Minor Salivary Gland Tumors: A Retrospective Study of 37 Cases

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Abstract

Salivary gland tumors represent 2% - 6.5% of all head and neck tumors. Since salivary gland tumors have various clinical features and histological types, it is often difficult to diagnose and treat them. The aim of this study was to conduct a retrospective clinical-statistical analysis of 37 minor salivary gland tumors (MSGTs) treated in the Department of Oral Surgery at Nihon University School of Dentistry at Matsudo over a 16-year period. The frequencies and distributions of sex, age, occurrence site, preoperative examination (fine needle cytology and/or biopsy), treatment, and prognosis of the tumors were analyzed and compared with previous reports. The average age at diagnosis was 58.1 years (range 22 - 91 years). The peak occurrence of tumors was in the sixties (10 cases, 27.0%), followed by the forties (8 cases, 21.6%) and the seventies (7 cases, 18.9%). The average age of patients with benign tumors was 56.8 years (range 22 - 91 years). For malignant tumors, the average age was 61.7 years (range 42 - 81 years). The male-female ratio was 1:2.1 for all minor salivary gland tumors, 1:2.4 for benign tumors, and 1.5:1 for malignant tumors. There were 27 (73.0%) benign and 10 (27.0%) malignant tumors. Pleomorphic adenoma (PA) was the most common tumor (24 (64.9%) cases). Most salivary gland tumors originated from the palate (21 cases, 56.8%); the second most common site was the buccal mucosa (14 cases, 37.8%). For most patients, an intraoral mass was the primary presentation, and the second most common symptom was swelling. Surgical treatment was performed for all cases, both benign and malignant tumors. There was one case of local recurrence of PA, as well as one of multiple lung metastases after surgery for adenoid cystic carcinoma. Diagnosis and treatment of MSGTs are often delayed because the pa-

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tients have few symptoms, and it is difficult to distinguish benign from malignant tumors. More cases need to be examined in the future, and the diagnostic accuracy of imaging and histopathological diagnosis needs to be improved.

Keywords
Minor Salivary Gland Tumor, Clinical Statistics, Intraoral, Japanese

1. Introduction

Salivary gland tumors represent 2% - 6.5% of all head and neck tumors [1] [2]. Minor salivary gland tumors (MSGTs) are uncommon, representing 25% of all salivary gland tumors [3]. Since salivary gland tumors have various clinical features and histological types, it is often difficult to diagnose and treat them.

Most clinical-statistical studies of salivary gland tumors involved major and minor salivary gland tumors, making it difficult to evaluate their real frequency and site distribution. Although the incidence of MSGTs is lower than that of major salivary gland tumors, the constituent ratio of malignant tumors in MSGTs is relatively high. [4] And there are few reports about MSGTs in Japan [5] [6] [7] [8]. The aim of this study was to report a clinical-statistical analysis of 37 MSGTs treated in the Department of Oral Surgery at Nihon University School of Dentistry at Matsudo over a 16-year period.

2. Materials and Methods

A retrospective study of all of cases diagnosed as benign and malignant MSGTs and treated was performed. The data were collected from the medical records of patients treated at the Department of Oral Surgery of Nihon University School of Dentistry at Matsudo from April 2006 to March 2022. A total of 37 cases that were classified based on the histological classification of salivary gland tumors, published by the World Health Organization (WHO) in 2017 [9], were considered.

The frequencies and distributions of sex, age, occurrence site, clinical symptoms, treatment, and prognosis of the tumors were analyzed and compared with previous reports.

The present study was approved by the Ethics Committee of Nihon University School of Dentistry at Matsudo, Japan (EC 21-19-028-2).

3. Results

1) Age and sex distributions (Table 1)

The average age at diagnosis was 58.1 years, with a range of 22 - 91 years. The peak occurrence of tumors was in the sixties (10 cases, 27.0%), followed by the forties (8 cases, 21.6%) and the seventies (7 cases, 18.9%). The average age of patients with benign tumors was 56.8 years (range 22 - 91 years). For malignant tumors, patient age ranged from 42 to 81 years, with an average age of 61.7
years. The male-female ratio for all minor salivary gland tumors was 1:2.1, with that for benign tumors being 1:2.4 and that for malignant tumors being 1:1.5.

2) Histological types (Table 2)
The sample was made up of 37 cases. There were 27 (73.0%) benign and 10 (27.0%) malignant tumors. The ratio of benign tumors to malignancies was 2.7:1. Pleomorphic adenoma (PA) was the most common tumor and accounted for 24 (64.9%) cases, followed by mucoepidermoid carcinoma (4 cases, 10.8%) and adenoid cystic carcinoma (3 cases, 8.1%), ductal papillomas (2 cases, 5.4%), with one each of cystadenoma, adenocarcinoma NOS, carcinoma ex pleomorphic

### Table 1. Age and sex distribution of minor salivary gland tumors.

| Age(years) | Benign tumors | Malignant tumors | Total |
|------------|---------------|------------------|-------|
|            | Male | Female | Male | Female | |
| 20 - 29    | 1    | 3      | 0    | 0      | 4    |
| 30 - 39    | 0    | 1      | 0    | 0      | 1    |
| 40 - 49    | 3    | 3      | 1    | 1      | 8    |
| 50 - 59    | 0    | 2      | 1    | 0      | 3    |
| 60 - 69    | 2    | 3      | 1    | 4      | 10   |
| 70 - 79    | 1    | 5      | 0    | 1      | 7    |
| 80 - 89    | 0    | 2      | 0    | 1      | 3    |
| 90 - 99    | 0    | 1      | 0    | 1      | 1    |
| Total      | 7    | 20     | 3    | 7      | 37   |

### Table 2. Distribution of the minor salivary glands tumors, according to histological type and sex.

| Pathological type       | Sex | Total |
|-------------------------|-----|-------|
| Benign tumor            |     |       |
| Pleomorphic adenoma     | 18  | 6     | 24  |
| Ductal papillomas       | 1   | 1     | 2   |
| Cystadenoma             | 0   | 1     | 1   |
| Malignant tumor         |     |       |
| Mucoepidermoid carcinoma| 3   | 1     | 4   |
| Adenoid cystic carcinoma| 3   | 0     | 3   |
| Adenocarcinoma, NOS     | 0   | 1     | 1   |
| Carcinoma ex pleomorphic adenoma | 0 | 1 | 1 |
| Clear cell carcinoma    | 0   | 1     | 1   |
| Total                   | 25  | 12    | 37  |
adenoma, and clear cell carcinoma. The details of ductal papillomas were intra-ductal papilloma and 1 case of inverted ductal papilloma.

3) Locations of the tumors (Table 3)

Most of the salivary gland tumors originated from the palate (21 cases, 56.8%), and the second most common site was the buccal mucosa (14 cases, 37.8%). The upper lip and the floor of the mouth had the same incidence (1 case each, 2.7%). For benign tumors, the most arising location was the palate (15 cases, 55.6%), and the second most common area was buccal mucosa (11 cases, 40.7%). For malignant tumors, the most arising location was the palate (6 cases, 60.0%), and the second most common area was buccal mucosa (3 cases, 30.0%).

4) Clinical symptoms (Table 4)

### Table 3. Pathological classifications and locations of minor salivary gland tumors.

| Pathological type                | Plate | Buccal mucosa | Upper lip | Floor of mouth |
|----------------------------------|-------|---------------|-----------|----------------|
| **Benign tumor**                 |       |               |           |                |
| Pleomorphic adenoma              | 15    | 8             | 1         | 0              |
| Ductal papillomas                | 0     | 2             | 0         | 0              |
| Cystadenoma                      | 0     | 1             | 0         | 0              |
| **Malignant tumor**              |       |               |           |                |
| Mucoepidermoid carcinoma         | 3     | 1             | 0         | 0              |
| Adenoid cystic carcinoma         | 1     | 2             | 0         | 0              |
| Adenocarcinoma, NOS              | 1     | 0             | 0         | 0              |
| Carcinoma ex pleomorphic adenoma | 1     | 0             | 0         | 0              |
| Clear cell carcinoma             | 0     | 0             | 0         | 1              |
| **Total**                        | 21    | 14            | 1         | 1              |

### Table 4. Signs and symptoms of minor salivary gland tumors.

| Pathological type                | Mass | Swelling | Pain | Ulcer | Discomfort |
|----------------------------------|------|----------|------|-------|------------|
| Pleomorphic adenoma              | 17   | 6        | 1    | 0     | 0          |
| Ductal papillomas                | 2    | 0        | 0    | 0     | 0          |
| Cystadenoma                      | 1    | 0        | 0    | 0     | 0          |
| Mucoepidermoid carcinoma         | 2    | 1        | 0    | 1     | 0          |
| Adenoid cystic carcinoma         | 1    | 0        | 1    | 0     | 1          |
| Adenocarcinoma, NOS              | 1    | 0        | 0    | 0     | 0          |
| Carcinoma ex pleomorphic adenoma | 1    | 0        | 0    | 0     | 0          |
| Clear cell carcinoma             | 1    | 0        | 0    | 0     | 0          |
| **Total**                        | 26   | 7        | 2    | 1     | 1          |
For most patients, an intraoral mass was the primary presentation as the chief complaint of the patient, and the second most common symptom was swelling. Other symptoms were pain in 2 cases (PA and adenoid cystic carcinoma), ulcer in 1 case (mucoepidermoid carcinoma), and discomfort in 1 case (adenoid cystic carcinoma). Mass was found in all tumors. Symptoms of benign tumors were mostly swelling or mass, but pain was observed in only one case (3.7%). For malignant tumors, pain, ulcer, and discomfort were observed in one case each (10%).

5) Treatment and Outcomes

Surgical treatment was performed for all cases, both benign and malignant tumors. There was a case of local recurrence of PA, as well as a case of adenoid cystic carcinoma with multiple lung metastases after surgery. A case of local recurrence of PA underwent reoperation, and now the prognosis was good. Since the progression of multiple pulmonary metastases of ACC was slow, no additional treatment was given and the patient is still under follow-up.

4. Discussion

Salivary gland tumors are a heterogeneous and rare group of lesions, especially when affecting the minor salivary glands [10]. They have a high malignancy rate [11] [12] [13] [14]. Therefore, collecting clinical data is important. The present study included a total of 37 patients treated between 2006 and 2022 with primary surgery at our department.

1) Age

The average age at diagnosis was 58.1 years, and the peak occurrence of tumors was in the sixth decade in the present study. MGST affect mainly adult patients from the 3rd through the 7th decades, with a peak prevalence from the 5th to 6th decades of life [3] [5] [6] [7] [10] [15]-[20]. Benign tumors occurred in all age groups; the average age was 56.8 years. Malignant tumors occurred after the 4th decade; the average age was 61.7 years. It has been reported that the patient age group of malignant tumors is older than of benign tumors [5] [7] [8] [12] [14] [19]. The present data are in agreement with these findings.

2) Sex

Most studies have shown that MSGTs are more common in females than males [5] [6] [7] [8] [10] [14] [17] [19] [21] [22] [23]. In the present study, the male-female ratio for MSGTs was 1:2.1, in accord with other studies [7] [17]. It has been suggested that the sex distribution of SGTs is related to ethnic variations [7] [14] [18]. The present study found a higher proportion of benign tumors among females than did the previous reports, with a ratio of 1:2.4 [7]. The male-female ratio for malignant tumors was 1:1.5. Some studies have reported that malignant tumors are more common in males than females [10]. Other reports stated that there was no sex predilection for benign and malignant minor salivary gland tumors [8].

3) Histological types

In the present study, 73.0% of MSGTs were benign, and 27.0% were malig-
nant. Other studies have reported that 40% - 72% were benign tumors. Benign salivary tumors are more common than malignant ones at departments of oral and maxillofacial surgery. However, data from cancer centers show more malignant tumors than benign ones [10]. PA is the most common, accounting for 33.2% - 65.9% of all minor salivary gland tumors. In the present study, PA accounted for 64.9% of all tumors, similar to previous reports [11].

Of malignant tumors, mucoepidermoid carcinoma was the most common. Some authors reported that mucoepidermoid carcinoma and adenoid cystic carcinoma malignant tumors were frequent [10] [11] [14] [19] [20] [21]. Both benign tumors and malignant tumors were similar to previous reports.

4) Locations of the tumors

The palate was the most common site for MSGTs in the present study, with a high frequency of involvement (56.8%), followed by the buccal mucosa (37.8%). Other studies have also reported that the palate was the most common site for MSGTs, and that approximately 40% - 80% of all tumors occurred in the palate [7]. This was consistent with the present data. There was no difference in the occurrence site between malignant and benign tumors in the present study. Both were the most frequent in the palate, followed by the buccal mucosa. This finding is in agreement with other studies [7] [10] [11] [14] [22]. However in previous reports [8] [10] [11] [14] [15], malignant tumors also occurred in the floor of mouth and lips. In this study, except for the palate and buccal mucosa, only one case was found in the floor of mouth.

5) Signs and symptoms

An intraoral mass was the most frequent of the signs and symptoms in the present study. The second most common symptom was swelling, but this may be the result of differences in patients’ descriptions, since “mass” and “swelling” may be the same symptom. Pain, ulcer, and discomfort were relatively uncommon, and their frequency did not differ significantly between benign and malignant lesions. Chaudhry et al. [16] reported that pain is a common presenting symptom of patients with malignant MSGTs; this was not found in the present study.

6) Treatment and Prognosis

The standard treatment of minor benign salivary tumors is surgical excision. In the present study, surgical treatment was performed for all benign tumors. There was a case of local recurrence of PA. On histopathological diagnosis of the resected specimen, the resection margins were negative, but recurrence was observed during follow-up 3 months after the operation, and additional resection was performed. It is recommended that the surrounding bone be removed when a PA occurs in the palate because of the possibility of recurrence or malignant transformation [23].

The treatment of malignant salivary tumors is surgical excision. Almost all malignant salivary gland tumors have low radio-sensitivity and high resistance to anticancer agents. They undergo additional treatment such as postoperative
radiotherapy or chemotherapy when they have marked local infiltration, or where the excisional margin is incomplete. Therefore, we believe that complete resection at the first surgery is important. In the present study, there was a case of adenoid cystic carcinoma with multiple lung metastases after surgery. The patient is being observed without additional treatment, because the tumor progressed indolently. Since there is a high probability of remote metastasis, such as to the lungs, follow-up should include examination of the entire body, and long-term follow-up is necessary. Diagnosis and treatment of MSGT's are often delayed because the patients have few symptoms, and it is difficult to distinguish benign from malignant tumors. Therefore, it is necessary to continue the study by examining more cases in the future and improve the diagnostic accuracy of imaging diagnosis and histopathological diagnosis.

5. Conclusion

This was a clinical-statistical analysis of 37 MSGTs treated in the Department of Oral Surgery of Nihon University School of Dentistry at Matsudo over a 16-year period. The frequency was the higher incidence of benign tumors, especially of PA. There was a tendency for female predominance and a higher incidence of palatal involvement.

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Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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