

Salivary gland tumor incidence in adult patients in a tertiary hospital in Mexico from 2008 to 2019

Incidencia de tumores de glándulas salivales en pacientes adultos en un hospital de tercer nivel en el Estado de México del año 2008 al 2019

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Abstract

Objective: To determine the incidence of salivary gland tumors in a population of a tertiary hospital in the State of Mexico, and to describe demographic variables. **Method:** An observational, cross-sectional and retrospective study of salivary gland tumors reported in a tertiary hospital in the State of Mexico in the period 2008-2019 is presented. **Results:** A prevalence of 0.049% was found. There was no difference between sex in the studied population. Benign salivary gland tumors were the most frequent (86.7%). The age range most affected was 51-60 years. The most frequently found tumor was the pleomorphic adenoma, followed by Warthin's tumor. There was 13.33% of sialolipomas, and one myoepithelioma. There were no cases of sublingual gland tumors or minor salivary glands. **Conclusion:** Tumors of the major salivary glands are infrequent tumors; population cases from a central Mexican state and their demographic characteristics are presented to contribute to the information found in local and international literature.

Keywords: Salivary glands. Tumor. Incidence. Mexico. Mexicans.

Resumen

Objetivo: Determinar la incidencia de los tumores de glándulas salivales en una población de un hospital de tercer nivel en el Estado de México y describir variables demográficas. **Material y métodos:** Se presenta un estudio observacional, transversal y retrospectivo de los tumores de glándulas salivales reportados en un hospital de tercer nivel en el Estado de México en el periodo 2008-2019. **Resultados:** Se encontró una prevalencia del 0.049%. No hubo diferencia entre sexos en la población afectada. Los tumores de glándulas salivales benignos fueron los más frecuentes (86.7%). El rango de edad mayormente afectado fue el de 51-60 años. El tumor más frecuentemente encontrado fue el adenoma pleomorfo, seguido por tumor de Warthin. Se presentó un 13.33% de sialolipomas y un mioepitelioma. No se presentó ningún caso de tumores de glándulas sublinguales ni glándulas salivales menores. **Conclusión:** Los tumores de glándulas salivales mayores son tumores infrecuentes, se exponen casos de población de un Estado del centro de México y sus características demográficas para contribuir a la información encontrada en literatura local e internacional.

Palabras clave: Glándulas salivales. Tumor. Incidencia. México. Mexicanos.

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Introduction

Salivary gland tumors are rare tumors, estimated to represent 3% of all head and neck tumors¹. The World Health Organization (WHO) created a classification for these tumors in 1982, with the latest updates in 2005 and 2017². These updates included adding soft tissue lesions and other epithelial lesions to the classification of benign tumors³.

Various global studies from 1999 through 2019 have reported a mean incidence rate of malignant tumors of 0.25% vs a 68.3% incidence rate of benign tumors. When focusing on studies conducted in Mexico, the mean incidence rate of malignant tumors is 0.41%, with benign tumors accounting for 66.0% (Table 1).

Among different types of salivary gland tumors, benign tumors are the most common (Table 1). The most frequent histopathological type of tumor is pleomorphic adenoma¹⁴. The parotid gland¹⁵ is the most widely affected gland by neoplasms, with a female predominance (2:1), except for Warthin's tumor, which happens to be more common in men¹⁶.

In 19 studies of 8049 cases, the mean number of tumors analyzed per study was 456 (range, 59-2508). The rate of benign tumors was 71.5%, with a male-to-female ratio of 1.0:0.9 and a mean age of 46.9 years. Different incidence rates of involvement in various glands were observed, eg, the parotid gland, submaxillary gland, sublingual gland, minor salivary glands, pleomorphic adenoma, and Warthin's tumor showed incidence rates of 72.9%, 16.5%, 0.5%, 14.3%, 59.8%, and 13.6%, respectively. The parotid gland had the highest incidence rates reported, while the sublingual gland had the lowest ones (Table 2).

Incidence has been reported to vary due to sample size and the period analyzed by each study, even within the same population, as seen in the data from Mexico (Tables 1 and 2). Additionally, due to the rarity of this condition, epidemiological studies are scarce²⁵. Therefore, this study aims to report data from a 10-year period to determine the incidence rate of salivary gland tumors in a tertiary referral center from the State of Mexico.

Material and methods

Study design

This is an observational, cross-sectional, and retrospective study.

Table 1. Incidence of major salivary gland tumors

Study	Year of study	Malignant tumors (%)	Benign tumors (%)	Country
Pinkston ⁴	1999	0.093	84.3	United States
Ledesma-Montes ⁵	2002	1.2	64.2	Mexico
Ansari ⁶	2007	0.019	68.4	Iran
Toranzo-Fernández ⁷	2008	0.11	55.9	Mexico
Mejía-Velázquez ⁸	2012	0.15	67	Mexico
Eti ⁹	2012	0.079	62.1	Turkey
Araya ¹⁰	2015	0.002	70.3	Chile
Bobati ¹¹	2017	0.58	69	India
Sotelo-Gavito ¹²	2018	0.16	77.2	Mexico
Reinheimer ¹³	2019	0.08	65.3	Brazil

Data mining and analysis

The study reviewed a total of 60 261 pathology records from a tertiary referral center in Mexico, specifically searching for records classified as major salivary gland tumors from 2008 through 2019. Diagnoses were corroborated with results records. Data mined included patient age, gender, affected gland, and tumor histology features.

Statistical analysis

Descriptive statistics were used for data presentation, and data were analyzed using Excel® and SPSS® (version 23.0, SPSS Inc., Chicago, IL, United States).

Results

In a tertiary referral center in Mexico, a total of 60 261 pathology specimens were studied over a 10-year period. Within these specimens, 30 tumors were categorized as major salivary gland tumors, and their histopathological results matched 1 of the classifications provided by the WHO. This accounts for an incidence rate of 0.049%.

The studied included 15 men and 15 women with salivary gland tumors. However, regarding benign tumors, there was a slight male predominance (53.8%) of cases. On the other hand, regarding malignant tumors, there was a female predominance, with 3 cases being reported in women vs only 1 case in men (Table 3).

Table 2. Characteristics of retrospective clinical trials

Author	Population	Year	No. of tumors studied	Benign tumors (%)	Women (%)	Men (%)	Parotid gland involvement (%)	Submandibular gland involvement (%)	Sublingual gland involvement (%)	Minor salivary glands involvement (%)	Pleomorphic adenoma (%)	Warthin tumor (%)	Mean age, or age range
Pinkston ⁴	United States	1999	248	84.30	NR	NR	86	14	0	0	65.60	NR	55.4
Maalita ¹⁷	Jordan	1999	221	68.40	38.47	61.53	70.13	10.40	0.45	19.00	47	NR	40-49
Vargas ¹⁸	Brazil	2002	124	80	60	40	70.96	24.19	0	4.83	67.70	10.48	48
Ito ¹⁹	Brazil	2005	496	67.50	NR	NR	67.70	9.47	0	22.70	54.20	NR	NR
Ansari ⁶	Iran	2007	130	68.40	60	40	63	23	0	14	65.40	NR	39
Toranzo-Fernández ⁷	Mexico	2007	106	55.90	53.85	46.15	NR	NR	NR	NR	NR	NR	NR
Subhashraja ²⁰	India	2008	684	62	48.83	51.17	60.52	16.95	0.58	21.92	53	3	43
Shishegar ²¹	Iran	2011	392	68.20	49.50	50.50	58.9	59	NR	NR	58	6	44.57±14.65
Trenkic ²²	Serbia	2012	139	73.38	52.52	47.48	84.70	5.76	0	10.07	49.02	47.06	51.2±13.9
Etir ⁹	Turkey	2012	235	62.13	51.06	48.94	82.38	17.62	0	17.87	67.12	21.23	NR
Mejía-Velázquez ⁸	Mexico	2012	319	67.04	63.80	36.20	78	20.70	3.60	17.70	65.50	7.30	42
Boza-Mejías ²³	Peru	2012	62	91.94	46.77	53.23	93.55	6.45	0	0	58.06	4.84	41.69
Luksic ²⁴	Croatia	2012	779	64.20	49.70	50.30	65.30	6.60	0.09	27.20	66.80	22.80	52
Bradley ¹⁵	United Kingdom	2013	1065	86.19	NR	NR	81	9	0	9	71	22	NR
Wang ¹⁶	China	2014	2508	77.11	49.60	50.40	69.70	10.48					
Araya ¹⁰	Chile	2015	279	70.30	58.10	41.90	67	17.60	0	18.70	53.80	8.20	53.3±19.09
Bobati ¹¹	India	2017	59	69.16	64.40	35.59	84.21	NR	NR	3.39	60.71	5.07	43
Sotelo-Gavito ¹²	Mexico	2018	79	77	51%	49	72	18	0	10	72	10	52.13
Reinheimer ¹³	Brazil	2019	124	65.30	54.83	45.17	57.20	11.20	2.41	29.00	59.67	3.22	53

NR: not registered.

Table 3. Type of tumor affecting by gender

Sex	Histological type of tumor														
	Benign								Malignant						Total
	Pleomorphic adenoma	%	Warthin tumor	%	Lipoma	%	Myoepithelioma	%	Mucoepidermoid carcinoma	%	Ex pleomorphic carcinoma	%	Hodgkin's lymphoma	%	
Man	7	23.33	3	10	3	10	1	3.33	0	-	1	3.33	0	-	15 (50%)
Woman	5	16.67	6	20	1	3.33	0	-	1	3.33	0	-	2	6.67	15 (50%)
Total	12	40	9	30	4	13.33	1	3.33	1	3.33	1	3.33	2	6.67	30 (100%)

Table 4. Type of tumor affecting by age range

Age ranges	Histological type of tumor														
	Benign								Malignant						Total
	Pleomorphic adenoma	%	Warthin tumor	%	Lipoma	%	Myoepithelioma	%	Mucoepidermoid carcinoma	%	Ex pleomorphic carcinoma	%	Hodgkin's lymphoma	%	
21-30	0	-	0	-	0	-	1	3.33	1	3.33	0	-	0	-	2 (6.67%)
31-40	3	10	0	-	0	-	0	-	0	-	1	3.33	0	-	4 (13.33%)
41-50	5	16.67	1	3.33	1	3.33	0	-	0	-	0	-	1	3.33	8 (26.67%)
51-60	4	13.33	5	16.67	1	3.33	0	-	0	-	0	-	1	3.33	11 (36.67%)
61-70	0	-	3	10	2	6.67	0	-	0	-	0	-	0	-	5 (16.67%)
Total	12	40	9	30	4	13.33	1	3.33	1	3.33	1	3.33	2	6.67	30 (100%)

Regarding the age at which tumors were diagnosed, the most affected age range was 51 to 60 years, followed by 41 to 50 years. Most pleomorphic adenomas were diagnosed in the 41 to 50 age group (41.7%), while most Warthin's tumors were diagnosed in the 51 to 60 age group (55.6%). Sialolipomas were found in older age ranges, with 50% (2) occurring in the 61 to 70 age group, and myoepithelioma being diagnosed in the 21 to 30 age group. Malignant tumors and non-Hodgkin lymphomas were diagnosed in patients in their 6th decade of life, while mucoepidermoid carcinoma and carcinoma ex pleomorphic adenoma were diagnosed in the 3rd and 4th decades of life, respectively (Table 4).

Out of the 30 major salivary gland tumors reported, 26 (86.7%) were benign tumors, and 4 (13.3%) were malignant tumors. Among the former, pleomorphic adenoma was the most frequent, accounting for 40% of all tumors, followed by Warthin's tumor, representing 30% of all tumors reported (Table 2). Additionally, 1 myoepithelioma (3.3%) and 4 sialolipomas (13.3%) were reported. Among malignant tumors, 2 non-Hodgkin lymphomas, 1

mucoepidermoid carcinoma, and 1 carcinoma ex pleomorphic adenoma were diagnosed (Table 5).

The parotid gland was the most widely affected major salivary gland (76.7%), followed by the submaxillary gland (23.3%). In this study, no tumors were reported in the sublingual gland or minor salivary glands. The most common tumor in both the parotid and submaxillary glands was the pleomorphic adenoma. Warthin's tumor was predominantly found in the parotid gland, while sialolipomas occurred equally in both the parotid and submaxillary glands. The sole myoepithelioma found was located in the parotid gland. Among malignant tumors, only carcinoma ex pleomorphic adenoma was found in the submaxillary gland, while the rest were found in the parotid gland (Table 5).

Discussion

The present study reported an incidence rate of salivary gland tumors lower compared to former studies reported in the Mexican population, such as

Table 5. Type of tumor affecting each salivary gland

Salivary gland	Histological type of tumor														Total
	Benign									Malignant					
	Pleomorphic adenoma	% Warthin tumor	% Lipoma	% Mioepithelioma	% Mucoepidermoid carcinoma	% Ex pleomorphic carcinoma	% Hodgkin's lymphoma								
Parotid	9	30	8	26.67	2	6.67	1	3.33	1	3.33	0	-	2	6.67	23 (76.67%)
Submaxillary	3	10	1	3.33	2	6.67	0	-	0	-	1	3.33	0	-	7 (23.33%)
Total	12	40	9	30	4	13.33	1	3.33	1	3.33	1	3.33	2	6.67	30(100%)

Ledesma-Montes et al., Mejía-Velázquez et al., Toranzo-Fernández et al., and Sotelo-Gavito et al. who reported incidence rates of 1.2%⁵, 0.15%,⁸ 0.11%,⁷ and 0.08%¹², respectively. However, the incidence rate found is comparable to that of authors from other countries, such as Etit et al. in the Turkish population (0.096%)⁹ and Reinheimer et al. in Brazil (0.08%)¹³, and it is even higher than that reported by Araya et al. in Chile at 0.002%¹⁰.

The study found no significant differences between sexes regarding salivary gland tumors. In the case of benign tumors, there was a slight male predominance, which differs from most studies that report an overall female predominance for this type of tumor. Some studies do report male predominance (53.3%²³) or in lower incidence rates^{16,20,21,24}, while others report female predominance. For instance, Ma'aita et al. reported a male predominance of 61.53%, while Mejía-Velázquez and Bobati reported a female predominance (63.88%⁸, and 64.4%¹, respectively).

Differences exist in the reported ages and the way they are presented in the literature. In the current study, the 6th decade of life is the most commonly affected, which is consistent with the findings made by Pinkston et al., Trenkic et al., Araya et al., Sotelo-Gavito et al., and Reinheimer et al., who reported mean ages of 55.4 years⁴, 51.2 years²², 53.3 years¹⁰, 52.13 years¹², and 53 years, respectively¹³.

The incidence rate of benign tumors is similar to that found by Bradley et al. in the United Kingdom, Pinkston et al. in the United States, and Vargas et al. in Brazil (> 80%)^{4,15,18}. Only 1 study reported a higher incidence rate of benign salivary gland tumors than the present study, with a 91.2% reported by Boza-Mejías et al.²³. The incidence rates reported among the Mexican population vary from 55.9% in Mejía-Velázquez's study up to 77.2% in Sotelo-Gavito's study^{7,12}.

The salivary gland most commonly affected was the parotid gland, which is consistent with all the studies compared. However, there are different rates of involvement reported in the literature, ranging from 57.2% in the Peruvian population up to 93.5%^{13,23} in the Brazilian population. The second salivary gland most widely affected is the submaxillary gland, and the incidence rates also varies widely in the literature. Our value is similar to the one reported by Mejía-Velázquez et al. at 20.7%⁸. However, other studies have reported incidence rates ranging from 5.76%²² up to 59%²¹. However, most of the studies found report incidence rates of the submaxillary gland between 10% and 20%^{4,9,10,17,20}. In the present study, no tumors were found in the sublingual gland or in the minor salivary glands. Former studies have reported incidence rates < 4%, or even < 1% for the involvement of these glands^{8,17,20,24}.

Pleomorphic adenoma was the most common tumor in all studies, including the present one. The incidence rate found in this study is consistent with that found by Ma'aita et al. (47%¹⁷), and Trenkic et al. (49%²²), which are among the lower incidence rates ever reported. The overall incidence rate of pleomorphic adenoma can be up to 72% in other studies, like Sotelo-Gavito et al. reported¹². Warthin's tumor is often reported as the 2nd most frequent tumor. Trenkic et al. and Etit et al. report incidence rates of 47.06% and 21.23%, respectively^{9,22}, while other studies report lower incidence rates ($\leq 10\%$) (Table 2).

Mioepitheliomas are rare, with only 1 case presented in this study, which is similar to other studies reporting just 1 case, just like our study^{10,18,20}. Other authors report 2 cases^{13,22}, while only 1 author reports 12 cases in his analysis of 392 salivary gland tumors²¹. As for sialolipomas, which are considered non-epithelial tumors, some studies report 2 cases in men out of

279 tumors¹⁰, 3 cases in the parotid gland out of 62 tumors²³, another study reported 4 cases out of 235 tumors⁹, 1 sialolipoma in 106 cases⁷, or 1 in 130 tumors⁶.

There has also been variability in the presentation of malignant tumors in major salivary glands in various studies, with incidence rates ranging from 8.06%²³ up to 44.1%⁷, although the mean incidence rate of the studies presented in table 3 is 29%. In the study conducted by Boza-Mejía et al., which had the highest incidence rate of benign salivary gland tumors, they reported 2 cases of adenocarcinoma and 2 cases of squamous cell carcinoma, each accounting for 3.23%²³. In Bradley et al.'s study, a lower incidence rate of malignant salivary gland tumors was reported vs the table 3 mean. They reported that the most common malignant tumor was mucoepidermoid carcinoma, being the parotid gland the most widely damaged¹⁵. Additionally, Pinkston et al. reported a lower incidence rate of malignant salivary gland tumors, and similarly, they reported that mucoepidermoid carcinoma is the most common one accounting for 8.1% of the reported tumors and primarily affecting the parotid gland⁴.

However, the presentation of mucoepidermoid carcinoma in the 3rd decade and adenoid cystic carcinoma in the 4th decade of life, as well as the 2 cases of Hodgkin lymphoma in the current study, may be due to the fact that the hospital unit where data was collected is not specialized in the management of cancer patients.

Conclusions

Despite reporting all cases over a 10-year period, this retrospective study found a lower incidence rate of salivary gland tumors vs the medical literature currently available. There was a male predominance in most diagnoses, except for Warthin's tumor, where females predominated. These findings differ from what is generally presented in the literature. Due to the limited number of cases studied, rare histopathological varieties such as mioepithelioma (1) and sialolipomas (4) were identified. Further prospective and cohort studies are needed to identify the risk factors associated with different histopathological types of these tumors. Additionally, investigating the rates of recurrence of salivary gland tumors and the presentation of other tumor types in the same patient may reveal correlations that contribute new information to established diagnoses.

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Conflicts of interest

None declared.

Ethical disclosures

Protection of human and animal subjects. The authors declare that no experiments were performed on humans or animals for this study.

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors have obtained the written informed consent of the patients or subjects mentioned in the article. The corresponding author is in possession of this document.

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