Intraoral Minor Salivary Gland Tumours: A Retrospective Study from a Dental and Maxillofacial Surgery Centre in Salem, Tamil Nadu

Maya Ramesh · Ramesh Krishnan ·
George Paul

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Abstract Minor salivary gland tumors (MSGTs) constitute a heterogeneous group of neoplasms with variation in histopathology. These are rare neoplasms usually occurring in the palate. Dental examination may provide an opportunity for early detection.

Aim This study was undertaken to do an epidemiological survey of minor salivary gland tumours reporting to a single dental and maxillofacial surgery centre and to determine the correlation of the histopathologic characteristics with the clinical features.

Materials and Methods A retrospective survey of the histopathological findings of 1,020 consecutive biopsy reports in a single dental and maxillofacial surgery centre was done to identify cases of MSGT. The results were tabulated based on various criteria.

Results In our study, only 8 tumors were benign (26.67 %), and 22 tumors were malignant (73.33 %). Mucoepidermoid carcinoma (MEC) was the most common tumor (15 of 30). Pleomorphic adenoma was most common benign MSGT in our series (7 of 30). This was followed by adenoid cystic carcinoma (6 of 30). Palate was the most common site (13 of 30) followed by buccal mucosa (5 of 30) and lip (4 of 30).

Conclusions Unlike many previous studies, malignant salivary gland tumours were predominant. MEC was the most common malignant tumour in our study similar to many other studies. The palate was the most common site for minor salivary gland neoplasms.

Keywords Minor salivary gland tumours · Benign tumour · Malignant tumour

Introduction

There are very few published reports on the incidence of minor salivary gland tumours (MSGTs) in the Indian population. Ironically all of them are from Tamil Nadu and Pondicherry [1–3]. While this study is from the same state, it is from a different region and from a private establishment rather than a Government Institute. While MSGTs constitute only 25 % of salivary gland tumours which in turn accounts for only 2–6.5 % of head and neck neoplasms [4], the clinician is quite likely to encounter them in the oral cavity from time to time.

Materials and Methods

The present study is based on the histopathological and clinical reports of a single dental and maxillofacial centre in Salem, Northwest Tamil Nadu. The histopathological records of 1,020 consecutive biopsy reports of over a 6 year period starting from January 2006 to March 2012 available as records in a dental and maxillofacial centre and the 30 cases thus obtained were retrospectively studied (0.029 %). The histopathological specimens reported as MSGT were reviewed with respect to age, gender, duration

M. Ramesh (✉)
Department of Oral Pathology, VMSDC, NH-47 Sankari Main
Road, Ariyanoor, Salem 636308, Tamil Nadu, India
e-mail: mayaramesh96@gmail.com

R. Krishnan
Department of Pedodontics & Preventive Dentistry, VMSDC,
Salem, Tamil Nadu, India

G. Paul
Dental Poly Clinic and Maxillofacial Centre, Salem,
Tamil Nadu, India

of the lesions at the time of presentation, clinical features and anatomic location of the tumors. They were classified on the basis of the 2005 WHO recommendation. Hematoxylin- and eosin-stained slides were examined and re-evaluated.

Results

Types of Tumours (Tables 1)

Out of the 30 cases in our study, only 8 tumors were benign (26.67 %), and 22 tumors were malignant (73.33 %). Pleomorphic adenoma was the most common benign intra-oral salivary gland tumour in our series (87.5 %, 7 of 8). In our study there was only one female patient with monomorphic adenoma (1/8, 12.5 %), the type being basal cell adenoma. Of the 22 malignant tumours, mucoepidermoid tumour was the most common MSGT (68.2 %, 15 out of 22). Out of the 15 cases, 4 (26.6 %) were males and 11 (73.4 %) were females. Among the mucoepidermoid carcinomas (MECs), 5 were of the central variety. Among the five central MECs, three cases occurred in the maxilla and two cases in the mandible and one maxillary case was a clear cell variant. Of these 4 (57.1 %) were males and 3 (42.9 %) were females. This was followed by adenoid cystic carcinoma (ACC, 27.3 %, 6 out of 22). Among the ACCs, 2 (33.3 %) were males and 4 (66.6 %) were females. In our study there was only one female patient of Polymorphous low Grade Adenocarcinoma (PLGA) (1/22, 4.55 %).

Anatomic Location of the Tumors (Table 2)

The anatomic location of the tumors was distributed between the hard palate, buccal mucosa, lips and floor of

mouth. Palate was the most common site, 43.3 % (13 of 30) followed by buccal mucosa, 13.3 % (5 of 30) and lip, 3.3 % (4 of 30). There were variations in the proportion of malignant and benign tumors at different sites, for example, 8 out of 13 (61.54 %) palatal tumors were malignant compared to 3 out of 5 in the buccal mucosa (60 %). In the lip, all the tumours were malignant (100 %).

Duration of Symptoms

The majority of patients (22 out of 30) presented to health professionals within the first year while the other 8 reported within 2 years of development of signs or symptoms. Two of the MECs were recurrence after a previous surgery.

Clinical Presentation

Most of the patients presented with an intraoral swelling. Ulceration and pain were present in 10 of the 30 cases (33.3 %). Lymph nodes were enlarged only in two of the cases, namely one MEC and one ACC. In both the cases, the lymph nodes were found to be positive for malignant cells.

Age and Gender Distribution (Table 3)

In our study of 30 patients with MSGTs, 10 (35.4 %) were males and 20 (64.6 %) were females.

The median age of diagnosis was 44.2 years with a range of 15–86 years. The peak occurrence of tumors was in the fifth decade for males and third decade for females. The median age of patients with benign tumors was 44.9 years (range 25–70 years) with a slight male (5 of 8) predilection.

Table 1 Types of tumours

Sl. nos.	Type of tumour	Number of cases	Average age	Males	Percentage	Females	Percentage	Age range	Percentage	Tenderness
1	Pleomorphic adenoma	7/8 (87.5 %)	47.9	4	57.1	3	42.9			
2	Monomorphic adenoma	1/8 (12.5 %)	50.6	0	0	1	100			
3	ACC	6/22 (27.3 %)	38.3	2	33.3	4	66.6			
4	MEC	15/22 (68.2 %)	45.8	4	26.6	11	73.4			
5	PLGA	1/22 (4.55 %)	49	0	0	1	100			
6	Salivary gland tumours	30	46.32	10	35.4	20	64.6			
		22 Malignant								
		8 Benign								
7	Benign	8	44.2					25–70	26.67	0
8	Malignant	22	43.9					10–88	73.33	10
9	Total	30							100	

Table 2 Anatomic location of the tumours

Sl. nos.	Location	Number of cases	Number of malignancies	Percentage
1	Palate	13	8	26.7
2	Lip	4	4	13.3
3	Floor of the mouth	1	0	0
4	Retromolar area	2	0	0
5	Buccal mucosa	5	3	10
6	Jaws	3	3	10
7	Commissure of lip	2	0	0
8	Intraoral tumours	30	18	60

Table 3 Age and gender distribution

Ages	Number of males	Number of females	Total number of patients
10–20	0	1	1
20–30	2	4	6
30–40	0	5	5
40–50	2	1	3
50–60	4	3	7
60–70	2	3	5
70–80	1	1	2
80–90	0	1	1
Total	10	20	30

For all malignant tumors, patient age ranged from 10 to 88 years with a median age of 43.9 years. For MECs, the average age was 45.8 years with 11 females and 4 males out of 15 patients.

For ACC, the average age was 38.3 years with two males and four females.

Discussion

Intraoral MSGTs in our study represented 0.029 % (30 out of 1,020) of all biopsies done in the Maxillofacial Centre. Review of the literature revealed that the relative frequency of MSGT from institutional studies ranges from 0.03 to 1.9 % [3]. MSGTs in the study in Chennai represented 1.52 % of all oral biopsies. Minor salivary gland carcinomas are a heterogeneous group of malignancies for which the etiology is unknown [5]. Unlike in the major salivary glands, where only a minority (20–30 %) of tumors are malignant, most (50–60 %) tumors arising from the minor salivary glands are malignant [5]. Many occur within the oral cavity, although other sites in the upper aerodigestive tract, such as the oropharynx and nasal/paranasal cavity, are reported. The age range of patients with intraoral salivary gland neoplasm in this study was 10–88 years. It has been noted that malignant salivary gland tumors increase steadily in frequency, from late in the second decade of life and reach a peak in the

sixth decade [5]. In the present study, the frequency of malignant salivary gland tumors was not significantly different between age groups. But in our study, the most common tumours in decreasing order were MEC followed by pleomorphic adenoma and ACC. This was similar to the study of the Libyan population by Jaber [5] in 2006 and the Indian study by Vani and Ponnaiah [3] in 2010.

The present study confirms the previous reports that the palatal mucosal glands are more frequently involved than any other group of minor salivary glands in the oral cavity. And our figure of 43.3 % was comparable to the study done in Chennai [3].

In the present study, 7 out of 30 (23.3 %) were classified as pleomorphic adenoma and this frequency was less than many of the previously reported studies (Table 1). This low prevalence, as before, may be explained by socio geographic variations among patients. In the present sample, intraoral pleomorphic adenoma was found over a wide age distribution with a peak frequency in the fourth decade. Isacson and Shear [6] reported a peak frequency in the third decade and Lucas [7] reported a peak frequency in the fifth and sixth decades. In many published studies, pleomorphic adenoma was the predominant tumor [7–9]. The present study shows that non-pleomorphic benign tumors are rare, being 1 out of 8 (12.5 %) of all benign lesions. According to Waldron et al. [9], monomorphic adenomas constituted approximately 19 % of benign lesions

followed by cystadenoma (9 %) and sialadenoma papilliferum (2 %).

Review of Literature (Table 4)

Table 4 Review of Literature

Sl. nos.	Year	Authors	Country	Total number of cases	Percentage of malignancies
1	1961	Chaudhry et al. [10]	USA	94	54
2	1962	Smith [11]	USA	37	35
3	1966	Reynolds et al. [12]	USA	25	64
4	1966	Bardwill et al. [13]	USA	61	82
5	1968	Luna et al. [14]	USA	68	81
6	1970	Crocker et al. [15]	USA	38	32
7	1983	Isacsson and Shear [6]	South Africa	201	28
8	1985	Eveson and Cawson [8]	UK	336	46
9	1986	Chau and Radden [16]	Australia	98	38
10	1988	Waldron et al. [9]	USA	426	42.5
11	1991	Ellis et al. [4]	USA	3355	48.7
12	1995	Loyola et al. [17]	Brazil	164	38
13	1996	Rivera-Bastidas et al. [18]	Venezuela	62	62
14	1997	Kusama et al. [19]	Japan	129	38
15	1998	Jones et al. [20]	UK	145	71
16	2002	Jansisyanont et al. [21]	USA	80	76.3
17	2005	Jaber [5]	Libya	75	61.3
18	2007	Wang et al. [22]	China	737	53.9
19	2008	Copelli et al. [23]	Italy	43	60.6
20	2011	Vani and Ponnaiah [3]	India	185	75
21		Present study	India	30	73.3

According to Chaudhry et al. [10], the benign tumors were more than four times as prevalent as the malignant ones, and the ratio of malignant-to-benign tumor was 1:1 in the report by Eveson and Cawson [8]. In our study, the percentage of malignant tumours were 73.3. However other studies also showed 70 % and above; similar to our study [3, 4].

The frequency of intraosseous MSGT (3, 10 %) found in the present study is within the global range of 0.2–11 % [3]. All three MEC cases occurred in the maxilla.

Intraoral swelling with or without ulceration was the most frequent sign and symptom of MSGTs in our study. Ill-fitting dentures, change in sensation and difficulty in speech were relatively uncommon and were not seen in our study. Pain was the most important presenting symptom, the number of malignant lesions causing pain being (10),

33.3 %. The suggestion that pain is a common presenting symptom of patients with malignant MSGTs [10] was evident in our study also.

The palate with 42–75 % has been cited as the most common site for MSGTs. Other sites of involvement include the lips (4–21 %), buccal mucosa (5–16 %), tongue, floor of mouth (4–12 %), and retromolar area (3–7 %) [24]. In our study, the locations of the tumors were the hard palate, buccal mucosa and lips in decreasing order. In the present study, the age of patients ranged from 12 to 82 years (mean 45.77 years), with peak incidence in the fifth and sixth decades of life for both genders (Table 2). A higher mean age for malignant tumors was not noticed in our study.

Analysis of gender predilection showed a female predominance in the present study (Table 3) (ratio 1:1.73). Female predilection has been reported by most studies, and only a few studies have reported equal distribution. The trend was different with malignant tumors, where male predilection was noted [4].

Within India and especially in the state of Tamil Nadu and adjoining region, the study by Subashraj et al. (Pondicherry) [1] found a lower frequency of 39 % malignant MSGT, whereas it was 70 % according to Doirairajan et al. (Chennai) [2] and 75 % according to Vani and Ponnaiah (Chennai) [3].

Of the malignant tumors, MEC, ACC, and polymorphous low grade adenocarcinoma (PLGA) were the most frequently encountered, of all MSGT. The literature shows that the proportion of MEC ranges from 7–46 % of all MSGT and from 17–68 % of malignant MSGT [3]. In our study, MEC was the most common malignant histological subtype, followed by ACC and PLGA. MEC was the most common tumour (50 %) and the average age of occurrence was fourth decade of life in our series. The palate, followed by buccal mucosa and labial mucosa were the most frequently affected sites. The literature shows that the lower lip was the second most frequent site for MEC [3].

The second most common malignant MSGT was ACC, the average age being 38.3, which accounted for 20 % of all MSGT. The palate was the most common area of ACC in our series. In other literatures, the mean age of diagnosis was 46 years (range 21–63 years) with peaks in the fifth and sixth decades of life [3].

Reports suggest that PLGA, which is becoming increasingly recognized, is a common malignancy. Waldron et al. [9] found that 26 % of MSGTs were PLGAs and in an African population, it was found to be the most common intraoral salivary gland malignancy. However most studies published prior to 1984 do not report PLGA as a recognized entity. These differences may be due to ethnic or environmental factors. The reasons for this are not apparent, but may be due to the differences in

interpretation of diagnostic criteria or different study backgrounds [5]. PLGA was the third most common malignant tumor in our study, average age being 49 years, accounting for 3.3 % of all MSGT, in agreement with other series.

A review of 41 series reveals that MEC and ACC both occurred with almost equal frequency either as a first (51 and 49 %, respectively) or second (41 and 46 %, respectively) most frequent tumor, and the third most common tumor was either adenocarcinoma not otherwise specified (42 %) or PLGA (34 %) [3].

Various treatments have been proposed from surgery or radiotherapy alone, or a combination of both. Most authors recommend surgery as the prime treatment with the aim to achieve oncological excision margins [5]. Beckhardt et al. reported a significantly higher relapse rate and worse outcome for patients diagnosed with ACC [25]. The presence of perineural invasion, a common finding in ACC, is likely to contribute to a worse outcome in these patients [5].

Conclusion

The present study shows that the intraoral MSGTs vary widely in clinical features and these should be taken into account by medical and dental practitioners in the differential diagnosis when assessing intraoral pathology. More studies will help to understand their prevalence, site, gender, age and histological typing in relation to the prognosis. The patients should be referred to multi-speciality head and neck clinics following diagnosis. These studies will be helpful in the proper diagnosis and appropriate management of tumours of minor salivary glands.

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