Ameloblastic Fibro-Odontoma of the Mandible with Emphasis on Evaluation Using Cone Beam Computed Tomography

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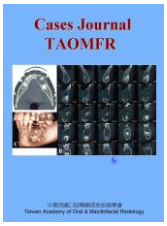

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Although head and neck neoplasms are uncommonly occurred in children, odontogenic tumors are relatively generally affected the pediatric population. The first two most frequent pediatric odontogenic tumors were odontoma and ameloblastoma respectively [1, 2]. Ameloblastic fibro-odontoma (AFO) is a rare mixed odontogenic tumor with epithelial and mesenchymal components. AFO, usually linked with unerupted tooth (or tooth germ), manifests clinically as a painless swelling either in the posterior mandible or posterior maxilla [3]. In addition, AFO is frequently detected radiographically as a well-demarcated mixed radiolucence and radiopaqcity lesion [3].

With reference to the recent classification of The World Health Organization (WHO), AFO is defined as a tumor composed of odontogenic epithelium embedded in cellular ectomesenchymal tissue that resembles dental papilla, with varying degrees of inductive change and dental hard tissue formation [4]. This clinical report aimed to present a case of AFO occurring in the posterior mandible of an 11-year-old girl with emphasis on evaluation using cone beam computed tomography (CBCT).

CASE PRESENTATION

An 11-year-old Taiwanese girl visited the local dental department with the chief complaint of a swelling mass over lower left posterior region for months and was referred to Kaohsiung Municipal Ta-Tung Hospital. The patient denied any systemic diseases and histories of food or drug allergies. Local heat, tenderness and paresthesia were not complained. On extraoral examination, mild facial asymmetry was noted. Intra-oral examination revealed a bony hard swelling, measured approximately 2.0 × 2.5 cm in diameter, with intact mucosa coverage over the buccal side of the left mandible extending from tooth

73 to 75; 74 was removed due to the high mobility and a decompression button set up (Fig. 1).

Figure 1 Intra-oral examination revealed a bony hard swelling covered with intact mucosa over the buccal side of the left mandible extending from tooth 73 to tooth 75. Noting that tooth 74 was removed with a decompression button was set up.

Periapical radiography revealed a well-defined unilocular radiolucent image with scalloped margin containing radiopacities over apical region of tooth 74, and tooth 75 (Fig. 2A).

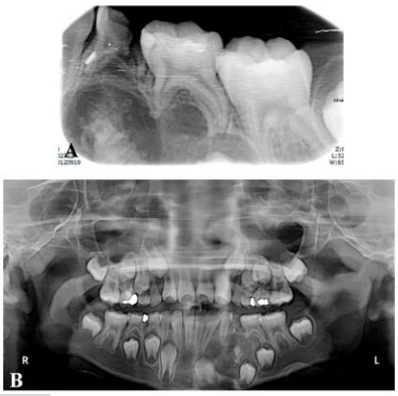

Figure 2 Periapical radiography revealed a well-defined unilocular radiolucent image with scalloped margin containing radiopacities over the apex of tooth 74 and 75 (A). Panoramic radiography showed a well-circumscribed monolocular radiolucent bony destruction containing foci of radiopaque material and encasing the tooth bud 34, which was pushed downward to the inferior border line of the left mandible (B).

Panoramic radiography showed a well-circumscribed monolocular radiolucent bony destruction containing foci of radiopaque material and encasing the tooth bud 34, which was pushed downward to the inferior border line of the left mandible, extending from the distal side of tooth 32 and the impacted tooth 33 to the mesial root of tooth 75 and the impacted tooth 35, measuring about 3.0 × 3.0 cm in maximum dimension (Fig. 2B).

Additionally, both the axial (Fig. 3A) and sagittal views (Fig. 3B) reconstructed from CBCT of the lesion revealed a well-circumscribed expansile unilocular radiolucence with varying amount of radiopaque foci and multiple impacted teeth over the left posterior mandible. The 3-dimension of CBCT reconstruction further delineated the detailed margin of this expansile bony defect containing radiopacities in the left mandible (Fig. 3C).

Based upon the aforementioned clinical and radiographic findings, the clinical impression was a benign odontogenic lesion; highly suspect AFO, over the left posterior mandible. Then, the patient was again referred to our institution for surgical treatment. Under general anesthesia, the lesion was totally enucleated, and teeth 73, 75, and 34 were all extracted (Fig. 4A). The surgical specimen was submitted for pathologic examination.

Macroscopic examination of the tumor mass, measured about 4.5 × 2.5 cm in diameter, showed an irregular capsule surface and the unerupted tooth 34 was also noted (Fig. 4B). Histologically, the decalcified section of the lesion showed areas of cell-rich mesenchymal stroma, cords, and follicles of ameloblastic epithelium together with area consistent with complex odontoma (Fig. 5A &B). Hence, the microscopic diagnosis of AFO was rendered for the current neoplasm. The postoperative course of the patient was uneventful, who has been undergone routine follow-up and no sign of recurrence has been noted.

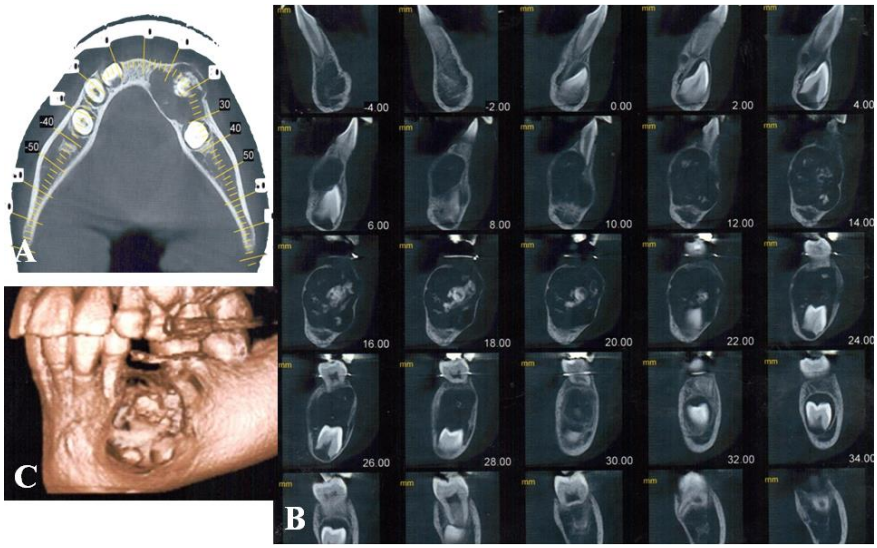

Figure 3 Axial (A) and sagittal (B) views reconstructed from cone beam computerized tomography (CBCT) of the lesion. 3-dimension of CBCT reconstruction delineated the detailed margin of this expansile bony defect containing radiopacities (C).

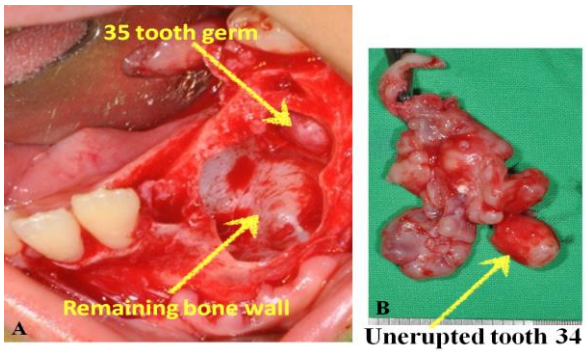

Figure 4 The lesion was enucleated, and teeth 73, 75, and 34 were all extracted (A). The surgical specimen showed an irregular capsule surface and the unerupted tooth 34 (B).

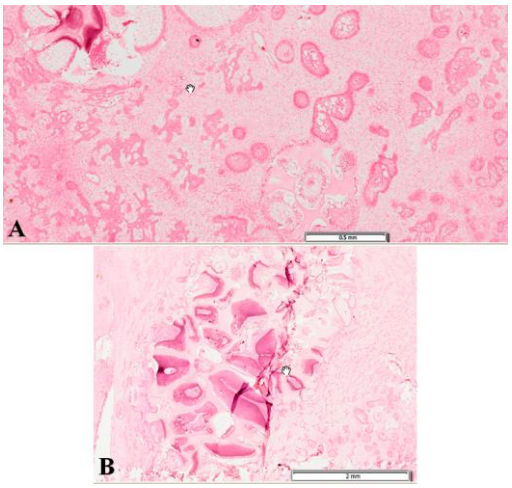

Figure 5 The decalcified section of the surgical specimen showed areas of cell-rich mesenchymal stroma, cords, and follicles of ameloblastic epithelium (A, Hematoxylin-eosin stain) together with area consistent with complex odontoma (B, Hematoxylin-eosin stain).

COMMENTS

AFO is an uncommon benign mixed odontogenic tumor of the jaws, which is, as shown in this case, most commonly found in the posterior mandible and maxilla [5]. This lesion is defined by the WHO as neoplasm consisted of proliferating odontogenic epithelium embedded in cellular ectomesenchymal tissue which is similar to dental papilla, with varying degrees of inductive changes and development of dental hard tissue [4]. To date, controversy still exists whether AFO is an overt neoplasm or belongs to a stage in the development of odontoma [6, 7] with, on one hand, some scientists regarded ameloblastic fibroma and AFO correspond to different stages of the same disease entity, and it will subsequent develop finally to a mature odontoma. On the other hand, the WHO classification defines the ameloblastic fibroma, AFO, and odontoma are different disease entities. Moreover, the residual/recurrent cases of ameloblastic fibroma demonstrate no verification that ameloblastic fibroma will further mature to become a more differentiated odontogenic lesions, for instance, AFO or odontoma. Also, AFO occurs in patients with younger mean age than the ameloblastic fibroma, and additionally, both the ultrastructural and immunohistochemical investigations demonstrate difference between ameloblastic fibroma and AFO [8]. Taken together, based on their clinical and radiological features from literatures, some of the AFOs are most likely true neoplasms whilst others seem to be developing odontomas.

Most recently, Buchner et al. [3] presented a comprehensive analysis of a large number of AFO patients (totally 114 cases) which reported that AFOs are significantly more common in males (male-to-female ratio to be 1.85:1) and in the mandible. The mean age was 9.6 years ranged from 8 months to 26 years. Most of the lesions were unilocular and less than 10% were multilocular; additionally, most lesions were mixed radiolucent-radiopaque, and only about 5% were radiolucent. Nearly all lesions (approx. 92%) were associated with unerupted tooth/teeth. The clinical and radiologic characteristics of the current case are mostly consistent to this comprehensive review, with the exception that the female is affected in the present case.

Clinically, the differential diagnosis of AFO should consider benign intraosseous jaw lesions showing mixed radiographic patterns, such as odontoma, calcifying epithelial odontogenic tumor, calcifying odontogenic cyst, and adenomatoid odontogenic tumor as well as ameloblastic fibrosarcoma or ameloblastic fibroodontosarcoma. In the current case, both the clinical and radiologic findings of the conventional radiographies (periapical and panorex), and particularly, CBCT suggest a benign mixed odontogenic tumor of AFO. Furthermore, the CBCT image, in the present case, proved to be an important complement to the conventional radiographic examination as it could provide additional data not evident on the 2-dimensional conventional imaging [9]. It is because both the extent of the lesion and its effects on adjacent structures were noted to be better delineated with CBCT. Moreover, the presence of calcifications, a significant radiographic characteristic of AFO, was more clearly visible on CBCT compared with the conventional imaging [9]. Histopathologically, the present case was characteristic of an AFO, demonstrating both the pathological features as an ameloblastic fibroma together with mature odontoma [10].

The treatment of choice of the present case was enucleation as most AFO lesions can easily be detached from the surrounding bone [11]. As most lesions

are coronal to an unerupted or a displaced tooth, the deciduous tooth is usually extracted along with the tumor, as demonstrated in the present case. When associated with a permanent tooth, if possible, the tumor is enucleated permitting the involved adult tooth to erupt. However, when the lesion is encasing the permanent tooth, as found in the present case, both the tumor and the tooth should be removed to reduce the potential risk of future recurrences.

The recurrence of AFO is typically associated with insufficient surgical removal or when tumor remnants persist in the margins of enucleation. Then, these tumor remnants may proceed to develop into either a lesion of recurrence or a lesion of malignant transformation. Malignant transformation of AFO to an ameloblastic fibrosarcoma or an ameloblastic fibro-odontosarcoma, despite very uncommon, has indeed been reported in English language literatures [12-14]. Most recently, Reiser et al. reported a 6-year-old girl with ameloblastic fibro-odontosarcoma, which developed gradually over duration of 3 years, probably from a formerly undiagnosed AFO [15].

CONCLUSION

Hereby, we documented a less common mixed odontogenic lesion of AFO affecting the mandible of a Taiwanese girl, with emphasis on radiographic evaluation using CBCT, which can provide better demonstration and 3-dimension of such an expansile lesion with an effect on adjacent dentition. Conservative surgery such as complete enucleation is the treatment of choice; however, since potential malignant transformation is probable; therefore, regular and long-term follow-up is strongly suggested for the present case.

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