

Original Research Article

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Histopathological spectrum of salivary gland lesions in Ajmer region, Rajasthan, India

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ABSTRACT

Background: There is a wide spectrum of salivary gland lesions with morphological and clinical diversity which makes it a difficult task for histopathological interpretation. Tumours of salivary glands are uncommon accounting for 3-10% of the total tumors of head and neck region and less than one percent of all tumours. The aim of this study was to recognize various histomorphological patterns of salivary gland lesions, their frequency, age, gender and site wise distribution.

Methods: This study was carried out from June 2015 to May 2018 in the department of Pathology, JLN Medical College and associated Group of Hospitals, Ajmer. Total 121 cases of salivary gland lesions were included. Specimens were processed and stained by Hematoxylin and Eosin stain followed by histopathological examination.

Results: Out of total 121 cases, 43.8% were non-neoplastic and 56.2% were neoplastic. In non-neoplastic lesions predominant was chronic sialadenitis (50.9%) followed by mucocele (28.3%) which commonly seen in submandibular gland (47.17%). Among neoplastic cases, 79.4% were benign and 20.6% were malignant lesions. Neoplastic lesions commonly were seen in parotid (75%). Pleomorphic adenoma was the commonest benign tumour (81.4%). Mucoepidermoid carcinoma was the most common malignant salivary tumour. Benign tumours were common in third and fourth decades, whereas malignant tumours were more common in fifth and sixth decades. Male predominance was seen in overall salivary gland lesions.

Conclusions: Histopathological examination is mandatory in the diagnosis of salivary gland lesions because of their wide spectrum of histomorphology.

Keywords: Mucoepidermoid carcinoma, Pleomorphic adenoma, Salivary gland lesions, Sialadenitis

INTRODUCTION

Salivary glands are important structures which secrete saliva, that take part in food digestion process. Saliva, apart from carrying some digestive enzymes, also has concentration of antibodies that participate in the body's defense system. The constant flow of saliva in the mouth reduces the accumulation of bacteria on intra oral structures, therefore reducing the chances of infection. Its lubrication effect makes speech and mastication easy and comfortable.¹

The spectrum of salivary gland lesions is wide and the relative incidence of neoplastic versus non neoplastic lesions is variable in different studies. The non-neoplastic conditions range from an inflammatory disorder of infectious, granulomatous or autoimmune etiology to obstructive, developmental and idiopathic disorders. These often present clinically as tumours and may have pathological feature similar to some of the neoplasm.²

Salivary gland tumours can show a striking range of morphological diversity between different tumour types

and sometimes within an individual tumour mass. In addition, hybrid tumours, dedifferentiation and the propensity for some benign tumours to progress to malignancy can confound histopathological interpretation. These features, together with the relative rarity of a number of tumours, can sometimes make diagnosis difficult.³

The global annual incidence when all salivary gland tumours were considered varied from 0.4-13.5 cases per one lakh population.⁴ In India overall incidence of Salivary gland tumours can be ascertained from the cancer registry established by Indian council of medical research. However the geographic area and population covered by these registries are small and perhaps unrepresentative of the Indian population. In addition there is limited published literature on Salivary gland tumours in Indian population.⁵

Between 64% to 80% of all primary epithelial salivary gland tumors occur in the parotid gland with most located in the superficial (lateral) lobe; 7-11% occur in the submandibular glands; fewer than 1% occur in the sublingual glands and 9-23% occur in minor glands.⁴

Females are more frequently affected, but there is some gender variation according to the tumour type. The average ages of patients with benign and malignant tumours are 46 and 47 years, respectively, and the peak incidence of most of the specific types is in the sixth and seventh decades.⁴ Among all patients, the most common tumour type is pleomorphic adenoma, which accounts for about 50% of all tumours. Warthin's tumour is second in frequency among benign tumours. In most large studies mucoepidermoid carcinoma is the most common malignant tumor.⁴ The aim of this study was to recognize various histomorphological patterns of salivary gland lesions, their frequency, age, gender and site wise distribution.

METHODS

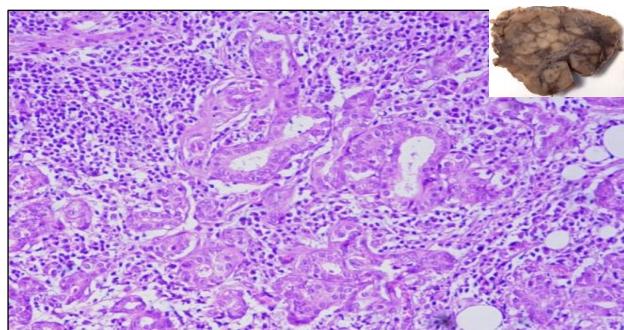
The present study "Histopathological Spectrum of Salivary gland lesions in Ajmer region Rajasthan." is carried out in the Department of Pathology, J.L.N. Medical College Ajmer and Associated Group of Hospitals, during a period of 3 years, retrospectively (from June 2015 to May 2016) and prospectively (from June 2016 to May 2018). Total 121 cases, of which 38 are retrospective and 83 are prospective cases of salivary gland Lesions are studied. For retrospective cases the blocks were retrieved and fresh sections were cut, stained and re-examined. Information regarding age, sex, chief complaints, clinical examination and radiological investigations were recorded from the Histopathological section of Department.

For prospective study the records of all the surgically resected salivary gland specimens received in department of pathology, J.L.N. Medical College, Ajmer were

included, which are properly labelled, preserved in 10% buffered formalin, and accompanied with completed requisition form containing the patient's identification, age, sex, clinical details. Specimen processed and stained with routine Haematoxylin and Eosin stain and special stains like Periodic Acid Schiff and Mucicarmine stain etc. The lesions were differentiated in nonneoplastic and neoplastic lesions. The neoplastic lesions were classified according to WHO histological typing of salivary gland tumors.

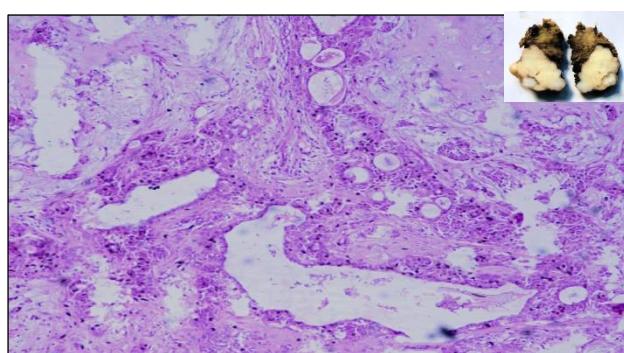
RESULTS

In our study total of 18631 specimens were received for histopathological examinations, during the period of three years from June 2015 to May 2018 of which 121 specimens of salivary gland lesions included according to inclusion criteria, representing 0.65%. In the study out of total 121 cases, 53 cases (43.80%) were diagnosed as non-neoplastic lesions and 68 cases (56.2%) as neoplastic lesions. Chronic sialadenitis (Figure 1) was the commonest non neoplastic salivary gland lesion accounting for 23 cases (43.39%) followed by mucocele 15 cases (28.84%).



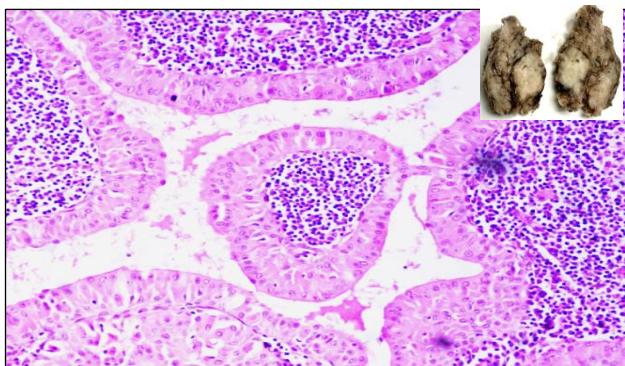
Inset showing cut surface gray brown to gray white lobulated, firm.

Figure 1: Chronic sialadenitis: showing lymphoplasmacytic infiltrate and areas of periductal fibrosis (H&E, 200X).



Inset: cut surface gray white area, solid homogenous; rest gray brown fibrofatty.

Figure 2: Pleomorphic adenoma: composes of epithelial and mesenchymal elements with tubular and ductal, cystic, squamoid and chondroid differentiation (H&E, 100X).

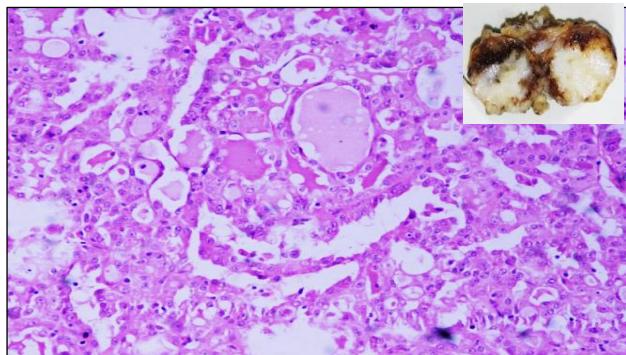


Inset showing cut surface, single well demarcated firm gray brown to gray white area (2x1.5 cm).

Figure 3: Warthin's tumor: consist of blunt papillary projection exhibits double layers of oncocytic lining cells and underlying lymphoid stroma, cystic spaces contains eosinophilic secretion (H&E, 200X).

Among all salivary gland lesions, pleomorphic adenoma (Figure 2) was the single commonest lesion with 44 cases (36.36%). In neoplastic lesions, benign tumours were far ahead in frequency than malignant with 54 cases (79.41%) out of total 68 cases of tumours. Out of total 54 cases of benign neoplastic lesions, pleomorphic adenoma was the commonest, (44 cases, 81.4%) followed by

Warthin's tumour (7 cases, 13%). Warthin's tumor was found exclusively in parotid gland with M:F ratio 6:1 (Figure 3).



Inset showing cut surface soft, smooth, gray white with peripheral gray brown area.

Figure 4: Mucoepidermoid carcinoma: Cystic spaces surrounded by mucinous cells with areas of squamous component (H&E 200X).

Out of total 14 malignant neoplastic lesions, Mucoepidermoid carcinoma (Figure 4) was the most common malignant salivary gland tumour (7 cases, 50%) followed by Adenoid cystic carcinoma (3 cases, 21.42%) (Table 1).

Table 1: Incidence and age wise distribution of salivary gland lesions.

Lesions	Age group in years							Total	Percentage (%)
	0-10	11-20	21-30	31-40	41-50	51-60	61-70		
Chronic sialadenitis	-	3	10	6	2	-	2	23	19
Mucocele	3	7	3	1	-	-	1	15	12.4
Sialolithiasis	-	2	-	1	-	1	-	4	3.3
Acute sialadenitis	-	2	1	1	-	-	-	4	3.3
T.B. sialadenitis	-	1	1	-	-	-	-	2	1.65
Ranula	-	1	-	1	-	-	-	2	1.65
Lympho-epithelial cyst	-	1	-	-	1	-	-	2	1.65
Hydatid cyst	-	-	1	-	-	-	-	1	0.82
Non neoplastic	3	17	16	10	3	1	3	53	43.80
Pleomorphic Adenoma	1	4	16	13	6	3	1	44	36.36
Warthin's tumour	-	-	-	2	2	3	-	7	5.78
Oncocytoma	-	-	-	-	-	1	-	1	0.82
Sialolipoma	-	-	-	-	-	-	1	1	0.82
Myoepithelioma	-	-	-	-	1	-	-	1	0.82
Benign neoplastic	1	4	16	15	9	7	2	54	44.63
Mucoepidermoid Ca	-	-	1	1	1	3	1	7	5.78
Adenoid cystic Ca	-	-	-	-	1	1	1	3	2.48
Ca Ex-PA	-	-	-	-	-	1	-	1	0.82
Acinic cell carcinoma	-	-	-	-	1	-	-	1	0.82
Squamous cell carcinoma	-	-	-	-	-	-	1	1	0.82
Adenocarcinoma	-	-	-	-	-	1	-	1	0.82
Malignant neoplastic	-	-	1	1	3	6	3	14	11.57
Total	4	21	33	26	15	14	8	121	100

The peak incidence of Non-neoplastic lesions was observed in 2nd and 3rd decade with mean age 27.09±13.62 years. Benign tumours were common in 3rd and 4th decades (mean age 37.26±13.69 years), whereas malignant tumours were commonest in 6th decade (mean age 53.57±12.67 years). Male predominance was seen in overall salivary gland lesions with exception in non neoplastic lesions in which M:F ratio was 1:1.21. The M:F ratio of benign neoplastic lesions was 1.45:1 and in malignant neoplastic lesions was 1.33:1 (Table 2).

Table 2: Gender wise distribution of salivary gland lesions.

Lesions	Male	Female	Total	M:F ratio
Chronic sialadenitis	10	13	23	1:1.3
Mucocele	9	6	15	1.5:1
Sialolithiasis	2	2	4	1:1
Acute sialadenitis	-	4	4	0:4
T.B. sialadenitis	1	1	2	1:1
Ranula	1	1	2	1:1
Lymphoepithelial cyst	1	1	2	1:1
Hydatid cyst	-	1	1	0:1
Non neoplastic	24	29	53	1:1.21
Pleomorphic adenoma	25	19	44	1.31:1
Warthin's tumour	6	1	7	6:1
Oncocytoma	-	1	1	0:1
Sialolipoma	-	1	1	0:1
Myoepithelioma	1	-	1	1:0
Benign neoplastic	32	22	54	1.45:1
Mucoepidermoid Ca	3	4	7	1:1.33
Adenoid cystic Ca	2	1	3	2:1
Ca Ex-PA	-	1	1	0:1
Acinic cell carcinoma	1	-	1	1:0
Squamous cell carcinoma	1	-	1	1:0
Adenocarcinoma	1	-	1	1:0
Malignant neoplastic	8	6	14	1.33:1

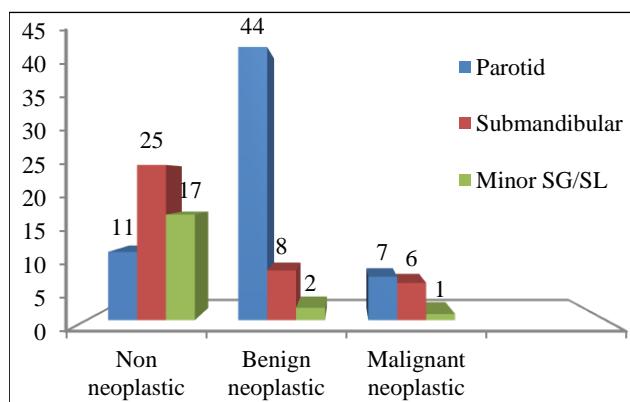


Figure 5: Distribution of salivary gland lesions according to site.

In site wise distribution of lesions, out of 121 cases overall commonest site involved was parotid with 62 cases (51.24%). Non-neoplastic lesions were more commonly seen in submandibular gland (47.17%) followed by minor salivary glands (32.08%). Neoplastic lesions were most commonly distributed in parotid gland (75%), followed by submandibular gland (20.6%) and minor salivary glands (4.4%), in decreasing order of frequency (Figure 5).

DISCUSSION

In our study out of 121 cases of salivary gland lesions we found 43.8% non-neoplastic lesions and 56.2% neoplastic lesions which correlate with studies of Mohan H et al (2011) and Ankur et al (2014).^{2,6} In our study Non-neoplastic lesions are comparatively more than other studies that of Laishram R et al (2013) with 25% and Kumar M A et al (2017) with 27.27% non-neoplastic cases.^{7,8} Peak age range was 11-40 years (Mean age 27.09±13.67 yr.) in non-neoplastic salivary lesions and most common site involved was submandibular gland which is concordant with studies of Mohan H et al (2011), Ankur et al (2014) and Soni D et al (2016).^{2,6,9} There is a female preponderance in our study (M:F=1:1.21) which correlates with the study of H. Mohan et al (2011) and Devi KR et al (2016).^{2,10}

Chronic sialadenitis was the commonest non neoplastic salivary gland lesion accounting for 23 cases (43.39%) which correlates with studies of, Mohan H et al (2011) (43%) and Ankur et al (2014) (47.62 %).^{2,6} Peak age incidence seen in 20-40 year which is also correlates with H. Mohan et al (2011) (20-40 years) and Ankur et al (2014) (20-50 year).^{2,6} Gender wise distribution shows female preponderance (M: F ratio 1:1.3) which is similar to Mohan H et al (2011) (M:F ratio of 1:1.2).² There were four cases of Sialolithiasis (7.54% of non-neoplastic cases) all were accompanied with chronic sialadenitis.

In this study 15 cases (28.84% of the non neoplastic group) of mucocele were diagnosed. They were seen mainly in 1st to 3rd decades (Age range 1 to 63 year) with male predominance (M:F ratio 1.5:1). They were all present in minor salivary glands, predominantly on lower lip with 13 cases (86.67%), one case on hard palate and one on floor of mouth. It correlates with studies of Mohan H et al (2011) (19% of non- neoplastic group, peak age of 10-40 year with male predominance and 80% in minor salivary gland) and Lopez P et al (2005) (age range 5-65 year with male predominance, M:F=1.57:1 and 94% cases of lower lip minor salivary gland).^{2,11}

In present study we found a diverse range of non-neoplastic cases with two cases of lymphoepithelial cyst, four cases of acute sialadenitis, two cases each of ranula and TB sialadenitis and one interesting case, hydatid cyst of salivary gland.

In neoplastic lesions we observed that benign tumours (79.4%) predominate over the malignant (20.6%), correlating with the findings of Pablo et al (2002), Fonseca et al (2012), Venugopal M et al (2016).¹²⁻¹⁴ Benign tumours are seen in a lower age group (mean age 37.36 ± 13.69 yr.) as compared to malignant tumours (mean age 53.57 ± 12.67 yr.), comparable with studies of Ankur et al (2014) and Soni D et al (2016).^{6,9}

In the present study, there is a male preponderance in neoplastic lesions (M:F ratio, 1.43: 1) which correlates with other studies that of Ahmed et al (2002), Shrestha S et al (2014) and Amin et al (2017).¹⁵⁻¹⁷

Among the salivary gland tumours, parotid was most commonly involved (75%) followed by submandibular gland (20.6%) which correlates with the studies of Pablo et al (2002), Amin et al (2017) and Rewusuwan et al (2006).^{12,17,18}

Pleomorphic adenoma was the most common tumour accounting for 81.48% of all benign tumours and 64.7% of overall tumours, concordant with the results of Soni D et al (2016), Pablo et al (2002) and Mohd Ayub et al (2008).^{9,12,19}

Parotid gland was commonest site of pleomorphic adenoma (77.27%) followed by submandibular gland (18.18%), similar with studies of Amin et al (2017), Rewusuwan et al (2006) and Bashir S et al (2013).^{17,18,20} There is a peak incidence in the middle age group (21-50 yr.), with male preponderance (M:F ratio, 1.31:1) like studies of Laishram et al (2013) and Amin et al (2017).^{7,17}

Warthin's tumour was second common benign tumor (12.96%) concordant with studies of, Ankur et al (2014), Laishram et al (2013), and Pablo et al (2002).^{6,7,12} Peak age incidence was seen in 31-60 years with M:F ratio of 6:1. Age wise distribution correlates with that of Ankur et al (2014), Laishram et al (2013), and Soni D et al (2016).^{6,7,9} Male predominance observed in this study is more pronounced, than other studies and the cases of Warthin's tumour were limited to parotid gland (100%).

One case of myoepithelioma accounting for 1.47% of all tumours and 1.85% of benign tumours was seen, correlating with Pablo et al (2002) (one case out of 124 tumors), Fonseca et al (2012) (3 cases out of 369 benign tumour).^{12,13} One case of sialolipoma was found comprising 1.47% of all tumours and 1.85% of benign tumours comparable to studies by Shrestha S et al (2014) (one case in parotid gland out of 66 benign tumours i.e. 1.51%). Amin et al (2017) (1 case out of 96 total tumours, 2.78% of benign tumours).^{16,17}

One case of oncocytoma was found comprising 1.47% of all tumours and 1.85% of benign tumours correlating to studies by Rewusuwan S et al (2006) (1 case out of 180 primary tumour i.e. 0.56%), Bashir et al (2013) (one case out of 80 tumors i.e. 1.25%).^{18,20}

The present study shows seven cases of mucoepidermoid carcinoma (10.29% of all tumors and 50% of malignant), comparable with other studies that of Soni D et al (2016), Pablo et al (2002) and Iqbal MS et al (2013).^{9,12,21} Age incidence of mucoepidermoid carcinoma in 21-70 year age group with female predominance, correlated with studies of Laishram et al (2013), Pablo et al (2002), Fonseca et al (2012).^{7,12,13} Parotid gland was the most common site for Mucoepidermoid carcinoma followed by submandibular gland as of Soni D et al (2016) and Venugopal M et al (2016).^{9,14}

In this study 3 cases of adenoid cystic carcinoma were found accounting for 4.41 % of all tumours and 21.43 % of malignant tumours, which correlates with studies of, Laishram, et al (2013, Soni D et al (2016 and Pablo et al (2002).^{7,9,19} There is male predominance in adenoid cystic carcinoma with M:F ratio of 2:1. One case of Carcinoma ex - pleomorphic adenoma was seen comprising 1.47% of all tumours and 7.14 % of malignant tumours correlating with studies of Laishram et al (2013) and Pablo et al (2002).^{7,12} One case of Acinic cell carcinoma was seen accounting for 1.47% of all tumours and 7.14% of malignant tumours Ankur et al (2014) and Soni D et al (2016).^{6,9} One case of squamous cell carcinoma was observed in a 70 year old male patient in parotid gland accounting for 1.47% of all tumours and 7.14% of malignant tumour similar to findings of Laishram et al (2013) and Soni D et al (2016).^{7,9} In the present study 1 case of Adenocarcinoma (NOS) was seen in 60 year male in submandibular gland comprising of 1.47% of all tumours and 7.14% of malignant tumours, which correlates with study of Pablo et al (2002) and Amin et al (2017).^{12,17}

CONCLUSION

The findings of this study were more or less similar to those in the published literature. There was distinctively high frequency of non-neoplastic lesions in comparison to many other studies. Several studies have reported a significant difference in the global distribution of salivary gland tumors, due to factors not clearly known.

However, some authors have attributed the differences in incidence to racial factors, the pathology centers of sample collection and the duration of the studies. There is limited epidemiological data of incidence of salivary gland lesions in tertiary care centers of Rajasthan. For that we need more population based formal studies to be carried out in this part of the globe to define the epidemiology of salivary gland neoplasm. There is a wide spectrum of salivary gland lesions with morphological and clinical diversity.

Histopathological examination of salivary gland lesions is the most important method in differential diagnosis of non-neoplastic and neoplastic lesions and in establishing the final diagnosis and deciding the final course of management.

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