ORIGINAL ARTICLE

CLINICOPATHOLOGICAL PATTERNS OF MINOR SALIVARY GLANDS TUMORS: A 16- YEAR RETROSPECTIVE REVIEW CARRIED OUT AT A SINGLE INSTITUTE IN NORTHERN JORDAN

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ABSTRACT

Minor salivary gland tumors (MSGTs) are relatively uncommon head and neck tumors with variable histological types. The primary purpose of this study was to explore the clinicopathological patterns of minor salivary gland neoplasms in the north Jordanian population and to compare them with findings from previous literature.

In this study, histopathological records of minor salivary gland tumors treated from January 2004 to December 2019 at the oral and maxillofacial surgery department, Prince Rashid Bin Al-Hassan Hospital, Irbid, Jordan, the main hospital in northern Jordan were reviewed.

The final sample showed 30 MSGTs ,17 of which were benign (56.66 %), and 13 were malignant (43.34%). There was male predominance (60%) with a male to female ratio of 1.5:1. Patient's ages ranged from 25 to 86 years, with a median age of 49.37 ± 18.70 years. Pleomorphic adenoma was the most frequent tumor, making up 50% of all MSGTs. Adenoid cystic carcinoma was the 2nd frequent tumor representing 26.68 % of all MSGTs. The Palate (47%) was the most commonly involved site, followed by the upper lip (30%)

The current review found that MSGTs are uncommon in the northern Jordan population, and there is a relatively higher percentage of benign tumors. Unlike most previous studies, males were more affected than females

Keywords: *Minor salivary gland tumors, northern Jordan, Pleomorphic adenoma, adenoid cystic carcinoma, palate*

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INTRODUCTION

Minor salivary gland tumors (MSGTs) are a rare heterogeneous group of lesions that occur in the submucosal layer of the oral cavity and oropharynx. They account for less than 20% of all salivary gland neoplasms.[1] MSGTs have a wide spectrum of overlapping histopathological features which in addition to the rarity of these tumors create a difficult and challenging situation for both surgeons and histopathologists in the diagnosis.

The development of MSGTs involves interaction between genetic and environmental factors. The exact etiology is unclear and controversial; however, recent research found that environmental factors like cigarette smoking increases the risk of salivary gland tumors.[2]

Previous studies of salivary neoplasms in Jordan encompass both the major and the minor salivary glands or major salivary glands alone.[3,4] The present study is the first to focus merely on the intraoral MSGTs in Northern Jordan.

This study aimed to evaluate the clinicopathological characteristics of minor salivary gland neoplasms in the north Jordanian population and to compare them with findings from previous studies. The retrieved data were reviewed and analyzed according to the frequency, histological type, and any possible correlation with the patient's age, sex, and location of these tumors.

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MATERIAL AND METHODS

A retrospective study was done by reviewing histopathological reports of minor salivary gland tumors treated from January 2004 to December 2019 at the department of oral and maxillofacial surgery, Prince Rashid Bin Al-Hassan Hospital, Irbid, the main hospital in northern Jordan. Records with incomplete data or lacking specific diagnosis were excluded. The following data were obtained from patients records: patients age, sex, tumors histopathological types, and location. Data were then processed and analyzed using Microsoft Excel 2010.Approval for this study was obtained from the Research and Ethical Committee of the Jordanian Royal Medical Services.

RESULTS

Thirty three patients had been diagnosed as minor salivary gland tumors (MSGTs). Three of which were excluded because of incomplete data or lacking specific diagnosis. Out of these,17 were benign (56.66 %), and 13 were malignant (43.34%). Further details may be seen in table 1. Table 2 shows the frequency and location of MSGTs according to histological type.

Fig 1 depicts proportions of intraoral locations of minor salivary gland tumors. It can be seen that the palate was the most affected intraoral site (47% % of the cases) followed by upper lip (30%), lower lip (7%), buccal mucosa (7%), retromolar (3%), maxilla (3%), and mandible (3%).

Regarding the palatal MSGTs,57.14% (8/14) were benign, whereas 42.86% (6/14) were malignant. The majority 88.89% (8/9) of MSGTs occurring in the upper

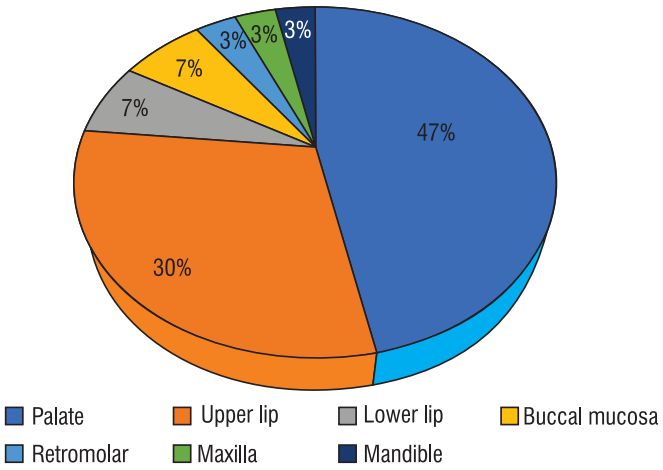

Fig 1: Frequency of intraoral locations of minor salivary gland tumors

TABLE 1: ANALYSIS OF AGE AND SEX IN BENIGN AND MALIGNANT MINOR SALIVARY GLAND TUMORS

	Number of Patients	Age range(years)	Mean age(years) ±std
Benign MSGTs	17 patients	25-73	40.53 ± 15.22
	10 Males	25-73	40.60 ± 16.40
	7 Females	26-70	40.43± 14.63
Malignant MSGTs	13 patients	34-86	60.92±16.77
	8 Males	34-86	52.75±15.28
	5 Females	61-84	74 ±9.35

TABLE 2: FREQUENCY AND LOCATION OF MINOR SALIVARY GLAND TUMORS ACCORDING TO HISTOLOGICAL TYPE

	Palate	Upper lip	Lower lip	Buccal mucosa	Retro-molar	Maxilla	Mandible	Total
Pleomorphic adenoma	7	7	1					15
Monomorphic adenoma		1						1
Myoepithelioma	1							1
Adenoid Cystic carcinoma	3	1	1	1		1	1	8
Mucoepidermoid carcinoma	2			1				3
polymorphous adenocarcinoma					1			1
Carcinoma ex pleomorphic adenoma	1							1
Total	14	9	2	2	1	1	1	30

lip were benign. There were two intraosseous MSGTs of adenoid cystic carcinoma, one involved the maxilla, and the other affected the mandible. The latter was the only tumor in this study that developed distant metastasis. Table (2) illustrates the distribution and frequency of the minor salivary gland tumors in the oral cavity according to histological type.

DISCUSSION

In the current study, 60 % of patients were males, and 40 % were females with a male: female ratio of 1.5:1. This ratio is inconsistent with most previous studies, which showed female predominance.[5-11] Moreover, A few studies showed no gender predilection .[12,13] This difference might represent genetic or geographical variations.

The patient's ages in this review ranged from 25 to 86, with an average of 49.37 ± 18.70 years; the mean age for benign tumors was lower than that of malignant tumors. This agrees with previous studies which reported that malignant MSGTs commonly occur in older patients than benign MSGTs.[5,14-16]

MSGTs can occur anywhere in the oral cavity. In the current paper, the palate, which is a rich site of minor salivary glands, was the most commonly involved site in 47% of MSGTs, which is consistent with previous studies from different regions.[1,4-10,12-14]

Benign tumors constituted 57.14% of the palate tumors in this series, which is in agreement with previous studies, which indicates that a higher proportion of benign tumors were seen in the palate than malignant tumors.[4-8] The upper lip was the second most affected site (30 % of all MSGTs) in this review, consistent with other researchers' findings.[1,5] Results showed that most upper lip tumors (88.89%)were benign, which is similar to the findings by Waldron et al,[5] who reported that 86% of upper lip MSGTs were benign, however, other authors found that buccal mucosa was the 2nd most common location affected by MSGTs.[6-8]

Our findings showed that benign MSGTs (56.66 %) were more common than malignant MSGTs (43.34%), which is consistent with several studies that recorded more benign MSGTs than malignant MSGTs.[1,4,8,17] On the other hand, some studies have found that malignant MSGTs were more common than benign MSGTs.[12,19] It is reasonable to assume that researches from oncology departments usually report more malignant than benign cases.

In this study's benign MSGTs group, pleomorphic adenoma (PA) was the most common benign MSGTs, accounting for 88.24% of benign MSGTs and 50% of all MSGTs.This finding is in agreement with many previous studies.[1, 4-10,12-15,18] An interesting finding in this review was that PA cases were distributed equally between the palate and the upper lip, which is contrary to previous researches reporting the palate alone as the most common location for PA.[1,4-10,12-16]

Other benign intraoral MSGTs in the current review were only 2 cases (11.76%)out of all 15 benign MSGTs , one of them was diagnosed as monomorphic adenoma involving the upper lip, which is considered as the common site for monomorphic adenoma,[8,14] and the other case was diagnosed as myoepithelioma, a rare salivary gland neoplasm primarily involves the parotid gland, that shares some histopathological features with PA. In contrary to PA, myoepithelioma lacks chondromyxoid stroma and has a more aggressive clinical course. Likewise, Fabio et al.[8] reviewed 546 MSGTs and found only one case of myoepithelioma. In the present review, no cystadenoma or oncocytoma cases were identified. consistently, Yih et al.[17]reviewed 213 MSGTs and did not record cystadenoma or oncocytoma. Similar findings were also reported by Al-Khatib et al.[4], Gbotolorun et al.[13], Sarmento et al.[19], revealing the rarity of these types of tumors.

Adenoid Cystic Carcinoma (ACC)was the most common malignant tumor in the present review accounted for 61.54 % of malignant intraoral MSGT and 26.68 % of all intraoral MSGTs. Al-Khatib et al.[4] also found that ACC was the most common malignant intraoral MSGTs. Similar results were also reported by M. Toida et al.[20],EC Otoh et al.[21],Kruse et al.[22],Belulescu IC[23].

The incidence of ACC metastasis to the distant organ (i.e., lung) was higher than spread to the regional cervical lymph nodes. In this study, one patient developed distant metastasis to the vertebral column after six months of being diagnosed with intraosseous (mandible) ACC. Unfortunately, the patient died after 16 months of the initial treatment. B. Bianchi et al.[24]studied 67 patients with intraoral minor salivary glands ACC and reported distant metastasis in 20 cases (29.85 %) of the 67 patients. Thus, it is essential to rule out distant metastasis during the follow up of ACC patients.

In contrast to some previous studies which found that Mucoepidermoid carcinoma (MEC) was the most common malignant intraoral MSGT.[5,6, 11,12] MEC was the 2nd most common malignant intraoral MSGT in the present study, accounting for 23.08% of malignant tumors and 10 % of all MSGTs.

Polymorphous adenocarcinoma, previously called polymorphous low-grade adenocarcinoma, creates a diagnostic challenge for histopathologists due to the similar and overlapping histopathological features with other tumors such as PA and ACC.[25]In the current review, only one case of malignant intraoral MSGT was diagnosed as polymorphous adenocarcinoma. Furthermore, Kusama et al.[15]did not identify any polymorphous adenocarcinoma in their series of 129 MSGTs.On the contrary, other researchers reported that polymorphous adenocarcinoma was more common than reported in literature; Fabio Ramoˆa Pires et.al.[8] found that out of their 241 malignant intraoral minor salivary gland tumors, 28 (12%) were polymorphous adenocarcinoma. moreover, polymorphous adenocarcinoma was reported as the most common malignant intraoral MSGT in a

study conducted by van Heerden WF.[26]

Carcinoma ex pleomorphic adenoma (CXPA) is a rare, aggressive salivary gland tumor that is hypothesized to arise from an already diagnosed PA lesion.[27] Yih et al.[17] reviewed 213 intraoral MSGT and found only 2 cases of carcinoma ex PA. Similarly, in this review, we recorded one case of CXPA for A 47 years old male patient who had an ulcerative mass on the right palate diagnosed 5years before attending our clinic as PA. Unfortunately, the patient refused treatment at that time and sought treatment only when the lesion became larger in size and painful. Therefore, it is vital to increase the patients' awareness of oral lesions as patients' delay in seeking treatment affects the prognosis and quality of life.

CONCLUSION

It is evident from this study that MSGTs are uncommon in northern Jordan population. The findings show that benign MSGTs were more frequent than malignant MSGTs.Pleomorphic adenoma and adenoid cystic carcinoma were the most common benign and malignant MSGTs respectively. Unlike most previous studies, males were more affected than females. The palate was the most common site for MSGTs. Long term follow up of patients with minor salivary gland tumors is essential to detect early recurrence or metastasis.

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CONTRIBUTIONS BY AUTHORS
All authors contributed substantially