Case Report

Ameloblastic Fibro-Odontoma in a 7-Month-Old Infant: A Case Report

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KEY WORDS

Mandibular Disease;
Odontogenic tumor;
Jaw neoplasm;

Received July 2016;
Received in Revised form November 2016;
Accepted December 2016;

ABSTRACT

Ameloblastic fibro-odontoma is a relatively rare, benign odontogenic tumor that usually occurs in children and adolescents with unerupted teeth. This article reports an ameloblastic fibro-odontoma in the anterior mandible as a "bump on her gum" in a 7-month-old girl. This is the first case under 9 months old reported to date. Radiographic and histologic findings as well as the treatment are discussed.

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Cite this article as: Mashhadiabbas F., Shamloo N., Jafari M., Vafadar Sh. Ameloblastic Fibro-Odontoma in a 7-Month-Old Infant: A Case Report. J *Dent Shiraz Univ Med Sci.,* 2017 September; 18(3): 234-236.

Introduction

Odontogenic tumors constitute 7% of all oral pathologic lesions found in children and adolescents. Ameloblastic fibro-odontoma (AFO) accounts for 1 to 3% of all odontogenic tumors, with the same percentage increasing to 4.6% in children. [1-4]

AFO is a relatively rare, benign, and slow-growing odontogenic tumor. [3, 5-6] The lesion occurs in the posterior region of the jaws in both males and females with the average age of 10 years. [1, 3-4, 6]

The lesion usually causes a painless expansion of the affected bone and is generally associated with unerupted teeth. [5-6] Radiographically, the lesion shows a well-circumscribed radiolucent area containing various amounts of radiopaque material of irregular size and form. [4, 7-9] AFO consists of proliferating odontogenic epithelium shaped as nests, islands, strands and long anastomosing cords embedded in a cellular ectomesenchymal tissue resembling dental papilla with varying degrees of inductive change and dental hard tissue. [2, 5-6] AFO is usually treated by enucleation and curettage without recurrence and its prognosis is excellent. [5-6, 10] Usually an odontogenic cyst or tumor causes failure of eruption of a single primary tooth. [7] This report describes an AFO in the anterior of the mandible of a 7-month-old girl, accompanied by an unerupted primary tooth.

Case Report

A 7-month-old girl was referred to the oral and maxillofacial surgeon for evaluation of a swelling on her gum that had been noticed by her parents when she had been smiling. There was no history of local trauma or infection. Intra-oral examination showed alveolar bone expansion in the anterior mandible. The overlying mucosa was intact, showing a smooth surface with a bluish hue. There was no erupted tooth in her mouth. An occlusal radiograph revealed a well-defined radiolucent lesion in the anterior part of the mandible as well as displaced adjacent primary incisor teeth (Figure 1a).

There existed expansion and thinning of buccal cortex adjacent to the lesion. An excisional biopsy was performed under general anesthesia. The lesion was enucleated with curettage of the margins up to the normal bone. A specimen was submitted for pathologic examination. Microscopically, the lesion was composed of pieces of loose cellular connective tissue reminiscent of dental papilla with cords, nests and islands of cuboidal to columnar odontogenic epithelial cells, with reverse polarity in the periphery. Cystic and microcystic degeneration in the nests and islands of

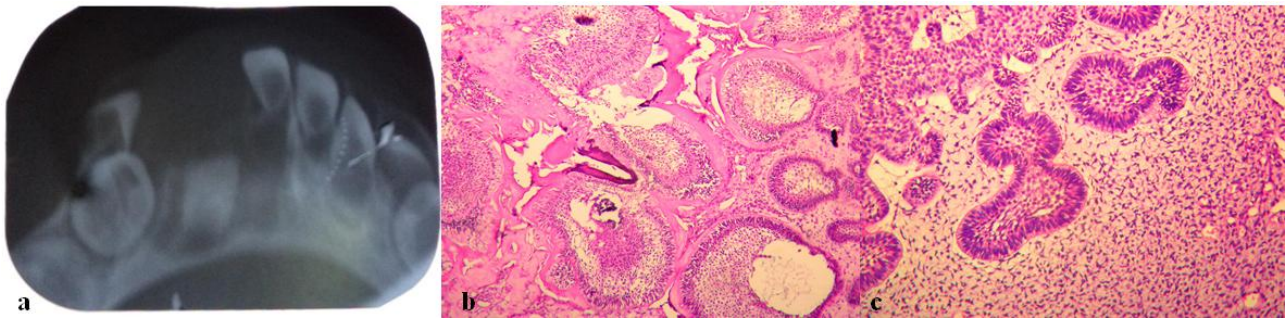

Figure 1a: Occlusal radiography shows a radiolucent lesion in the anterior part of mandible. **b:** Nest or islands of epithelial odontogenic within loose to cellular mesenchymal connective tissue background with tooth-like structures (x100). **c:** High power field (x200) of ameloblast-like nest with anastomosing appearance in cellular stroma of dental papilla-like connective tissue.

the epithelial cells was observed (Figure 1b and 1c).

Some foci of tooth structures composed of tubular dentin and enamel matrix were also seen. These findings were consistent with the diagnosis of AFO. One year after surgery, based on clinical appearances, no recurrence is seen and soft tissue is normal.

Discussion

AFO has traditionally been classified as a benign mixed odontogenic tumor. The term "ameloblastic fibro-odontoma" represents a histologic combination of ameloblastic fibroma and odontoma. [8-9] The etiology of AFO is unclear. Some researchers have suggested that ameloblastic fibroma is precursors of AFO, evolving into an odontoma. [5]

Most authors now agree that ameloblastic fibro-odontoma is a separate entity but it can be histologically indistinguishable from immature complex odontoma. [11] Ameloblastic fibro-odontoma is relatively rare and usually occurs in people younger than 20 years of age. The average age of patients is 11.5 years. [1-2, 4, 8, 12] Based on literature review the youngest case with AFO reported until now is that of a 9-month-old boy, [13] while only one case older than 30 years of age has to date been reported. [8] Our case is the first report of AFO occurring in a patient younger than 9 months of age. The site of occurrence of AFO was the anterior mandible. Although uncommonly found in the anterior mandible, an AFO according at this site tend to be in a younger patient with an average age of 3.3 years. [12] There is no gender predilection and the reported AFOs are most often located in the posterior segment of the mandible. [6, 11]

Painless swelling and failure of tooth eruption are the most common presenting complaints. Asymptomatic cases are usually discovered when radiographs are made to determine the reason for failure of a tooth to erupt. [6, 8]

Radiographically, the ameloblastic fibro odontoma shows a well-circumscribed radiolucent area containing a variable amount of radiopaque material of irregular size and form. The ratio of radiopaque to radiolucent areas differs from one lesion to another. [5, 8-9] The case presented in this report had a well-defined radiolucent area with no radiopaque structures, making it difficult to differentiate it from that of other odontogenic lesions. [6] The differential diagnosis in this case consisted of ameloblastoma, ameloblastic fibroma and ameloblastic fibro-odontoma.

Microscopically, the lesion is composed of strands, cords and islands of odontogenic epithelium embedded in a cell-rich, primitive ectomesenchyme resembling the dental papilla with variable amounts of irregular formation of enamel, dentin and a cementum-like material, as was seen in this case. [2, 5-6, 9] Surprisingly in this case, cystic and microcystic degeneration was seen in nests and islands of odontogenic epithelium, this being in fact a common feature of ameloblastoma. [14] Clinical and radiographic differential diagnoses of ameloblastic fibro-odontoma include odontoma, ameloblastoma, odontoameloblastoma, and ameloblastic fibroma. [2]

Conservative surgical excision is an accepted treatment for this lesion. In most cases, the impacted tooth associated with the tumor is removed at the same time. There is a low potential for recurrence. [6] Despite the low potential recurrence of the tumor, a 5-year-long follow-up is recommended. [5] In this case, the treatment consisted of enucleation and curettage of the bony walls followed by extraction of the primary

incisors. After one-year follow-up, there was no sign of recurrence; nonetheless, further examinations have been recommended.

Conclusion

Until now, the youngest reported patient with AFO was a 9-month old boy. The present article reports an AFO in the anterior mandible of a 7-month-old girl. The lesion was treated by enucleation with curettage of the margins up to the normal bone. One year after surgery, no recurrence is seen and soft tissue is normal.

Conflict of Interest

The authors disclose no potential conflicts of interest.

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