Intraoral minor salivary gland neoplasm: a single institution experience of 80 cases


Abstract. From March 1991 to February 2001, 80 cases of minor salivary gland tumours were diagnosed in the Department of Oral and Maxillofacial Surgery at the University of Maryland at Baltimore (Baltimore, MD, USA). Data extracted from a retrospective chart review included age, sex, symptoms, site, histological diagnosis, treatment and outcome. Pleomorphic adenoma was the most common benign tumour and makes up 89.5% of all benign tumours. The percentage of malignancy (76.3%) was much higher than that found in other studies. Of the malignant tumours, 54.1% were mucoepidermoid carcinomas. This study differs from many previous reviews that were published by pathologists rather than a surgical unit.

Introduction
Salivary gland tumours are uncommon. The reported incidence is approximately 3% of all head and neck neoplasms. Tumours of minor salivary gland origin account for only 10–15% of all salivary gland neoplasms. The percentage of minor salivary gland tumours is higher in the African series, although one of these papers is from a maxillofacial unit, which may see minor salivary gland tumour more than major salivary gland tumour. Most studies include major and minor salivary gland tumours together and there are few articles that study minor salivary gland tumours separately. The papers that have reported minor and major salivary gland tumours separately were done so from outside of the USA.

The purpose of this study is to report our experience with minor salivary gland tumours at the University of Maryland at Baltimore (UMAB), Department of Oral and Maxillofacial Surgery.

Materials and methods
The records of all patients with minor salivary gland tumours treated in the Department of Oral and Maxillofacial Surgery, UMAB during March 1991 to February 2001 were reviewed. A total of 80 patients with intraoral minor salivary gland tumours were identified.

Criteria for inclusion were biopsy-proven minor salivary gland tumours that occurred in the oral cavity. Most patients with intraoral minor salivary gland tumours received initially incisional biopsy from the private oral and maxillofacial surgeons. All patients had incisional biopsy performed before receiving the definitive treatment.

Results
A total of 80 minor salivary gland tumours were identified in 49 female patients and 31 male patients. The female to male ratio was 1.6:1, with a female to male ratio of 1.9:1 with the malignant tumours and 1:1.1 with the benign tumours.

All patients were investigated in a retrospective manner by an extensive chart review. Age, sex, symptoms, duration of symptoms, location, race, histology, and modalities of treatment were examined and compared to those in the existing literature.

Key words: minor salivary gland tumour; intraoral.

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respectively. For the malignant neoplasms, the median age for males and females was 52 and 54, respectively. The median age for males and females with minor salivary gland tumour was approximately the same: 54 and 55, respectively. The median age for those with malignant minor salivary gland tumours was 6 years younger than for those with benign tumours (54 years for malignant and 60 years for benign). By histological type, the median age for those with mucoepidermoid carcinoma was younger than for those with other histological types (48.5 years for mucoepidermoid carcinoma, 58 years for pleomorphic adenoma, 63 years for adenoid cystic carcinoma and 56 years for polymorphous low-grade adenocarcinoma). Six of the malignant tumours (6 out of 61) (10%) occurred in children aged 9–16 years and four of these were mucoepidermoid carcinomas; all of the benign tumours occurred in adults.

Race was reported in 77 out of 80 patients—71.3% were Caucasian and 22.5% were black. The distribution of minor salivary gland tumours by race indicated that the majority of patients who had pleomorphic adenoma, mucoepidermoid carcinoma, and polymorphous low-grade adenocarcinoma were Caucasian, while there was an equal Caucasian to black predilection with adenoid cystic carcinoma (Table 1).

The most common complaint that brought patients to see either the dentist or oral and maxillofacial surgeon was a painless swelling in the mouth (61.25%). Of these patients, 20% were without symptoms and the tumour was found on routine dental examination. The majority of patients with benign minor salivary gland tumours (68.4%) had symptoms for more than 1 year before seeking treatment (Fig. 1). Of patients with malignant minor salivary gland tumours, 50.8% had symptoms less than 1 year before receiving treatment. Of the patients with malignant tumours, 27.9% had experienced symptoms for longer than 1 year and 13.1% experienced no symptoms (found on routine dental examination), respectively (Fig. 2).

The majority of the patients with minor salivary gland tumours who received treatment at UMAB presented with malignant minor salivary gland tumours. Of these (76.3%) (61 out of 80) were classified as malignant and (23.7%) (19 out of 80) were classified as benign. The commonest sites for minor salivary gland tumours were the palate, buccal mucosa and upper lip, which accounted for 77.5% of cases. The palate was the most common site for all minor salivary gland tumours (53.8%) and (60.5%) (26 out of 43) of palatal tumours were malignant. The buccal mucosa was the second most common site for malignant salivary gland tumours (Table 2). The most common sites for malignant minor salivary gland tumours were the palate, buccal mucosa, maxilla, and retromolar fossa at 42.6%, 21.3%, 8.2%, and 8.2%, respectively.

There were 19 patients who had benign minor salivary gland tumours of which two (10.5%) were recurrent tumours that were initially treated elsewhere. Pleomorphic adenoma was the most common minor salivary gland tumour (17 out of 19 cases, 89.5% of benign minor salivary gland tumour) and accounting 21.3% of all minor salivary gland tumours. The palate was the most commonly involved site of pleomorphic adenoma (88.2%, 15 out of 17 cases of pleomorphic adenoma). The other site of presentation for pleomorphic adenoma was the upper lip. There was one patient who had basal cell adenoma of the palate and one with canalicular adenoma of the upper lip.

Mucoepidermoid carcinoma was the most common malignant lesion noted in this study. There were 33 tumours diagnosed as mucoepidermoid carcinoma,
accounting for 41.3% of all tumours and 54.1% of all malignant tumours. There were 13 males and 20 females in this mucoepidermoid carcinoma group. The palate and buccal mucosa were the most common sites of mucoepidermoid carcinoma (15 and six cases, respectively). Furthermore, there were four cases located intraorally (three cases in the mandibular retromolar region and one case in the maxillary premolar region). The mucoepidermoid carcinomas were further classified into low grade (16 cases), intermediate grade (eight cases), high grade (seven cases) and an unknown grade (two cases).

Polymorphous low-grade adenocarcinoma was the second most common malignancy. The incidence of polymorphous low-grade adenocarcinoma was 14.8% (9 out of 61 cases) of the malignant minor salivary gland tumours and 11.3% (9 out of 80 cases) of all tumours. The buccal mucosa and palate were the most common sites for polymorphous low-grade adenocarcinoma and the remaining locations were the upper lip and maxilla.

Adenoid cystic carcinoma was the third most common site of malignant minor salivary gland tumours. There were seven patients who had adenoid cystic carcinoma, accounting for 11.5% of malignancies and 8.8% of all minor salivary gland tumours. The palate was the most common site. The other sites where adenoid cystic carcinoma presented were the maxilla and upper lip.

All benign minor salivary gland tumours were treated surgically by wide local excision, except for two patients who did not receive any treatment due to their advanced age and poor general health. The commonest treatment modality for malignant minor salivary gland tumours (70.8%) was surgery alone. Surgery consisted of wide local excision and composite resection with or without neck dissection. The other treatment modalities for malignant salivary gland tumours were combined treatment (Table 3). There were five patients who either received treatment at other institutions or refused treatment.

There were no recurrences in benign minor salivary gland tumours with a follow-up from 3 months to 5 years. There were 61 patients with malignant tumours, of which 48 had low- or intermediate-grade cancers. These included low- and intermediate-grade mucoepidermoid carcinoma, polymorphous low-grade adenocarcinoma, clear cell carcinoma, low-grade adenocarcinoma, basal cell adenocarcinoma, mucous-producing cyst adenocarcinoma and acinic cell carcinoma. Three patients were lost to follow-up; of the remaining 45 patients, 42 remain cancer-free (93.3%) with a variable follow-up ranging from 3 months to 9 years. One woman with persistent intermediate-grade mucoepidermoid carcinoma, which recurred following four surgeries, radiation therapy and injections with p53 adenovirus, died of other causes.

Two patients remain alive with persistent disease. One male patient had a massive, neglected low-grade adenocarcinoma with persistent tumour at the skull base. One woman had polymorphous low-grade adenocarcinoma that recurred locally and in the neck. Following neck dissection and maxillectomy, the tumour again recurred in the maxilla and retropharyngeal nodes; following further excision, it recurred in the orbit and she has refused further treatment.

There were 13 patients with high-grade tumours, either high-grade mucoepidermoid carcinoma or adenoid cystic carcinoma. Of the seven patients with high-grade mucoepidermoid carcinoma, four (57%) had lymph node metastases at presentation. Two patients (15.3%) died because of disease: one male with adenoid cystic carcinoma with lung, brain, and bone metastases, and one female with high-grade mucoepidermoid carcinoma of locoregional recurrence after surgery plus radiation and further salvage surgery. One patient with adenoid cystic carcinoma died 1-week postoperatively of a presumed pulmonary embolism. One patient with high-grade mucoepidermoid carcinoma is alive with pulmonary metastases unresponsive to chemotherapy. Thus nine out of 13 (69%) of individuals with high-grade malignancies are alive without cancer but the follow-up period is short—7 years 6 months is the longest—this is very inadequate for adenoid cystic carcinoma.

**Discussion**

Most studies including this one have shown that minor salivary gland tumours are more common in females than males, with a ratio range from 1.2:1 to 1.9:1.21

The age range of the patients was 9–90 years. There was no statistically significant difference in the ages of females and males in the benign and malignant minor salivary gland tumour groups. The present study showed that the median age of the patients with malignant minor salivary gland tumours is 6 years younger than those with benign tumours. This may have been due to the large proportion of mucoepidermoid carcinomas. On the other hand, the study by Waldron et al. revealed that the patients with benign tumours were

<table>
<thead>
<tr>
<th>Treatment</th>
<th>No. of cases</th>
<th>% of malignant minor salivary gland tumours</th>
</tr>
</thead>
<tbody>
<tr>
<td>Wide local excision</td>
<td>32</td>
<td>53.0</td>
</tr>
<tr>
<td>Composite resection</td>
<td>8</td>
<td>13.0</td>
</tr>
<tr>
<td>Composite resection + neck dissection</td>
<td>3</td>
<td>4.8</td>
</tr>
<tr>
<td>Surgery + radiation + chemotherapy</td>
<td>11</td>
<td>18.0</td>
</tr>
<tr>
<td>Chemotherapy and/or radiation</td>
<td>2</td>
<td>3.2</td>
</tr>
<tr>
<td>Other</td>
<td>5</td>
<td>8.0</td>
</tr>
<tr>
<td>Total</td>
<td>61</td>
<td>100</td>
</tr>
</tbody>
</table>

*RMF: retromolar fossa.*

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**Table 2. Anatomical distribution of benign and malignant minor salivary gland tumours (n=80)**

<table>
<thead>
<tr>
<th>Site</th>
<th>Malignant</th>
<th>Benign</th>
</tr>
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<tbody>
<tr>
<td></td>
<td>No. of cases</td>
<td>% by location</td>
</tr>
<tr>
<td>Palate</td>
<td>26</td>
<td>60.5</td>
</tr>
<tr>
<td>Buccal</td>
<td>13</td>
<td>100</td>
</tr>
<tr>
<td>Upper lip</td>
<td>4</td>
<td>66.7</td>
</tr>
<tr>
<td>RMF*</td>
<td>5</td>
<td>100</td>
</tr>
<tr>
<td>Maxilla</td>
<td>5</td>
<td>100</td>
</tr>
<tr>
<td>Mandible</td>
<td>4</td>
<td>100</td>
</tr>
<tr>
<td>Lower lip</td>
<td>4</td>
<td>100</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td><strong>61</strong></td>
<td></td>
</tr>
</tbody>
</table>

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**Table 3. Treatment of malignant minor salivary gland tumours**

Intraoral minor salivary gland tumour 259
vesting egland tumours may result from a ‘har-
percentage of malignant minor salivary
malignant minor salivary gland
cystic carcinoma is the most common
previous surveys have reported that adenoid
this study, although a number of pre-

salivary gland tumours from most

cell carcinoma is similar.

This may be due to our racial population
reported in the study by I
high percentage of benign tumours were
5 years younger than those with malig-

salivary gland tumours from most

carcinoma. Furthermore, the study by

The percentage of malignant minor

malignant minor salivary gland tumours from most
studies was quite similar, representing
34.8% to 65%11,16,17,20,21. However, a
high percentage of benign tumours were
reported in the study by ISACSSON & SHEAR12, which they attribute to an
increased proportion of black patients in
their study group. They stated that black
people are affected by pleomorphic
adenoma about 3.5 times more often than Caucasian people.

In this study, the percentage of malig-
nant minor salivary gland tumours was
76.3%, which corresponds well to the
studies of SPIRO & POGER14,18. The high
percentage of malignant minor salivary
gland tumours may result from a ‘har-
esting effect’, with the majority of our
referrals from oral and maxillofacial sur-
geons who treat the benign minor sali-
vary gland tumours and refer the
malignant minor salivary gland tumours to
our institution.

The frequency of pleomorphic
adenoma in this study was 21.3%, which
was lower than that found in other
studies. The incidence of pleomorphic
adenoma has been reported to range
from 40.8–50% up to 70% (Table 4)4,12.

The incidence of mucoepidermoid
carcinoma in this study (41.3%) was
higher than in most published
studies, accounting for 8.6–33.9%.5,11,12,17,18,20,21 Mucoepidermoid
carcinoma was the most common malig-
nant salivary gland tumour reported in
this study, although a number of pre-
vious surveys have reported that adenoid
cystic carcinoma is the most common
malignant minor salivary gland
tumour1,11,17–19. This difference in the
histological distribution may be related to
the histological criteria that was used
for the diagnosis of adenoid cystic carci-
noma. Series prior to 1984 did not
recognize polymorphous low-grade
adenocarcinoma as an entity and it was
frequently diagnosed as adenoid cystic
carcinoma. Furthermore, the study by

Eveson & Cawson noted that the inci-
dence of mucoepidermoid carcinoma in
British and western European studies
was considerably lower than that in
reports from the USA, which might
represent a true geographic variation11.

The frequency of polymorphous low-
grade adenocarcinoma in the present
study was 11.3% of all minor salivary
gland neoplasms which was the same
range reported in other studies (1.4–
15.7%)3,12,20,21. However, in some
reports polymorphous low-grade adenoc-
carcinoma was not found or reported as
a small number. This was probably
because there was overlap of histological
features between polymorphous low-
grade adenocarcinoma and adenoid
cystic carcinoma and some studies were
reported before the polymorphous low-
grade adenocarcinoma was first intro-
duced as a distinctive neoplasm in 19849,11.

The anatomical distribution of minor
salivary gland tumours found in the
present study is slightly different from
that in other studies. In this study, the
palate, buccal mucosa, and maxilla were
the most predominant sites, while in
other major surveys the palate, upper
lip, and buccal mucosa were the pre-
dominant sites. The palate was reported
to be the predominant site of occurrence
and is the location of occurrence in
between 40–55%4,15.21 of all cases; this
has been reported to be as high as 75%.
This study reports that 53.8% of all
minor salivary gland tumours occurred
in the palate and that 60.5% (26 out of
43) were malignant; other studies
reported that approximately 50% of all
minor salivary gland tumours occurred
in the palate and 50% were malig-
nant2,7,11,21. It is not possible to differ-
entiate malignant from benign tumours
clinically. A total of 27.95 (one in four)
malignant tumours were present for more
than 1 year and 13.1% (one in seven)
were asymptomatic. Therefore all
suspected minor salivary gland tumours
require biopsy to avoid further delay in
diagnosis.

Although two of our 19 patients with
benign tumours had recurrent lesions,
we encountered no recurrence following
adequate local surgery. In our 61 malig-
nant tumours, only five were found to
have clinically proven lymph node met-
astases (8.2%): four high-grade muco-
epidermoid carcinomas and one poly-
morphous low-grade adenocarcinoma.

Two patients died of cancer, one of
distant metastases and one of locore-
gional recurrence. One patient died peri-
operatively; four patients have persistent
disease, three with local recurrence (one
died of other causes) and one with lung
metastases. A total of 54 patients (54 out
of 61; 88.5%) remain alive without obvi-
ous cancer, although the follow-up
period is short. Histological type, grade,
stage and site are all important determi-
ants of prognosis and aggressive sur-
gery with wide margins is the best

The information from this report was
unique and different from other studies
because it came from a maxillofacial
oncology unit rather than a pathology
unit.

References

Table 4. Distribution and percentage (shown in parentheses) of minor salivary gland tumours in the present study and previously reported series

<table>
<thead>
<tr>
<th>Variable</th>
<th>Present study</th>
<th>Rivera–Bastides</th>
<th>Van heerden &amp; Raubenheimer</th>
<th>Isacsxon &amp; Shear</th>
<th>Eveson &amp; Cawson</th>
<th>Waldron et al.</th>
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<tr>
<td>Total no. cases</td>
<td>80</td>
<td>62</td>
<td>70</td>
<td>201</td>
<td>335</td>
<td>426</td>
</tr>
<tr>
<td>Total no. benign</td>
<td>19</td>
<td>34</td>
<td>34</td>
<td>145</td>
<td>180</td>
<td>245</td>
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<td>PA</td>
<td>17 (21.3)</td>
<td>24 (38.7)</td>
<td>34 (48.6)</td>
<td>140 (70)</td>
<td>143 (42.7)</td>
<td>174 (40.8)</td>
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<td>CA</td>
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<td>3 (4.8)</td>
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<td></td>
<td>20 (4.7)</td>
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<td></td>
<td>5 (1.2)</td>
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<tr>
<td>Total no. malignant</td>
<td>61</td>
<td>28</td>
<td>36</td>
<td>56</td>
<td>155</td>
<td>181</td>
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<tr>
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<td>33 (41.3)</td>
<td>18 (29.0)</td>
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<td>13 (6.5)</td>
<td>30 (8.9)</td>
<td>65 (15.3)</td>
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<td>6 (9.7)</td>
<td>9 (12.8)</td>
<td>21 (10.4)</td>
<td>44 (13.1)</td>
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<td>3 (4.3)</td>
<td>15 (7.5)</td>
<td>40 (11.9)</td>
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<td>AceC</td>
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<td></td>
<td>6 (1.8)</td>
<td>15 (3.5)</td>
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<tr>
<td>UC</td>
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<td>PLGA</td>
<td>9 (11.3)</td>
<td>11 (15.7)</td>
<td>11 (10.5)</td>
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<td>1 (1.4)</td>
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