mass was firm, nontender to palpation, and not adherent to the overlying skin. No bruits or pulsations were detected. Intraoral examination showed expansion of the right buccal and lingual cortex, thus obliterating the buccal vestibule. A panoramic radiograph showed a well-defined multilocular radiolucency involving the right mandible. There was tooth displacement with respect to 47 and 46, and root resorption of the latter [Figure 1]. The computed tomography (CT) scan of the right mandible showed multilocular hypodensity with medial and lateral cortical expansion with perforation of the buccal and lingual cortical plates [Figures 2-5]. The laboratory investigation values were within normal limits. Incisional biopsy was subsequently performed at the anterior border of the ramus and body of the right mandible. The histopathological examination revealed an epithelium lined by ameloblast-like cells invading into the connective tissue (cystic capsule), suggestive of a mural variant of ameloblastoma [Figure 6]. An immunohistochemistry examination was done, which showed CD34 expression, Ki-67 nuclear staining, and MMP-2 expression, consistent with the mural variant of ameloblastoma [Figures 7-9]. On the basis of the clinical, radiological, and histopathological findings, a diagnosis of mural ameloblastoma of the right mandible was made. The treatment plan was for segmental resection with wide margins, to avoid a recurrence and reconstruction procedure using bone grafts or reconstruction plates and the subsequent long-term follow-up.

Discussion

Ameloblastoma is one of the most common tumors of the jaw, which are usually benign and locally aggressive epithelial neoplasms of odontogenic origin. Although locally invasive, they are considered to be benign neoplasms derived from the odontogenic epithelium.[4] The term 'ameloblastoma' includes several clinical, radiographical, and histological types. On the basis of the clinical behavior and prognosis, three types of ameloblastomas can be distinguished: (1) The 'conventional

Figure 1: Orthopantomograph showing a multilocular lesion over the right side of the mandible causing expansion and thinning of the cortical plates extending from the body of the mandible to involve the entire ramus

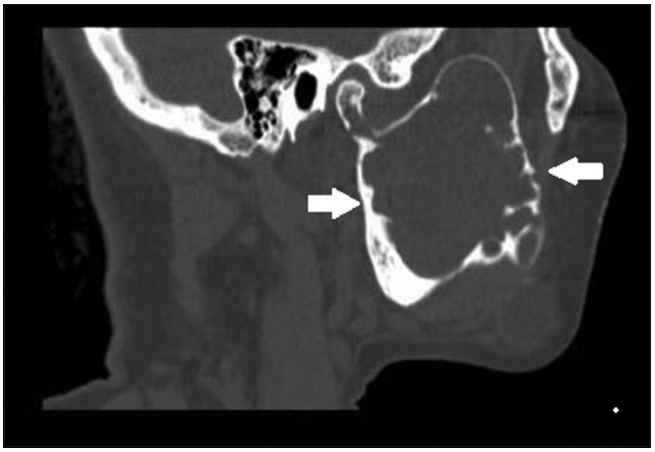

Figure 2: Computed tomography saggital view showing multiple locules on the right side of the mandible with expansion and thinning of both cortical plates and perforation in the region of the anterior border of the ramus

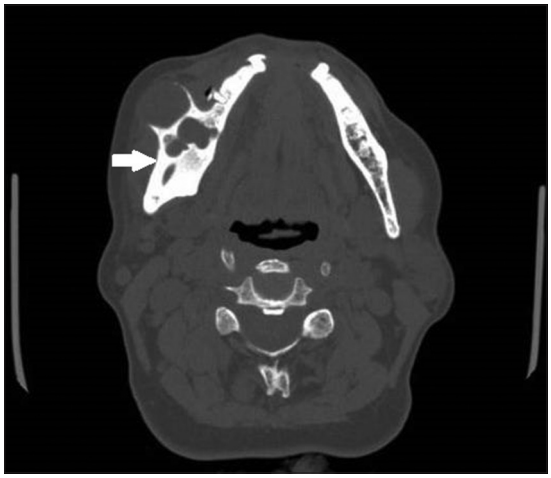

Figure 3: Computed tomography axial view showing multiple locules on the right side of the mandible with expansion and thinning of both buccal and lingual cortical plates

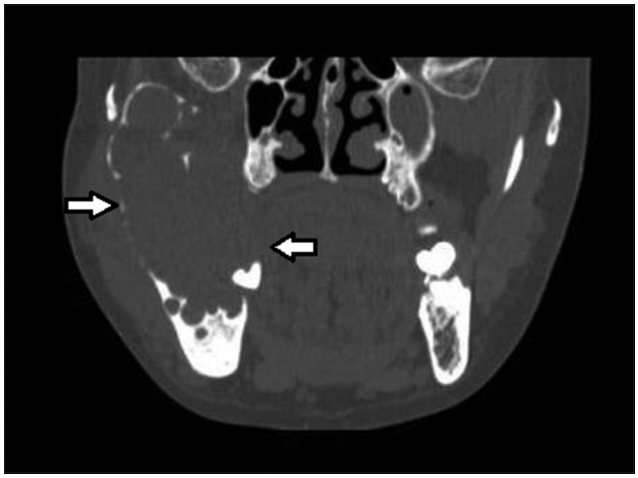

Figure 4: Computed tomography coronal view showing multiple locules on the right side of the mandible with expansion and thinning of both buccal and lingual cortical plates

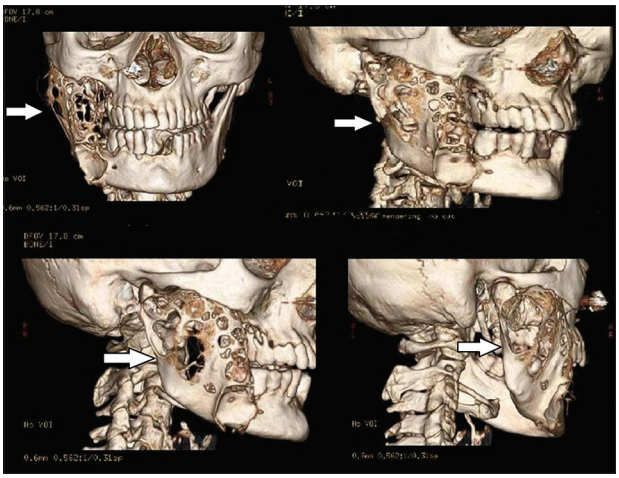

Figure 5: 3D CT view showing multiple locules over the right side of the mandible causing expansion and thinning of the cortical plates extending from body of the mandible to involve the entire ramus

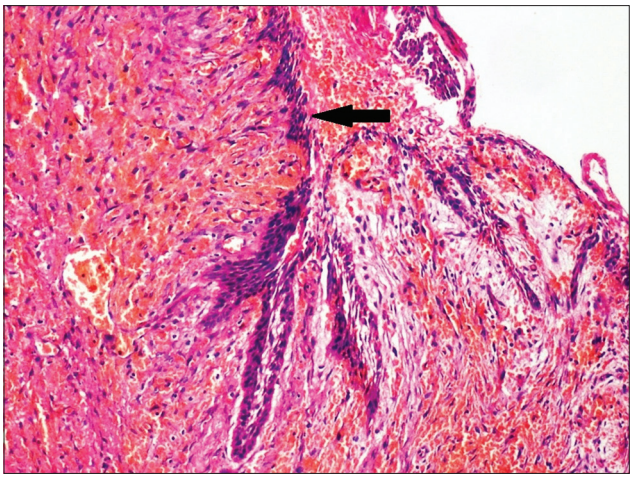

Figure 6: Histopathological section showing the epithelium lined by ameloblast-like cells invading into the connective tissue (cystic capsule), suggestive of a mural variant of ameloblastoma (H and E, Stain ×10)

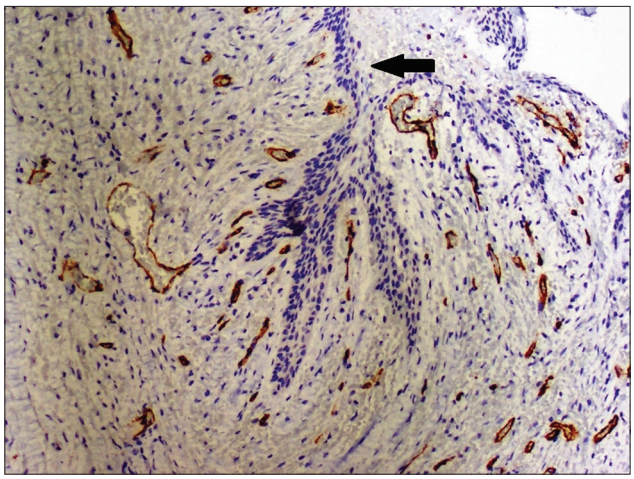

Figure 7: Immunohistochemistry picture showing CD34 expression in the mural variant of ameloblastoma (×10)

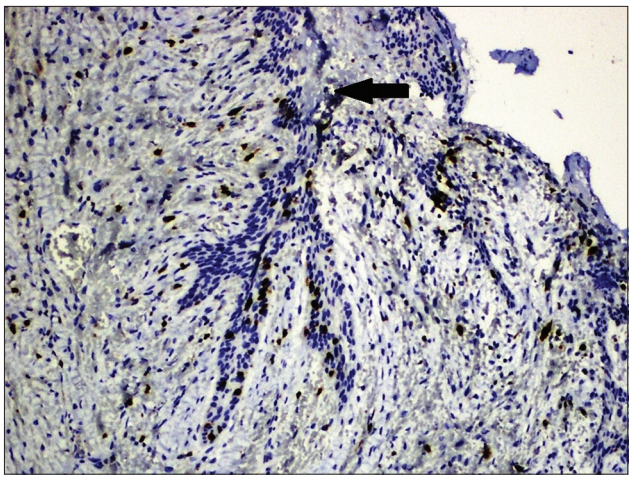

Figure 8: Immunohistochemistry picture showing Ki-67 nuclear staining in the mural variant of ameloblastoma (×10)

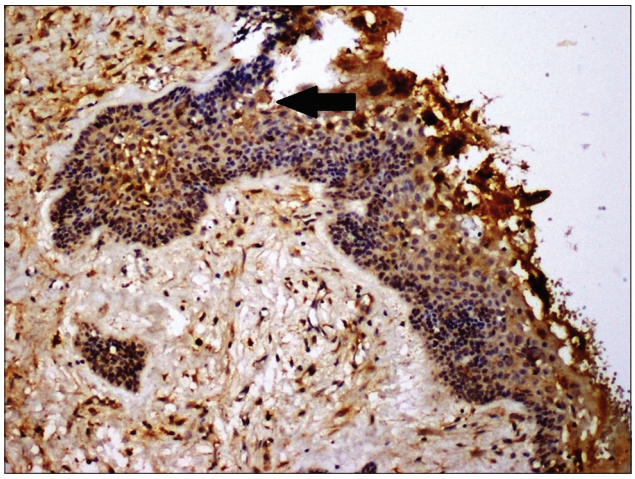

Figure 9: Immunohistochemistry picture showing MMP-2 expression in the mural variant of ameloblastoma (×10)

or classical', intraosseous, solid or multicystic ameloblastoma; (2) The unicystic ameloblastoma; and (3) The peripheral ameloblastoma. The current concept of categorizing ameloblastoma into solid or multicystic (the classic intraosseous ameloblastoma), unicystic, and peripheral subtypes appears to be gaining increasing support, because this classification criteria has a direct bearing on the biological behavior of these variants.[5] It is no longer appropriate to discuss ameloblastomas generally, as if all cases are essentially similar. It represents approximately 1% of the oral tumors, and 80% of the ameloblastomas occur in the mandible, with the remaining 20% in the upper jaw. The ramus of the mandible is the site of predilection, but the lesion in our case report was located at the border, between the ramus and body of the affected mandible. In approximately 40% of the cases there is an associated unerupted tooth, often the mandibular third molar.[5,6]

Ameloblastomas may remain asymptomatic before a facial swelling develops. Ameloblastoma may present on conventional radiographs as a unilocular or multilocular

corticated radiolucency, resembling a cyst. The presence of the bony septae may result in a honeycomb appearance. Buccal and lingual expansion is more common in ameloblastoma than in keratocystic odontogenic tumors. The resorption of dental roots may or may not be a feature. The radiographic differential diagnosis include a variety of odontogenic cysts, keratocystic odontogenic tumors, odontogenic myxoma, as well as non-odontogenic tumors and cysts, such as, central giant cell granuloma and simple bone cyst. CT and MRI are employed to investigate a solid tumor, to determine the edges and consistency of the tumor, which may be helpful in establishing its extent.[7,8] This advantage of such advanced imaging modalities was exhibited by the CT finding in our case report, which demonstrated medial and lateral cortical expansion with perforation of the buccal and lingual cortical plates [Figures 2-5].

Although ameloblastoma typically appears as an aggressive, often asymptomatic and slow growing tumor, it sometimes causes symptoms such as swelling, dental malocclusion, pain, and paresthesia of the affected area. It spreads by forming pseudopodes in the marrow spaces without concomitant resorption of the trabecular bone. This make, the margins of the tumor indistinct on radiographs or during surgery and the tumor frequently recurs on account of inadequate surgical removal. The appearance of septae on the radiograph usually represents differential resorption of the cortical plate by the tumor and not the actual separation of the tumor portions.[9,10] On account of its slow growth, recurrences of ameloblastoma generally present many years and even decades after the primary surgery. When treated inadequately, malignant transformation may occur. In most cases, an ameloblastoma has a characteristic, but not diagnostic radiographic appearance. The neoplasm usually appears radiologically as a unilocular radiolucent area or a multilocular radiolucent area, with a honeycomb appearance. The resorption of the adjacent tooth roots is not uncommon and in many cases an unerupted tooth, most often a mandibular third molar, is associated with the tumor.[11]

There are six different histopathological variants of ameloblastoma; desmoplastic, granular cell, basal cell, plexiform, follicular, and acanthomatous types. The histopathological type characterized by an epithelium lined with ameloblast-like cells invading into the connective tissue (cystic capsule), is suggestive of a mural variant of ameloblastoma, as in our case report.

The immunohistochemistry result in our case, which showed expression of CD34, MMPs, and Ki-67 is considered to be associated with interactions between epithelial cells and mesenchymal components in normal and neoplastic odontogenic tissues and these molecules might play a role in the regulation of tumor progression in ameloblastomas as well as regulation of developmental processes in tooth germs.[10,11]

Ameloblastoma has the tendency to recur, metastasize, and undergo malignant transformation. Recurrence seems to depend on several factors such as, the method of treatment of a primary lesion, extent of the lesion, and the site of origin.[12] Recurrence rates also vary for different procedures used to treat primary lesions, several authors have found a recurrence rate of 55 to 90% for all ameloblastomas treated conservatively (enucleation and curettage). However, the incidence of recurrence following radical resection is 5 to 15%.

Surgery is thus the treatment of choice for ameloblastomas.[13] Treatment options range from conservative surgery to radical procedures, which include radiotherapy, curettage, and enucleation. Surgery includes removal of at least 1 cm of normal bone beyond the tumor margins. Soft tissue borders at the time of resection may also be examined by frozen sections, to ensure that complete removal of the tumor has been achieved.[14]

Conclusion

Ameloblastoma is considered to be benign, but a locally invasive odontogenic tumor with a high rate of recurrence is usually diagnosed late because of its poor symptomatology and low prevalence. Nonetheless, the main success factor associated with treatment is the early diagnosis and categorization into subtypes hinged on the proper correlation of radiological and histopathological features. Resection with a wide safe margin is the treatment method of choice for solid/multicystic (mural) ameloblastomas, to minimize the incidence of recurrence.

References

1. Shteyer A, Lustmann J, Lewin-Epstein J. The mural ameloblastoma: A review of the literature. J Oral Surg 1978;36:866-72.
2. Josell SD, Reiskin AB, Gross BD. Dentigerous cyst with mural ameloblastoma. J Am Dent Assoc 1979;99:634-6.
3. Rapidis AD, Angelopoulos AP, Skouteris CA, Papanicolaou S. Mural (intracystic) ameloblastoma. Int J Oral Surg 1982;11:166-74.
4. Schofield ID. Intramural and mural ameloblastoma. J Can Dent Assoc 1981;47:792-4.
5. Reichart PA, Philipsen HP, Sonner S. Ameloblastoma: Biological profile of 3677 cases. Eur J Cancer B Oral Oncol 1995; 31:86-99.
6. Torres-Lagares D, Infante-Cossío P, Hernández-Guisado JM, Gutiérrez-Pérez JL. Mandibular ameloblastoma. A review of the literature and presentation of six cases. Med Oral Patol Oral Cir Bucal 2005;10:231-8.
7. Gümgüm S, Hoşgören B. Clinical and radiologic behaviour of ameloblastoma in 4 cases. J Can Dent Assoc 2005;71:481-4.
8. Mahadesh J, Rayapati DK, Maligi PM, Ramachandra P. Unicystic ameloblastoma with diverse mural proliferation-a hybrid lesion. Imaging Sci Dent 2011;41:29-33.
9. Chauhan DS, Guruprasad Y. Plexiform ameloblastoma of mandible. J Clin Imaging Sci 2011;1:61.
10. Guruprasad Y, Chauhan DS, Babu R. Unicystic ameloblastoma of maxilla. J Cranio Max Dis 2012;1:44-8.

11. Nadendla LK. Unusual imaging appearance of unicystic ameloblastoma. Contemp Clin Dent 2012;3:475-7.
12. Kowkabi M, Razavi SM, Khosravi N, Navabi AA. Odontogenic tumors in Iran, Isfahan: A study of 260 cases. Dent Res J 2012;9:725-9.
13. Nagalaxmi V, Sangmesh M, Maloth KN, Kodangal S, Chappidi V, Goyal S. Unicystic mural ameloblastoma: An unusual case report. Case Rep Dent 2013;2013:957418.
14. Nagata K, Shimizu K, Sato C, Morita H, Watanabe Y, Tagawa T. Mandibular ameloblastoma in an elderly patient: A case report. Case Rep Dent 2013;2013:145282.

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