Pediatric Ameloblastic Fibroma: A Case Report

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ABSTRACT

Aim: To highlight the potential difficulties in diagnosing neoplastic lesion of the head and neck in children within autism spectrum.

Background: Ameloblastic fibromas are a variation of odontogenic tumors that are located in the posterior mandible in 70% of cases. The tumors may be either unilocular or multilocular when observed radiographically. Ameloblastic fibromas tend to have well-defined, scalloped margins radiographically but may also be corticated. In close to 75% of all cases, an impacted tooth is associated with the lesion. Although not confined to patients who are in their first or second decades of life, ameloblastic fibromas most often arise in this population.

Case description: A 9-year-old male diagnosed with autism presented with tenderness in the left mandible. The parents were able to elucidate the child's problem as tingling rather than pain. The patient was referred for histopathological diagnosis and treatment. After evaluation and biopsy, the lesion was identified as a pediatric ameloblastic fibroma. He subsequently underwent the conservative approach of marsupialization and curettage without complication. Six-month follow-up revealed no evidence of recurrence and normal eruption patterns of the succedaneous teeth in the affected area.

Conclusion: In this specific case, the conservative approach to treatment appears to have been appropriate. This may not be the appropriate course for every case; as such, each case will have an individualized approach. Earlier recognition with careful inspection can reduce potential complications.

Clinical significance: We might be missing early diagnosis of ameloblastic fibroma and other significant orofacial neoplasms in patients who are nonverbal or nondescriptive such as those with autism. Moreover, careful inspection of radiographic and clinical signs cannot be overemphasized.

Keywords: Autism, Marsupialization, Nonverbal, Pediatric ameloblastoma.

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INTRODUCTION

This case report illustrates the development, diagnosis, and treatment of an ameloblastic fibroma in a 9-year-old patient. An ameloblastic fibroma is an odontogenic tumor composed of epithelial and mesenchymal tissue that, while benign in its initial presentation, has a high likelihood of malignant recurrence as an ameloblastic fibrosarcoma posttreatment.¹ While ameloblastic fibromas are similar to and often mistaken for ameloblastomas, they are different in several ways. An ameloblastoma is a tumor composed only of epithelial tissue, but an ameloblastic fibroma contains both epithelial and mesenchymal tissues.² The mesenchyme will cause a hematoxylin and eosin (H&E) stain to turn more pink due to its reaction with the eosin dye.³ An ameloblastic fibroma may also be distinguished histopathologically from an ameloblastoma by its smaller islands of ameloblastic epithelium.² An ameloblastic fibroma may present with or without a definite capsule and usually appears radiographically as a solid, smooth soft tissue mass.⁴

It should be noted whether an ameloblastic fibroma does recur as an ameloblastic fibrosarcoma; it is the mesenchymal component that is malignant.¹ Ameloblastic fibromas have the highest incidence in the posterior mandible in tandem with impacted teeth.⁵ These odontogenic tumors therefore occur frequently during the first and second decades of life, without either gender eliciting a higher predisposition than the other.⁶ Tumors originating in the mandible often have a less guarded prognosis than those of the maxilla, as the proximity of the maxillary sinuses and nasopharynx allow maxillary tumors to progress undetected for longer periods of time.⁷ Radiographically, an ameloblastic fibroma presents as either a unilocular or a multilocular radiolucent lesion.⁵ The radiographic margins tend to be well defined, and they may be

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corticated or scalloped.⁸ An impacted tooth is associated with the lesion in about 75% of cases.⁸ The tumor is present in the posterior mandible in 70% of cases.⁹

CASE DESCRIPTION

The patient initially presented in 2015 (Fig. 1) as a 7-year-old Caucasian male diagnosed with high-functioning autism. His medical history includes a dermoid cyst removed from the back of the head at 18 months of age and an umbilical hernia correction at 4 years of age. In April 2016, the patient was hospitalized for adenovirus. Patient is not on any medications and is currently up to date on vaccinations.

Between 2015 and 2017, the patient was treated with class III composite restorations, sealants, and the removal of space maintainers. During September 2017, the patient presented for restorations on #C and #H. The patient complained of "tingling in the area of lower left tooth," corresponding to #K, thereby prompting a new panoramic radiograph to be taken (Fig. 2). Upon evaluation, a consultation with an oral and maxillofacial surgery (OMFS) was sought due to the appearance of the soap bubble radiolucency.

Fig. 1: Panoramic radiograph, October 2015

Fig. 2: Panoramic radiograph, September 2017

Fig. 3: $\times 10$; H&E stain: (A) Islands and cords of odontogenic epithelium; (B) Hyalinized fibrous connective tissue; and (C) Myxoid fibrous connective tissue

Fig. 4: $\times 20$; H&E stain: (D) Islands of epithelium peripheral columnar cells and (E) Cellular mesenchymal tissue resembles early dental papilla

Fig. 5: $\times 20$; H&E stain showing odontogenic islands in a variably hyalinized and myxoid stroma: (F) Hyalinized areas with less cellularity and (G) Uniform cells in myxoid stroma, lacking the atypia and hypercellularity seen in an ameloblastic fibrosarcoma

Clinically, some buccal expansion and slight sponginess to the buccal plate was palpated. Patient reported tenderness at the lower left mandible upon palpation. The patient was subsequently referred to the UF Health Oral Surgery and Oral Pathology Departments to rule out a cyst vs a more benign aggressive lesion.

The patient had developed an ameloblastic fibroma in his lower left mandible near unerupted tooth #37 [Fédération Dentaire Internationale (FDI) tooth numbering system]. The differential diagnosis originally included central giant cell lesion, deciduous cyst, ameloblastoma, and odontogenic keratocyst. Upon presentation for treatment, a cone beam computed tomography from C-1 to C-5 showed “a well-defined, multilocular, corticated

radiolucency with scalloped borders in the left posterior mandible extending from the interradicular region of 36 to the mesial tooth bud of 38 (FDI tooth numbering system) and from the alveolar crest of tooth buds 38–37 (FDI tooth numbering system) of the inferior third of the mandible.” The radiologic impression postulated a benign neoplasm of odontogenic origin. In November 2017, an excisional biopsy (Figs 3 to 5) of the site was performed, which showed a benign odontogenic neoplasm comprising “islands of ameloblastic epithelium in myxoid and hyalinized fibrous, highly cellular connective tissue which resembles dental pulp and contains spindle-shaped fibroblasts.” The epithelial and mesenchymal components in these configurations are indicative of an ameloblastic fibroma, and lesion was subsequently diagnosed as an ameloblastic fibroma.¹⁰

After a definitive diagnosis was made, the ameloblastic fibroma was treated by direct curettage in January 2018. Upon sulcular incision and exposure of the mandibular alveolar ridge, a handpiece and round bur were used to unroof bone over the lesion. No cystic cavity was noted, but rather solid white granulomatous tissue was seen throughout, which is consistent with the central giant cell granuloma. While the inferior alveolar nerve was not visualized, the impacted tooth #37 (FDI tooth numbering system) was visualized. The procedure was performed without complications and the specimen was submitted for examination by a pathologist to confirm the original diagnosis of ameloblastic fibroma.

The patient recovered well and impacted #37 (FDI tooth numbering system) was preserved. Upon presentation in August 2018, the patient demonstrated no facial asymmetry and normal intraoral presentation. Radiographic evaluation showed normal radiographic findings despite invasive surgery.

DISCUSSION

The case presented shares many similarities with other reports on ameloblastic fibromas but has its own unique symptoms and characteristics. In one similar report, a 15-year-old female patient presents with swelling in the posterior mandible, which somewhat mirrors the chief complaint of the patient in this particular case.¹¹ Histopathologically, the case also shows similarities because the lesion was found to contain “highly cellularized connective tissue stroma comprising odontogenic epithelium arranged in the form of strands, chords, and follicles of varying shape and size.”¹¹ The patient was also treated by direct curettage under general anesthesia. However, several distinguishing characteristics were described in the case report that set it apart from the case in this paper. First, an “orthopantomogram showed a huge radiolucent lesion involving the body of the mandible from distal to 45 to the ramus of the mandible.”¹¹ This lesion is much larger than that of the one observed in the subject of this paper. This could be a product of the lesion having more time to progress as the patient is several years older than this paper’s study patient. Furthermore, the case report notes that “46, 47, 48 were not clinically seen” and “serous discharge from a small opening distal to 45” was observed.¹¹ These differences could also be attributed to the age disparity between the two patients, due to both the age of onset and any effects the respective lesions would have on the natural schedule of the mandible’s growth and development.

Another similar case details a 1-year-old patient presenting with “swelling in her posterior mandible” that was noted to be hard to palpation.¹² Like the case in this paper, no pus or drainage was observed, and “the buccal cortical plate expansion in the region of swelling was detected.”¹² The lesion was also treated with surgical excision and direct curettage.¹² This case does, however, differ from that of this paper’s subject in a number of ways. Its case report describes “a large well defined unilocular radiolucent lesion extending from the right deciduous canine to second molar region.”¹² This paper’s subject was found to have a multilocular lesion, which was confined to the posterior mandible from #36 to the tooth bud of #38 (FDI tooth numbering system). The age difference between the two patients must be considered. The mandible of this paper’s subject has had several more years to develop and is much larger, and therefore the size cannot be accurately compared without taking into account measurements of the lesions and the overall size of each mandible. This paper’s subject was also found to have a multilocular lesion as opposed to a unilocular lesion. The parents of the 1-year-old patient were said to have initially refused treatment, only to return 3 months later. This patient’s lesion was described as being markedly more aggressive than the subject of this paper, whose lesion developed gradually for at least 2 years before detection.

Another report on a 3-year-old female describes a similar case in which swelling in the right mandibular body was present and a panoramic radiograph showed “a large multilocular, radiolucent lesion with scalloped margins.”¹³ These characteristics are similar to those found in the case studied in this paper. The lesion was also noted to have “extended from the right first deciduous molar area to the ascending ramus and coronoid process.”¹³ While the age disparity and subsequent difference in the size of each mandible may again be cited as possible factors, it should be emphasized that the coronoid process’s involvement represents a departure from not only the subject of this paper’s case, but any of the other cases examined in this paper. Another notable difference is that the

Fig. 6: Panoramic radiograph, August 2018

“radiograph also revealed expansion of the cortex and resorption of the roots of the lower right second deciduous molar.”¹³ While expansion of the cortex has been previously seen, root resorption was not seen at all.

The goal of comparing and contrasting similar cases with the subject of this paper is to illustrate how an ameloblastic fibroma can have many different characteristics and begin at different points in a person’s lifetime. The intention was not only to show specific signs and symptoms but also to illustrate that an ameloblastic fibroma is a complex condition and can present with a multitude of characteristics. While this paper does extrapolate the causes of each case’s unique features, it only offers a background or context from which to postulate possible reasons for them, and in no way explains them. The similarities and differences among each ameloblastic fibroma case report support the need for increased attention to this particular odontogenic tumor. Further research is required to pinpoint the exact cause of an ameloblastic fibroma and the mechanisms by which one progresses.

CONCLUSION

Initially, there was guarded prognosis of retaining #37, #36, and the tooth bud of #38 (FDI tooth numbering system). At postop and 3-month evaluation, #37 was assessed and confirmed for vitality and continued eruption. The tooth buds of #38 and #36 (FDI tooth numbering system) were also successfully retained (Fig. 6). This case study demonstrates the successful management and outcome for a young patient. The pathology was initially undetected and progressed slowly for approximately 2 years before its eventual discovery. The delay in diagnosis may be attributed to the fact that the patient has a compromised ability to communicate symptoms due to autism. Furthermore, this case reinforces the importance of following the American Academy of Pediatric Dentistry (AAPD) guidelines of taking a first panoramic radiograph of the mixed dentition.

Great diligence must be exercised in radiographic evaluation of all patients, especially pediatric patients, as pathology consistent with growth and development may occur in the first two decades of life. When dealing with odontogenic tumors, special attention must be given to these patients as many lesions may be prone to recurrence. The patient will continue to be monitored, as an ameloblastic fibroma has a recurrence rate of 18–43.5%.¹⁴ Furthermore, one third of ameloblastic fibrosarcomas presents as the recurrence of a previously treated ameloblastic fibroma.¹

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