On Some Considerations of Ameloblastoma
(Case Report)

by

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I Introductory

It is the established fact that oral cavity of humans is very susceptible to various

tumors. This can be explained that the growth process and cell differentiation of oral
cavity are natally quite complicated and also it is exposed to various physiological
stimulations, coupled with the fact that oral cavity is anatomically liable to sustain
external injuries. Further, as the jaw bones possess teeth there are known some
tumors peculiar to oral cavity in connection with its odontogenesis. They are what is
known as odontogenic neoplasmas which include follicular cyst, ameloblastoma and
odontoma (radicular cyst is also sometimes included) according to different opinions of
the scholars. However, of these odontogenic neoplasmas what we may consider to be
genuine tumor beyond any refutation is ameloblastoma. Although it is not often that the
case of ameloblastoma is clinically encountered, it holds out a few points of interest in
that it may often recur despite its benignity in patho-histological aspect and, therefore,
a mere conservative treatment of extirpation curettage is not sufficient. But, at the
same time, a dental surgeon hesitates to administer a maxillotomy on the patients under
age, especially those of pre-marriage women.

In the present study, the authors report on 20 cases of ameloblastoma patients
with whom they have come into contact in their clinical operations in a dental hospital
attached to Nihon University School of Dentistry.

II Designations

The name 'adamantinoma' was introduced by FALKSON in 1879 but as early as
1877 Busch gave his finding that this tumor is caused by the entrance of ectoderm.
In the same year, KOLAEKEK thought that this tumor was due to the proliferation of
oral mucous membranes. However, it is generally believed that FALKSON definitely
established the fact that this tumor is formed by the epithelial, cells corresponding to
the enamel organs at a developmental period, designating it as 'adamantinoma'. Ac-
cording to THOMA,1) MALASSEZ (1885) is also one of those who had used the designation
adamantinoma in the earliest time. Indeed, there is no other tumor which is designated
with so many names as 'adamantinoma'. Sometimes such designations as multilocular
cystoma, central epithelioma and, in rare cases, soft odontoma are used. In his report
ENDO2) gives the synonyms of adamantinoma as many as nineteen, MEAD3) gives six and

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CAHN\(^6\) the same number of synonyms. The latter attributes the multiplicity of synonyms to the fact that the cause and behavior of adamantinoma are hardly understood. MEAD mentions that the term multilocular cystoma is quite inappropriate in that there are other diseases with multilocular effects. However, the term ‘adamantinoma’ is generally used in practice but it does not contain any hard tissue made of enamel, the whole substance being derived from the epithelia of enamel organ. For this reason, the term can not be said to be scientifically accurate. It is on this theory that CHURCHILL (1934) proposed an alternative term ‘ameloblastoma’ which has been adopted by American Pathological Society, finding its way into the recent publications. In Japan, these two terms adamantinoma and ameloblastoma are in use intermixedly.

### III Statistical Data

The case of ameloblastoma is one of those odontogenic neoplasmae which are of comparatively rare occurrence. OI²\(^5\) and his associates report eight (8) cases of ameloblastoma (8.8\%) out of ninety (90) odontogenic neoplasma complaints. After a statistical survey over 3,287 odontogenic neoplasma complaints, ISHII\(^6\) reports fifty-one (51) cases of ameloblastoma (4.6\%). Of these neoplasma complaints the highest percentage is occupied by the cases of radicular cyst (86.1\%) and the lowest is those of odontoma (1.6\%). As is attested by these figures, the case of ameloblastoma is not one with which a dentist comes into daily contact.

**Difference by sex:**

In terms of sex distinction, forty (40) cases are males and forty-eight (48) are females with 10\% difference higher in the latter (ISHII). On the other hand, OTAKE\(^7\) gives a reverse picture in which thirty-two (32) cases are males while twenty (20) are females. With reference to twenty (20) cases under review by the present authors, no particular distinction in terms of sex is noticed. MASAKI\(^8\) reports seventeen (17) cases of males and ten (10) cases of females. SMALL\(^9\) reports 987 cases of ameloblastoma out of 1,036 neoplasma complaints collected from his own clinical experience and other data, in which 52\% is males and 48\% is females.

**Distinction by age:**

OTAKE gives 30.7 years of age as an average age when ameloblastoma is usually treated at hospital, with the female a few years younger than the male. 26 years of age is reported to be the stage at which the patient becomes aware of his ameloblastoma complaint. According to ISHII, twenty (20) cases took place at age level from 13 to 20 (29\%), seventeen (17) cases from 21 to 30 (24.6\%) and sixteen (16) cases under the level of 12 (23.2\%).

**Start of initial treatment:**

As a set of data which calculates the approximate age at which the ameloblastoma patient receives treatment, OTAKE gives twenty-four (24) cases as receiving the initial treatment sometimes between 21 to 30 years of age (27.6\%), twenty (20) cases from 31 to 40 (23\%) and fourteen (14) cases from 13 to 20 (16\%).

Our own examinees revealed the average age at the initial treatment as 29.2 years of age, an age ranging running from 15 to 68 years of age. Fig. 1 shows the ameloblastoma of this 68-year old patient in which a gigantic swell is prominent. Fig. 2 is its microscopic finding.

The following table gives a three-way comparison of surveys made by OTAKE,
ISHII and SMALL that give the occurrence of ameloblastoma cases in terms of jaws. These figures conclusively point to the higher occurrence of ameloblastoma in lower jaw. Only one (1) case is the upper jaw ameloblastoma in twenty (20) of our own collection.

**Locality of occurrence:**

In terms of localities where the cases of ameloblastoma are found, the following breakdown is reported by SMALL.

With regard to our own cases, the majority of them has occurred in the molar and ramus ascending regions.

<table>
<thead>
<tr>
<th></th>
<th>OTAKE's survey</th>
<th>ISHII's survey</th>
<th>SMALL's survey</th>
</tr>
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<tbody>
<tr>
<td>No. of cases</td>
<td>50</td>
<td>124</td>
<td>925</td>
</tr>
<tr>
<td>Upper jaw</td>
<td>4</td>
<td>12</td>
<td>173 (22%)</td>
</tr>
<tr>
<td>Lower jaw</td>
<td>46</td>
<td>112</td>
<td>752 (78%)</td>
</tr>
</tbody>
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<table>
<thead>
<tr>
<th></th>
<th>Upper jaw</th>
<th>Lower jaw</th>
</tr>
</thead>
<tbody>
<tr>
<td>Molars</td>
<td>47% (21)</td>
<td>Molars &amp; ramus ascending region 70% (170)</td>
</tr>
<tr>
<td>Ramus mandibulae &amp; floor of cavum nasi</td>
<td>33% (15)</td>
<td>Premolar region 20% (49)</td>
</tr>
<tr>
<td>Premolar region</td>
<td>9% (4)</td>
<td>Raphe region 10% (27)</td>
</tr>
<tr>
<td>Canine region</td>
<td>9% (4)</td>
<td></td>
</tr>
<tr>
<td>Palate region</td>
<td>2% (1)</td>
<td></td>
</tr>
</tbody>
</table>
Fig. 3 shows a case of ameloblastoma on the upper front region of a 38-year-old female and Fig. 4 is a roentgenogram of the same. Its tissue image is found to be somewhat atypical (Fig. 5).

Fig. 3

Fig. 4

Fig. 5
IV Derivations

As is the case with other tumors in general, the real derivation of ameloblastoma has not been definitely established. However, it is generally attributed to the enamel epithelium in fetal period. But as there are reported some cases in which ameloblastoma occurred in other part of the body, it is not always possible to determine the exact locality of derivation.

CAHN mentions that though ameloblastoma is caused by epithelia which are closely related to the dentition, it is not known whether the cells responsible for the ameloblastoma are related to enamel or to other region of the tooth band. To mention a few of the possible derivation of ameloblastoma that have been given by the eminent researchers in the field. BUSCH ascribes this to a part of ectoderm which has been lost in the formation of jaws, KOLACZEK to the proliferation and growth of the epithelium of oral mucous membrane which has invaded into the jaw in the state of cord, KROMPECHER gives its derivation in the basal-cell tumor of oral epithelium and FALKSON refers to the supernumerary germs of enamel organs or their variation in the degenerated state as a motive for ameloblastoma. KEYEL\(^{10}\) gives a hypothesis after his survey on the four cases of ameloblastoma found among the negroes that it may be due to the residue of enamel in the 4th molar. BENAGIANO\(^{11}\) attributes its usual derivation from dental germs of the lower 3rd molar to the fact that ameloblastoma is mostly confined in the angle of the lower jaw. Opposing to a supposition that ameloblastoma is derived from the epithelium of oral mucous membrane, BENAGIANO points out that even after completion of the growth of teeth the ameloblastoma may develop due to MALASSEZ's epithelial debris.

MEAD gives such factors as the transposition of MALASSEZ’s epithelial debris, abnormal states of enamel due to inflammation or fracture, growth of odontogenic epithelia in other bodily parts and proliferation of maxillary epithelium to be responsible for the case of ameloblastoma. BLAND-SUTTON gives his belief that it is derived from the oral mucous membrane or periodontium.

THOMA enumerates the abnormal states of enamel or its composition, cell remains from enamel during or after the odontogenic process, epithelium of odontogenic cyst, surface epithelium wrapping the jaws and transposition of epithelia in other bodily parts among the factors that give rise to the ameloblastoma. Of them, a factor of odontogenic cyst deserves special notice in that CAHN\(^{12}\) (1933) and CHURCHILL (1934) respectively reported on the same finding. In this country also the identical conclusion has been put forward by ENOMOTO et al\(^{13}\). THOMA\(^{14}\) thinks that PARTSCH’s Neben Höhle contains a latent possibility of causing the ameloblastoma.

IV Classification

The usual classification of ameloblastomae is two-fold; those which are adamantinoma solidum and those which are cysticum. There are some scholars who divide them further into those of adamantinoma cysticum and those of adamantinoma non-cysticum, those of toothed and those of toothless, unilocular and multilocular, or benign central epithelioma and multilocular jaw cyst. Although those of unilocular and those of multilocular can be detected by means of roentgenography, we will have recourse to histological techniques to classify those of solidum or cysticum.

With regard to our own cases under review, all the ameloblastoma cases have been those of cysticum. According to SMALL, 365 cases (78%) of ameloblastoma cysticum and
100 cases (22%) of ameloblastoma solidum are reported out of 465 case histories. On the whole, the ameloblastoma of cysticum type takes place more frequently than that of solidum type. This may be explained by the fact that the ameloblastoma of solidum type turns itself into that of cysticum type with the passage of time. This fits with a finding of MEAD that the ameloblastoma of solidum type is most frequently found in its transition to that of cysticum. SMALL adopts a two-fold division of follicular and plexiform types. Fig. 6 is a picture of the ameloblastoma of solidum type and Fig. 7 shows a case of ameloblastoma which is unilocular clinically but also has the cyst portion from the histological point of view.

V Malignity

As has been touched in the foregoing section, the ameloblastoma is very slow in its development but it is apt recur when it is treated with a mere restorative operation. This propensity to recur is the basis for a clinical interpretation that the ameloblastoma occupies a position between malignant and benign tumors. Some scholars warn against a drastic operation such as jaw joint resection which disturbs the functional and cosmetical aspects of a patient. On the other hand, there are others who advocate a jaw joint resection on grounds that it is effective as a complete cure. Reports are numerous on the recurrences of the ameloblastoma which have tended towards a malignant tumor, causing a metastasis into other bodily parts or cancerous change. One of our own cases is a woman of 44 years old whose buccinator was completely encroached upon even after her ameloblastoma had been extirpated as sequestrum of the jaw bone. As it is a case of some interest, we give a brief extract from her history card on our file.
Case Report

Sex: Female.
Age: 44 years old.
Occupation: Barber.

The patient came to our hospital with a chief complaint that she suffered from tumors in the right angle of lower jaw and buccal region. She had complained of the same symptom at the age of nineteen and was recommended to undergo an osteotomy. However, she had the tumor removed by means of curettage and received radiations by the quartz lamp for about ten times. At the same time, her right lower molar was extracted without any subsequent prosthetic treatment. About two years prior to her visit to us (42 years old) her remaining 14 teeth with the exception of $7654$ and $7$ were extracted because of a loose feeling of them. A denture plate was placed afterwards but constant pain in her mucous membrane of gums made her receive penicillin injection of 150 million units, four X-ray radiations and two radium radiations. Despite these cares, an enlargement of her right buccal region had developed with an accompaniment of pain and this was the reason for her coming to our hospital.

Diagnosis:

The bone structure and nutritional condition of the patient are found to be fair. A tumor of indefinite shape is seen extending from her right buccal region to the lower jaw, which is out of symmetry. The said tumor has some gloss and is reddish with accompanying hot-feeling. It precludes our touch of the submandibular lymphatic nods. Stenostomy of slight degree is noticed and $7654$. 

Fig. 8
Fig. 9
and 7 are either quite loose or extruding. A cavity of egg-size is formed in the right lower molar region and surrounding soft tissues have raised themselves. The use of an explorer has indicated the roughness of bone surface. Her complaint of pain is constant and lack of appetite is reported. A roentgenogram of the said region (Fig. 8) reveals an extensive shadow-gram in the right jaw angle and what is considered to be a right lower 3rd molar is impacted in the ramus ascendens (Fig. 9).

A histological examination made on a test material taken from the diseased region indicates the case of ameloblastoma. Based on these findings, a diagnosis is made as ameloblastoma and secondary bacillary infection.

**Treatment:**
In view of a high inflammation of the tumor, an antiphlogistic treatment was indicated. As the patient did not favor a hospitalization, she came to the hospital at a specified time of the day and received the administration of antibiotics and exchange of lavage tamponade according to a routine method.

**June 25** Abscess formation and its fluctuation was noticed (Forced hospitalization was enforced).

**June 26** Pus discharge by incising 3 centimeters along the right lower jaw. Post-operational care included the guanoflacin, trypsin, greenpole, etc.

**July 17** Another incision was effected around the previous incised spot for the further discharge of pus.

**July 26** The inflammatory tumor still persisted around the buccal region, and two sequestrums with the size of a thumb were removed from the lower jaw angle.

**July 31** Another two sequestrums were extirpated from the oral cavity.

**Aug. 13** As some undulation took place around the previous incisions, they were extended to the length of 12 cm.

**Aug. 26** The ramus ascendens in the right lower jaw was removed in the complete state of sequestrum, together with the impacted right 3rd molar. Abscess-like prominences with the sizes ranging from the small-finger tip to the thumb developed on and around the incisions. Turning to the violet color in a few days of development, they fell in necrosis. This process took place for several cycles repeatedly. An examination of the necrotic tissues in the buccal region induced us to suspect the cases of atypical adamantinoma and complication of endothelioma and cancer (Figs. 10, 11, 12). From this time on, the patient complained a lack of appetite and her debility became pronounced on the whole.

**Sep. 16** We observed the state of carcinomatous cachexia but the patient insisted on being dehospitalized and sent home. She died three days later.

This is an example of the case in which an early adamantinoma (19 years of age) could not be entirely checked by a conservative treatment but the tendency had been towards sequestration. However, the malignity of adamantinoma is rather infrequent. Siegfried and Wunderer give the total number of malignant cases of adamantinoma to be 10, taken from both his own patients and literature. Small gives 2.0% out of 1036 adamantinoma cases as malignant.
It goes without saying that an adequate treatment of the adamantinoma should be referred to oral surgery. It is maintained that this tumor is non-sensitive to the X-ray treatment. THOMA divides the operation into three aspects, 1) extirpation, 2) osteotomy of surrounding regions, and 3) bone resection. However, with a simple

**VI Therapy**

It goes without saying that an adequate treatment of the adamantinoma should be referred to oral surgery. It is maintained that this tumor is non-sensitive to the X-ray treatment. THOMA divides the operation into three aspects, 1) extirpation, 2) osteotomy of surrounding regions, and 3) bone resection. However, with a simple
extirpation there will be a danger that the case may recur at a later age. But it is fortunate that it usually takes a fairly long time for a recurrence, sometimes running anywhere from 20 to 30 years in duration. Therefore, the clinician may derive profit when he subjects the case of adamantinoma to a periodic examination. In our own experience, one recurrence has taken place out of 12 arthrotyomy cases of the one-side lower jaw, 2 cases of partial extirpation and 6 cases of complete extirpation and partial resection. Figs. 13 and 14 are pictures of the adamantinoma (25-year-old male), which is subjected to a periodic inspection following an additional curettage but no recurrence is reported for three years to date. Fig. 15 is a picture of the adamantinoma (22-year-old male) which developed on the lower jaw. Discharge of pus was noticed in the vestibule of the mouth (vestibulum oris). After the administration of apicotomy, no tendency for a recurrence is suspected for two and half years to date.
VII References

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