Ameloblastic fibroma and related lesions: a clinicopathologic study with reference to their nature and interrelationship

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BACKGROUND: Ameloblastic fibroma (AF) and related lesions constitute a group of lesions, which range in biologic behavior from true neoplasms to hamartomas. The aim of this study was to elucidate the nature and interrelationship of this group of lesions.
METHODS: Clinical and pathological studies were undertaken retrospectively on 13 cases of AF and seven cases of ameloblastic fibro-odontoma (AFO). Thirty-three complex odontomas and 33 compound odontomas were also included for comparative purpose. Relevant follow-up data were recorded and the literature was reviewed.
RESULTS: The majority of patients with AF (nine cases, 69.2%) were over the age of 22 years with frequent involvement (76.9%) of the posterior mandible. Tumors recurred in four of 11 patients with follow-up information and two recurrent tumors showed malignant transformation. There was no case in this series that could be designated as the so-called ameloblastic fibrodentinoma, apart from one recurrent AF in which further maturation to form only tubular dentin materials was identified. AFO tended to occur at a younger age group with an average of 9.6 years. Recurrence was noted in two of five patients with follow-up data and both recurrent lesions showed limited growth potential and further maturation into a complex odontoma. Significant differences were noted in the age and site distribution between the complex and the compound odontomas.
CONCLUSION: Whilst the majority, if not all, of AFs are true neoplasms with a potential to recur and/or of malignant transformation, some, especially those occurred during childhood, could represent the primitive stage of a developing odontoma. Our data also suggests that some AFOs are hamartomatous in nature, representing a stage preceding the complex odontoma.

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Introduction

Ameloblastic fibroma (AF) and related lesions are defined by WHO as 'neoplasms composed of proliferating odontogenic epithelium embedded in a cellular ectomesenchymal tissue that resembles dental papilla, and with varying degrees of inductive change and dental hard tissue formation' (1). This group of lesions is also sometimes referred to as mixed odontogenic tumors and usually includes AF, ameloblastic fibrodentinoma (AFD) and ameloblastic fibro-odontoma (AFO) (2–5). Further, of particular interest concerning the histogenesis of these mixed odontogenic tumors is a group of non-neoplastic malformations containing fully calcified or mineralized dental tissues, the odontomas (3, 5). Despite numerous efforts however, there is still considerable confusion concerning the nature and interrelationship of these mixed odontogenic tumors and related lesions (2, 3). Because of the obvious difficulties in distinguishing between the true tumor and a developing odontoma that may at one stage exhibit the histologic appearances of an AF or an AFO, their overall nature as neoplasms or as various sequential stages of non-neoplastic developmental anomaly could not be readily determined. Furthermore, there is no consensus on whether these tumors should be categorized as separate entities or whether they represent different stages in the maturation of the same entity.

In order to shed some light on the nature and the interrelationship of this group of odontogenic lesions, we intended to report a total number of 86 cases of the above-mentioned lesions from Chinese patients and to document their clinical and pathological background.

Materials and methods

Cases diagnosed as AF, AFO, complex/compound odontomas or under other related diagnostic terms were

retrieved from the files of the Department of Oral Pathology, School of Stomatology, Peking University, China during the period 1949–2002. Standard hematoxylin and eosin stained sections were reviewed and the lesions reclassified according to the WHO Histological Typing of Odontogenic tumors (1992)(1). Thirteen cases of AF and seven cases of AFO were diagnosed. We did not find any cases that could be designated as AFD, apart from one recurrent AF in which further maturation to form only tubular dentin materials was identified. The term odontoma denoted lesions that contained all fully calcified dental tissues, and included two types, the complex (33 cases) and compound (33 cases) odontoma. In several cases, the recurrent tumor showed apparent maturation or malignant transformation, their designation into different lesion groups was based on the clinical and histological features of the primary tumor. Clinical data including age, sex, anatomic site, duration, radiographic features, clinical impressions, treatment and available follow-up were recorded.

Results

Ameloblastic fibroma

Clinical findings

The clinical features of 13 cases of AF were summarized in Table 1. The patient age at first presentation ranged from 6 to 51 years (mean: 26 years), and peaked at the third decade (seven of 13, 53.8%). Seven patients were male and six female. All 13 tumors occurred in the mandible, with 10 in the posterior part of the mandible and three involving both anterior and posterior parts. In eight of 10 patients with available radiographs, well-defined multilocular radiolucencies were noted (Fig. 1a). Unilocular lesions were only found in two cases (Fig. 1b). Six patients were initially treated by enucleation or curettage, six cases were extirpated with

Table 1 Clinical features of ameloblastic fibromas

No	Age[a]/ sex	Location[b]	Symptoms and radiographic findings	Size (cm)	Treatment and follow-up
1	28/F	R mandible: first premolar to ramus	Swelling. Multilocular radiolucency, second molar unerupted and displaced	8 × 4.5	Segmental resection 7 month after surgery, NSR
2	23/F	L mandible: second premolar to ramus	Swelling. Multilocular radiolucency	5 × 3	Enucleation 17 month after surgery, NSR
3	15/F	L mandible: first premolar to first molar	Swelling. Unilocular radiolucency	3 × 2	Segmental resection 1 year after surgery, NSR
4	21/M	Mandible: R first molar to L lateral incisor	Swelling. Unilocular radiolucency	4.5 × 2	Enucleation 2 year after surgery, NSR
5	51/F	L mandible: incisor to second molar	Swelling. Multilocular radiolucency	4 × 3	Segmental resection 5 year after surgery, NSR
6	24/F	R mandible: canine to second molar	Swelling and loose first molar. Multilocular radiolucency	3.5 × 2	Segmental resection 10 year after surgery, NSR
7	24/M	L mandible: angular and ramus region	Swelling. Radiograph not available	Unknown	Segmental resection REC (8 year), enucleation, histology identical to AF 14 year after second surgery, NSR
8	36/M	R mandible: molar to ramus	Swelling, loose teeth and difficulty to open mouth. Multilocular radiolucency	7 × 6	Hemimandiblectomy. Lost to follow-up
9	13/M	R mandible: first molar to ramus	Swelling. Multilocular radiolucency	5 × 3.5	Segmental resection 22 year after surgery, NSR
10	24/M	Mandible: R first molar to L first molar	Swelling. Multilocular radiolucency	5 × 2.5	Enucleation. Lost to follow-up
11	44/F	R mandible: premolar region	Swelling. Radiograph not available	2 × 2	Enucleation REC (8 year), sub-mandibulectomy, histology identical to AF REC (14 year), surgical details unknown, histologic evidence of sarcomatous transformation, OFS, lost to follow-up
12	29/M	L mandible: first molar to ramus	Swelling. Radiograph not available	5 × 4	Enucleation REC (2 year), surgical details unknown, histologic evidence of sarcomatous transformation, OFS, lost to follow-up
13	6/M	L mandible: second premolar to ramus	Swelling, pain. Multilocular radiolucency	4 × 2.5	Enucleation REC (21 month), enucleation, histology identical to AFD 7 year after second enucleation, NSR

[a]Age at first diagnosis.
[b]R, right; L, left.
REC, recurrence; NSR, no sign of recurrence; AF, ameloblastic fibroma; AFD, ameloblastic fibrodentinoma; OFS, odontogenic fibrosarcoma.
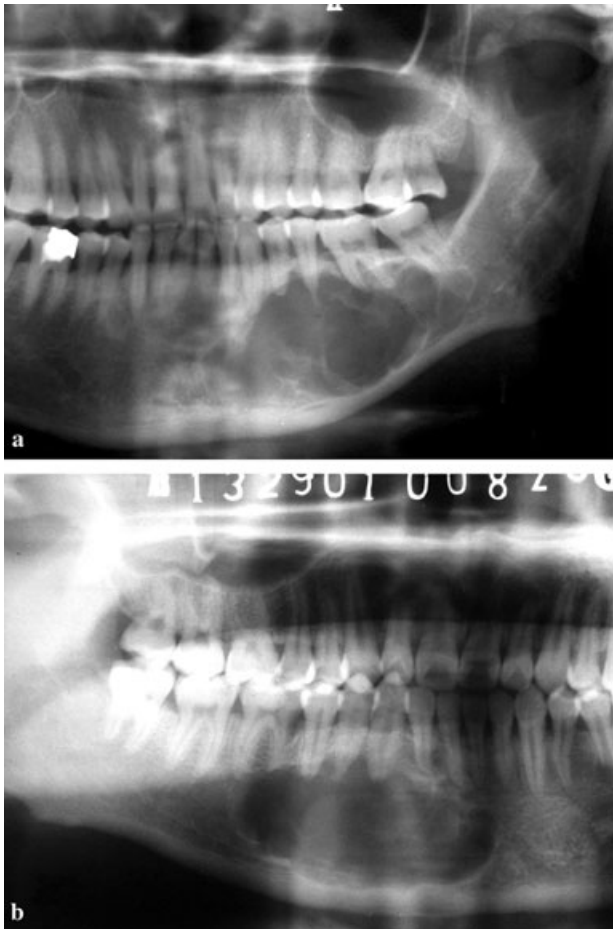
Figure 1 Radiographs of ameloblastic fibroma showing multilocular (a) and unilocular (b) radiolucencies in the mandibles.

segmental ostectomy of the jaw and one patient was subject to hemimandiblectomy. Follow-up data were available on 11 patients and two patients were lost to follow-up. The length of follow-up ranged from 5 months to 22 years. Four patients (case nos. 7, 11, 12 and 13) had recurrences (36.4%) and three of which were first time recurrence. Malignant transformation was identified in two of the four recurrent tumors (case nos. 11 and 12) and one of which did not show signs of malignancy until the second recurrence. Both malignancies were diagnosed as odontogenic fibrosarcomas. Further maturation from AF into AFD was noted in one recurrent tumor (case no. 13). The other case showed identical histological features to its primary tumor (case no. 7).

Pathological findings
On histological examination, all lesions showed strands, cords, and islands of odontogenic epithelium in a primitive connective tissue stroma that closely resembles the dental papilla (Fig. 2a). Although a cell-free zone and/or a zone of hyalinization were occasionally found at the epithelial-mesenchymal interface (Fig. 2b,c), no hard tooth structures were detected in all the primary tumors of this group. Of the four recurrent tumors, one tumor retained its previous histologic appearance (case no. 7), whereas one recurrent tumor demonstrated typical tubular dentin formation without the co-existence of enamel (case no. 13; Fig. 3). Two cases of odontogenic fibrosarcoma were identified as recurrences in previously diagnosed cases of AF (case nos. 11 and 12). Both latest recurrent tumors showed unequivocal features of malignancy in the mesenchymal component, i.e. anaplastic fibroblasts with abundant, often atypical
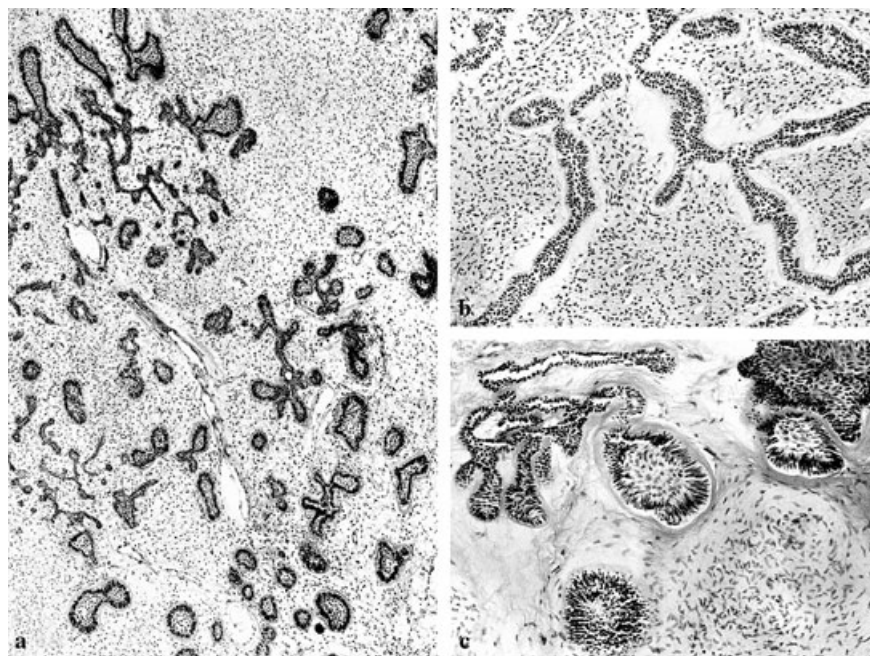

Figure 2 Histological features of ameloblastic fibroma show strands, cords, and islands of odontogenic epithelium in a primitive connective tissue stroma resembling dental papilla (a). A cell-free zone (b) or a zone of hyalinization (c) is occasionally found at the epithelial–mesenchymal interface.
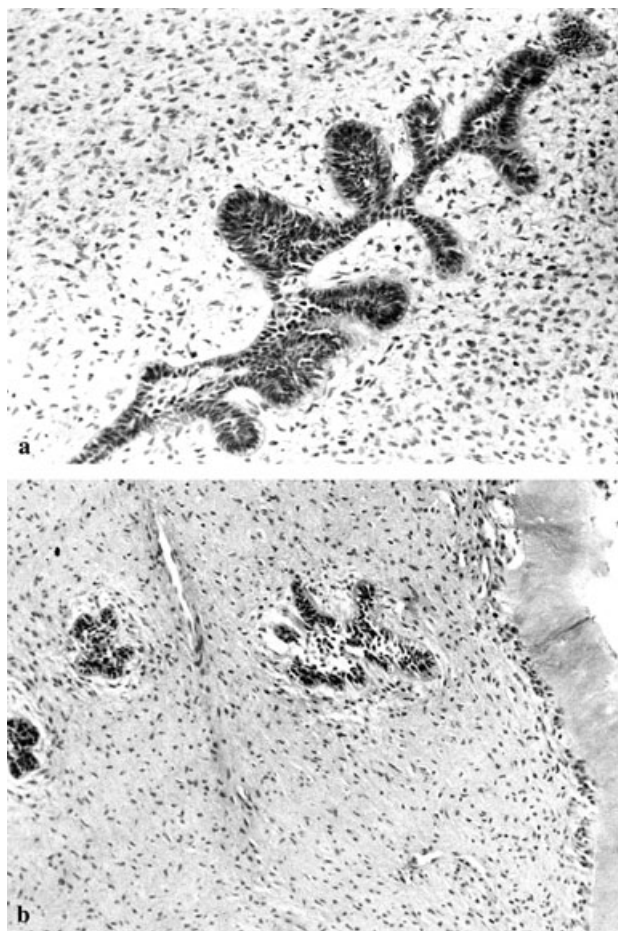
Figure 3 The primary tumor (case no. 13) showing features identical to an ameloblastic fibroma (a). The recurrent lesion 21 months after initial surgery containing typical tubular dentin identical to AFD (b).

Figure 4 Sarcomatous transformation of ameloblastic fibroma (case no. 12): the recurrence 2 years after the first surgery showing sarcomatous transformation of the mesenchymal components, i.e. marked cellular pleomorphism and atypical mitoses (a). The primary tumor showing typical features of an ameloblastic fibroma (b).

mitotic figures. The epithelial component was disappeared completely in both tumors, giving a histologic appearance of the fibrosarcoma (Fig. 4a). The trace of an odontogenic origin was only evident when examining their primary or previously recurrent lesions, in which typical features of AF could be recognized and both epithelial and mesenchymal components showed no signs of cytological atypia (Fig. 4b).

Ameloblastic fibro-odontoma
Clinical findings

Seven cases were diagnosed as AFO in the present series (Table 2). The average age of patients was 9.6 years. The tumors occurred more often in the mandible (five cases) than in the maxilla (two cases), with all the mandibular cases involving the posterior areas. Radiographs showed that the tumors were all unilocular radiolucencies with various amounts of radiopaque material of irregular size and shape (Fig. 5). Five patients were initially treated by enucleation or curettage, one by segmental resection and one by hemimandiblectomy. Recurrence was noted in two of the five patients with follow-up data. The length of follow-up ranged from 1 month to 18 years. Of the two recurrent cases, one (case no. 17) was from a 2-year-old boy who
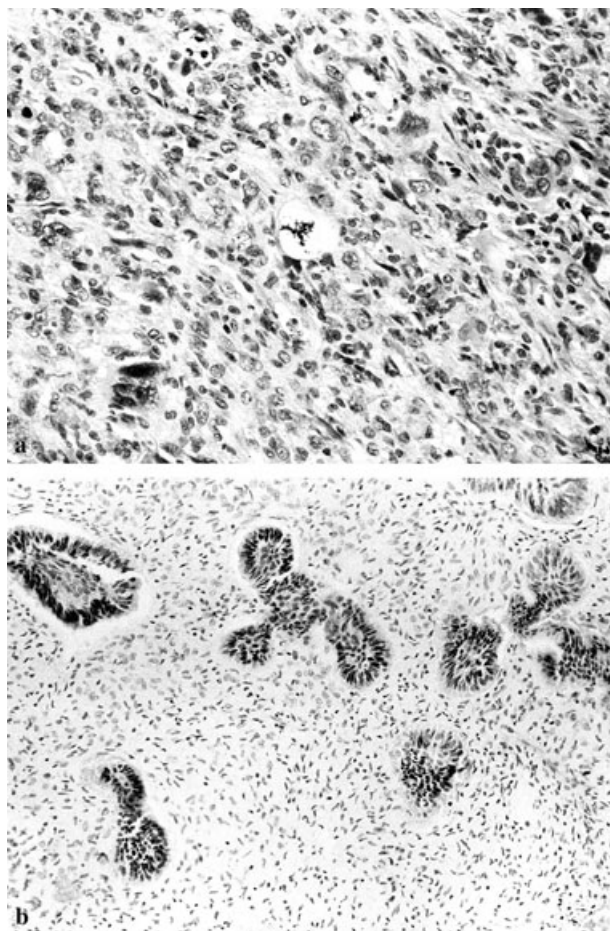
had a mixed radiolucent and radiopaque lesion adjacent to two developing tooth germs in the right anterior part of the maxilla (Fig. 6a). The lesion was initially enucleated and diagnosed as AFO histologically (Fig. 6b). One year later, an ill-defined, mixed radiolucent-radiopaque area was detected near the previous surgical site. To obviate the possible damage of the adjacent developing tooth germs, the patient was not treated until 7 years later. The radiograph at this time showed a well-defined radiopaque mass with a radiolucent border (Fig. 6c). The curetted materials this time were proved to be a complex odontoma (Fig. 6d). Following 10 years of the second surgery, there was no sing of recurrence. Another recurrent case (case no. 18) involved a 6-year-old girl who had a well-defined, radiolucent-radiopaque lesion in the right posterior mandible (Fig. 5). The lesion was initially curetted with preservation of the developing first molar and a diagnosis of AFO was confirmed. A recurrent lesion was noted 5 years later and second curettage was performed. The histologic diagnosis this time was a complex odontoma. No recurrence was detected 9 years after the second surgery.

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Table 2 Clinical features of ameloblastic fibro-odontoma

No	Age[a]/Sex	Location[b]	Symptoms and radiographic features	Size (cm)	Treatment and follow-up
14	9/F	R maxilla: first molar region	Impacted first molar. Radiolucent and radiopaque lesion over the crown of the impacted tooth	2 × 2	Enucleation 2.5 year after surgery, NSR
15	1.5/M	R mandible: first and second deciduous molars	Swelling. Radiolucency with radiopaque mass	2 × 1.5	Enucleation 12 year after surgery, NSR
16	18/F	R mandible: first premolar to ramus	Swelling. Unilocular radiolucency	6 × 3	Hemimandiblectomy. Lost to follow-up
17	2/M	R maxilla: incisor region	Swelling and pain. Radiolucent and radiopaque lesion	2 × 1	Enucleation REC (1 year), untreated 7 year later, enucleation, histology identical to complex odontoma 10 year after second surgery, NSR
18	6/F	R mandible: first molar to ramus	Swelling. Unilocular radiolucency with radiopaque mass	5 × 3	Enucleation REC (5 year), enucleation, histology identical to complex odontoma 9 year after second surgery, NSR
19	16/M	R mandible: first premolar to first molar	Swelling and pain. Radiolucent and radiopaque lesion	3 × 2	Segmental resection 1 month after surgery, NSR
20	15/M	L mandible: molar and angular region	Swelling and pain. Radiolucent and radiopaque lesion	3 × 2	Enucleation. Lost to follow-up

[a]Age at first diagnosis.
[b]R, right; L, left.
REC, recurrence; NSR, no sign of recurrence.
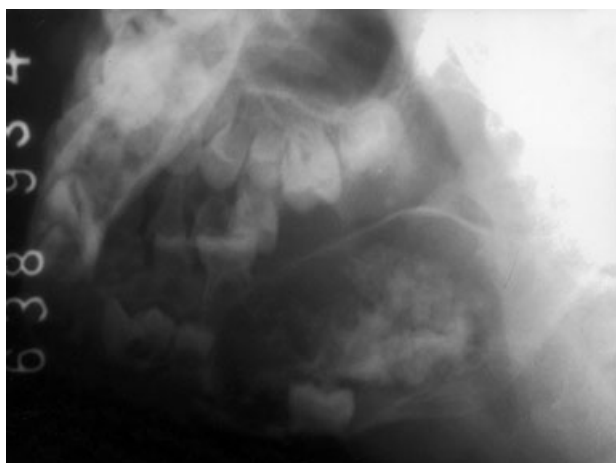
Figure 5 Radiographs of ameloblastic fibro-odontoma showing a well-defined unilocular radiolucency with radiopaque materials of irregular shape in the center area (case no. 18).

Pathological findings

Microscopically, this group of tumors showed similar features of AF, but also revealed inductive changes leading to the formation of both dentin and enamel (Fig. 7). Histologic review of the primary tumors of the two recurrent cases demonstrated features identical to AFO, whereas the specimens from the recurrence contained only calcified dentin and enamel elements arranging in a pattern similar to complex odontoma (Fig. 6b,d).

Odontoma
Complex odontoma

The clinical details of 33 cases of histologically verified complex odontoma was summarized in Table 3.

Radiographically, all lesions presented as well-defined radiopaque areas, often with a radiolucent border. Conservative excision was the treatment of choice for all cases except for one patient who was treated by partial resection of the mandible because of a relatively large size. Follow-up of the patients was not routinely undertaken because of the limited growth potential of the lesions.

Compound odontoma

The diagnosis of 33 cases of compound odontoma was confirmed by reviewing both radiographic and histologic features of the lesions. The main feature of this type of odontoma was the presence of many tooth-like structures. The clinical details of the compound odontoma was shown in Table 3. Radiograghs of the lesion usually showed numerous tooth-like radiopaque masses situated in a well-defined radiolucent area. Similarly, all patients were treated by enucleation and were not routinely followed after the surgery.

Discussion

It is widely accepted that the fully developed and calcified odontoma is a hamartoma, rather than a neoplasm (1, 6). The debate is whether the remaining lesions, often grouped as AF and related lesions, are in fact neoplasms, or stages in the development of the odontoma (2, 3) Cahn and Blum (7) postulated that an AF could develop eventually into an odontoma if the lesion had been allowed to remain. This would imply that all AF, AFD and AFO merely represent various stages of the same lesion, and will mature over time resulting in ultimately the formation of an odontoma (7–9). However, this 'continuum concept' has not been widely accepted for the following reasons. Considerable numbers of residual or recurrent cases of AF have
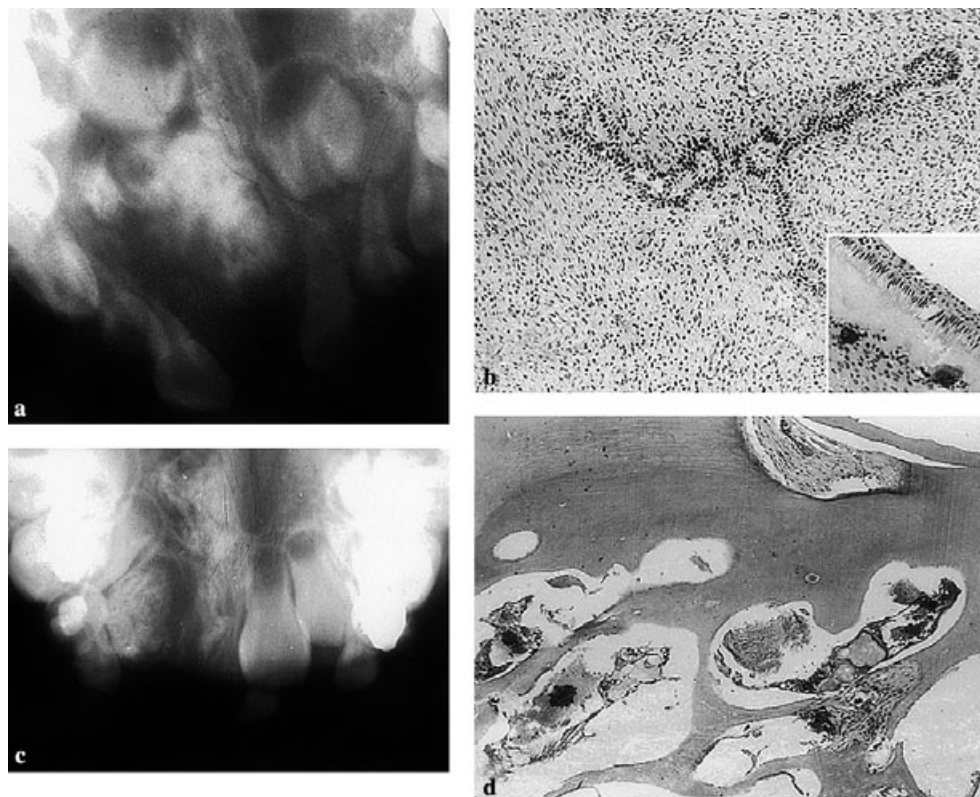
Figure 6 A mixed radiolucent and radiopaque lesion is seen in the right anterior part of the maxilla in close association with two developing tooth germs (a) (case no. 17). The primary lesion containing areas of ameloblastic fibroma with dental hard tissue formation (insert) is identical to AFO (b). Eight years following the initial enucleation, a recurrent lesion of a well-defined radiopaque mass with a radiolucent border is noted in the previous surgical site (c). The curetted materials contain only calcified dentin and enamel elements arranging in a pattern similar to complex odontoma (d).
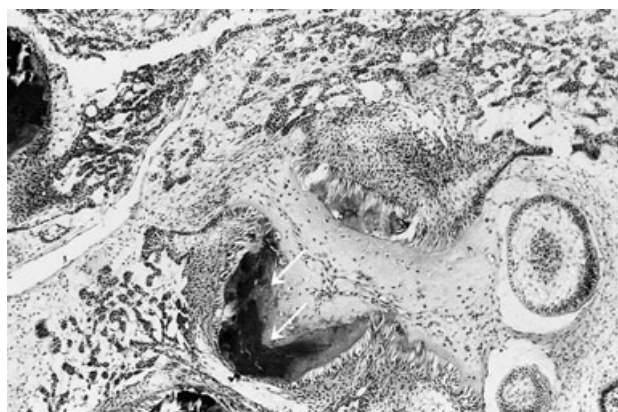

Figure 7 Ameloblastic fibro-odontoma showing similar features of ameloblastic fibroma, but also containing dentin and enamel matrix (arrows). Note the dentinoid materials containing entrapped mesenchymal cells are seen in the lesion.

shown no evidence of further maturation into a more differentiated odontogenic lesion, such as an AFO or an odontoma (10, 11). Furthermore, AFs are known to occur at ages well beyond completion of odontogenesis (2, 3). Our data showed that AF occurred mostly in the third decade with only four patients being younger than 22 years. When compared with those of other reports (mean: 9–15.5 years) (2, 3, 5, 12), the average age at time of diagnosis (26 years) in the present series was significantly higher. A similar age distribution of AF from the Chinese patients (mean: 24 years) was also reported(13). Although this may indicate possible geographic or racial differences in patient age of AF, it is also possible that many tumors from the present series had in fact existed for quite some time before diagnosis because of the relatively lower regular dental attendance in the Chinese population. The fact that all patients of AF in our series had presenting signs of jaw swelling and many tumors had reached a considerable size when first seen lends support for the later possibility. These cases may well be severed as examples of possible treatment delay, and yet the tumors were incapable of progressing into more differentiated ones. In addition, our data also demonstrated that AF occurred at a much older age than did the AFO (mean: 9.6 years). Therefore, these findings do not support the concept of AF developing into AFO and eventually into complex odontomas.

The neoplastic nature of AF is often suggested by the fact that some of these tumors could recur following surgery (12, 14–16) and that malignant transformation from a pre-existing AF has been occasionally reported (17–22). Of the four recurrent AFs presented here, two revealed unequivocal evidence of sarcomatous transformation of the previously benign mesenchymal tumor

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Table 3 Clinical features of odontomas

	Complex	Compound
Age		
0–9	1	4
10–19	14	22
20–29	7	3
30–39	5	4
40–49	2	0
50–59	2	0
60–69	1	0
70–79	1	0
Mean age (years)	25.9	15.1
Gender		
Male	16	15
Female	17	18
Location		
Mandible	23	9
Incisor-canine	3	8
Premolar	1	1
Molar	19	0
Maxilla	10	24
Incisor-canine	3	21
Premolar	1	3
Molar	6	0
Signs and symptom		
Asymptomatic	10	14
Swelling	19	4
Pain	14	0
Impacted teeth	4	15

component. As reported by many other authors (17–19, 21, 22), the epithelial elements within the malignantly transformed tumors disappeared completely in both tumors, thus showing histologic features identical to a fibrosarcoma. It is interesting to note that one AF of the present series involving a 6-year-old boy did show further maturation with typical tubular dentin formation in its recurrent tumor. This example indeed supports the views expressed by several authors (2, 3, 5, 11) that some AFs, especially those occurred during childhood, could represent the primitive or first stage of a developing complex odontoma. In other words, there exist two lesions that have the histologic appearance of AFs, the neoplastic AF and one stage of a developing odontoma. The problem is that in any given case we cannot distinguish between the actual neoplasm and an odontoma undergoing maturation. Analysis of our own data leads us to believe that majority of the histologically verified AFs is probably separate neoplastic entities, whereas a small number of lesions with a similar histology of AF in a child could represent a developing odontoma.

The average age of patients with AFO in our series was 9.6 years, with all seven patients being younger than 18 years. Six of seven cases occurred in the posterior mandible. This is in general agreement with other reported series (2, 3, 5). AFO has been reported to have a non-aggressive, hamartomatous behavior with little tendency to recur. Sporadic recurrences of AFO have been attributed to the inadequate surgical removal at the time of initial treatment (23, 24). The two recurrent AFOs presented here also appeared to be caused by incomplete excision of the primary lesions. The two

lesions, involving a 2-year-old boy (case no. 17) and a 6-year-old girl (case no. 18), were relatively large in size and were associated with contiguous developing tooth germs. To avoid the damage of the developing tooth germs, a conservative enucleation had to be applied in both cases. Interestingly, the subsequent follow-up of both recurrences in fact highlighted the limited growth potential of the lesions and their possible nature as immature or developing complex odontomas. Following 8 and 5 years of initial surgery respectively, both recurrent lesions did not appear to increase in size and instead became fully calcified and were histologically proven to be complex odontomas. In literature, however, it is not difficult to find reported cases of definitively neoplastic AFOs that demonstrate long-term and sustained growth without maturation into a complex odontoma (9). Thus, one could suppose that different lesions were being described under the same term of AFO, with some being hamartomatous in nature and others being a true de novo neoplasm. Histologic distinction between these lesions is difficult. The age of patients and the size of the lesion at initial discovery could be the important factors in judging the behavior and the nature of the lesions. Based on our observation as well as the previous reports (3, 5), AFO occurs primarily in persons under 20 years of age, and overall, has a less aggressive behavior with little tendency to recur in comparison with AF.

The AFD is a rare entity and its very existence is not completely accepted (1, 3, 4). Indeed, AFD is considered, by some authors (4), to occupy a stage between the AF and AFO based on the extent of histodifferentiation. Our present series confirmed its rarity and interestingly depicted an AF showing further maturation to form tubular dentin in its recurrent lesion. This particular case involving a 6-year-old boy could be severed as an example that there may be a maturation spectrum from the AF to the AFO with the AFD as an intermediate form. However, this does not necessarily suggest that all AFs will differentiate over time into an AFO or an odontoma.

Although odontomas are generally included in the classification of odontogenic tumors, most authorities will concede that these lesions are more properly considered to be malformations (hamartomas) rather than true neoplasm (1, 3, 5). Some authors suggest that for practical purposes the so-called complex and compound odontomas can be considered the same (14). However, our present data indicates that significant differences between the two types merit their continuing separation as two entities. The complex odontoma tended to occur at an older age (mean: 25.9 years) than the compound variant (mean: 15.1 years). The predominance of posterior mandible involvement (57.6%) was evident in the complex odontoma, whereas a predilection for the maxillary incisor-canine region (63.6%) was noted in the compound odontoma. Thus, our data as well as those of previous reports(3, 5, 25, 26) suggest that the two malformations are pathogenetically different. The complex odontoma, having an expected higher age at time of diagnosis and a similar predilection for

the posterior mandible area as compared with AFO, may well represent the terminal stage of the hamartomatous lesions coded as AFOs. In fact, the disordered arrangement of odontogenic epithelium and ectomesenchyme as well as the irregularly formed primitive dentin and enamel matrix in an AFO make it unlikely that any attempt at producing tooth-like structures in the lesion could prevail. The compound odontoma, on the contrary, hardly showed any clinicopathologic correlation with AFO. We concur with the idea that its pathogenesis is different from that of the complex odontoma and its occurrence may be the result of 'multiple schizodontia' because of a local hyperactivity of the dental lamina (3).

In summary, the present study indicates that AFs are commonly seen in adults past the tooth-developing age (over 22 years), which suggests that the majority of AFs are neoplastic in nature. Its tendency to recur and to undergo malignant transformation also denotes its neoplastic character. However an AF being diagnosed in childhood could raise the possibility to represent the initial and most primitive stage of a developing complex odontoma. All the AFOs presented here were cases developing under the age of 22 years. The sequential age difference and the common site predilection observed between the AFO and the complex odontoma suggest that at least some AFOs are hamartomatous in nature, representing a stage preceding the complex odontoma. It has to be emphasized, however, that at present we are unable to differentiate a hamartomatous lesion from a neoplasm among this group of lesions merely on histologic grounds. Experiments are currently underway in our laboratory to examine the clonal status of AF and its related lesions using techniques of clonal analysis (27, 28) in order to further clarify this heterogeneous group of lesions.

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