

# Odontogenic Myxoma

## A Clinicopathologic Study of 25 Cases

Tie-Jun Li, PhD; Li-Sha Sun, MDS; Hai-Yan Luo, MDS

● **Context.**—Odontogenic myxoma is an uncommon tumor that has the potential for extensive destruction of the jaws.

**Objective.**—To document the clinical, pathologic, and behavioral features of odontogenic myxomas.

**Design.**—Histologic and immunocytochemical examinations were performed on odontogenic myxomas from 25 Chinese patients. Clinical and available follow-up data were analyzed.

**Results.**—In the present series, 13 were male and 12 female. The age at diagnosis ranged from 6 to 66 years, with a mean age of 28.8 years. Twelve tumors involved the mandible and 13 occurred in the maxilla, with a predilection for posterior areas. The posterior maxillary tumors frequently (9/10) involved the maxillary sinus. Of the 23 cases with radiographic records, 22 lesions presented with a multilocular appearance. Although 80% of the mandibular lesions showed a well-defined border, only 33.3% of the

maxillary tumors were well-defined. Histologically, odontogenic myxomas were mainly composed of spindled or stellate-shaped cells in a mucoid-rich intercellular matrix. Tumors containing noticeable fibrous components were evident in 13 cases. Apart from 5 cases treated conservatively by enucleation, the remaining 20 cases were treated by relatively radical procedures, including block/segmental resection and partial or total maxillectomy or mandibulectomy. Follow-up data were available on 22 patients and only 1 patient initially treated by enucleation had a recurrence.

**Conclusions.**—Odontogenic myxomas have a very bland histologic appearance that lacks atypia and may easily lead to misdiagnosis. The tumors are infiltrative with no capsulation and may recur after inadequate surgery.

(*Arch Pathol Lab Med.* 2006;130:1799–1806)

Myxomas are benign, but locally invasive, neoplasms that rarely appear in the skeleton. When they do occur in osseous sites, they are found almost exclusively in the jaws.<sup>1,2</sup> The World Health Organization<sup>3</sup> and many authorities<sup>1,4</sup> consider the jaw myxoma to be an odontogenic tumor on the basis of its site, which is almost exclusive to the tooth-bearing portions of the jaws; the common occurrence in youth or in association with missing teeth; the structural resemblance to dental mesenchyme; and the sporadic presence of islands of odontogenic epithelium. Although these evidences are largely circumstantial, an origin from the odontogenic apparatus appears highly likely. Since first described by Thoma and Goldman<sup>1</sup> in 1947, myxomas of the jaws have continually been the subject of much debate. Apart from its disputable origin from odontogenic tissues, the discussion has mainly been on therapy with recommendations varying from enucleation to extensive surgery followed by radiotherapy.<sup>5</sup> Moreover the discussion has been complicated by the possibility that other tumor entities have been included in some series of myxomas, mimicking these by myxomatous degenera-

tion.<sup>6</sup> This article describes the clinicopathologic presentation of a large series of odontogenic myxomas in China for a period of 20 years and compares this information with that available in the literature.

### MATERIALS AND METHODS

After reviewing the patient details, clinical information, and histology, 25 cases fulfilling the criteria of odontogenic myxoma were identified from the files of the Department of Oral Pathology, School of Stomatology, Peking University during the years 1985 through 2005. For inclusion in this series, all cases were primary intraosseous tumors of the jaws. The tumors were composed of rounded and angular cells lying in an abundant mucoid stroma, as defined by the World Health Organization.<sup>3</sup> Most tumors contained little collagen, but some of these tumors consisting of isolated thick collagen bands were classified as myxofibromas. Gingival masses with similar histologic features but not showing bone involvement were not included in the series. Clinical outcome and follow-up data were evaluated by consulting individual clinical reports and pathology files. In addition, the patients or close relatives were contacted and asked to complete a questionnaire regarding the current condition of the patient. The surgical specimens had been routinely fixed in 10% neutral formalin (18–48 hours), processed, and embedded in paraffin. In some cases, decalcification of the specimen was performed in 10% formic acid for 5 to 72 hours. Serial sections of 4- $\mu$ m thick were cut and used for hematoxylin-eosin, periodic acid-Schiff (with or without diastase digestion), alcian blue (pH 2.5), and immunocytochemical staining. Immunocytochemical staining was performed using a standard streptavidin-biotin-peroxidase complex method (LAB-SA kits, Zymed Laboratories, South San Francisco, Calif). Details of primary antibodies used are listed in Table 1.

Accepted for publication May 4, 2006.

From the Department of Oral Pathology, School and Hospital of Stomatology, Peking University, Beijing, PR China.

The authors have no relevant financial interest in the products or companies described in this article.

Reprints: Tie-Jun Li, PhD, Department of Oral Pathology, School and Hospital of Stomatology, Peking University, 22 S Zhongguancun Ave, Haidian District, Beijing, PR China 100081 (e-mail: litiejun22@vip.sina.com).

**Table 1. Technical Data of Immunocytochemical Staining\***

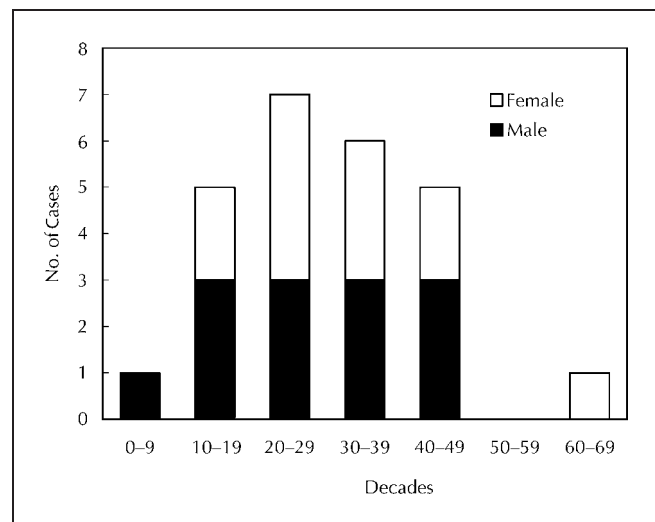
| Antibodies (clone)                    | Pretreatment  | Dilution |
|---------------------------------------|---------------|----------|
| Pankeratin (AE1/AE3)                  | Citrate HIER  | 1:100    |
| Keratin 19 (RCK108)                   | Trypsin (20') | 1:100    |
| S100 (4C4.9)                          | None          | 1:50     |
| Vimentin (V9)                         | None          | 1:50     |
| Desmin (ZC18)                         | None          | 1:200    |
| Smooth muscle actin (1A4)             | None          | 1:100    |
| Neuron-specific enolase (E27)         | None          | 1:50     |
| Glial fibrillary acid protein (ZCG29) | None          | 1:50     |

\* All antibodies were purchased from Zymed Laboratories, South San Francisco, Calif. HIER indicates heat-induced epitope retrieval.

## RESULTS

### Clinical Findings

The clinical features of patients are summarized in Table 2. Of the 25 patients, 13 were male and 12 female. The age at diagnosis ranged from 6 to 66 years, with a mean age of 28.8 years. The majority of the cases (23 cases [92%]) were diagnosed between the second and fifth decades, with only 1 case being younger than age 10 years and 1 case older than age 50 years (Figure 1). Twelve tumors involved the mandible and 13 occurred in the maxilla. For the purpose of analysis, the location of the lesion was recorded according to the center of the lesion. Of the 12 mandibular tumors, 9 (75%) involved the premolar and molar region, 6 of which extended to the ramus; 2 (16.7%) occurred in the ascending ramus; and the remaining 1 case was located in the canine and incisor area. One large mandibular tumor crossed the midline. Ten (76.9%) of the 13 maxillary tumors occurred in the premolar and molar region, 9 of which extended into the maxillary sinus. Only 3 maxillary tumors involved the anterior portion, 1 in the incisors region and the other 2 in the canine-premolar region, respectively (Figure 2). Although 1 of the anterior maxillary tumors extended from the left second premolar to the right first incisor, involving the sinus and crossing the midline, the other 2 were relatively small lesions and were situated between the roots of teeth. All the posterior tumors in both the mandible and maxilla tended to be large and destructive with frequent involvement of the ramus or the maxillary antrum. Swelling, ranging in duration from 1 month to 17 years, was the principal finding in all cases. Six patients also complained of pain, and 2 mandibular cases showed sign of paraesthesia of the lower lip. Nosebleed was recorded in 3 maxillary cases. Mobility of the associated teeth was noted in 7 cases. Radiographs were available in 23 patients. Twenty-two of 23 lesions presented with a multilocular appearance. These lesions were characterized by the presence of fine or "wispy" bony trabeculae within the radiolucent defects (Figure 3, a and b). The borders of the lesion were well defined in 12 (54.5%) cases and poorly defined in 10 (45.5%). Although 8 (80%) of 10 mandibular lesions showed a well-defined border, only 4 (33.3%) of 12 maxillary lesions were judged as well-defined lesions. The 1 unilocular lesion was a well-defined radiolucency involving the mandibular molar and angle region (Figure 3, c). As radiolucencies with soap bubble or honeycombed appearance were the principal findings in most cases, 3 lesions of the present series showed a mixed radiolucent-radiopaque feature, 1 of which revealed a sunray or sunburst appearance similar to that seen in an osteosarcoma (Figure 3, d). Marked cortical expansion was noted in 13 lesions, tooth/root dis-

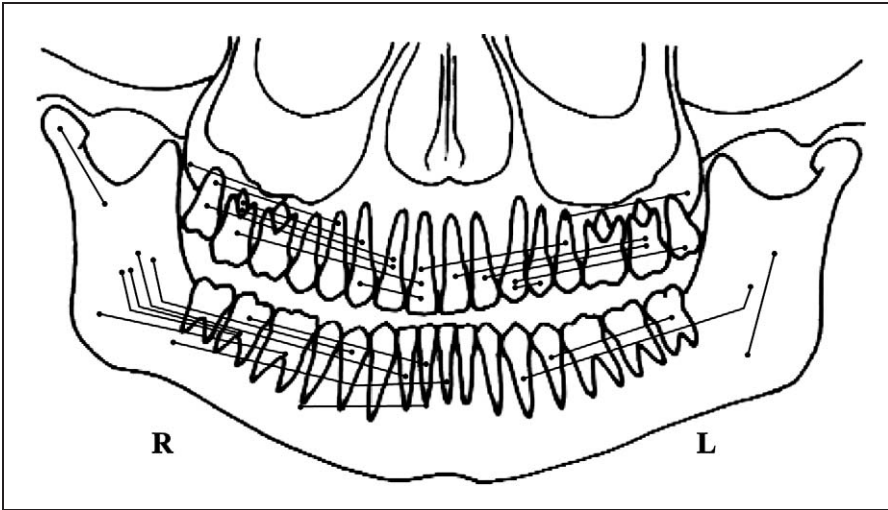


**Figure 1.** Age and sex distribution of odontogenic myxoma.

placement in 11, impacted teeth in 6, and root resorption in 3.

### Pathologic Features

Gross pathology of myxoma specimens from the jaws usually showed a whitish-grey glistening or gelatinous mass with minimal true encapsulation (Figure 4, a). Histologically, all odontogenic myxomas were mainly composed of spindle or stellate-shaped cells in a mucoid-rich intercellular matrix (Figure 4, b), but histologic variations in different tumors or areas did exist. A search was made for the following characteristics: cellularity, pleomorphism, mitotic figures, multinucleation, encapsulation, permeation of bone, vascularity, lobulation, odontogenic epithelium, and degree of fibrosis calcification. The majority of the tumors was monotonous and hypocellular with a preponderance of spindle or stellate-shaped cells. Cytoplasmic processes were often long and anastomosed with other cell processes. Nuclei were usually small, inconspicuous, and hyperchromatic. Although binucleated cells were present in 13 tumors (Figure 4, c), mitosis and multinucleation were uncommon. There was little evidence of encapsulation in the present series and the margins were ill defined with increased osteoclastic activity in the peripheral bone (Figure 4, d). Sparsely scattered residual bony trabeculae were frequently observed in 14 (56%) of tumors (Figure 4, e). Vascularity was minimal and inconspicuous with only 2 tumors exhibiting a marked increase of blood vessels. Also infrequent were lobulation (3 cases) and nests of odontogenic epithelium (2 cases). The quantity of fibrous



**Figure 2.** Anatomic location and extension of 25 odontogenic myxomas.



**Figure 3.** Radiographs of odontogenic myxomas showing multilocular radiolucencies with fine bony trabeculae in the defect in a mandibular tumor (a) and a maxillary tumor (b). b, The border of the maxillary lesion in the premolar and molar area is poorly defined. c, A unilocular radiolucency involves the mandibular molar and angle area associated with an unerupted tooth. d, A mixed radiolucent-radiopaque lesion in the mandible showing a similar sunray appearance.

tissue varied considerably in each case and in each specimen. In most cases, increased fibers showed a very loose arrangement or could be seen only in the periphery of the tumor or the tumor nodules. Tumors containing noticeable fibrous component and/or thick collagenous bands were evident in 13 cases. The mucoid-rich extracellular matrix showed positive staining for Alcian blue (Figure 4, f) and

periodic acid-Schiff. Immunostaining of the tumor cells showed uniform positivity with vimentin and patchy staining with smooth muscle actin. Cells were negative for desmin, neuron-specific enolase, glial fibrillary acid protein, and S100. Keratins (both pankeratin and CK19) was consistently expressed by the epithelial nests in the 2 myxomas in which epithelium was present (Figure 4, g).

**Table 2. Clinical Features of Odontogenic Myxomas**

| Case No. | Age, y/Sex* | Location  | Duration | Symptoms and Radiographic Findings   | Treatment and Follow-up†  |
|----------|-------------|---|----------|--|---|
| 1        | 40/F        | R mandible: lateral incisor to ramus            | 1 mo     | Swelling, pain, mobility of the posterior teeth; well-defined multilocular radiolucency, cortical expansion, and root resorption   | Segmental resection 5 mo after surgery, NSR   |
| 2        | 66/F        | R mandibular condyle and upper ramus            | 17 y     | Swelling, pain; poorly defined multilocular radiolucency, cortical expansion   | R Hemimandilectomy 2 y after surgery, NSR, lost to follow-up                                      |
| 3        | 32/M        | Mandible: R 2nd premolar to L 1st incisor       | 1 mo     | Swelling; well-defined multilocular radiolucency, displacement of the anterior teeth   | Enucleation and curettage 7 y after surgery, NSR  |
| 4        | 28/M        | L maxilla: 2nd premolar to maxillary tuberosity | 3 mo     | Swelling, mobility of the posterior teeth, sinus involvement; radiograph not available   | Enucleation and curettage 8 y after surgery, NSR  |
| 5        | 20/M        | L mandible: cuspid to ramus                     | 1 y      | Swelling, pain, mobility of the posterior teeth; well-defined multilocular radiolucency, cortical expansion, impacted and displaced 2nd and 3rd molars                           | Segmental resection 1 y after surgery, NSR, lost to follow-up                                     |
| 6        | 22/F        | R maxilla: incisor to 2nd premolar              | 3 y      | Swelling; poorly defined multilocular radiolucency, cortical expansion, sinus involvement  | Partial maxillectomy 3 y and 4 mo after surgery, NSR  |
| 7        | 17/F        | L mandible: ramus region                        | 4 mo     | Swelling, pain; radiograph not available   | Segmental resection 2 y and 6 mo after surgery, NSR   |
| 8        | 46/M        | Mandible: R 3rd molar to L lateral incisor      | 2 y      | Swelling, pain, transient paraesthesia of lip; well-defined multilocular radiolucency, cortical expansion, displacement of teeth   | Segmental resection 4 y after surgery, NSR  |
| 9        | 33/M        | R maxilla: cuspid to 2nd molar                  | 5 mo     | Swelling; ill-defined multilocular radiolucency, root resorption, sinus involvement  | Enucleation and curettage REC (6 mo), partial maxillectomy 2 y and 7 mo after second surgery, NSR |
| 10       | 18/F        | R maxilla: lateral incisor to 3rd molar         | 1 y      | Swelling, nosebleed; poorly defined multilocular radiolucency, cortical expansion, sinus involvement   | Partial maxillectomy 12 y after surgery, NSR  |
| 11       | 24/M        | Maxilla: R 1st incisor to L 2nd premolar        | 1 mo     | Swelling, nosebleed; poorly defined multilocular radiolucency, displacement of teeth, sinus involvement  | Partial maxillectomy 3 y after surgery, NSR   |
| 12       | 12/M        | R mandible: 2nd molar to ramus                  | 2 mo     | Swelling; well-defined multilocular radiolucency, cortical expansion, impacted 1st molar, displacement of teeth  | Segmental resection 3 y after surgery, NSR  |
| 13       | 36/F        | R mandible: 2nd molar to ramus                  | 2 y      | Swelling, pain; poorly defined multilocular radiolucency, cortical expansion   | Segmental resection 3 mo after surgery, NSR, lost to follow-up                                    |
| 14       | 15/M        | L maxilla: 1st incisor to 2nd molar             | 2 mo     | Swelling, mobility of the posterior teeth; poorly defined mixed radiolucent-radiopaque lesion, multilocular, cortical expansion, sinus involvement, impacted and displaced molar | Partial maxillectomy 1 y after surgery, NSR, lost to follow-up                                    |
| 15       | 18/M        | R maxilla: lateral incisor to 2nd molar         | 2 y      | Swelling, nosebleed; poorly defined multilocular radiolucency, cortical expansion, sinus and orbit involvement, impacted and displaced 3rd molar                                 | Maxillectomy 6 y and 4 mo after surgery, NSR  |
| 16       | 7/M         | R mandible: 1st molar to mandibular angle       | 6 mo     | Swelling; well-defined unilocular radiolucency, impacted molar   | Enucleation and curettage 7 y after surgery, NSR  |
| 17       | 22/F        | R mandible: 1st premolar to ramus               | 6 mo     | Swelling; well-defined multilocular radiolucency with radiopaque bone spicules showing sunray/sunburst appearance, cortical expansion, impacted and displaced molar              | Block resection 7 y after surgery, NSR  |
| 18       | 21/F        | R maxilla: 1st molar to maxillary tuberosity    | 2 mo     | Swelling, mobility of molars; poorly defined multilocular radiolucency, sinus involvement  | Partial maxillectomy 2 y after surgery, NSR   |



**Table 2. Continued**

| Case No. | Age, y/Sex* | Location                                | Duration | Symptoms and Radiographic Findings  | Treatment and Follow-up†                            |
|----------|-------------|---|----------|---|---|
| 19       | 37/M        | R mandible: 1st incisor to 2nd molar    | 2 y      | Swelling; well-defined multilocular radiolucency, cortical expansion, root resorption                                   | Curettage 11 y after surgery, NSR                   |
| 20       | 30/F        | L maxilla: lateral incisor to 2nd molar | 3 yr     | Swelling; well-defined multilocular radiolucency  | Partial maxillectomy 2 y after surgery, NSR         |
| 21       | 24/F        | R maxilla: 1st premolar to 3rd molar    | 7 mo     | Swelling, pain, mobility of the posterior teeth; well-defined multilocular radiolucency, sinus involvement              | Partial maxillectomy 3 y after surgery, NSR         |
| 22       | 40/M        | L maxilla: cuspid to 1st premolar       | 4 y      | Swelling; well-defined multilocular radiolucency causing root deviation   | Block resection 10 y and 6 mo after surgery, NSR    |
| 23       | 41/F        | R maxilla: 1st incisor to cuspid        | 1 y      | Swelling; well-defined multilocular radiolucency, cortical expansion, displacement of teeth                             | Block resection 4y and 6 mo after surgery, NSR      |
| 24       | 32/F        | L mandible: 1st premolar to 3rd molar   | 1 y      | Swelling, paraesthesia of lip, mobility of the posterior teeth; well-defined multilocular radiolucency, root resorption | Segmental resection 3 y and 8 mo after surgery, NSR |
| 25       | 42/M        | L maxilla: 1st premolar to 3rd molar    | 7 y      | Swelling; ill-defined mixed radiolucent-radiopaque lesion, multilocular, cortical expansion, sinus involvement          | Partial maxillectomy 3 mo after surgery, NSR        |

\* Age at first diagnosis.

† NSR indicates no sign of recurrence; REC, recurrence; R, right; L, left.

**Treatment and Follow-up**

In 5 cases, the primary treatment was conservative consisting of careful enucleation followed by curettage of the bony tumor bed. Four patients showed no sign of recurrence after being followed 7 to 11 years. One patient with a maxillary tumor initially treated in a conservative way exhibited recurrent tumor 6 months after the surgery. The recurrent tumor caused further destruction of the bone with involvement of the maxillary sinus and was then treated by partial maxillectomy. No recurrence was detected 2.5 years following the second operation. Three patients underwent block resection and none of them manifested recurrence after a follow-up period of 7 to 10.5 years. The majority of the patients (17 cases) in this series were treated by relatively radical procedures, including segmental resection (7 cases), partial maxillectomy (8 cases), maxillectomy (1 case), and hemimandibulectomy (1 case). Follow-up data were available on 14 patients and the remaining 3 were lost to follow-up after being followed for 3 months to 1 year. Apart from 2 recent patients who were only followed for 3 and 5 months, respectively, the postoperative observation period of the other 12 patients ranged from 2 to 12 years (mean, 3.9 years) and none of them developed recurrence.

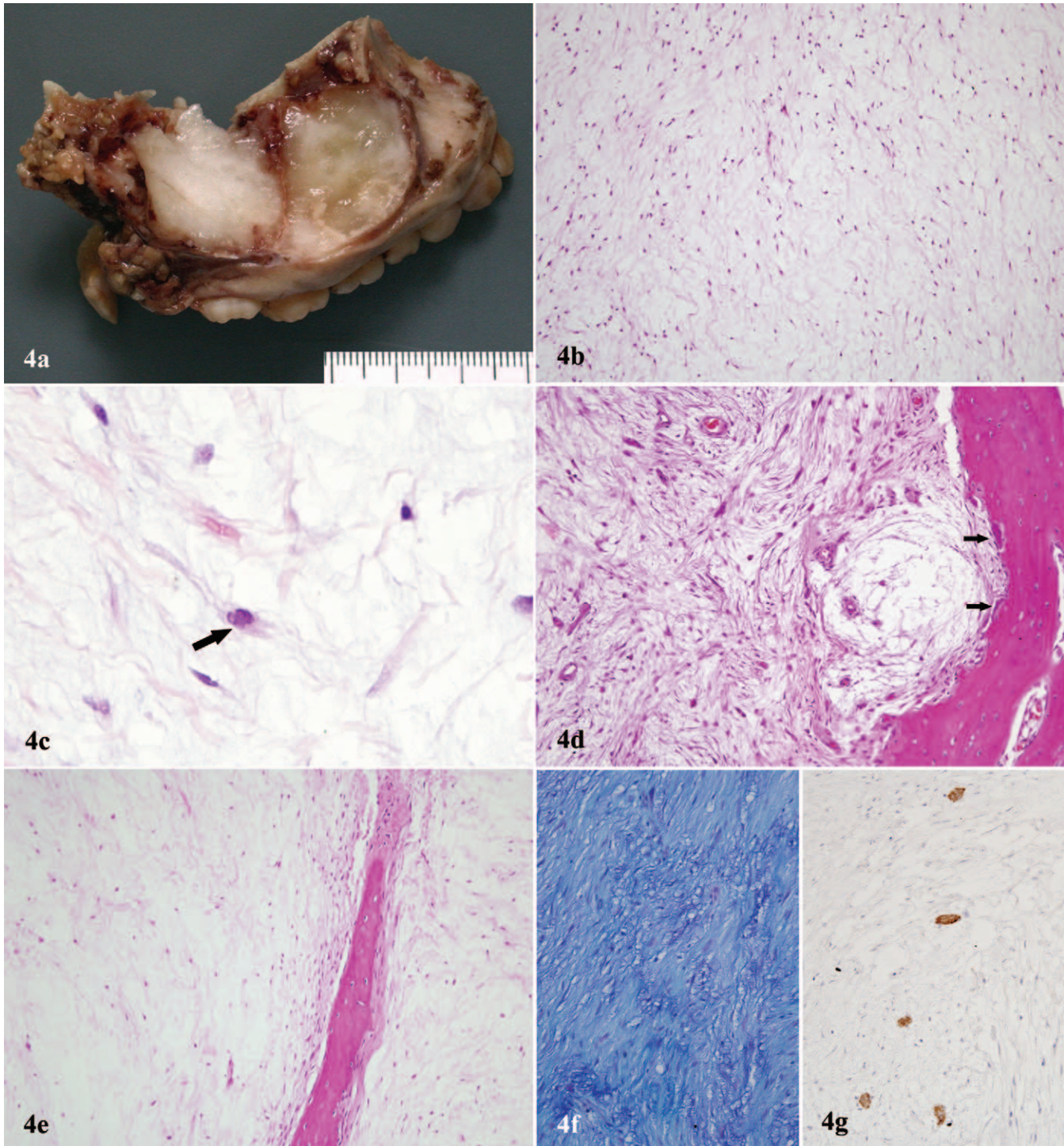
**COMMENT**

Mucoid tumors of soft tissue represent a heterogeneous group of lesions that exhibit significant differences in biologic behavior, ranging from harmless to malignant neoplasms.<sup>7</sup> As an osseous entity, however, odontogenic myxomas are found predominantly in the bone of the jaws and are considered slow-growing tumors with the potential for extensive bone destruction, cortical expansion, and a relatively high recurrence rate.<sup>1,2,4,8,9</sup> A review of English-language literature between 1965 and 1995 by Kaffe et al<sup>10</sup> disclosed a total number of 164 cases of odontogenic myxomas with relevant information of the age, gender, and

location. The mean age of the patients at the time of diagnosis is 29.6 years, ranging from 1 to 73 years with 75% occurring in the second to the fourth decades. The tumors occur more often in females (100 cases [61%]) than in males (64 cases [39%]) and are located in the mandible in two thirds (109 cases) and in the maxilla in one third (55 cases) of cases. The age distribution of the present series was in keeping with this review, with the majority of the patients (92%) being older than the age of 10 years and younger than 50 years. However, our data showed no sex predilection, which is in agreement with several other studies.<sup>3,11-14</sup> It is also apparent from this study that there was an approximately equal number of maxillary and mandibular cases. This concurs with findings from some reports,<sup>11,14,15</sup> but there are studies in the literature revealing more mandibular tumors.<sup>5,6,12</sup> Such wide variations in both gender and location may indicate geographic variations. Although our data on gender and location of odontogenic myxomas are comparable with a demographic study of 759 odontogenic tumors in a Chinese population,<sup>14</sup> there are several studies from the African continent reporting a marked gender bias with male-female ratios ranging from 1:2 to 1:3 and a strong predilection for the mandible as compared with the maxilla (2.5:1).<sup>6,16,17</sup>

Despite the differences with respect to location, most odontogenic myxomas occur in the premolar and molar region of both jaws. In our series, posterior tumors in both the maxilla and mandible tended to be larger and more destructive with frequent involvement of the maxillary sinus (9 of 10 cases) and the mandible ramus (6 of 9 cases). Three of 4 anterior tumors were small lesions between roots of teeth. Twenty-three of 25 tumors occupied only 1 side of the jaws (right or left). This concurs with the notion that odontogenic myxoma rarely crosses the midline.<sup>10,18</sup>

The radiographic features of odontogenic myxomas described in the literature are varied, ranging from small unilocular lesions to large multilocular tumors that often



**Figure 4.** *a*, Transected surgical specimen of a maxillary myxoma. The whitish glistening tumor mass fills a bony cavity with minimal true encapsulation. *b*, Histologically, odontogenic myxoma is characterized by proliferation of spindled and stellate cells in a mucoid-rich matrix (hematoxylin-eosin, original magnification  $\times 100$ ). *c*, High-power view showing a binucleated cell (arrow) in the tumor (hematoxylin-eosin, original magnification  $\times 400$ ). *d*, Increased osteoclastic activity (arrows) is evident in the peripheral bone adjacent to the tumor and there is little evidence of capsulation in the periphery of the tumor (hematoxylin-eosin, original magnification  $\times 200$ ). *e*, Residual bony trabeculae are scattered in the tumor (hematoxylin-eosin, original magnification  $\times 100$ ). *f*, The extracellular matrix is alcian blue positive (alcian blue, pH 2.5, original magnification  $\times 200$ ). *g*, Immunostaining shows keratin reactivity in the epithelial nests in the tumor (streptavidin-biotin, original magnification  $\times 200$ ).

displace teeth or less frequently resorb roots of teeth.<sup>10,19</sup> Borders may be well demarcated or ill defined.<sup>10</sup> In contrast to the previous reports, which often indicate that nearly half of the tumors are unilocular, all but 1 lesion in the present series with radiographic records were de-

scribed as multilocular tumors. Radiolucencies, separated by many bony trabeculae and resulting in a soap bubble or honeycomb appearance, were common features in the majority of the cases, except for 3 tumors exhibiting a mixed radiolucent-radiopaque feature. More than one half



(54.5%) of the lesions were well defined, but poorly defined borders were seen in the remaining tumors. There seemed to be no correlation between the borders of the lesion and the amount of bony trabeculae within the lesion, but maxillary tumors (66.7%) were more likely to be ill defined in nature compared with mandibular lesions (20%). Radiographically, myxomas may present similar features of an ameloblastoma or a central giant cell granuloma. One maxillary tumor of this series with a mixed radiolucent-radiopaque appearance also exhibited a similar radiating pattern that may mimic an osteosarcoma. These facts do pose potential difficulties in reaching a proper diagnosis merely on radiographic studies. A biopsy is, therefore, necessary to ascertain an accurate diagnosis.<sup>10,16</sup>

Odontogenic myxoma is usually characterized by cortical expansion of the jawbones and, in maxillary lesions, by frequent extension into the maxillary sinus.<sup>4,10,15,18</sup> Our results strongly support these statements. Thirteen (56.5%) of 23 cases with radiographic records showed a marked cortical expansion and 10 (76.9%) of the 13 maxillary tumors extended into the maxillary sinus. According to some studies, root displacement rather than resorption is the rule of jaw myxomas.<sup>4,10,19</sup> This is supported by the results of the present study as tooth/root displacement was noted in 11 (47.8%) of cases but root resorption in only 3 (13%).

The specimens of the present series appeared macroscopically as an infiltrative mass of mucoid or gelatinous material. All tumors were composed of loosely arranged, spindle-shaped and stellate cells many of which had long tapering cytoplasmic processes. The intercellular background was mucoid. Binucleated cells were frequently encountered, but cellular pleomorphism, mitotic figures, and multinucleation were rare. Most myxomas in this series contained little collagen, but in 13 cases areas with moderate amount of fibrous components were observed. When collagen fibrils are prominent, these tumors are often designated as myxofibromas,<sup>3,18</sup> which are probably identical to what has previously been reported as the simple type of odontogenic fibroma.<sup>20</sup> Residual bony trabeculae are also 1 of the characteristic microscopic features of odontogenic myxomas, with more than half of our cases manifesting this feature. It is believed that the mixed radiolucent-radiopaque appearance on radiography may be due to these residual bone structures within the tumor.<sup>5,10,21</sup> Some other infiltrative and bone-destroying odontogenic tumors, mainly ameloblastomas, almost never contain residual bony trabeculae except its infiltrative margins.<sup>3</sup> Immunocytochemical findings of this study were consistent with the results reported by Green et al<sup>22</sup> and Lo Muzio et al.<sup>21</sup> All tumor cells were found to be positive for vimentin, and a fraction of the tumor cells showed positivity for smooth muscle actin. All lesions were negative for desmin, neuron-specific enolase, glial fibrillary acid protein, and S100. S100 positivity in a minority of odontogenic myxomas has been reported by Lombardi,<sup>23</sup> but several other studies<sup>21,22,24</sup> as well as the present study fail to identify S100 reactivity in the tumors. Thus, these findings are compatible with a fibroblastic or myofibroblastic cell type for the tumor.

Myxoid change in other neoplasms may mimic odontogenic myxomas.<sup>25</sup> Such appearances may be found in myxoid neurofibroma, myxoid lipoma, and chondromyxoid fibroma. Clinical correlation, microscopic features,

and immunocytochemistry should allow an appropriate diagnosis to be made. Dental follicles or papillae, normal developmental structures of odontogenesis that are occasionally removed in conjunction with unerupted or impacted teeth, may be misinterpreted as a myxoma.<sup>26</sup> Dental follicles usually contain odontogenic epithelial rests in the majority of cases, in a fibrous background showing a variable degree of myxoid change, and are often partially lined by reduced enamel epithelium, in contrast to myxomas. Radiographically, dental follicles are well-demarcated, thin, semicircular radiolucencies around unerupted teeth.<sup>26</sup> In contrast, odontogenic myxomas are larger, often expansile, and poorly defined radiolucent lesions. Dental papillae easily become separated from developing teeth during surgery or gross specimen examination and could be erroneously interpreted as odontogenic myxoma. However, dental papillae consist of well-circumscribed elliptical pieces of immature mesenchymal tissue, 1.5 cm or less in diameter, which may be rimmed by odontoblasts. The deposition of eosinophilic dentinoid tissue along the periphery of this myxoid tissue is another helpful differential point in support of dental papilla.<sup>26</sup>

The recommended treatment of choice for odontogenic myxomas is radical surgery<sup>16,27</sup> or conservative excision depending on tumor size.<sup>15,21,28</sup> All the cases in the present series were treated by surgery and only 1 of 22 patients with follow-up records recurred. Although the 17 patients treated by relatively radical procedures, such as block/segmental resection, partial maxillectomy, or hemimandibulectomy, showed no sign of recurrence, 1 of 5 conservatively treated (enucleation/curettage) patients exhibited recurrent tumor. In the literature, recurrence rates range from 10% to 33% with a reported average of 25%.<sup>4,11,21</sup> Recurrence rate differences are obviously related to the method of treatment, with conservative procedures resulting in more recurrences.<sup>4,13</sup> Odontogenic myxomas are not encapsulated and often infiltrate through bone without any well-defined borders.<sup>8,9,25</sup> Thus, complete surgical removal can be difficult, especially in the maxilla because of the proximity of vital structures and the more complex anatomy.<sup>4,25,29</sup> Our data indicate that recurrence rate can be greatly reduced by application of wide excision or resection of the lesion together with adjacent tissue. However, there are those who prefer limited conservative treatment, as a significant number of myxomas are treated conservatively by excision or curettage with no recurrence.<sup>5,21,28</sup> Indeed, in the present series, the conservative procedure was successful in achieving recurrence-free outcome in 4 of 5 cases. Some authors believe an initial conservative approach sparing uninvolved structures could be used to allow maximal preservation of function, reserving the more radical approach only for recurrences.<sup>5,30</sup> The prime reason for recurrence is thought to be related to incomplete removal rather than the intrinsic biologic behavior of the tumor.<sup>31</sup> Although there are few studies regarding this, radiotherapy and chemotherapy appear to be ineffective in controlling the recurrent lesions. It is suggested that patients be followed closely for at least 2 years because this is the most likely time for recurrence.<sup>11,32</sup>

This work was partly supported by research grant 30572048 from National Nature Science Foundation of China .

#### References

1. Thoma KH, Goldman HM. Central myxoma of the jaw. *Am J Oral Surg Orthod.* 1947;33:532-540.

2. McClure DK, Dahlin DC. Myxoma of the bone: report of 3 cases. *Mayo Clin Proc.* 1977;52:249–253.
3. Buchner A, Odell EW. Odontogenic myxoma/myxofibroma. In: Barnes L, Eveson JW, Reichart P, Sidransky D, eds. *Pathology and Genetics of Head and Neck Tumors*. Lyon, France: IRAC Press; 2005:316–317. *World Health Organization Classification of Tumours*.
4. Baker BF. Odontogenic myxoma. *Semin Diagn Pathol.* 1999;16:297–301.
5. Slootweg PJ, Wittkamp ARM. Myxoma of the jaws: an analysis of 15 cases. *J Maxillofac Surg.* 1986;14:46–52.
6. Adekeye EO, Avery BS, Edwards MB, Williams HK. Advanced central myxoma of the jaws in Nigeria: clinical features, treatment and pathogenesis. *Int J Oral Surg.* 1984;13:177–186.
7. Graadt van Roggen JF, Hogendoorn PCW, Fletcher CDM. Myxoid tumors of soft tissue. *Histopathology.* 1999;35:291–312.
8. Zohar Y. Proper treatment of odontogenic myxoma. *J Oral Maxillofac Surg.* 1989;47:546–548.
9. Wong GB. Large odontogenic myxoma of the mandible treated by sagittal ramus osteotomy and peripheral ostectomy. *J Oral Maxillofac Surg.* 1992;50:1221–1224.
10. Kaffe I, Naor H, Buchner A. Clinical and radiological features of odontogenic myxoma of the jaws. *Dentomaxillofac Radiol.* 1997;26:299–303.
11. Barros RE, Dominguez FV, Cabrini RL. Myxoma of the jaws. *Oral Surg.* 1969;27:225–235.
12. Van Der Weel I. *Disease of the Jaws: Diagnosis and Treatment*. Copenhagen: Munksgaard; 1991:206.
13. White DK, Chen SY, Mohnac AM, Miller AS. Odontogenic myxoma: a clinical and ultrastructural study. *Oral Surg.* 1975;39:901–917.
14. Lu Y, Xuan M, Takata T, et al. Odontogenic tumors: a demographic study of 759 cases in a Chinese population. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 1998;86:707–714.
15. Keszler A, Dominguez FV, Giannuzio G. Myxoma in childhood: an analysis of 10 cases. *J Oral Maxillofac Surg.* 1995;53:518–521.
16. Simon ENM, Merckx MAW, Vuhahula E, Ngassapa D, Stoelinga PJW. Odontogenic myxoma: a clinicopathological study of 33 cases. *Int J Oral Maxillofac Surg.* 2004;33:333–337.
17. Odukoya O. Odontogenic tumors: an analysis of 289 cases. *J Oral Pathol Med.* 1995;24:454–457.
18. Farman AG, Nortje CJ, Grotepass FW, Farman FJ, Wan Zyl JA. Myxofibroma of the jaws. *Br J Oral Surg.* 1977;15:3–18.
19. Peltola J, Magnusson B, Happonen RP, Borrmann. Odontogenic myxoma: a radiographic study of 21 tumors. *Br J Oral Maxillofac Surg.* 1994;32:298–302.
20. Dunlap CL. Odontogenic fibroma. *Semin Diagn Pathol.* 1999;16:293–296.
21. Lo Muzio L, Nocini PF, Favia G, Procacini M, Mignogna D. Odontogenic myxoma of the jaws: a clinical, radiological, immunohistochemical and ultrastructural study. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 1996;82:426–433.
22. Green TL, Leighty SM, Walters R. Immunohistochemical evaluation of oral myxoid lesions. *Oral Surg Oral Med Oral Pathol.* 1992;73:469–471.
23. Lombardi T. Comparative immunohistochemical analysis between jaw myxoma and mesenchymal cells of tooth germ. *Pathol Res Pract.* 1992;188:141–144.
24. Moshiri S, Oda D, Worthington P, Myall R. Odontogenic myxoma: histochemical and ultrastructural study. *J Oral Pathol Med.* 1992;21:401–403.
25. Halpenny W, Verey A, Bardsley V. Myxoma of the mandibular condyle: a case report and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2000;90:348–353.
26. Kim J, Ellis GL. Dental follicular tissue: misinterpretation as odontogenic tumors. *J Oral Maxillofac Surg.* 1993;51:762–767.
27. Schmidseider R, Groddeck A, Schunemann H. Diagnostic and therapeutic problems of myxomas (myxofibromas) of the jaws. *J Maxillofac Surg.* 1978;6:281–286.
28. Asamoia EA, Anyanlere AO, Olaitan AA. Pediatric tumors of the jaws in Northern Nigeria, clinical presentation and treatment. *J Cranio Maxillofac Surg.* 1990;18:130–135.
29. Happonen RP, Peltola J, Ylipaavalniemi P, et al. Myxoma of the jawbones: an analysis of 13 cases. *Proc Finn Dent Soc.* 1998;84:45–52.
30. Allphin AL, Moniglia AJ, Gregor RT, Sawyer R. Myxomas of the mandible and maxilla. *Ear Nose Throat.* 1993;72:280–284.
31. Batsakis JG. Myxomas of soft tissue and facial skeleton. *Ann Otol Rhinol Laryngol.* 1987;96:618–619.
32. Deron PB, Nikolovski N, denHollander JC, et al. Myxoma of the maxilla: a case with extremely aggressive biologic behavior. *Head Neck.* 1996;18:459–464.