

DESMOPLASTIC AMELOBLASTOMA

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CONTENT

- I. CASE REPORT
- II. DISCUSSION
- REFERENCE
- ABSTRACT

The ameloblastoma is a true neoplasm of enamel organ-type tissue which does not undergo differentiation to the point of enamel formation, and the term "adamantinoma" which implies the formation of hard tissue was replaced by "ameloblastoma" since this tumor is not associated with the formation of hard tissue.(1)

Recently I experienced a case of ameloblastoma which showed atypical radiographic appearance which could not be explained by any of the known bone destruction patterns of ameloblastoma. Rather the lesions looked like a fibro-osseous lesion.

The result of biopsy was quite strange not only to the radiologists but also to the surgeons who operated the lesion. The suspicion could only be explained by finding a report(2) by Japanese who experienced a similar case.

I. CASE REPORT

A 33-year-old Korean woman referred to

Pusan National University Hospital because of a recurrent bony swelling of right mandibular body. The patient's past history revealed that hard swelling of the right mandible was first discovered five years ago. She sought the care of an otolaryngologist who made a diagnosis of fibrous dysplasia. The patient had undergone the contouring of right mandible three years ago. Her medical history was VPC (ventricular premature contracture) on EKG, and otherwise unremarkable.

Clinical examination(Fig. 1) revealed a slow growing hard painless mass with buccolingual expansion of right mandibular body.



Fig. 1. Clinical picture shows buccal expansion of right mandibular body with normal overlying mucosa.

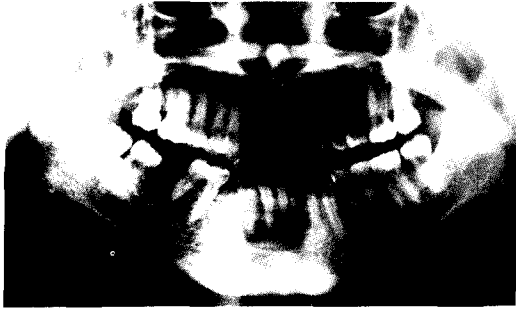


Fig. 2. Panoramic view shows ill-defined radiolucent-radiopaque mixed lesion extending from left mandibular central incisor to the extracted right mandibular first molar area.



Fig. 3. Occlusal view shows buccal expansion of the lesion.



Fig. 4. Periapical views show the radiolucent-radiopaque mixed lesion in detail which resembles a fibro-osseous lesion.

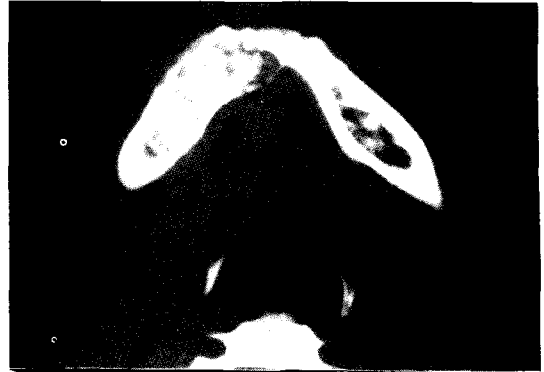


Fig. 5. The computed tomography shows the calcified materials within the lesion and the loss of buccal cortical plate which could be explained by the previous bony contouring surgical procedures.

The overlying mucosa was normal and there was slight facial asymmetry. The involved teeth was mobile but there was no apparent sensory disturbance of the right inferior alveolar nerve or evidence of lymph node enlargement.

Radiographic examination revealed an ill-defined radiolucent-radiopaque mixed lesion which extended from left mandibular central incisor to the extracted right mandibular first molar area.(Fig.2)

The lesion showed a buccal and lingual expansion(Fig. 3) and the involved teeth, #43-45, showed displacement with some loss of lamina duras(Fig. 4). The axial and coronal CT scans(Fig. 5) clearly showed the calcified materials within the lesion and the loss of buccal cortical plate which could be explained by the previous bony contouring surgical procedure. Radiographic impression was suggestive of a fibro-osseous lesion.

Since the bone biopsy revealed an ameloblastoma, the lesion was resected under general anesthesia after medical control of the VPC. The mandible was reconstructed immediately with Titanium-mesh application with



Fig. 6. Numerous follicles of ameloblastoma surrounded by bony structures. (HE stain, x25)

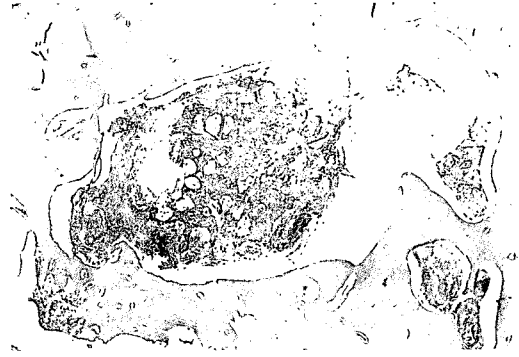


Fig. 8. Some areas of the tumor are arranged in sheet form. (HE stain, x25)

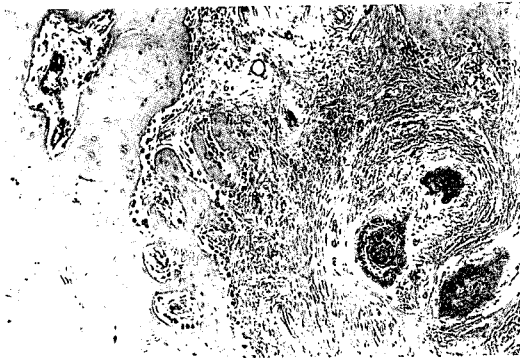


Fig. 7. Bony trabeculae surrounding the ameloblastoma nests are mature and interconnected. Rimming osteoblasts show proliferating activity. (HE stain, x100)

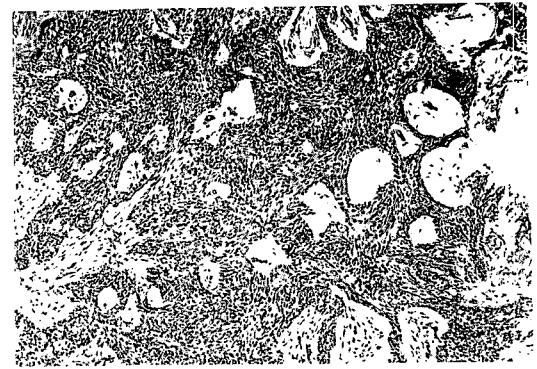


Fig. 9. High power view of Fig. 3. The tumor shows plexiform type ameloblastoma. (HE stain, x200)

iliac PMCB (particulated marrow cancellous bone) packing.

The pathologic features on gross examination, the resected mandible measured 5x3x1.2cm in dimensions. Discolored lesion was identified on the lingual surface of mandible and measured 4x2.2cm in its extents. Microscopically, numerous epithelial nests or islands surrounded by abundant bony tissue were seen (Fig. 6). The area of each epithelial nest was composed of loosely arranged cells and thus resembled the stellate reticulum. But most of them were more solid than the ordinary folli-

cular ameloblastoma and constituent cells showed squamoid differentiation. But keratinization or definite keratin pearl formation was not seen. This area was surrounded by a peripheral single row of columnar cells resembling ameloblasts in which the nuclei were polarized away from the basement membrane. Quite frequently a zone of hyalinization were seen immediately adjacent to and surrounding the follicles. Numerous bony trabeculae connected each other were lined by a number of active osteoblasts. Lamellar bones surrounding tumor nests were also sufficient (Fig. 7, 8, 9).

II. DISCUSSION

Ameloblastomas do not generally show evidence of induction(3), however, rare cases associated with enamel and dentin are referred to as odontoameloblastomas which is different from the ameloblastic fibro-odontoma in histologic pattern and clinical behavior(3-5). Slabbert et al.(3) reported an ameloblastoma with dentinoid induction and named it as dentinoameloblastoma.

Okada et al.(2) reported a case of ameloblastoma accompanied by prominent bone formation and remarked four characteristics of these lesions found in Japanese medical literature.

Our case seems to have similar radiographic features as the case described by Okada et al and meets the characteristics in that the lesion site is anterior portion of the jaw, originated in the central area, and solid ameloblastomas with squamous metaplasia and fibrous connective stroma. Also there are many bone trabeculae lined by a number of active osteoblasts which could be explained by the marked inductive change of the odontogenic epithelium in connective tissue.(6)

Ashman et al.(7) reported a case of desmoplastic ameloblastoma which is a rare histologic variant of ameloblastoma. This too frequently develops in the anterior mandible or maxilla and may mimic benign fibro-osseous lesions in their radiographic appearance. The histologic pattern is characterized by islands of ameloblastic-appearing columnar cells that surround spindle-shaped cells that resemble stellate reticulum and a stroma with marked desmoplasia. *Later I found some more reports of desmoplastic ameloblastoma which shows similar patterns as my case which made me think that this case and the cases mentioned by Okada et al(2) were also the desmoplastic*

ameloblastomas.

In view of the radiologists, it is quite a diagnostic dilemma that there are some ameloblastomas which show bone-forming fibro-osseous lesion patterns.

Though it is said(8) that the anterior portion of the jaw is not a common site for the formation of ameloblastoma and that tumors in this region are less extensive than those in the posterior portion, still it is the ameloblastoma which shows recurrence after treatment other than total resection.

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ameloblastoma는 일반적으로 경조직을 형성하지 않는 진성 종양으로 알려져 있으나 최근에 저자는 종양내에 골형성을 수반한 경우의 ameloblastoma를 경험하고 이것을 보고하였다. 이 드물게 나타나는 ameloblastoma는 desmoplastic ameloblastoma로 명명되고 있으며 방사선 사진상으로 fibro-osseous lesion 과 감별을 요한다.