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Abstract

A case of the so-called adenoameloblastoma developed in the right maxillary sinus of a 10-y-old girl was reported. The histological features of this tumor were discussed in detail. In the twenty cases of adenoameloblastoma, including the present case, reported in Japan up to the present, some statistic investigations have been made in regard to the clinical aspects.

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THE SO-CALLED ADENOAMELOBLASTOMA REPORT OF A CASE

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It is well-known that the so-called adenoameloblastoma is a rare odontogenic tumor and recognized as a histologic variant of ameloblastoma. More than fifty cases of this tumor have been described in the Western literature, while in Japan at least nineteen such cases have been reported up to the present. This article deals with another case encountered by the author.

CASE REPORT

Clinical findings: J. K., a 10-year-old Japanese girl, was admitted to the Okayama Red Cross Hospital, complaining of a swelling of the right cheek with slight tenderness, which persisted for about one month. Oral examination revealed a diffuse fluctuant nontender expansion in the right upper anterior region. X-ray examination disclosed the tumor occupying the right maxillary sinus and the permanent canine erupted in the opposite direction into the same sinus (Fig. 1).

At the operation the anterior part of the maxillary bone was found to be quite thin due to compression of the tumor. The tumor filled up the right maxillary sinus and embedded the most part of the mis-erupted canine. The inferior part of the maxilla was somewhat destroyed but there was no adhesion between the tumor and the bones, so that the tumor was easily extirpated together with the associated tooth. No recurrence is noticed at present about one year after operation.

Macroscopic findings of the extirpated tumor (Fig. 2): The tumor was pigeon's egg-sized, with a well-defined fibrous capsule, through which the apex of the canine extruded. The cut-surface presented a large cystic cavity, containing approximately 6 ml of a thin straw-colored fluid. The inner surface of the cavity gave a variegated, dark-red and grayish-white, roughly granular appearance. Most part of the wall was less than 5 mm in thickness and mainly composed of grayish-white, solid tissue with small hemorrhagic foci, while some areas in the lateral side of it appeared like a thin membrane.

Histologic findings: Serial paraffin sections stained with hematoxylin-eosin, van Gieson's stain, Mallory-Azan's stain, Pap's silver impregnation, mucicarmine, PAS-reaction with or without previous digestion by diastase and Kossa's stain were examined.

Microscopic examination revealed the typical pattern of the so-called adenameloblastoma. A few places of the cystic cavity were lined by the thin-layered stratified squamous epithelium similar to that of dentigerous cyst (Fig. 3). The solid part of the tumor was composed of sheets of the epithelial cells with scanty connective tissue stroma. Characteristic duct-like structures comprised a central lumen lined by tall columnar cells in a single layer scattered throughout the tumor (Fig. 4). The cytoplasm of these columnar cells was clear or finely granular, and the nuclei were located away from the central lumen (Fig. 7). The lumina contained some fibrillar or homogenous, eosinophilic materials attached to the free-margin of the columnar cells, occasionally in a cuticle-like fashion. In other places, the epithelial cells formed convoluted or whorl-like structures consisting of double strands of columnar cells without central lumen (Fig. 5). Eosinophilic homogenous material was also found in the narrow space between these strands facing each other. Infrequently, a direct continuity could be traced between the central lumen of the duct-like structure and the stromal tissue, in which degenerated blood vessel-like structure could be very rarely observed (Fig. 6).

The remaining epithelial cells were stellate or spindle-shaped, with clear cytoplasm and oval nuclei, forming solid sheets among the structures of columnar cells. The spindle-shaped cells often oriented as if they were stromal cells, and in looser areas they gave an appearance resembling stellate reticulum (Fig. 9). Besides these, the epithelial cells formed a few small clumps consisting of large polyhedral cells with eosinophilic cytoplasm and clear cellular boundaries, suggesting squamous metaplasia (Fig. 10). Few mitotic figures of the epithelial cells were observed.

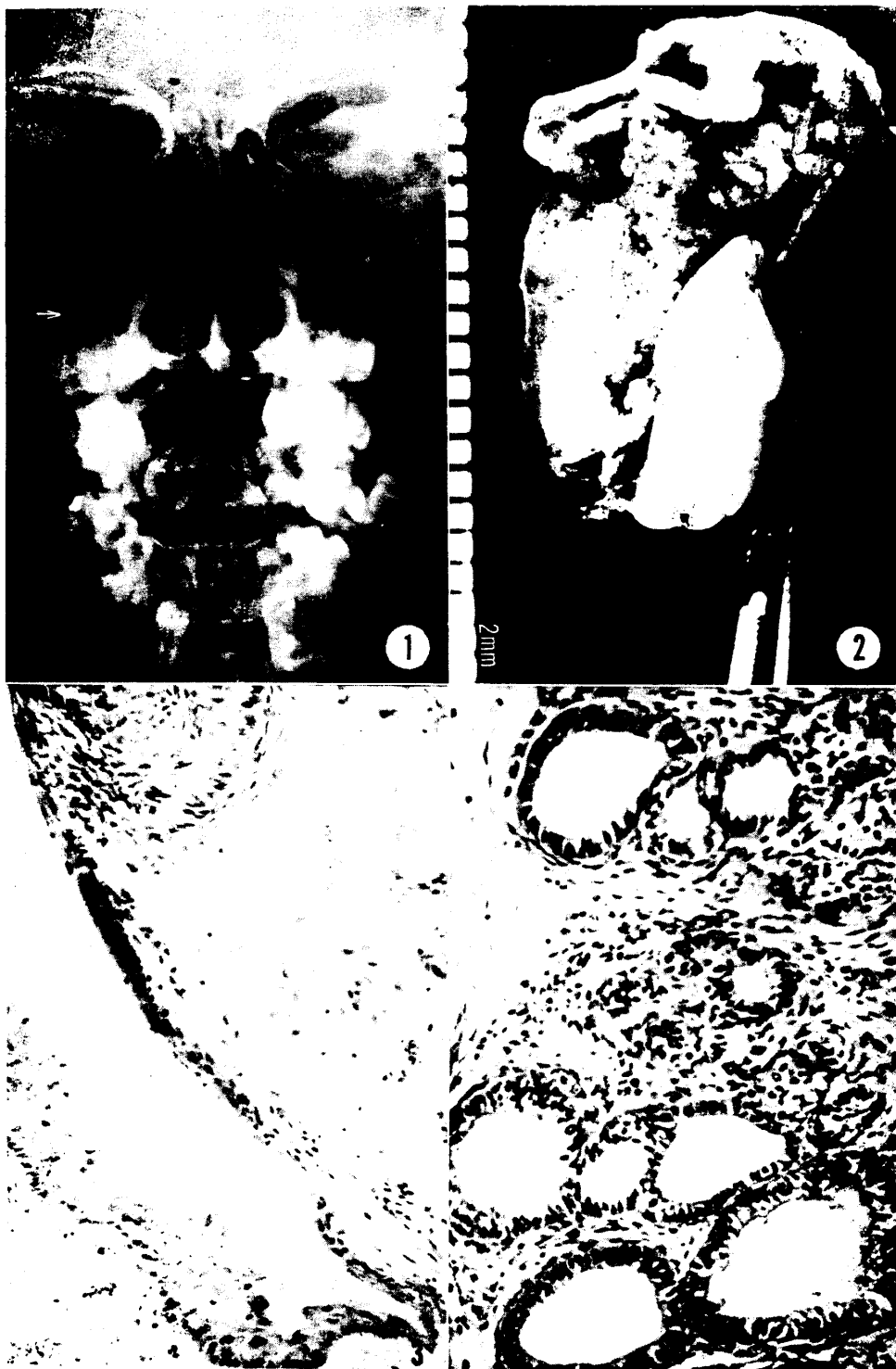
The stromal tissue occasionally underwent degeneration, forming small cystic spaces enclosed by slender strands of epithelial cells (Fig. 12). Within these stromal cysts some eosinophilic, homogenous or hyaline droplet-like ma-

Fig. 1 X-ray film taken before operation. The right maxillary sinus appears diffusely opaque and the canine erupted into the same sinus (↑) is observed.

Fig. 2 The cut-surface of the extirpated tumor, showing a large cystic cavity. The inner surface of the cavity gives roughly granular appearance. Note the association with the canine.

Fig. 3 The thinner portion of the cyst wall. The inner surface is lined by the thin-layered stratified squamous epithelium. This area shows a transition between the epithelial lining and the tumorous tissue. H-E. Low-power magnification.

Fig. 4 Note a number of the duct-like structures with scanty stroma, the most characteristic pattern. H-E. Low-power magnification.





terials were contained and in some places blood vessels and small hemorrhages were also found.

Small calcified bodies scattered throughout the tumor were found (Figs. 10, 11). They were small ovoid or irregularly fragmented, and the larger ones gave somewhat lamellar appearance. Infrequently, a few calcified bodies were observed within the duct-like lumen. Eosinophilic, non-calcified, droplet-like materials were also recognized within the epithelial sheets.

The eosinophilic material within the duct-like lumen was argyrophilic (Fig. 8), PAS-positive and weakly positive to mucicarmine, and stained reddish with van Gieson's stain and blue with Mallory-Azan's stain. Basement membrane could not be demonstrated between the columnar cells and the epithelial cells surrounding the duct-like structures. The solid areas seemed to be avascular but a few delicate argyrophilic fibers and some elements stained blue with Mallory-Azan's stain were observed among the epithelial cells. Within the cytoplasm of the epithelial cells, especially of the columnar cells, PAS-positive granules could be demonstrated, but no mucicarmine-positive substance. These PAS-positive granules disappeared after digestion by diastase. The calcified bodies revealed a positive reaction of Kossa's stain. Non-calcified eosinophilic materials within the epithelial sheets were argyrophilic and PAS-positive, and stained reddish with van Gieson's stain and bluish or partially dark-reddish with Mallory-Azan's stain. The calcification seemed to be deposition of inorganic substances around these non-calcified materials.

COMMENT

The so-called adenoameloblastoma has been recognized as a rare histologic variant of the ameloblastoma by many authors, though the designation has not been generally accepted. It has been described in the literatures under varying nomenclatures, such as pseudoadenoma adamantinum⁷, glandular adamantinoma¹⁶, epithelial tumors associated with developmental cyst³⁵, cystic complex composite odontome²², teratomatous odontoma⁴, pleomorphic adenoma-like

Fig. 5 A convoluted tubular structure showing invagination of the columnar cells. H-E. Medium-power magnification.

Fig. 6 A direct connection between the duct-like lumen and the stromal tissue. A degenerated blood vessel-like structure is observed within the lumen. H-E. High-power magnification.

Fig. 7 Medium-power photomicrograph demonstrating the duct-like structures consisting of single-layered columnar cells with nuclei placed away from the central lumen. Eosinophilic fibrillar material attached to the free-ends of the columnar cells is seen. H-E.

Fig. 8 The luminal content is argyrophilic, and attaches to the inner surface of the duct-like structures. Note the absence of basement membrane between the columnar cells and the spindle-shaped epithelial cells surrounding the duct-like structure. Pap's silver impregnation. Medium-power magnification.



tumor²⁶, tumor of enamel organ epithelium¹⁸, adenomatoid ameloblastoma^{10,15}, ameloblastic adenomatoid tumor⁵, pseudoadenomatöser Typ des Adamantinoms¹⁴ and so on. Since BERNIER and TIECKE² published a review of nine cases of this tumor as adenoameloblastoma, this term has been most commonly accepted^{1,3,6,8,9,17,19,23,27,30,31,38-42}.

In Japan, the first reference to this lesion was made by MASAKI²¹ in 1939, and then NAKAGO and KAWAKATSU²⁴ reported a further example as "ameloblastoma arose in follicular cyst". Since ITO *et al.*¹¹ reported a case as adeno-ameloblastoma, some cases have been added by several authors^{10,12,13,20,25,26,29,33,34,36,37}, and nineteen cases have been reported in Japan up to the present.

It is well-known that the adenoameloblastoma shows somewhat different clinical features than those of the usual ameloblastoma. It is believed in general that adenoameloblastoma occurs nearly twice as commonly in females, quite frequently in the second or third decade, and rather frequently in the upper jaw, especially in the anterior region, and that it is often associated with unerupted tooth. While the usual ameloblastoma predominantly affects male, commonly occurs in the fourth decade and most frequently develops in the lower molar region.

The investigation of nineteen cases collected in Japanese literature and the author's additional case may briefly be described as follows: Fourteen (78%) of eighteen cases, exclusive of two cases of MASAKI²¹ with incomplete clinical data, were females, and four (22%) were males. Seventeen (94%) of eighteen patients were between ten and twenty-two years of age, and another one was thirty years old male. Thirteen (65%) of twenty had the tumor associated with unerupted tooth. Thirteen (65%) had the lesion in the upper jaw and eleven of them had the tumor in the anterior region, while five of seven had in the anterior region of the lower jaw. Most of adenoameloblastoma are detected as a tumor in the jaw-bones. The cases recognized as tumor that occupied the maxillary sinus amounted to only five; namely, Case 5 of BERNIER and TIECKE², the case of KANEKO *et al.*¹², the case of LENTRODT and SHIMIZU¹⁵ and the case presented in this article.

Most authors have pointed out the resemblance of the clinical behaviors of

Fig. 9 A loose area of spindle-shaped epithelial cells, resembling stellate reticulum of usual ameloblastoma. H-E. Medium-power magnification.

Fig. 10 A small epithelial cluster consisting of large polyhedral cells with eosinophilic cytoplasm and clear cellular boundaries, suggesting squamous metaplasia. Arrow indicates a small calcified focus. H-E. High-power magnification.

Fig. 11 Small calcified bodies among the epithelial sheets. The larger ones give somewhat lamellar appearance. H-E. Medium-power magnification.

Fig. 12 The cystic spaces due to degeneration of stromal connective tissue, enclosed by slender strands of epithelial cells. H-E. Low-power magnification.

this tumor to those of the dentigerous cyst. Actually most cases have been clinically diagnosed as dentigerous cyst and some of the tumors really developed in association with dentigerous cyst. It is probable that this tumor may occur more frequently than reported, because it would be liable to be overlooked as simple dentigerous cyst.

This lesion is said to be benign one and a simple enucleation is the treatment of choice. No recurrent case among the established examples of the adenoameloblastoma has been reported, while the recurrence or invasive character of the usual ameloblastoma is not infrequent.

As regards the histogenesis of the adenoameloblastoma WILLIS⁴⁸ suggested the resemblance between this tumor and the pleomorphic tumors of the salivary glands, and STAFNE³⁵ speculated the origin to be of epithelial remnants in the closure of the globular and maxillary processes. Several investigators^{26, 28, 39} explained that the parenchyma of this tumor is derived from the primitive oral epithelium and that the latter is able to produce not only dental organ but also glandular tissue in the course of its differentiation. However, the glandular origin of this tumor has not been generally accepted. Most commonly it has been considered that this tumor is of odontogenic origin because of a close resemblance of its columnar cells forming the duct-like structures to the ameloblasts and of its frequent association with unerupted tooth. The actual origin of this tumor in the dental tissue is still undetermined. Theoretically it is possible to assume that this tumor has its origin in the enamel organ or its remnants, the primitive oral epithelium, or the lining epithelium of dentigerous cyst, as well as in the usual ameloblastoma, but none of them is completely satisfactory to explain the histogenesis of this tumor.

In the present case the tumor appeared as large cystic one and most of the cystic wall composed of the tumor tissue, while in a few areas the inner surface of the cavity was lined with non-neoplastic stratified squamous epithelium in a thin layer. The tumor tissue had likely proliferated to replace the lining epithelium of a dentigerous cyst. These findings may suggest that this tumor arises in the wall of the dentigerous cyst, but it is yet premature to conclude that the actual origin of the tumor is the lining epithelium of dentigerous cyst.

The histologic pattern of the adenoameloblastoma appears to be considerably different from that of the usual ameloblastoma. However, a thorough histologic examination of the present case revealed that the columnar cells forming the duct-like structures closely resembled the ameloblasts (Fig. 7), and, moreover, the solid areas included the features suggestive of the simulation of stellate reticulum (Fig. 9) and even squamous metaplasia (Fig. 10) as already described. These findings seem to suggest the origin of this tumor to be the remnants of enamel organ epithelium as LUCAS¹⁸ and BHASKAR³ stated. Nevertheless, it is

reasonable to separate adenoameloblastoma from usual ameloblastoma because of differences in clinical and histo-pathological respects.

On the characteristic duct-like structures no one supports the true glandular origin at present. LUCAS¹⁸ explained that these structures represent an abortive attempt at the formation of enamel organ in a fashion of cystic follicles and the columnar cells was polarized in the opposite direction within the epithelial clusters to the ameloblast-like cells in ameloblastoma, in which these cells are arranged in the periphery of the epithelial follicles, as an analogous differentiation to epithelial pearls in the keratinizing squamous cell carcinoma. The eosinophilic material within the duct-like lumen has been considered to be enamel-matrix or pre-enamel by many authors^{8, 18, 19, 22, 35, 41} because of its close relationship to the ameloblast-like cells and of the presence of acid mucopolysaccharides⁸. SHEAR³² suspected the homogenous material to be pre-dentine matrix.

ISHIKAWA and MORI¹⁰ demonstrated the direct transition between the duct-like lumen and the degenerated stroma and considered that the lumen corresponds to the stromal cyst, containing degenerated mesenchymal substances. Furthermore, they described in the same article that the flattened epithelium around the columnar cells of the duct-like structures presented a distinct positive reaction for alkaline phosphatase, showing an identical appearance as the stratum intermedium of the normal tooth germ, while the reaction was usually negative in the epithelial portion of ameloblastoma. The similar results were also presented by SHIMIZU and KOMORI³³. This fact is of notice because the stratum intermedium is believed to play an important role in normal amelogenesis.

The author also found an apparent connection between the duct-like space and the stromal tissue, and very rarely, even the vascular involvement within the abortive lumen (Fig. 6). The staining characteristics of the luminal contents actually suggested the presence of mesenchymal components. Argyrophilic fibrils in contact with the free-ends of the columnar cells gave an appearance just identical with the basement membrane (Fig. 8). Positive reaction with mucicarmine does not always imply the presence of mucous substance secreted by epithelial cells because mucicarmine-positive material is possibly able to originate from degenerated mesenchymal tissue.

The author holds the same view as do ISHIKAWA and MORI in regard to the interpretation that the duct-like lumen represents the stromal cyst. In view of this it is no wonder that the columnar cells are arrayed in a single layer like a gland in the middle of the epithelial sheets, although it is still obscure why few columnar cells are arranged in the periphery of the epithelial clusters.

In most of the cases so far reported a number of small calcified spherules were found scattered throughout the tumor, occasionally even within the duct-like lumen, and in some instances they formed larger masses. The genesis of

the calcification is still under discussion. LUCAS¹⁹ supposed that they might also be of enamel nature and OEHLERS²⁷ stated that they resembled enameloid tissue. CAHN⁴ believed them to be cementicles. MILES²² noticed the presence of both dentine-matrix and mesodermal pulp tissue in his case and regarded the tumor as cystic complex composite odontome. However, GORLIN and CHAUDHRY⁸ and ISHIKAWA and MORI¹⁰ were of the opinion that they were probably dystrophic calcification. In the present case the calcification appeared to be formed around the eosinophilic non-calcified materials derived probably from the degenerated epithelial and mesenchymal cells. Accordingly, the mode of the calcification is likely to be dystrophic.

Finally, the author would like to suggest the need for a change of nomenclature to a more appropriate term, for instance, "adenomatoid ameloblastoma" advocated by ISHIKAWA and MORI¹⁰.

SUMMARY

A case of the so-called adenoameloblastoma developed in the right maxillary sinus of a 10-y-old girl was reported. The histological features of this tumor were discussed in detail.

In the twenty cases of adenoameloblastoma, including the present case, reported in Japan up to the present, some statistic investigations have been made in regard to the clinical aspects.

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