A Clinical Case of Keratinizing and Calcifying Odontogenic Epithelial Tumor in the Upper Jaw

by

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In recent years, there have been published clinical reports dealing with cysts or tumors of the odontogenic character the extract of which revealed pathologic keratinizing or calcifying changes in the light of histopathologic examination [1, 6, 8, 9, 10, 11, 12]. These pathologic changes are said to give quite different histopathologic findings from those of the radicular cyst, follicular cyst or cystic ameloblastoma.

The case of an upper tumor to be described here is one which was diagnosed as a clinical osteofibroma but, upon its histopathologic examination, the extract was found to be what could be termed as keratinizing and calcifying odontogenic epithelial tumor.

Clinical Record

1. Patient: Y. M, aged 23, male, employed as a clerk in a company.
2. Initial examination: June 23rd 1965.
3. Chief complaint: A large swelling extending from the buccal region of 3 4 5 to the palate.
4. Familial background: Nothing particularly noteworthy.
5. Previous clinical history: About 2 years before, the patient underwent an operation for the duodenal ulcer.
6. Current clinical history: About one year ago, the patient came under a care by a general practitioner for a bean-sized tumor on the left upper jaw in the locality of cuspid. The tumor was resected and as a result, it had disappeared and cured. The patient had no further subjective symptom and when he was treated for the dental decay early this year, his attention was drawn to a swelling around 3 4 5 region. The patient was referred to us by the dentist who had attended him.
7. Overall findings: The patient is in good physical conditions and well-nourished. The face is symmetrical and no apparent abnormality is noted (Fig. 1). The submaxillary lymphatic node is not felt by palpation. There is no disturbance in opening the mouth. The result of our clinical laboratory test revealed nothing to be particularly noted.
8. Findings of the oral cavity: A swelling the size of indexfinger is detected on the buccal gingiva of left upper 3 4 5 region (Fig. 2). Boundaries of the swelling are fairly clear, the mucobuccal fold is not discolored and its hardness resembles that
of the bone structure. Undulation is sometimes noticed in parts of the gingiva. Although there is another swelling the size of walnut on the palate in the region of 2 3 4 5, there is no discoloration of the oral mucosa and it is as hard as the bone structure. No pain is complained of the buccal and palate sides (Fig. 3). Although there is noted no movement of the teeth in their socket in the swollen region, they appear to be somewhat pressed toward the palate. Pain is not felt to the touch. The electric diagnostic data are 6V for $\frac{3}{4}$, 11V for $\frac{4}{5}$ and 11V for $\frac{5}{2}$.

Roentgenographic and Histopathologic Findings

The roentgenographic image revealed the fact that the swelling was not communicated with the maxillary sinus. However, a radiopaque region is noted all over 2 3 4 5 in terms of X-ray occlusion method and X-ray intraoral method as well (Figs. 4, 5), with transparent spots here and there. The apical spots of 4 5 had been resorbed.

For operative purposes, we administered 105 mg opisthane, 60 mg histamine and 0.5 mg atropine sulfate in installments by way of pre-medication. The oral cavity was cleansed routinely and after a conduction anesthesia into the infraorbital foramen, maxillary tuberosity and large palatine foramen by the use of 10ml of 1% hydrochloric procaine in
association with an infiltration anesthesia in the neighborhood of the swelling, an oblique section was performed respectively on the left upper median line, distal side of 6 and cervical region, thus folding the periosteal membrane valve and chiseling off the bone structure to remove the swelling. The 3 4 5 teeth could not be conserved and were therefore extracted. After the extraction of these teeth, the periosteal membrane valve was unfolded and the suture was effected. For the prevention of a post-operative infection, the patient was medicated for a week with 1.5 grams of T.C. a day. He was discharged from the hospital two weeks after the operation and is currently in the third month of observation (Fig. 7)

From 14 region of the swelling, we had obtained a little amount of cheese-like extract (Fig. 6) and subjected the extract to the following histopathologic detection. It was routinely decalcified, embedded in paraffin wax and sliced for the microscopy by the use of H-E staining. As is known from Figs. 8 and 9, the center of the extract consisted of a group of degenerated keratinized epithelia and calcareous spots were found here and there. In the peripheral region, more or less the same finding was obtained but, here, cyst wall structures wrapped by the epithelia as is the case with odontogenic
cyst were partial observed (Figs. 10, 11, 12). In spots there were places where the granulation tissues embracing foreign body giant cells had proliferated themselves in contact with the degenerated epithelia, thus resorbing degenerated tissues (Fig. 13).

The bone structure in the cortex of jaw bone had been internally resorbed by the proliferation of these tissues and become quite thin but part of the sponginosa had remained within these tissues and was connected with degenerated epithelial nests deposited with the calcareous matter (Fig. 14).
Clinical literature is available on the abnormally keratinized or calcified tissue image obtained from a pathogenic cyst or tumor found in the jaw region.

PINDBOURG [1] designated such a keratinized image calcifying epithelial odontogenic tumor in his case report but his type of tumor is derived from the atrophied enamel epithelia and it is of a relatively rare incidence. THOMA and GOLDMAN [2], STOOPACK [3], IVY [4], WUNDERER [5], CHAUDHRY [6], VICKER [7] BHASKAR [8] and the Department of Oral Surgery, Tokyo University [9] have published similar clinical findings.

According to PINDBOURG, this tumor has a high percentage of local relapse and its image, consisting of multiple epithelial cells, produces a great deal of round interstices through the degeneration of intercellular substance and these interstices are full of eosin-affinity matters which will gradually become calcified.

On the other hand, GORLIN [10, 11] who gives a total number of 15 cases (11 of his own clinical encounter and 4 references) of calcifying odontogenic cyst and makes a distinction from the calcifying epithelial odontogenic tumor by PINDBOURG. According to the latter author, there is no significant difference in sex and age relative to the incidence of this cyst and of 15 cases of his reporting, 5 cases took place in the gingival region rather than in the jaw area.

Histologically, the inside of these cysts was covered with deep-layer epithelia with the so-called ghost cells here and there and according to the advance of degeneration, the boundaries between epithelia and connective tissues will become more obscure. Granulation tissues will invade among the ghost cells and calcium salts will deposit themselves on the ghost cells which have become further uniform [10, 11] GORLIN assigns the cause of this cyst to odontogenesis: 1) 3 of his 15 cases were definitely derived from the odontogenic epithelia of the developing or unerupted teeth and also, in other cases the cyst epithelia were composed of cells resembling the enamel epithelia?, 2) these cysts were made up of dentin-like substances and, 3) position of these cysts was in the region of jaws.

Further, GORLIN gave 3 similar cases in the lower jaw as keratinizing and calcifying odontogenic cyst [12]. He stated that the histologic features of these cysts were the abnormal keratinization and calcification of cyst epithelia, linking them derivationally with calcifying epithelial odontogenic tumor by PINDBOURG.
Our case described here bears a resemblance to those tumors and cysts reported by PINDBORG, GORLIN, GOLD and others respectively. In the roentgenographic images of PINDBORG, GOLD and others they were all X-ray transparent, whereas in our case there were X-ray transparent and radiopaque spots commingled here and there.

In the cysts reported by GORLIN and GOLD, there was found the fluid in the content with the single exception of the latter which gave a cheese-like extract. Our case gave only a cheese-like extract without a trace of content fluid altogether.

As for the appearance of ghost cells or corresponding cells, PINDBORG, GORLIN and GOLD did not recognize it. In our biopsy, we first noted epithelial tissues which were assumed to constitute cyst walls and here and there those cells resembling epithelial layers of odontogenic cysts were observed, thus enabling us to infer them to be of odontogenic origin.

Keratinized matter and shapes of calcified substance were quite irregular in our case which markedly differed from what was described by PINDBORG or GORLIN, the overall image being close to the description by GOLD.

As has been described above, we consider our case to be a close one to what was reported by GOLD in terms of roentgenographic findings, absence of content fluid, conditions of cyst walls and epithelial layers, keratinized matter and shapes of calcified substance, etc. As GORLIN pointed out a possibility of relapse of this kind of tumor, we shall continue our observation of this patient at regular intervals.

In passing, importance should be attached to the fact that, to cite a recent report by VICKERS et al., in which 6 out of 7 calcifying epithelial odontogenic tumors gave a significant amount of amyloid, the formation of amyloid often precedes the calcification in this type of tumor.

Conclusions

1. The case described here was clinically diagnosed as an osteofibroma and removed from a 23 year-old male from his left upper jaw.
2. In terms of various histopathologic examinations, the tumor was diagnosed to be a case of keratinizing and calcifying odontogenic epithelial tumor.
3. At this time of reporting, the patient has not suffered a relapse.

References


