Ameloblastic Fibroma: A critical evaluation of reported cases provides evidence of two types

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There is still controversy as to the clinical findings and behavior of ameloblastic fibroma (AF). To clarify the exact biological profile, we critically re-evaluated the clinical characteristics of 31 cases of AF through a review of the Japanese literature. AF can be divided clinically into two distinct types: neoplastic AF (24 cases) and pericoronal hamartomatous AF (7 cases). The former appears as an expansive multilocular radiolucent lesion within the posterior mandible in the second decade. A recurrence rate of 13% was estimated. Its tendency to recur and to develop at ages beyond completion of odontogenesis clearly denotes a neoplastic nature. On the other hand, the latter appears as an asymptomatic small unilocular radiolucency over the occlusal surface of an unerupted mandibular molar primarily in the first decade. Neither persistent growth nor recurrence was observed. There is no proof that pericoronal AF sequentially progresses into a neoplastic AF. However, since two lesions are histologically indistinguishable, clinical findings have to be considered initially in a management plan of AF.

Key words: ameloblastic fibroma, biological profile, odontogenic hamartoma, odontogenic tumor, pericoronal lesion

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Introduction

Ameloblastic fibroma (AF) is the prototype or the least histologically differentiated of so-called mixed odontogenic tumors (1-6). Although more than 150 cases, including Japanese reports, have been published (4), conflicting data exist in the literature on the clinical characteristics. For example, some were found incidentally by microscopic examination of the dental follicular tissue of unerupted teeth (7-9), whereas others appeared as a large expanding lesion suggestive of neoplasm (10-23). Most authors consider that AF rarely ever recurs (1-6), but Trodahl (24) recorded a 43% cumulative recurrence rate. It is still, at present, a matter of debate whether AF is a truly neoplastic entity or a developmental anomaly (hamartoma).

In this context, we attempt to clarify the biological profile of AF based on a literature survey. There can be no doubt that two different clinical types that have the same histologic features exist. It is clinically important to realize the entity of pericoronal AF has a non-aggressive and non-recurrent nature. In addition, the histopathologic diversity, the other remaining gap in our knowledge of AF, will be discussed briefly.

Review of the Literature

Through a review of the Japanese literature, we retrieved 31 well-documented cases of AF with each patient's clinical record and photomicrographs (25-54). All papers published in abstract form or without adequate photographs were deemed unacceptable (55). We also excluded a possible recurrent case, since the histological diagnosis of the primary tumor was uncertain (56).

True neoplastic AF (25-47)

The median age of the 24 patients was 14 years (range, 1-38); 11 patients were diagnosed in the second decade. One lesion was observed at birth (26). The tumor was equally encountered in both genders. Nineteen tumors (79%) occurred in the mandible and 13 in the posterior segment. None developed in the anterior maxilla.
Painless swelling was the first symptom in 21 cases (88%). Radiographically, 15 cases (71%) were associated with an impacted tooth and 12 (60%) showed a multilocular radiolucency. Three cases (13%) recurred at 3, 4 and 12 years after first operation, respectively (27, 45, 46). One patient was of particular interest since the tumor recurred twice (45). Another 13 cases of recurrent AF have been reported (24, 57-61). Although there are no specific clinicopathological findings to predict the recurrence, the mean time between initial treatment and recurrence was 4 years, ranging from 6 months (24) to 23 years (60). Therefore, careful follow-up of more than 4 years is mandatory. The higher frequency of association with the impacted tooth suggests that AF may have arisen from cells in the dental follicle.

Pericoronal hamartomatous AF (48-54)

The age of the 7 patients ranged from 1 to 11 years, with a median of 5 years. One lesion was found at birth (53). Four patients were female and 3 male. Six cases (86%) occurred in the mandible, especially in the posterior segment. Only one case developed in the anterior maxilla (54). Interestingly, two examples of peripheral AF were recorded (48, 53); however, it is our impression that they are not a true peripheral lesion but rather an erupted pericoronal AF occurring during tooth development, although inconclusive. All patients were asymptomatic and failure of tooth eruption was the major reason for seeking treatment. Five lesions (71%) were associated with a deciduous tooth and 2 with a permanent tooth. The radiographic finding in 4 cases (80%) was a unilocular radiolucency. No recurrence was observed. The dental follicular origin of pericoronal AF is apparent. The comparison of clinical findings between true neoplastic and pericoronal hamartomatous AF is summarized in Table 1.

Discussion

So-called mixed odontogenic tumors have been a topic of academic interest to oral pathologists. One of major interests that has been investigated is whether these tumors represent different stages in the histomorphological differentiation of the same entity (1-6). With regard to AF, it is generally agreed that some cases are true neoplasms, whereas others are hamartomas presenting the earliest stage in the developing odontoma (1-6). In this review, our critical evaluation of reported cases provided evidence of two clinical types: a true neoplastic AF and a pericoronal hamartomatous AF. However, it is not possible to state with certainty that pericoronal AF may maturate over time, eventually leading to the formation of an odontoma, unless such evidence is forthcoming.

Table 1: True neoplastic and pericoronal hamartomatous ameloblastic fibromas

<table>
<thead>
<tr>
<th>Clinical features</th>
<th>TNAF</th>
<th>PHAF</th>
</tr>
</thead>
<tbody>
<tr>
<td>No. of patients</td>
<td>24</td>
<td>7</td>
</tr>
<tr>
<td>Mean age (range)</td>
<td>14(1-38yr)</td>
<td>5(1-11yr)</td>
</tr>
<tr>
<td>Sex</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>12</td>
<td>3</td>
</tr>
<tr>
<td>Female</td>
<td>12</td>
<td>4</td>
</tr>
<tr>
<td>Maxilla</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Anterior</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>Anteroposterior</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>Posterior</td>
<td>4</td>
<td>0</td>
</tr>
<tr>
<td>Mandible</td>
<td></td>
<td></td>
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<tr>
<td>Anterior</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Anteroposterior</td>
<td>5</td>
<td>0</td>
</tr>
<tr>
<td>Posterior</td>
<td>13(54%)</td>
<td>5(71%)</td>
</tr>
<tr>
<td>Radiography</td>
<td>15(21%,71%)</td>
<td>7b</td>
</tr>
<tr>
<td>Impacted tooth</td>
<td></td>
<td></td>
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<tr>
<td>Multilocular</td>
<td>12(20%,60%)</td>
<td>1</td>
</tr>
<tr>
<td>Unilocular</td>
<td>8</td>
<td>4(5%,80%)</td>
</tr>
<tr>
<td>Swelling</td>
<td>21(88%)</td>
<td>0</td>
</tr>
<tr>
<td>Recurrence</td>
<td>3(13%)</td>
<td>0</td>
</tr>
</tbody>
</table>

TNAF : true neoplastic ameloblastic fibroma ; PHAF : pericoronal hamartomatous ameloblastic fibroma.

aData are not available in remaining cases.

bFive deciduous and two permanent teeth.

cRemaining two cases are peripheral lesions.
The concept of pericoronal AF has already been discussed in the literature (2, 7-9); however, its existence as a separate entity is not widely accepted. As mentioned above, this lesion develops during childhood (the period of normal odontogenesis) and has little or no proliferative potential. It does not recur. Indeed, in all cases leaving the underlying tooth in toto, the involved tooth was permitted to erupt, even when the lesion extended to the surgical margins. Similar clinical findings and behavior have also been documented in non-Japanese cases (2-4, 6, 62-73). Such characteristics would favor the interpretation of a hamartomatous nature. Therefore, oral surgeons must realize the non-neoplastic character of pericoronal AF and it should be treated ultraconservatively.

The most important problem is that a histologic distinction between two types of AF is impossible. The histopathology of AF is well-described and well-photographed (74, 75); however, it shows a more considerable histologic diversity than generally believed (76). The amount and morphology of both epithelial and ectomesenchymal components vary from case to case and from area to area within the same tumor. Consequently, some contain larger epithelial islands composed of a peripheral palisading of columnar ameloblastic cells and a central stellate reticulum-like area, whereas others contain only smaller double-layered dental lamina-like rests. The former shows the close resemblance to ameloblastoma and the latter may mimic the odontogenic fibroma. The amount of normal odontogenesis) and has little or no proliferative potential. It does not recur. Indeed, in all cases leaving the underlying tooth in toto, the involved tooth was permitted to erupt, even when the lesion extended to the surgical margins. Similar clinical findings and behavior have also been documented in non-Japanese cases (2-4, 6, 62-73). Such characteristics would favor the interpretation of a hamartomatous nature. Therefore, oral surgeons must realize the non-neoplastic character of pericoronal AF and it should be treated ultraconservatively.

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Summary

The clinical characteristics of reported cases of AF are sufficiently different to recognize two separate lines. These include a relatively innocuous benign tumor and a pericoronal hamartoma. However, two lesions do not comprise a continuum. Including a hamartomatous AF into a true AF category results in the erroneous biological profile of a neoplastic AF. Unfortunately, we have no histologic method for differentiating the two. Conversely, we cannot predict the clinical behavior of AF on histologic grounds. However, it is no longer acceptable to simply refer to these lesions as AF. All oral pathologists should add an informative phrase in their report correlated to the clinical findings.

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