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Ameloblastic Fibroma of the Maxilla, Report of a case

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Ameloblastic fibroma is a kind of odontogenic mixed tumor which shows remarkable proliferative changes. The tumor contains both, the epithelial and mesenchymal neoplastic elements; the former originating from outer enamel epithelium or the rest of dental lamina and the latter from dental papilla or its surrounding connective tissue.

Being relatively uncommon in occurance, its clinical and histological differentiation from ameloblastoma and ameloblastic odontoma is interesting but difficult. The following is a case of histologically proved ameloblastic fibroma developed in the maxilla.

Report of a case :

A patient, 10 year old Japanese girl was admitted to the dental hospital in June 6, 1975 with the chief complaint of painless swelling on the left side of the face (Fig. 1). Nothing particular was noticed in the past history of family background.

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The patient had had a trauma in about February 1975, on the left buccal region, resulting in echimosis for which no treatment was received. Two months later she noticed the swelling. On physical examination the naso-labial sulcus had almost disappeared because of the diffuse swelling which had a reddish colour and elastic firm consistency. There were neither signs of inflammation, orbital symptoms or nasal obstruction.

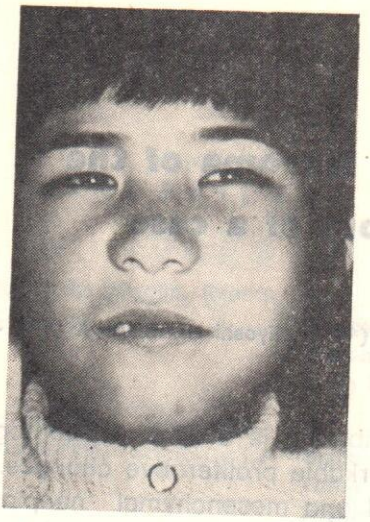

Fig. 1 : Swelling on the left side of the face.

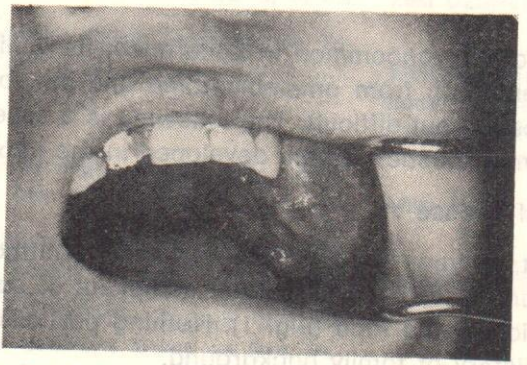

Fig. 2 : Intraoral appearance of the tumor.

Intraorally, except the diffuse swelling on the buccal gingiva (Fig. 2), the mucosa was healthy and there was no ulcer formation.

The neighbouring teeth diverted mesially and distally, being all in vital condition.

Roentgenographic examination :

The impacted second molar and adjacent large radiolucent area with well defined borders were clearly visible (Fig. 3).

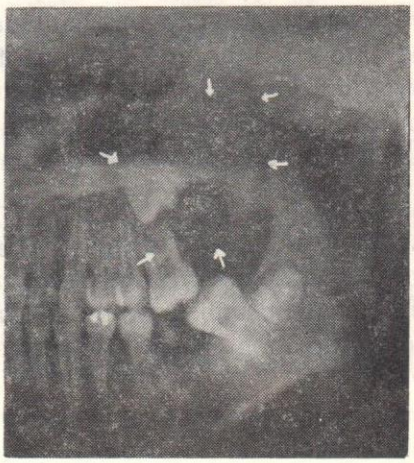

Fig. 3 : The cavity of the tumor on panaromic x-ray.

This cystic cavity had a fine bone structure with radiopaque material scattered here and there (Fig. 4). The lateral side of alveolar bone between first bicuspid and the first molar teeth was completely destroyed.

Fig. 4 : Dental x-ray of the involved area.

In August 8, 1975, under local anesthesia a biopsy was performed through an incision along the alveolar ridge. The result was reported as ameloblastic fibroma by the pathologist.

Treatment and course :

No active treatment was carried out for some time in order to maintain the normal limits of bleeding time which seemed to be disturbed in routine laboratory examinations. The patient then, in mid- January 1976 was admitted to inpatient department. Under general anesthesia through endotracheal intubation, the operation was performed by an intraoral approach. Surgical exploration disclosed a large, firm mass completely filling the maxillary sinus. The tumor was intact with orbital wall superiorly and tuber maxilla posteriorly.

After the enuclation of the tumor, the entire bony surface of the sinus was curetted and filled with gauze of antibiotic ointment. The prognosis was good and until now there was no evidence of recurrence. The follow-up roentgenograms showed the gradual decrease of the cavity.

Histopathological studies :

Cords and islands of epithelial cells, showing various developmental stages from dental lamina to enamel organ, were proliferated in the stroma of mesenchymal connective tissue. The cords or strands of cuboidal cells were two or three layers in thickness and resembled the dental lamina. The end portions of the cords frequently showed round or oval swellings and occosi onally han some resemblance to enamel organ. Rosette like arrangements of epithelial cells were observed in the transverse section of cords (Fig. 5).

The cells of central area had eosinophilic cytoplasms with intercellular bridges. Microcyst formation within the islands were often observed and there were little atypisms of odontogenic epithelial cells.

The connective tissue element took the form of a very cellular fibroblastic tissue resembling the dental lamina in the developing tooth, though in some areas of the connective tissue, there was cystic degeneration of mesenchymal tissue associated with a decrease of cellular population. Little atypism of the mesenchymal tumor cells were observed. The proportion of the epithelial element

to the mesenchymal element varied from one portion to another in the tumor tissue. There were some roundish calcified bodies in the tumor tissue, which we could not determine whether they were odontogenic products or not.

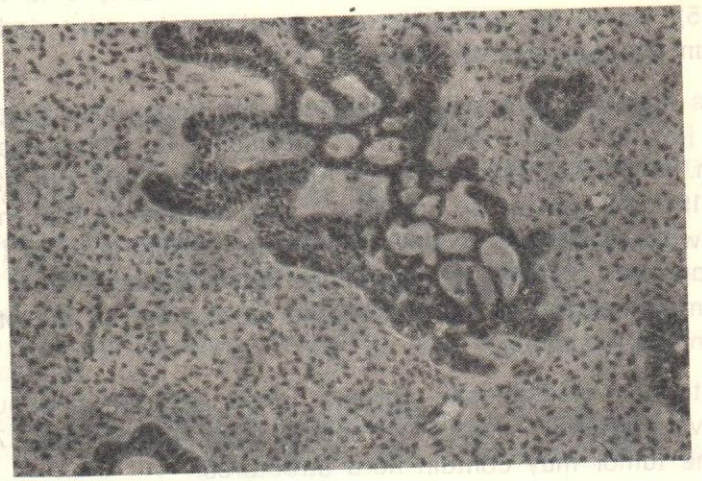

Fig. 5 : General microscopic appearance of the tumor (H. E. x 100).

Discussion :

The place and the name of this tumor has varied considerably in the classifications of odontogenic tumors since Broca (1).

Thoma and Goldman (15), employed the term «soft odontoma» and defined it as a mixed odontogenic tumor. It appeared with the name of «ameloblastic fibroma» in Bernier's classification (13). In 1958 Pindborg and Clausen (12), divided the odontogenic tumors into two, where ameloblastic fibroma was mentioned as epithelial tumor with inductive changes in connective tissue. This classification was mainly based on presumed theory of induction, that is, the inductive effect of embryologic epithelial tissue on the mesenchyma, as also noted by other workers (4, 13).

The incidence of ameloblastic fibroma is relatively small in comparison to ameloblastoma. In addition to the previously collected 28 cases by Carr and co-workers (3), 6 cases of ameloblastic fibroma were written up in Japanese literature between 1960. However the histomorphological and roentgenologic similarities of

ameloblastic fibroma to ameloblastoma may lead to erraneous diagnosis resulting in the radical resection of the involved jaw bone. In fact it has been stated before that roentgenologic differentation from unilocular ameloblastoma is impossible (5, 15).

Patients with ameloblastic fibroma are usually 5 to 20 years old (15), where as ameloblastoma developes in older individuals, with an avarage age of 38-39 (6).

As observed in previous literature, the tumor may develope on either jaw, but more frequently on the mandible, without sex predilection. In the statistical data of Armed Forces Institute of Pathology (1937-69), out of 24 patients it was located on the mandible in 21, while found only to develope on the maxilla in three (16). Out of 6 Japanese patients, 5 were males and one was female. In three of them te tumor was located on the maxilla and in the other 3 it was on the mandible.

Although ameloblastic fibroma cases reported previously did not have any hard tissue, more recently Cina (4) and Villa (17) noted that the tumor may contain hard structures.

It was suggested by Cahn and Blum (2) that ameloblastic fibroma may start producing calcified odontogenic structures and mature into ameloblastic odontoma, if allowed to remain. In fact, in Trodahl's (16) two recurrent cases, the maturational progression was evident. On the other hand according to Carr (3) and Mitsui (10) there was no histopathologic change in their cases of recurrence.

We identified in the tumor tissue some small calcified stuructures roentgenologically and histologically but could not distinguishe if they were odontogenic or not. In fact at present there is no positively efficient method to determine. We consider that there are some cases of ameloblastic fibroma maturing by aging towards ameloblastic odontoma and/or odontmoa, and unless the exact nature of these calcified structures determined it seems difficult to classify this tumor as separate entity, therefore it seems more beneficial at present to accept Thoma's classification (15).

ÖZET

10 yaşındaki bir kız hastanın üst çene büyük azı dişleri yakınında oluşan bir ameloblastik fibroma olgusu sunulmuş ve az görülen bu odontojenik tümör ile ilgili yayınlar etraflıca gözden geçirilmiştir.

SUMMARY

A case of ameloblastic fibroma is presented in which the tumor was located on the maxilla of a 10 year old female patient; and the literature related to this uncommon odontogenic tumor is reviewed in detail.

LİTERATÜR

1 — **Broca, P.** : Traite des Tumors, Paris, 1866, P. Asselin, p. 350.

2 — **Cahn, L. R., and Blum, T.** : Ameloblastic odontoma; case report critically analyzed, J. Oral Surg. 10: 169, 1952.

3 — **Carr, R. F., Halperin, V., Wood, C., Krust, L., and Schoen, J.** : Recurrent Ameloblastic Fibroma, ORAL SURG. 29: 85, 1970.

4 — **Cina, M. T., Dahlin, D. C., and Gores, R. J.** : Odontogenic Mixed Tumors, A Review of the Mayo Clinic Series, Proc. Staff Meet., Mayo Clin. 36: 664, 1961.

5 — **Gorlin, R. J., Chaudry, A. P., and Pindborg, J. J.** : Odontogenic Tumors. Classification, Histopathology and Clinical in men and domesticated animals, Cancer 14: 73, 1961.

6 — **Grenfell, J. W., and Maris, A. M.** : Ameloblastic Fibroma. Report of a case, ORAL SURG. 21: 403, 1966.

7 — **Kojima, T., Nagase, H., Okamoto, E., Kojima, M., Fujimoto, J., Shimizu, T.,** and Shimizu, T. : A Case of Ameloblastic Fibroma, originated in the maxilla, Jap. J. Oral Surg. 20: 385, 1974.

8 — **Matsumoto, Y., Inaba, O., and Yamomoto, S.** : A Case of Ameloblastic Fibroma, J. Jap. Stomat. 20: 829, 1971.

9 — **Mito, T., Kimura, N., Fujil, S., and Tanaka, S.** : A Case of Ameloblastic Fibroma, Jap. J. Oral Surg. 10: 309, 1964.

10 — **Mitsui, Y., Kimura, N., Mizuno, Y., and Tanaka, S.** : Discussion of Histopathologic Changes in a Case of Recurrent Ameloblastic Fibroma, Jap. J. Oral Surg. 12: 226, 1963.

11 — **Nii, S., Higa, S., and Sonoyama, N.** : A Case of Ameloblastic Fibroma, Jap. J. Oral Surg. 20: 42, 1974.

12 — **Pindborg, J. J., and Clausen, F.** : Classification of Odontogenic Tumors; A suggestion, Acta Odont. Scandinav. 16: 293, 1958.

13 — **Sedano, H. O.** : Ameloblastic Fibroma; Report of two cases, ORAL SURG. 17: 475, 1964.

14 — **Seo, M., Okada, T., Yamomoto, Y., and Ono, S.** : A Case of Ameloblastic Fibroma, Originated in the Maxilla, J. Jap. Stomat. 23: 239, 1974.

15 — **Thoma, K. H., and Goldman, H. M.** : Odontogenic Tumors; Classification based on observations of epithelial, mesenchymal and mixed varieties, Am. J. Path. 22: 433, 1946.

16 — **Trodahl, J. N.** : Ameloblastic Fibroma; A survey of cases from the Armed Forces Institute of Pathology, ORAL SURG. 33: 547, 1972.

17 — **Villa, V. G.** : Ameloblastic Fibroma in the Mandibl, ORAL SURG. 15: 82, 1962.