Response to letter to the editors "Re: Byung-Do Lee, Wan Lee, Kyung-Hwan Kwon, Moon-Ki Choi, Eun-Joo Choi and Jung-Hoon Yoon. Glandular odontogenic cyst mimicking ameloblastoma in a 78-year-old female: a case report. Imaging Science in Dentistry 2014; 44(3): 249-52."

Byung-Do Lee^{1,*}

¹Department of Oral and Maxillofacial Radiology and Wonkwang Dental Research Institute, College of Dentistry, Wonkwang University, Iksan, Korea

Thank you for your attention to our study and wonderful questions. Our reply is as below.

Question) The title of the article states that the case is about a glandular odontogenic cyst (GOC) mimicking ameloblastoma. The history (slow expansion, no symptoms) and the radiographic appearance of the lesion (lobulated, well-defined margin, erosion and perforation of the lingual cortex, root resorption of the adjacent teeth) may suggest ameloblastoma, but this set of diagnostic factors is not specific to this particular type of lesion. Therefore, we believe this really isn't a case of "mimicry". Would it not have been more appropriate to define this lesion as mimicry in a case where there was another feature specific to ameloblastoma?

Answer) It has been reported that GOC and ameloblastoma show similar radiological features such as a lobulated margin, perforation of the cortex, and root resorption of the adjacent teeth , although these are not specific features. Ameloblastoma is the one of most common benign tumors of the jaw, and extensive root resorption is a characteristic feature of ameloblastoma. We made a tentative diagnosis based primarily on the prevalence of this benign tumor. But histopathological findings revealed that this case was a GOC. Our title is "Glandular odontogenic cyst mimicking ameloblastoma" because ameloblastoma is the representative benign tumor of the posterior mandible.

Received March 24, 2015; Revised April 3, 2015; Accepted April 9, 2015 *Correspondence to : Prof. Byung-Do Lee

Department of Oral and Maxillofacial Radiology, College of Dentistry, Wonkwang University, #460 Iksan-daero, Iksan, Jeonbuk 570-749, Korea Tel) 82-63-859-2912, Fax) 82-63-857-4002, E-mail) eebydo@wonkwang.ac.kr Question) Another point to mention is that the authors refer to the radiographic appearance of multiple foci and the cortical perforation as being helpful in distinguishing the GOC from ameloblastoma. But this radiographic feature also occurs in ameloblastoma, so with all due respect, we do not understand how it could be helpful in distinguishing between the two types of jaw lesions radiographically.

Answer) GOC and ameloblastoma may show common radiographic features of cortical perforation. The GOC has two clinically important attributes: a high recurrence rate and an aggressive growth potential.¹

Our lesion showed multiple cortical perforation in the radiolucent lesion (26.0 mm \times 11.3 mm), which indicated some aggressiveness. Our view of the above distinction was based on this description from a previous study:

Radiological features which may be helpful in distinguishing multilocular GOC's from ameloblastomas include irregular loculations and a partially sclerotic border with foci of perforation.²

Question) As we read the article, we all agreed that it was not very clear when the histopathological examination was performed. It was either done on a biopsy taken before or during the operation or on the gross specimen, but it wasn't clear which. It is perfectly normal for a provisional diagnosis to be false, but we believe it would have been better if a biopsy were performed before the operation. If a biopsy was performed before surgery, can you clarify to us what biopsy method was used presurgically: a fine-needle aspiration biopsy or an incisional biopsy? Or was a frozen-section examination performed during the operation to decide if a more conservative or a more

Copyright © 2015 by Korean Academy of Oral and Maxillofacial Radiology

which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited. Imaging Science in Dentistry • pISSN 2233-7822 eISSN 2233-7830

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (http://creativecommons.org/licenses/by-nc/3.0)

radical surgical treatment was appropriate? According to a study by Aronovich and Kim, ~90% of benign oral and maxillofacial lesions are correctly diagnosed and treated during surgery by frozen section histopathology, compared to the definitive histopathology done after the operation.³ Lastly, was the gross specimen sent for histopathological examination to confirm the diagnosis?

Some researchers advocate that an ameloblastoma-no matter the type-should be treated radically to prevent recurrences.⁴ As for this case, it is not clear if the surgery was performed according to the ameloblastoma diagnosis. If that's the case, could a cyst not be distinguished from an ameloblastoma when the lesion was opened up? Did the authors consider the different treatments of these two lesions during the operation?

Answer) Another question was about whether we performed preoperative incisional biopsy, fine-needle aspiration biopsy, or frozen-section examination. We decided not to do a preoperative biopsy because the characteristics of the mass were suggestive of a benign tumor with locally aggressive behavior, which we mentioned in our report as "cortical perforation and erosion were also observed, suggesting its aggressiveness; however, the mild expansion of the lingual cortex represents the benign nature of the lesion."

We considered various treatment options such as enucleation, enucleation with peripheral ostectomy, and radical surgery such as segmental mandibulectomy. Most studies support radical surgery as a treatment for ameloblastoma to minimize the risk of recurrence of the tumor.³ However, considering the age of the patient, radical surgery such as segmental mandibulectomy was not advisable in this case because this operation takes a long time and should be accompanied by soft tissue and/or hard tissue reconstruction in almost all cases.

Several clinical studies have favored an enucleation operation with peripheral ostectomy as treatment for ameloblastoma, preserving mandibular continuity.⁴ Enucleation with peripheral ostectomy is thought to be an acceptable treatment option for a locally aggressive benign tumor such as ameloblastoma. We decided not to do preoperative biopsy because the histopathologic result could not change the treatment plan. After the operation, the gross specimen was sent for histopathologic examination to confirm the diagnosis, and the final diagnosis was glandular odontogenic cyst.

References

- 1. Macdonald-Jankowski DS. Glandular odontogenic cyst: systematic review. Dentomaxillofac Radiol 2010; 39: 127-39.
- Noffke C, Raubenheimer EJ. The glandular odontogenic cyst: clinical and radiological features; review of the literature and report of nine cases. Dentomaxillofac Radiol 2002; 31: 333-8.
- Antonoglou GN, Sándor GK. Recurrence rates of intraosseous ameloblastomas of the jaws: a systematic review of conservative versus aggressive treatment approaches and meta-analysis of non-randomized studies. J Craniomaxillofac Surg 2015; 43: 149-57.
- Shi S, Liu Y, Shan Y, Fu T, Zhao S. Enucleation combined with peripheral ostectomy: its role in the management of large cystic ameloblastomas of the mandible. J Craniomaxillofac Surg 2014; 42: 1659-63.