Ameloblastic fibroma: a case report in a 6 year old

M. Garcia-Pola Vallejo* / M. Gonsalez Garcia** / L. Villalain Alvarez*** /
M. Fresno Forcedello**** / J.S. Lopez-Arranz*****

A six-year-old boy was seen by his dentist for a tumor mass in the left mandibular region. The panograph revealed a multilocular radiotransparent lesion extending from the canine to the left mandibular ascending ramus with well defined borders. After biopsy, the lesion was enucleated via curettage of the bone bed. The lesion was diagnosed as ameloblastic fibroma. After six months, radiographs showed that the surgical defect had filled with new bone.
J Clin Pediatr Dent 25(3): 245-248, 2001

INTRODUCTION

Ameloblastic fibroma, first described in 1891 by Kruse[1] is a relatively infrequent tumor[2] accounting for between 2%[3] and 2.5%[4] of all odontogenic neoplasms. The lesion consists of a benign, mixed odontogenic tumor characterized by a proliferating odontogenic epithelium embedded in cellular ectomesenchymal tissue and resembling the dental papilla. Varying degrees of inductive change have been described.[5] Both the epithelial and mesodermal components are neoplastic.[6]

The present study describes an odontogenic tumor in a 6-year-old boy with non-pathognomonic clinical, radiological and macroscopic characteristics.

* Dra. M. Garcia-Pola Vallejo, Department of Oral Pathology, Faculty of Medicine, School of Dentistry, Oviedo University Spain.
** Dr. M. Gonsalez Garcia, Department of Oral and Maxillofacial Surgery, Faculty of Medicine, School of Dentistry, Oviedo University Spain.
*** Dr. L. Villalain Alvarez, Department of Oral and Maxillofacial Surgery, Faculty of Medicine, School of Dentistry, Oviedo University Spain.
**** Dr. M. Fresno Forcedello, Department of Pathology, Faculty of Medicine, Oviedo University Spain.
***** Dr. J.S. Lopez-Arranz, Department of Oral and Maxillofacial Surgery, Faculty of Medicine, School of Dentistry, Oviedo University Spain.

Send all correspondence to Dra. Maria-Jose Garcia-Pola Vallejo, C/ Julian Claveria s/n, Department of Oral Pathology, Faculty of Medicine, School of Dentistry, Oviedo University, 33006 Oviedo (Astrurias), Spain.

Telephone: 34-98-5103635. 98-5270918.
Fax: 34-98-5272305.
E-mail: mgpola@fade.es

CASE REPORT

A 6-year-old Spanish boy was seen by his dentist for a tumor mass in the left mandibular region. Orthopantomography revealed a multilocular radiotransparent lesion extending from the zone of the canine to the left mandibular ascending ramus. The patient was referred to the Service of Oral and Maxillofacial Surgery of Asturias Central Hospital (Spain) on October 10, 1999.

The medical history, systems exploration, family history and routine laboratory findings were unremarkable. Extraoral examination revealed a firm, tender mass in the left mandible (Figure 1). The skin was clear and there were no areas of facial inflammation. No palpable neck adenopathies were observed, and the rest of the head and neck exploration was essentially normal.

The intraoral examination in turn showed a firm tumor located in the left mandibular hemibody, extending from the canine to the ascending ramus, with reduced vestibular depth in the lower left canine and molar region. The oral mucosa overlying the lesion was normal.

The radiographic study revealed a large multilocular radiotransparency with well defined borders, surrounded by a radiopaque rim, extending from the left canine region to the ascending ramus of the mandible. Germ displacement was observed of the permanent left second premolar and first and second molars. Root resorption of the deciduous first and second molars was also seen in the left jaw, but not on the contralateral side (Figure 2).

On December 9, 1999, an intraoperative biopsy was obtained under general anesthesia, which diagnosed an odontogenic tumor exhibiting an ameloblastic component. The lesion was enucleated via curettage of the bone bed, with removal of 74 and 75, and of the germs of 34 and 36 included within the tumor. The patient was discharged from hospital on the following day.

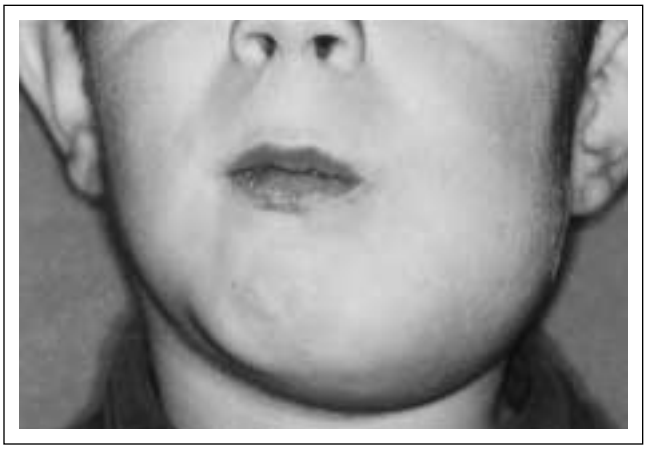

Figure 1. Clinical photograph of patent. Extraoral symmetry affecting left side of the face.

Figure2. Panoramic radiographic show a multilocular, expansible radiolucency extended from the left lower canine region to the ascending ramus.

Figure 3. Macroscopic appearance of the tumor.

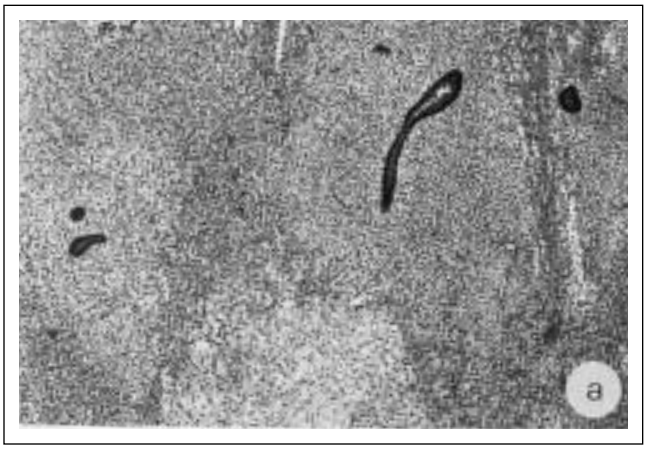

Figure 4a. The tumor is composed of a cell-rich mesenchymal tissue with proliferation of neoplastic epithelium (2.5X).

The surgical piece was rose-color, multinodular, segmented and of soft consistency (Figure 3), and measured approximately 3.5 x 2.5 x 2 cm.

The histopathological study revealed narrow strands and nests of odontogenic epithelium in a stroma of uniform mesenchymal connective tissue (Figure 4a). The connective component presented a very cellular mass of young fibromyxoid tissue, and was composed of stellate and elongated mesenchymal cells (Figure 4b). The epithelial area consisted of strands and islets that exhibited a peripheral layer of columnar and cuboidal cells which enclosed a small number of cells resembling stellate reticulum of the enamel organ in a developing tooth (Figure 5a). A hyalinized, pale eosinophilic band of varying thickness was seen surrounding the islets of odontogenic epithelium (Figure 5b). Neither the odontogenic epithelium nor the fibroblastic mesenchyma showed mitotic activity or cellular atypia. The lesion was diagnosed as ameloblastic fibroma.

After 6 months of follow-up, radiographs showed that the surgical defect had filled with new bone (Figure 6).

DISCUSSION

Central odontogenic fibroma appears to occur more frequently in children and young adults. Cases developing in newborn infants[7,8] and in adults up to 42 years of age have also been reported, the mean age being 14.6 years.[2] A case of congenital ameloblastic fibroma associated to other malformations has been described.[9]

The tumor is slightly more common in males than in females.[10] No racial predilection exists.[11] The mandible is affected four or five times more commonly than the upper jaw.[10,12]

Ameloblastic fibroma presents no pathognomonic clinical or radiological characteristics. The radiographic study shows either a unilocular or multilocular radiotransparent lesion, that is frequently associated with the crown of an unerupted tooth[10] or to an impacted tooth.[13,14] The lesion is usually located in the premolar-

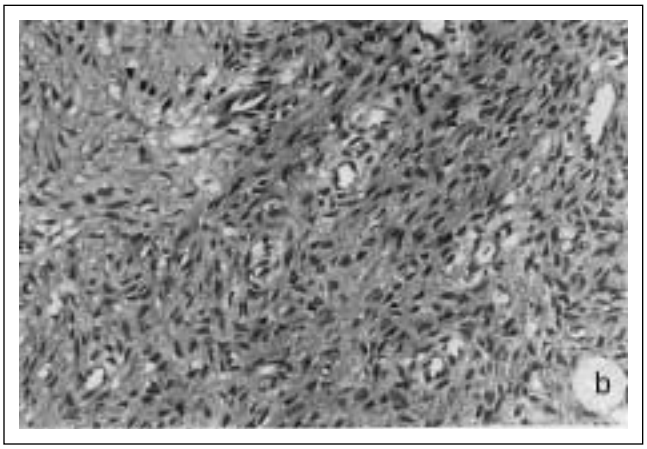

Figure 4b. A higher magnification (40X). The connective cells are rounded, angular, elongated and strellate.

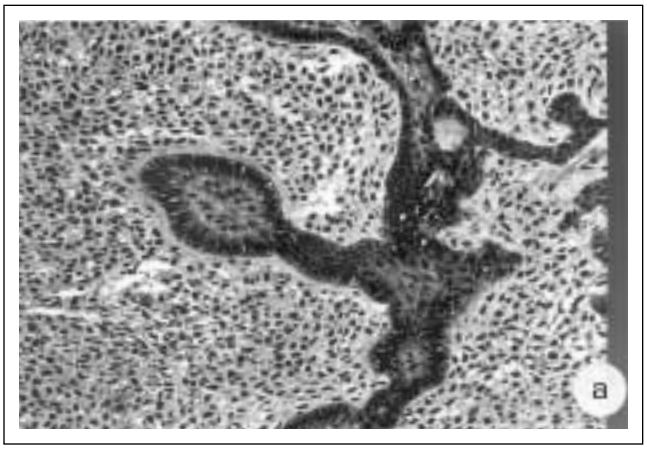

Figure 5a. Photomicrograph showing cords of epithelium with peripheral cells similar to those of theinner enamel epithelium, which resemble the early stages of development of the enamel organ (25X).

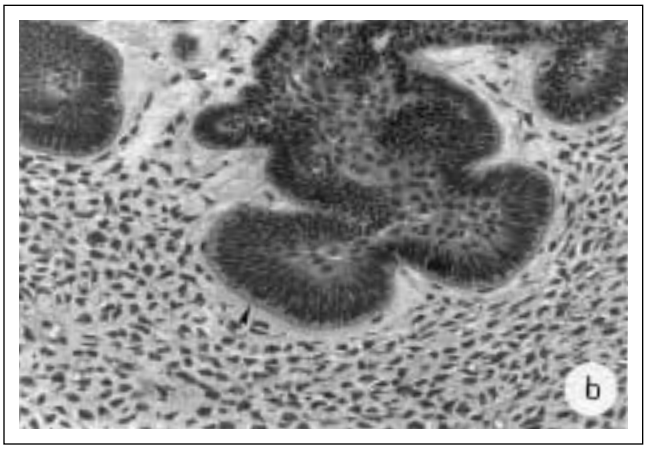

Figure 5b. Rimming the basal lamina around the islands of odontogenic epithelium are strands of eosinophilic acellular material, suggesting an induction of the adjacent mesenchyme (arrow) (40X).

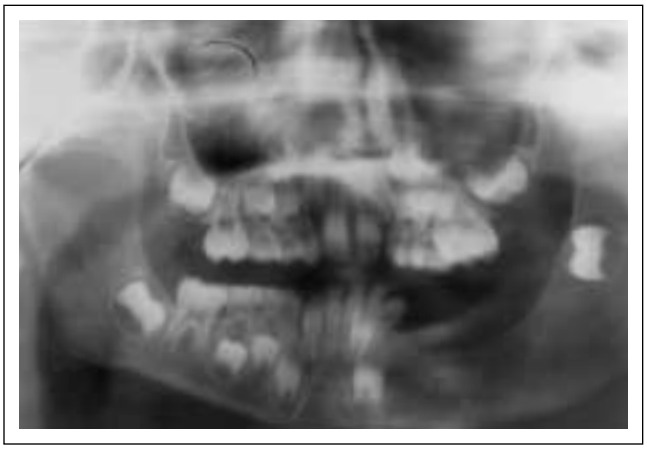

Figure 6. Panoramic radiographic taken at 6 months after surgery.

molar region.[5] The radiographic appearance of ameloblastic fibroma may be identical to that of numerous other cysts and tumors;[11] in this sense, radiographic differentiation from ameloblastoma (for example) may be impossible.[5]

The microscopic appearance of the mesenchymal component resembles the fibromyxoid tissue of the dental papilla.[4] Sometimes scattered foci of epithelial islets can be found, resembling the early stages of development of the enamel organ.[11] Many of these epithelial nests contain a central portion that is less cellular and exhibit a stellate morphology reminiscent of the stellate reticulum of the enamel organ in a developing tooth.[11]

The tumor may also exhibit signs of apparent induction, such as narrow cell-free zones and areas consisting of amorphous hyaline-like material surrounding epithelium.[6] Pindborg *et al.*[15] indicated that odontogenic tumors represent a range of conditions, the features of which probably depend on the stage of induction towards tooth formation reached prior to neoplastic[16] or hamartomatous proliferation.[17] It has been suggested that inductive changes may allow progression from one type of odontogenic tumor to another, e.g., ameloblastic fibroma may evolve towards an ameloblastic fibroodontoma.[18]

Two additional forms of ameloblastic fibroma have been described: a clinical variant (peripheral ameloblastic fibroma), and a histopathological variant (granular cell ameloblastic fibroma). Peripheral odontogenic fibroma has been defined as a peripheral odontogenic tumor representing the extraosseous counterpart of central odontogenic fibroma.[15] Granular cell ameloblastic fibroma is in turn composed of nests of odontogenic epithelium surrounded by granular cells.[5,19]

Ameloblastic fibroma has classically been considered an encapsulated tumor - this being a feature shared with other mixed odontogenic tumors.[20-22] As a result, surgical removal or enucleation is the treatment of choice.[23] However, the case described in the present

study involves a number of singular characteristics such as the absence of any consistent capsule, and the segmentation of the tumor mass that prevented habitual surgery (i.e., enucleation) - requiring exeresis and thorough curettage of the bone bed.

Approximately 20% of ameloblastic fibromas can recur after conservative removal. Such recurrences may result from incomplete initial removal,[24] and some surgeons recommend more aggressive surgical excision.[10] Regardless of the therapeutic approach adopted, a patient who has undergone treatment for an ameloblastic fibroma must be subjected to regular follow-up for at least 10 years, since in addition to its high recurrence rate, the possibility exists of malignant transformation.[25] Approximately 50% of all ameloblastic fibrosarcomas develop as a recurrence of ameloblastic fibroma.[10,26]

Reviews of the literature agree on the need for: (a) very accurate histological examination of the lesion; (b) a conservative approach to ameloblastic fibromas except in recurrent cases; and (c) long-term postoperative follow-up.[13,14]

CONCLUSIONS

The classical notion that these tumors are encapsulated and thus easy to remove is somewhat in conflict with the high recurrence rates observed and the potential for evolution towards ameloblastic fibrosarcoma. In our opinion, the best management approach in cases of ameloblastic fibroma in children is to surgically remove the lesion and furthermore perform careful curettage of the bone bed. Periodic follow-up examinations and radiographic studies are advised for at least 10 years after surgery.

REFERENCES

1. Kruse A. Uber die entwicklung cystichen geschwulste in unterkiefer. Arch F Pathol Anat 124: 137-8, 1898.
2. Slootgweg PJ. An analysis of the inter-relatioship of the mixed odontogenic tumors-ameloblastic fibroma, ameloblastic fibro-odontoma, and the odontomas. Oral Surg 51: 266-76, 1981.
3. Gorlin RJ. Odontogenic tumors. In: Gorlin RJ, Goldman HM. Oral Pathology. St Louis: Mosby Company, pp 481-515, 1970.
4. Bhaskar SN. Synopsis of Oral Patholology. 6ª ed. St Louis. CV Mosby pp 273-6, 1981.
5. Kramer IRH, Pindborg JJ, Shear M. Histologic typing of odontogenic tumours (ed 2). Berlin: Springer-Verlag, pp 3-10, 1992.
6. van Wyk CW, van der Vyver PC. Ameloblastic fibroma with dentinoid formation/immature dentinoma. J Oral Pathology 12: 37-46, 1983.
7. Mosbi EL, Russell D, Noren S, Barker BF. Ameloblastic fibroma in a 7 week-old infant: a case report and review of the literature. J Oral Maxillofac Surg 56: 368-72, 1998.
8. Young AH. Ameloblastic fibroma in an infant. J Oral Maxillofac Surg 43: 289-92, 1985.
9. Naidoo LC, Stephen LX. Congenital ameloblastic fibroma in association with oculoauriculovertebral spectrum. Int J Pediatr Otorhinolaryngol 43: 283-8, 1998.
10. Waldron ChA. Odontogenic cysts and tumors. In: Neville B, Damm D, Allen C, Bouquot J. Oral & Maxillofacial Pathology. Philadelphia, W.B. Saunders Company; pp 493-540, 1995.
11. Hoffman S, Jacoway JR, Krolls S. Intraosseous and parosteal tumors of the jaws. Washington, AFIP, pp 113-9, 1987.
12. Trodahl JN. Ameloblastic fibroma: a survey of cases from Armed Force Institute of Pathology. Oral Surg Oral Med Oral Pathol 33: 547-8, 1982.
13. Baroni Ch, Farneti M, Stea S, Rimondini L. Ameloblastic fibroma and impacted mandibular first molar. Oral Surg Oral Med Oral Pathol 73: 548-9, 1992.
14. Taçar F, Tümer C, Ayhan A, Çençift K, CertogluG. A mandibular radiolucency in a 9 year old. Oral Surgery Oral Medicine Oral Pathology 84: 231-3, 1997.
15. Pindborg JJ, Kramer IRH, Torloni H. Histological typing of odontogenic tumours, jaws cysts and allied lesions. Geneva: World Health Organization, pp 29-30, 1971.
16. Philipsen HP, Reichart PA, Praetorius F. Mixed odontogenic tumours and odontomas. Considerations on interrelationship. Review of the literature and presentation of 134 new cases of odontomas. Oral Oncol 33: 86-99, 1997.
17. Brethaux-Bardinon MP, Ferkadji N, Deffez JP. A propos des fibromes amèloblastiques. Rev Stomatol Chir Maxillofac 95: 75-7, 1994.
18. Becker J, Schuppan D, Philipsen HP, Reichart PA. Ectomesenchyme of ameloblastic fibroma reveals a characteristic distribution of extracellular matrix proteins. J Oral Pathol Med 21: 156-9, 1992.
19. Shiro B, Jacoway J, Mirmiran A, McGuirt W, Siegal G. Central odontogenic fibroma, granular cell variant. A case report with s-100 immunohistochemical and a review of the literature. Oral Surg Oral Med Oral Pathol 67: 725-30, 1989.
20. García-Pola Vallejo M, González García M, López-Arranz JS, Herrero Zapatero A. Adenomatoid odontogenic tumor arising in a dental cyst: report of unusual case. J Clin Pediatr Dent 23: 55-8, 1998.
21. Owens BM, Schuman NJ, Micer HH, Turner JE, Oliver FM. Dental odontomas. J Clin Pediatr Dent 21: 261-4, 1997.
22. Ozer E, Pabuccuoglu U, Gunbay U, Saioglu S, Aktas S. Ameloblastic fibro-odontoma of the maxilla: case report. J Clin Pediatr Dent 21: 329-31, 1997.
23. Dallera P, Bertoni F, Marchetti C, Bachini P, Campobassi A. Ameloblastic fibroma: a follow-up of six cases. Int J Oral and Maxillofac Surg 25: 199-202, 1996.
24. Monteil RA, Le Blas, Favot C, Loubiere R. Stereologic analysis of histologic parameters of a twice-recurrent ameloblastic fibroma. Oral Surg Oral Med Oral Pathol 61: 168-72, 1986.
25. Muller S, Parker DC, Kapadia SB, Budnick SD, Barnes EL. Ameloblastic fibrosarcoma of the jaws. A clinicopathologic and DNA analysis of five cases and review of the literature with discussion of its relationship to ameloblastic fibroma. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 79: 464-77, 1995.
26. Oliveira Nogueira T, Rodarte Carvalho Y, Blumer Rosa LE, Murilo Dos Santos L. Possible malignant transformation of an ameloblastic fibrosarcoma: a case report. J Oral Maxillofac Surg 55: 180-2, 1987.