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Clear cell-rich odontogenic tumor of the mandible in a 4-year-old child: A report of a diagnostically challenging case

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Odontogenic tumors;
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Clear cell odontogenic carcinoma (CCOC), initially called *clear cell odontogenic tumor*,[1] is a typical example of a clear cell-rich odontogenic tumor. CCOC is currently classified as a malignant odontogenic tumor.[2] Here, we reported a benign clear cell-rich odontogenic tumor that was histologically similar to CCOC.

A 4-year-old boy presented with a radiolucency in the mandible that was discovered incidentally on routine dental radiography. A panoramic radiograph showed a well-defined expansile radiolucent lesion in the right anterior mandible (Fig. 1A). Computed tomography revealed a 3.1-cm heterogeneously enhancing mass (Fig. 1B). Surgical excision was performed. Gross examination showed a well-encapsulated mass (Fig. 1C), and a fibrous capsule was identified by histologic examination (Fig. 1D). The mass was composed of nests and sheets consisting mainly of clear cells (Fig. 1E). Occasional tumor cells had slightly eosinophilic cytoplasm or a basaloid appearance. Some nests showed peripheral palisading of clear cells with reverse polarity, reminiscent of ameloblastoma (Fig. 1F). Cystic degeneration of solid nests was occasionally seen (Fig. 1G). Tumor cells showed no cytologic atypia. Clear cells were positive for periodic acid-Schiff but negative after diastase treatment, indicating the presence of cytoplasmic glycogen. Tumor cells were positive for cytokeratin 7 and p63. Whole-exome sequencing and RNA sequencing were performed by using the SureSelect Human All Exon V8 (Agilent Technologies, Santa Clara, CA, USA) and the TruSeq RNA Exome (Illumina, San Diego, CA, USA), respectively, as described previously;[3,4] genetic alterations commonly found in ameloblastoma (mitogen-activated protein kinase and Hedgehog pathway mutations) and CCOC (*EWSR1* rearrangements) were not detected. A definite diagnosis was not made. The patient was followed up for 8 years after surgery with no evidence of recurrence.

A varying number of clear cells can be observed in several types of odontogenic tumors, including calcifying epithelial odontogenic tumor, dentinogenic ghost cell tumor, CCOC, and so-called odontogenic carcinoma with dentinoid.[2,3] Among these tumors, CCOC is histologically characterized by a predominant population of clear cells. In addition, an entity called "clear cell ameloblastoma" has rarely been reported as a biphasic tumor consisting of an ameloblastomatous component mixed with an extensive clear cell component.[5] Therefore, CCOC or ameloblastoma was considered as a differential diagnosis for the present case. The morphologic features of tumor cells and their growth pattern were almost similar to those of CCOC; however, it was difficult to make a diagnosis of malignancy considering the encapsulated nature of the tumor. In some

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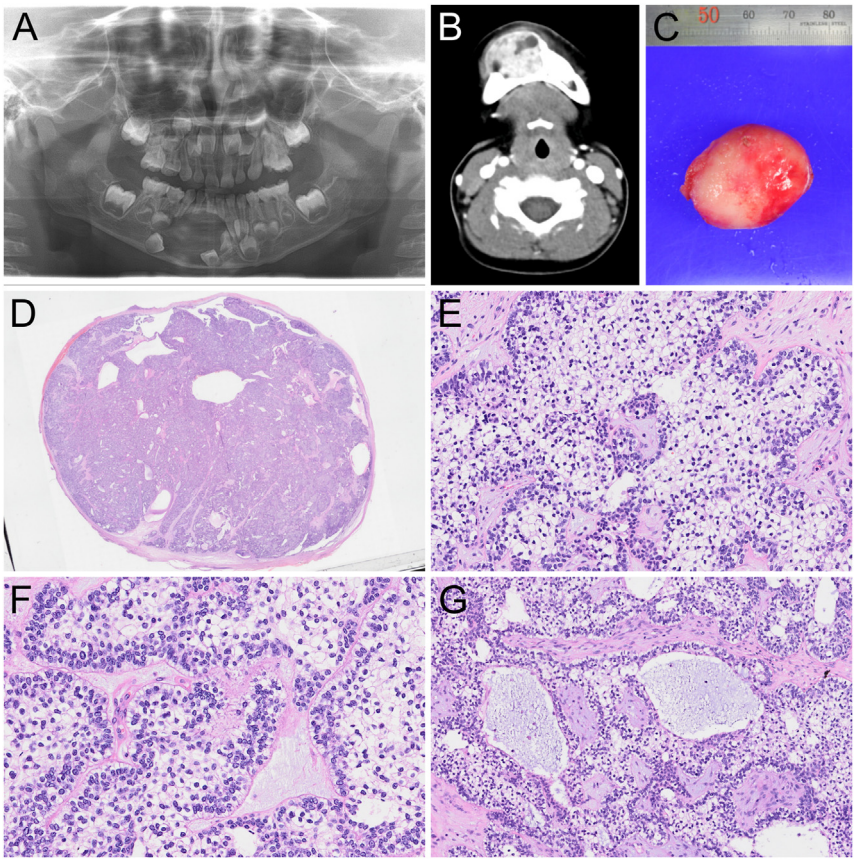

Figure 1 Radiological (A and B), macroscopic (C), and microscopic (D–G) findings of a clear cell-rich odontogenic tumor of the mandible. (A) A panoramic radiograph shows a well-defined radiolucent lesion in the right anterior mandible. (B) A computed tomography image reveals a heterogeneously enhancing expansile mass in the right anterior mandible. (C and D) A well-encapsulated tumor is identified by macroscopic (C) and microscopic (D) examination. (E) Tumor islands consist predominantly of clear cells. (F) The peripheral cells of tumor islands show reverse polarity. (G) Cystic degeneration of tumor islands is seen.

tumor islands, the peripheral cells showed ameloblastomatous features; however, the central epithelial cells had a morphology different from the stellate reticulum seen in the follicular subtype of ameloblastoma. Moreover, the results of the molecular analyses did not support a diagnosis of either CCOC or ameloblastoma.

Although not reaching a definitive diagnosis, this study suggests the potential limitations in the differential diagnosis of clear cell-rich odontogenic tumors when based on the current WHO classification.[2] It can be speculated that the present case may represent a benign counterpart/precursor of CCOC or a separate entity that is morphologically similar to but molecularly distinct from CCOC or ameloblastoma.

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

Acknowledgments

None.

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