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Ameloblastoma with dystrophic calcification: A case report with 3-dimensional cone-beam computed tomographic images of calcification

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ABSTRACT

This report presents a rare case of ameloblastoma with histopathologic and radiographic calcification, including 3-dimensional cone-beam computed tomographic (CBCT) images. A 22-year-old woman had hard swelling on the right mandible. Panoramic and CBCT images showed multilocular radiolucencies with internal calcification foci in the right mandible. Three-dimensional images clearly showed varying-sized radiopacities within the lesion from various angles. A histopathologic examination showed central squamous differentiation and more densely packed peripheral palisading ameloblastic cells. Many areas of keratin pearls and calcifications were also seen. Four previous reports have described 5 cases of ameloblastoma showing histopathologic calcification. This might be the first report to present the calcification of ameloblastoma on panoramic and CBCT images, especially on 3-dimensional images. ***(Imaging Sci Dent 2020; 50: 373-6)***

KEY WORDS: Cone-Beam Computed Tomography; Ameloblastoma; Calcification

Ameloblastomas are benign tumors of odontogenic epithelial origin, arising from odontogenic epithelium derived from remnants of the dental lamina and enamel organ, from the epithelial lining of an odontogenic cyst, or from basal cells of the oral mucosa.[1,2] Ameloblastoma is a neoplasm of the enamel organ type that has not undergone differentiation to the point of hard tissue formation.[1] Several microscopic subtypes of conventional ameloblastoma are recognized, but these patterns generally have little bearing on the behavior of the tumor. Large tumors often show a combination of microscopic patterns, including follicular, plexiform, acanthomatous, granular cell, desmoplastic, and basal cell types. Ameloblastomas were classified as conventional, unicystic, extraosseous/peripheral, and metastasizing ameloblastomas in the 2017 WHO classification.[3]

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The radiographic features of ameloblastoma are a unilocular radiolucent cavity with or without septa, multilocular radiolucent cavities with curved septa, and a honeycomb or soap bubble pattern of radiolucencies.[4]

Radiolucency with or without septa is the main radiographic feature of ameloblastoma, but we encountered a case of ameloblastoma with many small radiopaque foci in the radiolucent cavity on dental cone-beam computed tomography (CBCT). Pindborg and Weinmann first reported 2 cases of ameloblastoma with calcification in 1958.[5] Three additional cases of oral and maxillofacial ameloblastoma with calcification on histopathologic examination have been reported in the literature.[6-8]

This report aimed to present a rare case of ameloblastoma with histopathologic and radiographic calcification, along with 3-dimensional CBCT images.

Case Report

A 22-year-old woman visited the Department of Oral and Maxillofacial Surgery of Chonnam National University Dental Hospital in 2013. She had a painless hard swelling

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Fig. 1. Panoramic radiograph shows multilocular radiolucencies with faint dispersed radiopaque foci on the right mandible.

on the right mandible on visual examination and palpation. On intraoral examination, the enlarged right mandibular lesion was covered with normal-appearing mucosa and the involved right mandibular premolars and molars revealed a negative response to the mobility and percussion tests. On palpation, the intraoral right mandibular swelling was hard and slightly tender. She had noticed the right mandibular swelling 5 months before, and the swelling had gradually grown since then.

Panoramic (TrophyPan/Kodak 8000. Carestream Health Inc. Rochester, NY, USA) and CBCT examinations (MerCuray. Hitachi Medical Corp. Tokyo. Japan) were performed at the Department of Oral and Maxillofacial Radiology. Panoramic imaging showed varying-sized multilocular expansile radiolucencies, some perforation of the expansive buccal and lingual cortical bones including the mandibular inferior cortex, faint scattered radiopaque foci, loss of lamina dura, and external root resorption of the involved right mandibular premolars and molars (Fig. 1).

Dental CBCT images clearly showed many varying-sized calcifications within the lesion from various angles. Calcifications in the cavity lesion were clearly seen on 3-dimensional (3D) images at various angles (Figs. 2 and 3). The differential diagnosis included ameloblastoma and odontogenic keratocyst. Histopathologically, the lesion showed some central squamous differentiation and more densely packed peripheral palisading ameloblastic cells, with many areas of keratin pearls and calcifications (Fig. 4). The histopathologic diagnosis was acanthomatous ameloblastoma with dystrophic calcification. The patient underwent segmental mandibulectomy and reconstruction with a fibular osseous free bone graft on the lesion, and dental implant treatment finished in 2015.

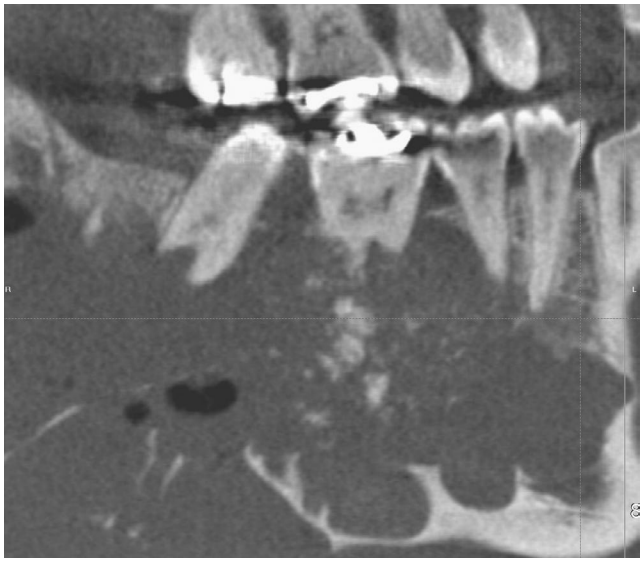

Fig. 2. Panoramic reconstruction cone-beam computed tomographic image shows varying-sized, scattered calcified foci in the lesion.

Discussion

Upon searching the picture archiving and communication system (PACS) of Chonnam National University Dental Hospital between November 2008 and August 2020 for ameloblastoma, this case of ameloblastoma with calcification was found among 82 cases of ameloblastoma. The small radiopacities indicating calcification could be found even on panoramic radiography in this case. The CBCT images showed many scattered definite radiopaque foci of calcification. Some calcifications measured approximately 0.3 mm and were found in clusters on the histopathologic examination (Fig. 4).

A PubMed search yielded 4 reports describing a total of

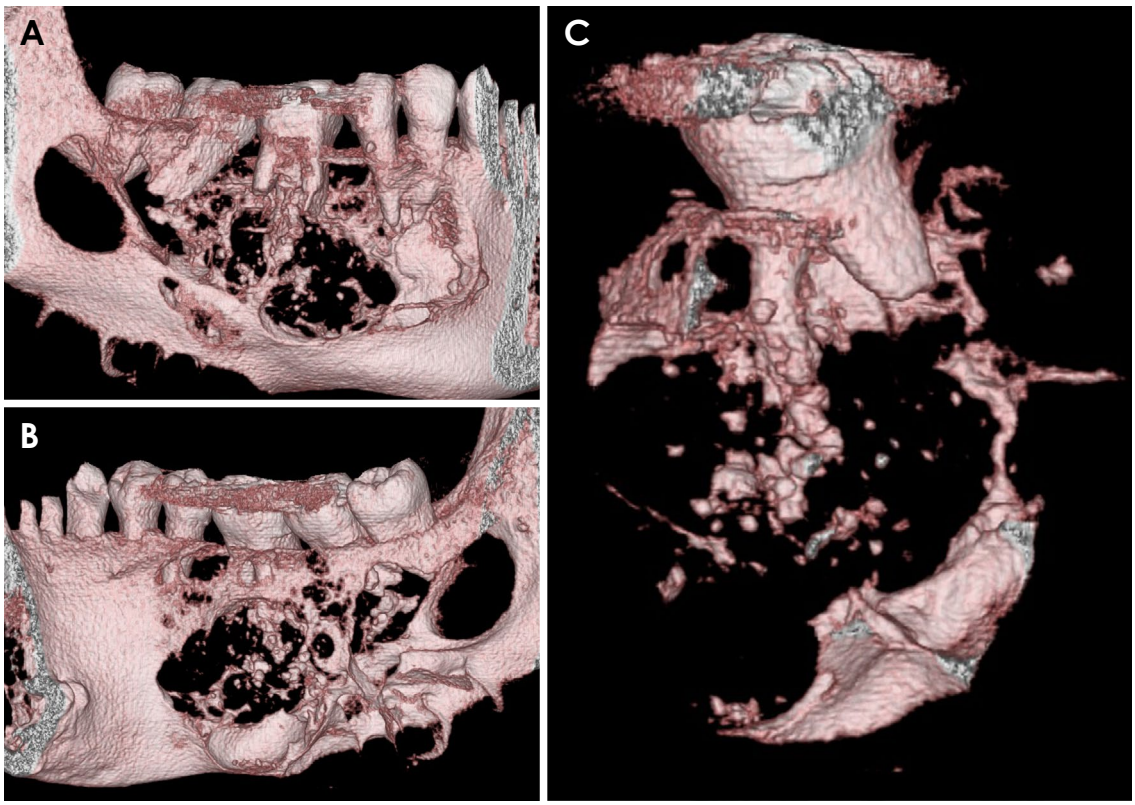

Fig. 3. A. Buccal view of a 3-dimensional (3D) image shows numerous discrete, varying-sized calcifications dispersed in a multilocular cavity on the right mandible. B. Lingual view of the 3D image. C. A blocked 3D image through the right mandibular first molar shows varying-sized calcifications in the cavity with resorbed buccal and lingual cortical borders.

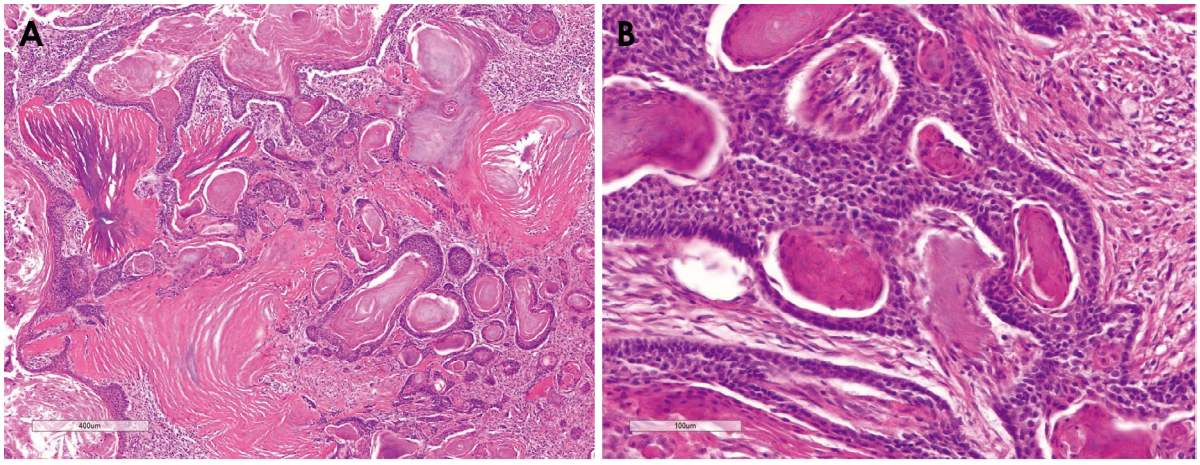

Fig. 4. A. Histopathologic exam shows numerous dystrophic calcifications and keratin pearls (H&E stain, original magnification ×40). B. Acanthomatous ameloblastoma showing squamous differentiation of epithelial nest (H&E stain, original magnification ×200).

5 cases of ameloblastoma with calcification in the oral and maxillofacial region.[5-8] Histopathologically, 5 cases (including the present case) were acanthomatous, while 1 case was follicular ameloblastoma.

Although calcification in ameloblastoma was first reported in 1958,[5] it is an uncommon feature. Some studies have suggested an explanation for the relationship between ameloblastoma and calcification.[9,10] Ameloblastoma and craniopharyngioma share a common embryonic origin from the oral ectoderm. Craniopharyngioma tends to be

cystic and to form ghost cells and calcifications.[9] Half of craniopharyngioma cases correspond histologically to calcifying odontogenic cyst (COC), 24% to ameloblastoma, and 8% show features of both calcifying odontogenic cyst and ameloblastoma.[10] These developmental and histologic relationships among craniopharyngioma, COC, and ameloblastoma might explain why this case of ameloblastoma was accompanied by calcification.

Previous reports regarding calcification of ameloblastoma[5-8] mainly presented histopathologic findings of calcification. However, in this case, calcification could be detected on panoramic radiography as faint radiopaque foci dispersed in the cavity, and CBCT images clearly showed somewhat round, varying-sized calcified foci. The demonstration of clear calcification on 3D images might be the most striking feature of this case. This was made possible by technical advancements in dental CBCT, which is capable of depicting submillimeter structures and the clustering of adjacent considerable-sized calcifications, as in this case.

In summary, the calcifications of ameloblastoma on panoramic and CBCT images, especially on 3D images, matched the histopathologic finding of calcification. This is the most striking feature of the present report, and future reports of ameloblastoma with calcification may be able to demonstrate finer images of calcifications in most cases thanks to the widespread availability of dental CBCT.

Conflicts of Interest: None

References

1. Masthan KM, Anitha N, Krupaa J, Manikkam S. Ameloblastoma. J Pharm Bioallied Sci 2015; 7: 167-70.
2. Neville BW, Damn DD, Allen CM, Chi AC. Oral and maxillofacial pathology. 4th ed. St. Louis: Elsevier; 2016. p. 653-62.
3. Evangelou Z, Zarachi A, Dumollard JM, Peoc'h M, Komnos I, Kastanioudakis I, et al. Maxillary ameloblastoma: a review with clinical, histological and prognostic data of a rare tumor. In Vivo 2020; 34: 2249-58.
4. Mallya SM, Lam EW. White and Pharoa's oral radiology: principles and interpretation. 8th ed. St. Louis: Elsevier; 2019. p. 413-9.
5. Pindborg JJ, Weinmann JP. Squamous cell metaplasia with calcification in ameloblastomas. Acta Pathol Microbiol Scand 1958; 44: 247-52.
6. Siar CH, Ng KH. Calcifying and keratinizing ameloblastoma of the maxilla. J Laryngol Otol 1991; 105: 971-2.
7. Kim JD, Choi JY. Ameloblastoma of the mandible and maxilla: CT findings. J Korean Radiol Soc 1995; 33: 351-6.
8. Lascane NA, Sedassari BT, Alves Fde A, Gallottini MH, de Sousa SC. Peripheral ameloblastoma with dystrophic calcification: an unusual feature in non-calcifying odontogenic tumors. Braz Dent J 2014; 25: 253-6.
9. Bernstein ML, Buchino JJ. The histologic similarity between craniopharyngioma and odontogenic lesions: a reappraisal. Oral Surg Oral Med Oral Pathol 1983; 56: 502-11.
10. Paulus W, Stöckel C, Krauss J, Sörensen N, Roggendorf W. Odontogenic classification of craniopharyngiomas: a clinicopathological study of 54 cases. Histopathology 1997; 30: 172-6.