Response to the Letter Entitled “Archegonous Cystic Odontoma Is Not Necessarily Primordial” by Ide et al.

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We welcome the commentary written by Ide et al. [1] on our paper published in the journal [2]. Cases like ours and the one presented by our colleagues from Japan bring forth thoughts and individual opinions not only on unusual odontogenic lesions but also uncommon microscopic observations made in otherwise conventional odontogenic pathologic processes. The example included and briefly discussed by Ide et al. [1] features histopathologic findings that are uncommon in the wall of dentigerous cysts. Although our example was not associated with an impacted tooth [2], we agree with Ide et al. [1] that their case includes most of the histopathologic findings seen in our case. However, in the absence of an impacted tooth and the presence of an epithelial cystic lining that could have been interpreted as ameloblastomatous, the classification of the lesion was initially, in the year 2001, problematic. This problem in classification was evident 14 years later when our case was presented at the Seminar of the Annual Meeting of the American Academy of Oral and Maxillofacial Pathology [3]. A wide variety of interpretations were received by anonymous oral pathologists and are summarized in Table 1. The senior author of our paper with the use of immunohistochemistry for calretinin supported the argument against the diagnosis of unicystic ameloblastoma, which may have had treatment consequences for the patient.

We gave the readers of our paper two possible diagnostic choices. Interestingly, Dr. Ide et al. provided four additional possible diagnostic choices (Table 2). Dr. Ide et al., however, should allow us to discard the incongruous term “odontogenic cyst of undetermined origin with hamartomatous components” (it is either odontogenic or not). Perhaps a more appropriate term would be “odontogenic cyst not otherwise specified with hamartomatous components”.

The letter by Dr. Ide et al. [1] is entitled “Archegonous cystic odontoma is not necessarily primordial”. We agree with their opinion but allow us to state that we never made that argument. Thus, the statement in the letter that reads “... which convinced the authors that it was a true POC associated with an aborted tooth (cystic aborted odontoma)” is not valid. Similarly, their following statement “the present case would seem to provide ... part of DC” is irrelevant.

We would like to make some comments on the literature cited by Dr. Ide et al. referenced in the paragraph containing the excerpt “analogous observations ... under various bizarre terminologies” [1]. The case of AF Gardner [4] from 1955 appears to be an example of a dentigerous cyst associated with the mandibular second premolar. The reference of the presence of “a tooth germ” made in the histopathology section of the report may represent an odontoma. We were unable to find the Japanese abstract by Eto et al. [5] in conventionally used search engines. Therefore, its validity could not be confirmed by us. The example by Alampally et al. [6] features only focal reverse polarization that may be interpreted as unicystic ameloblastoma. The numbers of figures in this publication is limited and are of suboptimal quality to justify the argument of the authors. The case by Rathod et al. [7], published in an open access peer reviewed journal may represent a dentigerous cyst with mucous metaplasia/prosoplasia in association with an odontoma. We agree with

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Table 1 The interpretations regarding our case of primordial odontogenic cyst (POC) or archegonous cystic odontoma (ACO) provided by anonymous oral pathologists during the Seminar of the Annual Meeting of the American Academy of Oral and Maxillofacial Pathology in 2015

	Diagnoses of attendees
1	Ameloblastoma ($\times 9$), cystic, with dentinoid, dentinoameloblastoma, plexiform
2	Ameloblastic fibro-dentinoma ($\times 2$)
3	Odontogenic hamartoma, with dentigerous cyst or cystic ($\times 2$)
4	Ameloblastoma, conventional (not cystic)
5	Possible early odontoma, ameloblastic fibro-odontoma with cystic ameloblastomatous features
6	Ameloblastic fibro-odontoma, cystic
7	Emerging ameloblastic fibrosarcoma, considered teratoma
8	Paradental cyst

Table 2 Alternative diagnoses provided by Ide et al. [1] regarding archegonous cystic odontoma (ACO)

1	Complex odontogenic cyst with mural hamartoma
2	Cystic odontogenic hamartoma
3	Cystic hamartoma with rudimentary odontogenesis or with immature odontogenic tissue
4	Odontogenic cyst of undetermined origin with hamartomatous components

Ide et al. [1] that the example published by Costa et al. [8] should be accepted as a cystic odontoma.

In their letter, Ide et al. [1] attempted to categorize different types of rudimentary odontogenesis. This theoretical approach can be used for better naming and subclassification of appropriate lesions. For the sake of discussion of such associations, we present an additional case of what we would consider a cystic odontoma or a dentigerous cyst with arrested tooth development. It involved an 11-year-old Caucasian male who presented with an asymptomatic, expansile radiolucent cystic lesion developing in the area of the third

mandibular molar (tooth #32) and expanding into the mandibular ramus (Fig. 1a). Microscopically, the submitted specimen was partially surfaced by generally thin, non-neoplastic and non-keratinizing, stratified squamous epithelium featuring neutrophilic and lymphocytic exocytosis (Fig. 1b, d). The cystic wall was composed by fibrocollagenous and fibromyxoid connective tissue exhibiting mild chronic inflammation (Fig. 1c, d). An odontoma characterized by dentin, cementum, an empty space apparently occupied by enamel lost during decalcification, and remnants of enamel matrix were observed (Fig. 2a–d). Adjacent to the hard

Fig. 1 **a** Panoramic radiograph showing a well-demarcated radiolucency developing in the area of tooth #32 and expanding into the mandibular ramus. The white arrows highlight the extent of the lesion. **b–d** Microscopic characteristics of the specimen depicting a cystic lumen surfaced by non-keratinizing stratified squamous epithelium of variable thickness featuring neutrophilic and lymphocytic exocytosis. The cystic wall consisted of fibrocollagenous and fibromyxoid connective tissue with chronic inflammation (H&E, original magnification **b** $\times 40$, **c** $\times 180$, **d** $\times 360$)

Fig. 2 **a, b** Hard tissue structures characterized by dentin, cementum and remnants of enamel matrix. **c, d** Interconnecting cords of odontogenic epithelium enveloping dentin. **e, f** Round and ovoid

calcifications and cords and nests of odontogenic epithelium embedded in the fibrous connective tissue (H&E, original magnification **a** $\times 14$, **b** $\times 110$, **c** $\times 150$, **d** $\times 180$, **e** $\times 110$, **f** $\times 320$)

structures and in the fibrous connective tissue there were multiple cords and nests of odontogenic epithelium as well as round and ovoid calcifications (Fig. 2c–f).

In conclusion, the observed histopathologic features in all cases included in Dr. Ide's et al. commentary and in our response, regardless if they represent temporal manifestations in cystic odontomas, unusual odontogenic cysts, or possible tumors with mural "hamartomatous" components should entice researchers to investigate the molecular events that drive dental abnormalities. Again, we would like to express our sincere appreciation to Dr. Ide and his team for sharing their thoughts and experience.

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