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A repeatedly recurrent desmoplastic ameloblastoma after removal and allobone graft: Radiographic features compared with histological changes

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ABSTRACT

A 40-year-old man suffered from a repeatedly recurrent desmoplastic ameloblastoma in the right maxillary anterior and premolar regions. During the first visit, the patient was provisionally histopathologically diagnosed with a developmental cyst, and it was confirmed to be unicystic ameloblastoma and resected. Four years later, the lesion recurred, and was diagnosed as a desmoplastic type of ameloblastoma and removed again. Then, 5 years after the second surgery, the lesion recurred again, and was diagnosed as a type containing a follicular pattern, recurrent ameloblastoma. A panoramic radiograph showed a multilocular and mixed radiolucent/radiopaque expansile lesion at the first visit, a unilocular cystic lesion confined to the premolar area at the second visit, and a small soap bubble appearance in the molar area in the final visit. Cone-beam computed tomographic images of the final recurrence of the tumor revealed multiple small cyst-like structures in the right maxillary anterior and posterior regions. (***Imaging Sci Dent 2013; 43: 201-7***)

KEY WORDS: Ameloblastoma; Recurrence; Cone-Beam Computed Tomography; Radiography, Panoramic

Ameloblastoma is an uncommon, slow-growing, and locally invasive benign tumor that constitutes 11% to 18% of odontogenic tumors that arise in both jaws.[1-5] It has several different histological and clinico-radiologic types with a high rate of recurrence despite being classified as a benign tumor.[6] Most ameloblastomas occur between age 30 and 60 years, peaking around age 35 and show no sex predilection.[7,8] Approximately 80% of ameloblastomas occur in the mandible, usually in the vicinity of the molars and ramus.[3,7-11] In one report, symptomatic ameloblastomas accounting for 35% of 60 patients were discovered incidentally on radiographic studies, and ameloblastoma usually appears as unilocular or typical multilocular cystic radiolucencies.[2]

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Desmoplastic ameloblastoma is rare, accounting for approximately 4% to 13% of ameloblastomas.[12-16] The main histological types of ameloblastoma are the follicular and plexiform patterns. The desmoplastic ameloblastoma is one of the other less common histological variants.[17] The desmoplastic variant of ameloblastoma usually appears in the anterior and premolar regions as a mixed radiolucent and radiopaque lesion, sometimes resembling a benign fibro-osseous lesion.[12]

Ameloblastoma is a locally aggressive tumor that may cause recurrence and in rare cases, malignant transformation, with repeated postsurgical reccurences.[18,19] Recurrence after incomplete removal is a feature of ameloblastomas and it is, therefore, recommended that desmoplastic ameloblastomas be treated by complete surgical resection.[18,20] An autogenous bone graft for subsequent reconstruction after resection prevents the site from becoming affected by a recurrence of the neoplasm.[21,22]

This report presents a case of a patient who was initially diagnosed with a developmental cyst resembling unicystic

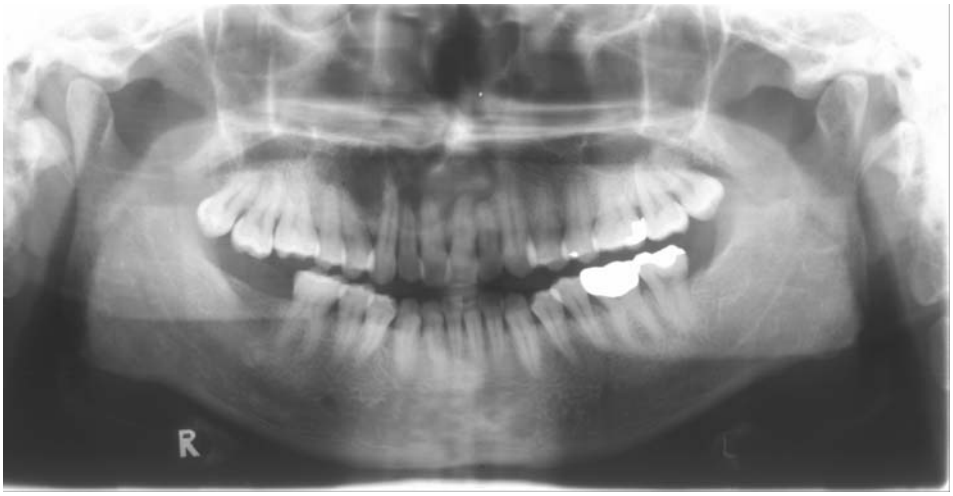

Fig. 1. Panoramic radiograph reveals a multilocular and mixed radiolucent/radiopaque expansile lesion extending from the right second premolar to the left canine of the maxilla, showing a soap bubble appearance in the middle third of the lesion, and an ill-defined radiopacity in the left third of the lesion.

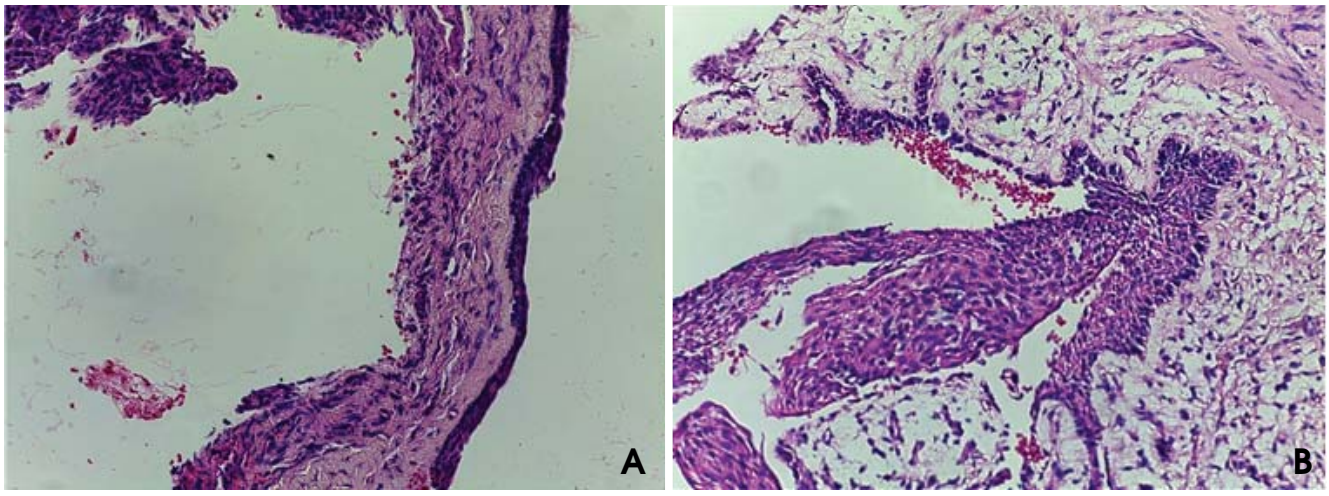

Fig. 2. A. The incisional biopsy shows a cystic structure lined by epithelium (H&E stain, 200×). B. The histopathologic examination after the first surgery on the tumor shows peripheral palisading of hyperchromatic epithelium and loose fibrous stroma (H&E stain, 200×).

ameloblastoma in the anterior and the right premolar maxillary regions and secondly with desmoplastic ameloblastoma at recurrence after enucleation and bone grafting, which repeatedly recurred. The lesion was successfully controlled with the third resection.

Case Report

A 40-year-old man visited the Chosun University Dental Hospital with a tender swelling on his anterior palatal area. The overlying mucosa appeared normal and all the teeth in the affected area were caries free and reacted positively to both thermal and electrical stimuli. His medical history was non-contributory.

A panoramic radiograph revealed a multilocular and mixed radiolucent/radiopaque expansile lesion extending from the right second premolar to the left canine of the maxilla, presenting a soap bubble appearance in the middle third of the lesion, and ill-defined radiopacity in the left third of the lesion (Fig. 1). The lesion showed an often curved and scalloped border extending to the middle portion of the roots of the upper anterior teeth. The lesion was diagnosed provisionally and radiographically as ameloblastoma with histological evidence of a developmental cyst showing a cystic structure lined by epithelium with incisional biopsy (Fig. 2A). After endodontic treatment of multiple upper anterior teeth, he underwent enucleation of the lesion and an allobone graft by an oral surgeon. Histopathologically, the enucleated mass showed unicystic ameloblastoma with peripheral palisading of hyperchromatic epithelium and loose fibrous stroma (Fig. 2B). The fistula formation process near the bone graft area was persistent.

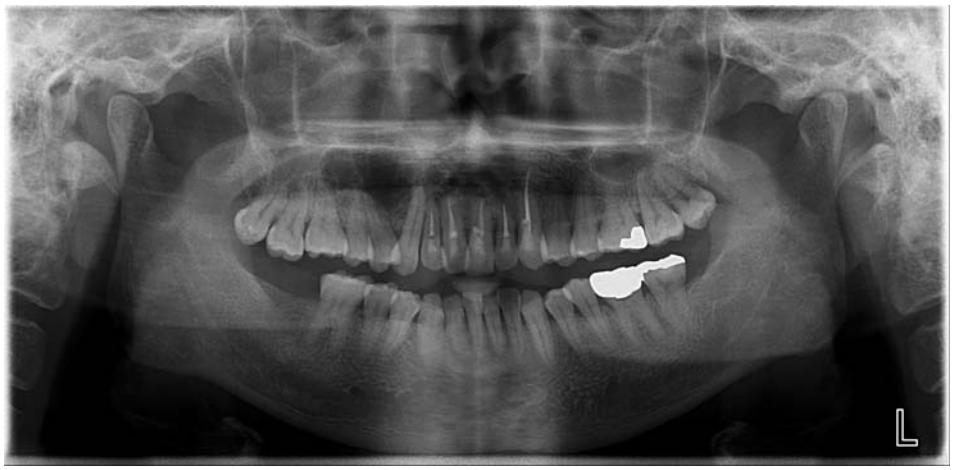

Fig. 3. A panoramic radiograph reveals a unilocular radiolucent cystic area confined to the premolar region of the right maxilla, 4 years after the first surgery.

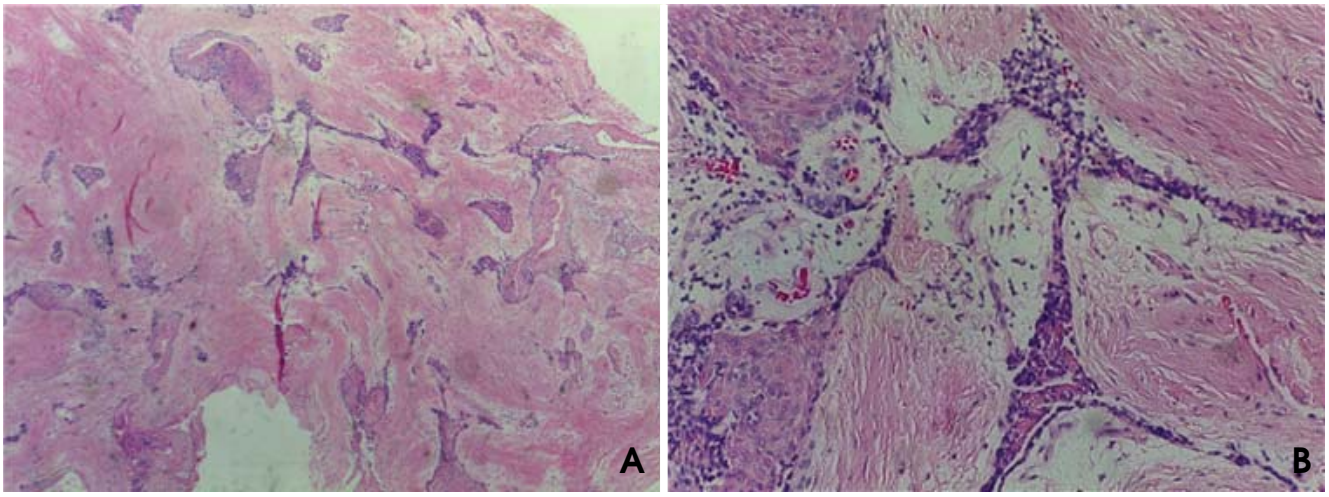

Fig. 4. A. Histopathologic examination 4 years after the first surgery shows the desmoplastic type of ameloblastoma (H&E stain, 40 ×). B. The histopathologic features contain small islands and thin cords of ameloblastic epithelium within a dense fibrous connective tissue stroma (H&E stain, 200 ×).

After being treated repeatedly with antibiotics and dressing for about 5 months, he finally underwent removal of the allobone.

Four years after the first surgery, a recurrent tumor was found in the same area. A panoramic radiograph showed a unilocular cystic radiolucency in the area corresponding to the right part of the primary lesion (Fig. 3), and re-enucleation of only the cystic lesion and Tutoplast grafting were performed. Histopathologically, the enucleated mass had changed from a developmental cyst to the desmoplastic type of ameloblastoma that contained small islands and thin cords of ameloblastic epithelium within a dense fibrous connective tissue stroma (Fig. 4).

The patient returned with the symptom of mild pain at the site of the operation 5 years after the second surgery. A panoramic radiograph revealed a soap bubble appearance in the periapical area of the right first and second molars (Fig. 5). On the axial cone-beam computed tomographic (CBCT) images, multiple small cyst-like structures were found adjacent to the maxillary sinus and in the anterior area, which were separated by normal bone. The lesion at the molar area was well demarcated from the maxillary antrum (Fig. 6). Sagittal CBCT images showed small loculations in the molar and anterior area, respectively, and they were separated by normal bone (Fig. 7). The tumor showed a relatively defined border, and it was adjacent to the maxillary sinus (Fig. 7A).

Finally, the tumor histology became a follicular pattern next to the desmoplastic type that contained small cystic tumor islands and thin cords of ameloblastic epithelium within connective tissue stroma (Fig. 8).

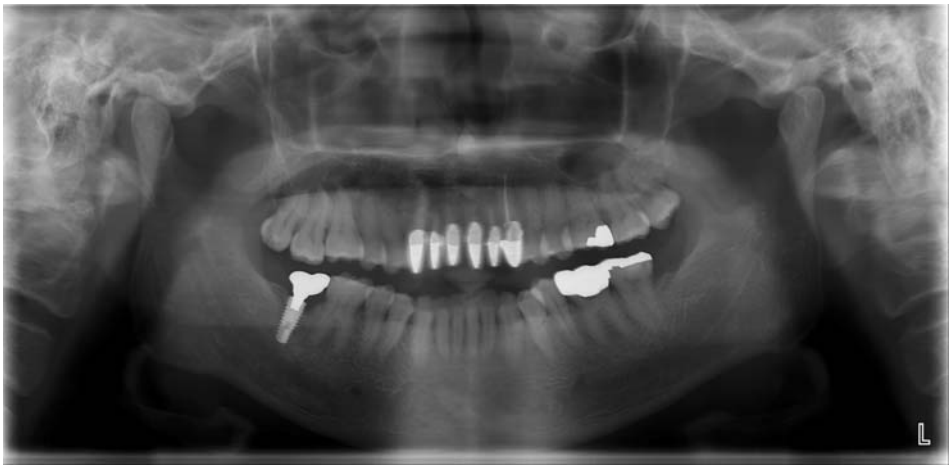

Fig. 5. Five years after the second surgery. The panoramic radiograph reveals a soap bubble appearance in the periapical area of the right first and second molars of the maxilla.

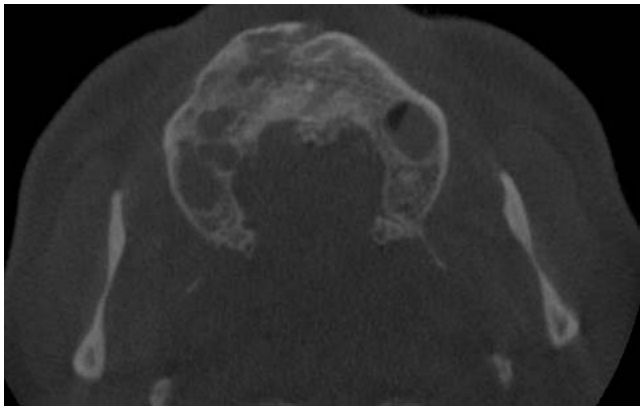

Fig. 6. An axial view of the CBCT image shows multiple small cyst-like structures in the right anterior and posterior regions. They are separated by normal bone. The lesion in the molar region is well demarcated, and separated from the maxillary antrum.

Discussion

In 1984, Eversole et al[17] first described the "desmoplastic ameloblastoma." Histologically, a desmoplastic ameloblastoma consists of an abundant and excessive stromal collagenization or desmoplasia in which irregularly shaped islands of odontogenic epithelium are scattered.[12] Waldron and El-Mofty[12] introduced the term "hybrid lesions" to describe the conditions in which desmoplastic ameloblastoma was located adjacent to follicular or plexiform ameloblastoma. Wakoh et al[23] presented a case of a patient demonstrating follicular-type ameloblastoma with desmoplasia, in whom radiological findings suggested the coexistence of a fibro-osseous lesion with a solitary cystic lesion and proposed it to be hybrid follicular/desmoplastic ameloblastoma.

Histopathologically, the original biopsy of our case showed features that were consistent with those of a developmental cyst suspected to be unicystic ameloblastoma. In the second histological study, the features of the enucleated mass had changed from a developmental cyst to a desmoplastic type of ameloblastoma. Finally, the tumor histology became a small cystic and follicular pattern next to the desmoplastic type of ameloblastoma. However, in the desmoplastic type of this case, a histologically fibro-osseous lesion-like pattern and hybrid pattern were not recognizable and radiologically, the relationship to the mixed lesion was also unclear.

Analyzing the radiographical features in order of histological changes, this tumor initially showed a multilocular radiolucent lesion with soap-bubble appearance in the right maxilla and concomitant radiopaque area in the left anterior region of the maxilla on the panoramic radiograph with the histopathologic features of a developmental cyst next to a unicystic ameloblastoma.

At the second visit, only panoramic radiography revealed a unilocular cystic radiolucency confined to the right maxillary premolar region; however, the biopsy result of the enucleated mass was desmoplastic ameloblastoma. Although Philipsen et al,[24] in their report of 2 cases of desmoplastic ameloblastoma, remarked that the presence of osteoplasia in desmoplastic ameloblastoma might correspond to the radiologic appearance of mixed radiolucency and radiopacity in some of the desmoplastic ameloblastomas, thereby presenting radiographic features of a fibro-osseous lesion. However, Sun et al[18] reported that desmoplastic ameloblastomas radiologically presented as radiolucent (44.4%) and mixed radiolucent/radiopaque (55.6%) in the cases they reviewed. Effiom and Odukoya[25] also reported that multilocular radiolucency accounted for 82.4% of cases in a desmoplastic series and the remaining

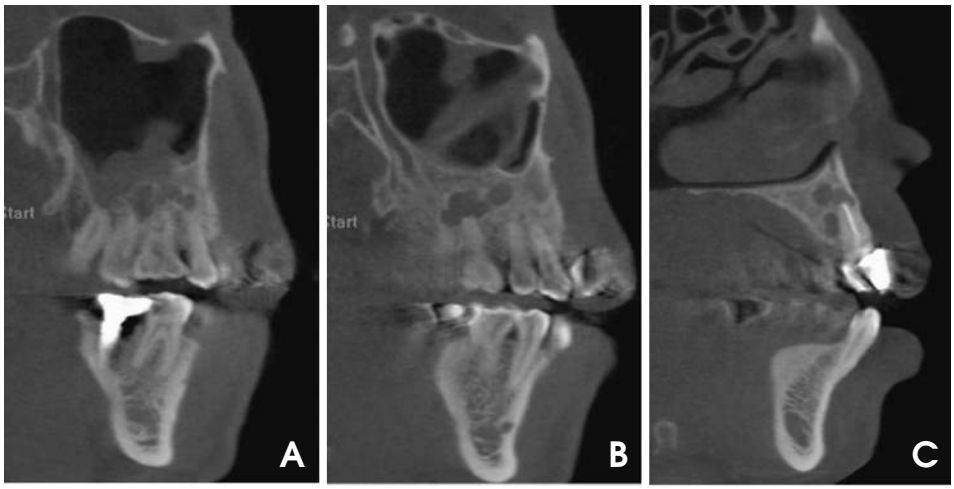

Fig. 7. Sagittal CT images (A; molar area B; premolar area C: anterior area) show small loculations in the molar and anterior region of the maxilla, respectively, separated by normal bone. The tumor shows a relatively defined border, and it is adjacent to the maxillary sinus.

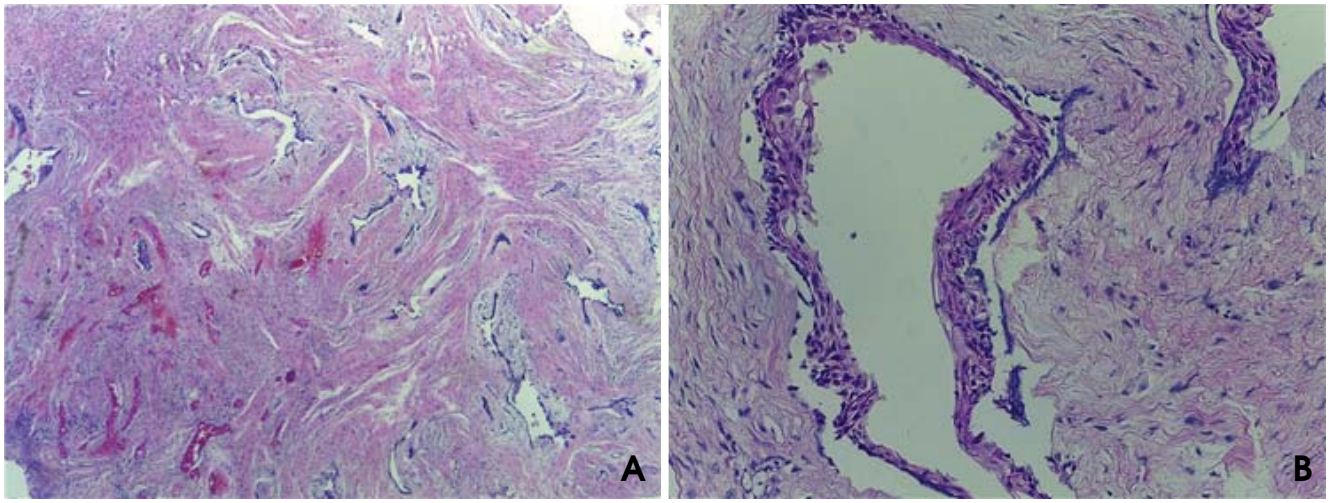

Fig. 8. A. The histopathologic examination 5 years after the second surgery reveals small cystic tumor islands and thin cords of ameloblastic epithelium within connective tissue stroma (H&E stain, 40×). B. A follicular pattern next to the desmoplastic type of ameloblastoma (H&E stain, 200×).

17.6% presented with a mixed radiolucent and radiopaque radiographic appearance, thereby mimicking a fibro-osseous lesion. Kawai et al[26] reported that desmoplastic ameloblastoma appeared either as a diffuse, poorly delineated, mottled radiolucent/radiopaque lesion similar to a benign fibro-osseous lesion or as a lesion with a honeycomb or soap-bubble appearance with indistinct borders. However, the relationship to the mixed lesion was not discovered in this case.

At the final visit, a panoramic radiograph revealed a small area of soap bubble appearance in the periapical area of the right maxillary first and second molars. An axial CBCT image showed that multiple small cyst-like structures adjacent to the maxillary sinus and in the anterior area, and they were separated by normal bone (Fig. 8). The sagittal CBCT images showed small loculations in the molar and anterior areas, respectively, and they were separated by normal bone (Fig. 7). The histological result was the desmoplastic type containing a small cystic and follicular pattern as recurrent ameloblastoma. The radiographic features of our final recurrent ameloblastoma showed agreement with what another author mentioned, that the recurrent tumor has a characteristic appearance of multiple small cyst-like structures with very coarse sclerotic cortical margins sometimes separated by normal bone.[1]

Regarding the behavior of desmoplastic ameloblastomas, Philipsen et al[24] suggested that desmoplastic ameloblastomas in the maxilla would be more aggressive than those in the mandible. Mendenhall et al[7] also suggested that

maxillary ameloblastomas tended to have a higher local recurrence rate because the thinness of the cortical bone was a less effective barrier to tumor invasion compared with the mandible. In this report, the tumor developed between the anterior and the right premolar regions of the maxilla.

Concerning the recurrence, it was mentioned in the WHO classification of odontogenic tumors that desmoplastic ameloblastoma possibly has a lower recurrence rate than other ameloblastomas.[27] In contrast, Keszler et al[28] reported that desmoplastic ameloblastoma showed a higher recurrence rate (21.4%) than the other type of ameloblastomas (10.1%). Desmoplastic ameloblastomas have the potential for recurrence because they fail to produce a capsule.[14,29] Sun et al[18] analyzed 115 cases of desmoplastic ameloblastoma from 35 published papers and reported that whereas enucleation provided a recurrence rate of 21.1%, resection reduced this rate remarkably to 3.1%. The average period until recurrence was 36.9 months. When applying conservative techniques such as enucleation, this recurrence originates from small fragments of the tumor left in situ and jeopardizes possible good results from immediate reconstruction of the surgical defect.[30,31] In this report, the patient's tumor recurred after conservative treatment and an immediate autobone graft.

At the first visit, our patient showed the histological feature of unicystic ameloblastoma. Actually, the surgeon thought of the tumor as a dentigerous cyst by clinical examination. Unicystic ameloblastoma may form from the epithelial lining of a dentigerous cyst and show radiographical similarities with dentigerous cysts.[1] Phillipsen and Reichart[32] found that 66 cases among 152 cases of unicystic ameloblastoma were initially diagnosed as dentigerous cysts from the literature and that unicystic ameloblastoma radiographically might appear not only as a unilocular, but indeed also as a multilocular bone defect.

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