Radiographic Feature of Ameloblastic Fibro-Odontoma : Report of Four Cases and Review of Literature

Yi-Wen Cheng^{*}, Li-Ling Huang^{*}, Yu-Hsun Kao^{*}, Edward Cheng-Chuan Ko^{*,†}, Yuk-Kwan Chen^{†,‡}, Yu-Feng Chen^{*}

^{*}Division of Oral and Maxillofacial Surgery, Department of Dentistry, Kaohsiung Medical University Hospital, Kaohsiung, Taiwan, R.O.C.

[†]School of Dentistry, College of Dental Medicine, Kaohsiung Medical University,

Kaohsiung, Taiwan, R.O.C.

[‡]Divisions of Oral Pathology, Department of Dentistry, Kaohsiung Medical University Hospital, Kaohsiung, Taiwan, R.O.C.

Abstract

Ameloblastic fibro-odontoma (AFO) has been defined by the World Health Organization (WHO) as a rare odontogenic tumor that is more prevalent in children. AFO usually occurs in the posterior region of the jaws, especially in the mandibular area. In radiographic images, most AFO demonstrates a welldemarcated circumscribed radiolucent lesion containing variable amounts of radiopaque material and usually involved with an unerupted tooth. The aim of this report is to present various types of radiographic images of AFO in the Taiwanese population. We classified these images into 4 groups as follows : 1. total radiolucent without radiopaque material, 2. few proportion of opacities, 3. large proportion of opacities, 4. single opaque mass from x-ray image. The present report is to demonstrate four different types of radiographic images of AFO and four case reports of Taiwanese population.

Key words: Ameloblastic Fibro-Odontoma, Radiographic Image, Odontogenic tumor.

Introduction

Ameloblastic fibro-odontoma (AFO) was first introduced by Hooker¹ in 1967 and was defined by the World Health Organization $(WHO)^2$ in 2005 as a neoplasm composed of proliferating odontogenic epithelium embedded in cellular ectomesenchymal tissue that resembles dental papilla, with varying degrees of inductive change and dental hard tissue formation.

AFO is generally considered to be uncommon, comprising 0% to 3.1% of odontogenic

tumors^{3, 4, 5}. AFO typically demonstrates a painless mass in the posterior mandible or maxilla with or without swelling^{2, 5-8} and occurs in patients' first decade of life⁴. Radiographically, the tumor commonly presents as a unilocular or multilocular mixed radiolucent mass with well-defined borders and various amounts of radiopaque material of irregular shapes and sizes. In addition, it is often discovered in association with an unerupted tooth.

This report presents four cases of different types of radiographic images.

Case 1 (Total radiolucent without radiopaque material)

A 15-year-old girl was referred to our OPD for evaluation of an impacted second molar in the left posterior mandible. She reported no pain, numbness, or swelling. A periapical film revealed a total radiolucent without radiopaque lesion with corticated margin in the left posterior mandible (Fig. 1). The lesion was enucleated intraorally after extraction of the permanent lower left second molar under local anesthesia. The surgical specimen was fixed in 10% formalin and subjected to pathological analysis with H&E stain.

The microscopic exam showed islands and strands of odontogenic epithelium surrounded by embryonic cellular connective tissue. The epithelial component formed islands or strands that consisted of stellate reticulum surrounded by a layer of ameloblast-like columnar cells. The mesenchymal component was connective tissue with fibroblast of dental papilla of the tooth germ (Fig. 2) (Fig. 3). The histopathological diagnosis of AFO was made.

Clinical follow-up and radiographic examinations were carried out, no recurrence was noted.

Case 2 (Few proportion of opacities)

A 8-year-old girl went to our OPD for a painless swelling over her left mandible. Clinical examination revealed facial asymmetry of the face. Intraorally, both buccal and lingual cortical expansion of the left mandible was observed. The panoramic radiography showed a multilocular radiolucent lesion contained with few numbers of small opacities extending from the lower left central incisor to the lower left first molar (Fig. 4). The tumor was enucleated and the lower left first premolar was extracted under general anesthesia. The histopathologic diagnosis was ameloblastic fibro-odontoma. The following up was unremarkable and there was no signs of recurrence.

Case 3 (Large proportion of opacities)

A 16-year-old male was referred to our OPD to treat his impacted lower right first premolar. The panoramic radiograph displayed a unilocular and well-demarcated lesion, and large proportion of opacities occupied the radiolucent area (Fig. 5). The permanent tooth was displaced to the mandibular body. Under local anesthesia, the retained primary tooth was extracted and the lesion was enucleated. The diagnosis was confirmed with AFO. The patient has been monitored and the lesion has not recurred.

Case 4 (Single opaque mass)

A 15-year-old female was brought to our OPD with her chief complaint of missing her lower right second molar. Intraoral examination revealed no bony expansion in the area. The radiograph showed a well-circumscribed unilocular radiolucency containing a single opaque mass as well as lower right second molar (Fig. 6). The clinical diagnosis was odontoma. The



Fig. 1. Periapical film of lower left first and second molar showed image with total radiolucent without radiopaque material.

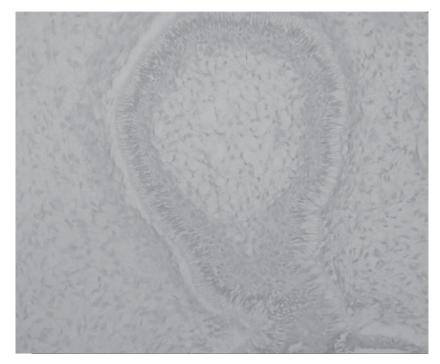


Fig. 2. Microscopic image. Ameloblastic island in loosen connective tissue with H&E stain (200X).

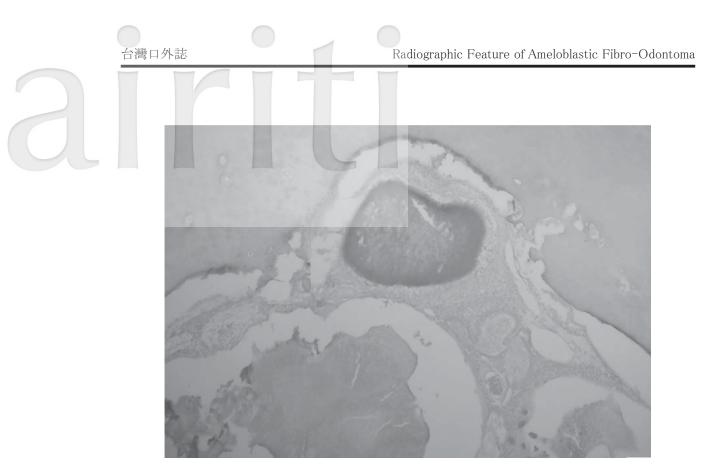


Fig. 3. Microscopic image. Enamel matric and cementum with H&E stain (100X).

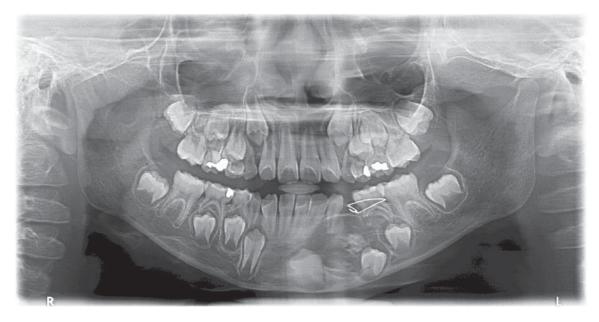


Fig. 4. Panoramic film showed multilocular radiolucent lesion contained with few proportion of small opacities extending from lower left central incisor to lower left first molar.

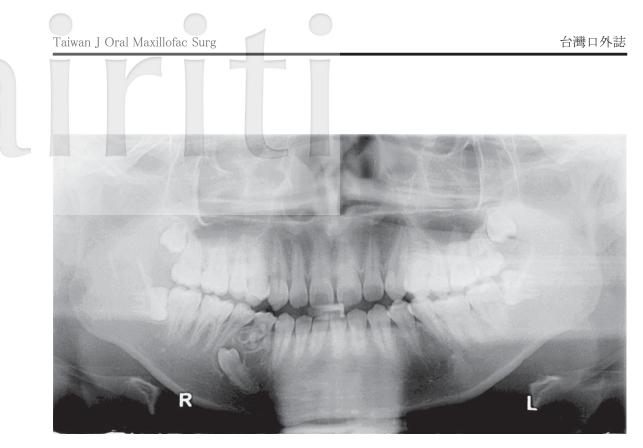


Fig. 5. Panoramic film showed unilocular radiolucent lesion contained with large proportion of opacities in the right mandible.



Fig. 6. Panoramic film showed unilocular radiolucent lesion contained with a single opacities over right mandibular third molar area.

patient was treated by enucleation of the lesion without tooth removal under general anesthesia. The post-operative course was uneventful. Histopathological examination of the specimen confirmed the diagnosis of AFO. The follow-up revealed no recurrence after operation.

Discussion

The AFO is classified as a mixed odontogenic tumor composed of odontogenic epithelium and ectomesenchymal elements. The term "AFO" represents a histological combination of ameloblastic fibroma and a complex odontoma. This lesion exhibits similar biological behavior to ameloblastic fibroma or complex odontoma.

AFO has been reported with a prevalence range of $0\%\sim3.1\%^{3, 4, 5}$ within odontogenic tumors, with the average age of diagnosis being in the first and second decades. According to the revised WHO classification, it is a benign tumor without invasive growth. Malignant transformation of ameloblastic fibro-odontoma has rarely been reported.

According to the literature review, most lesions of the AFO are mixed radiolucent and radiopaque. In terms of radiographic features of AFO, 94.8% are radiolucent and radiopaque (mixed type), and 5.2% are only radiolucent reported by Amos et al⁹. However, only few articles have reported the diverse type of radiopacity. We can classify the tumor into four divisions: 1. total radiolucent without radiopaque material, 2. few proportion of opacities, 3. large proportion of opacities, 4. single opaque mass. All our cases were associated with an unerupted impacted tooth.

Because of the low recurrence rate, the most common surgical treatment is conservative

enucleation or curettage^{1, 2}. However, some authors suggest that recurrence occurs from a residual lesion of inadequate surgical removal⁶.

The radiographic features of AFO usually include: radiolucent image with corticated margin and variant radiopaque content, attached with an unerupted tooth². These features of AFO are similar with other odontogenic tumors: complex odontoma, ameloblastoma or calcifying epithelial odontogenic tumor¹⁰. The incidence of AFO in odontogenic tumor is not high, but it should be taken into consideration that its characteristics are similar to other odontogenic tumors, and careful differential diagnosis must be made.

Conclusion

AFO is an uncommon lesion that belongs to the group of mixed odontogenic tumors. Most of the reported lesions are involved with an unerupted tooth. This study represents four cases for Asian population in Taiwan and provides various data of the clinical and radiographic features of AFO. The specific Taiwanese data of AFO can provide the information for the initial diagnosis. However, we still need more data for statistical analysis of our domestic population.

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台灣口外誌

造釉細胞纖維齒瘤之放射線影像: 四例報告及文獻綜述

鄭宜文* 黃麗玲* 高郁勛* 柯政全*,* 陳玉昆^{*,‡} 陳裕豐*

*高雄醫學大學附設中和紀念醫院口腔外科 「高雄醫學大學附設中和紀念醫院口腔病理科 "高雄醫學大學口腔醫學院牙醫學系

摘 要

造釉細胞纖維齒瘤已經被世界衛生組織定為一好發於兒童的罕見齒源性 腫瘤。造釉細胞纖維齒瘤主要出現在後牙區,特別是下顎後牙區。在放射線 影像上,造釉細胞纖維齒瘤的表徵大多為邊緣清楚的放射線透過性影像合併 有不透過性影像,且伴隨有未萌發的牙齒。本文報告的目的在研究台灣人口 中,造釉細胞纖維齒瘤在影像學上不同的表徵。我們將其分成四類:1.完全 放射線透過性影像;2.放射線透過性影像合併少量不透過性影像;3.放射線 透過性影像合併多量不透過性影像;4.single opaque mass放射線透過性影像 合併單一放射線不透過性影像。此報告提出台灣族群中造釉細胞纖維齒瘤的 影像特徵,以利臨床醫師在影像診斷上的參考,並且加上文獻上的回顧及提 出建議的治療方式。

關鍵詞:造釉細胞纖維齒瘤,放射線影像,齒源性腫瘤。

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Reprint requests to: Dr. Yu-Feng Chen, Department of Oral and Maxillofacial Surgery, Kaohsiung Medical University Hospital, 100 Tzi-You 1st Rd, Kaohsiung 80756, Taiwan, R.O.C.