Pattern of salivary gland tumors in Manipur, India: A 10 year study

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Abstract

Background: Salivary gland lesions, especially the neoplastic lesions constitute a highly heterogeneous histopathologic group. Several studies have reported a significant difference in the global distribution of salivary gland tumors, but no formal study has been carried out in this part of the globe. Objectives: To document the pattern of various salivary gland tumors in Manipur, a state in North Eastern India. Materials and Methods: This is a 10 years (2002-2011) retrospective study of all salivary gland specimens received at our referral teaching hospital in Manipur, India. All the histopathology slides of salivary gland specimens during the study period were reviewed and clinical details were obtained from the archives. Restaining of slides and fresh sections of tissue blocks were performed whenever required. Data thus collected were analyzed. Results: A total of 104 cases of salivary gland lesions were studied during the study period. Age ranged from 5 years to 78 years with an overall slight female preponderance (M:F = 1:1.08). Parotid (56.65%) was the commonest gland involved followed by submandibular gland (31.73%). Neoplastic lesions comprised of 78 (75%) cases and non-neoplastic lesions constituted 25% (26 cases). Among the neoplastic lesions, benign lesions (53.85%) predominated over malignant lesions (21.15%). Pleomorphic adenoma was the commonest benign neoplastic lesion and mucoepidermoid carcinoma was the commonest malignant tumor. Chronic sialadenitis was the predominant lesion in the non-neoplastic group. Conclusion: The principal site for salivary gland tumors was the parotid gland and pleomorphic adenoma outnumbered all the other tumors. Females are more affected in the malignant group.

Key words: Mucoepidermoid carcinoma, pleomorphic adenoma, salivary gland

Introduction

Salivary gland tumors are relatively uncommon compared to other tumors and is said to account for less than a 2% of all human tumors.[1] Salivary gland tumors correspond to approximately 3% to 10% of neoplasms of the head and neck region.[2] Neoplasms of salivary glands though uncommon, are of particular interest to histopathologists because of their varied histological and biological characteristics.[3] The majority (70%) of salivary gland tumors arise in the parotid gland. Although, the majority of minor salivary gland tumors are malignant, three fourths of parotid tumors are benign.[4] Malignant and benign salivary gland tumors may resemble each other grossly, if seen early in their clinical course. Furthermore, the histological bland nature of most malignant salivary gland tumors such as basal cell adenocarcinoma (BCAC), low grade mucoepidermoid carcinoma, acinic cell carcinoma and others, and the aggressive biological nature of benign tumors are typified by high rate of recurrence. In view of the fact that these tumors are treated on the basis of their histological and local findings, correct histological diagnosis is obligatory.[3] The goal of this study was to retrospectively analyze 78 neoplastic and 26 non-neoplastic lesions. Manipur is a state in North Eastern India where salivary gland cancers constitute about 1.16% of all diagnosed cancers.[5] This study was conducted to reveal the pattern of various salivary gland tumors in Manipur, a state in North Eastern India.

Materials and Methods

This was a retrospective study from January 2002 to December 2011. All the salivary gland specimens received at histopathology section of pathology department, Regional Institute of Medical Sciences (RIMS) hospital were included in the study. RIMS is a major tertiary health-care teaching institute offering histopathology services to the entire state of Manipur as well as the neighbouring states. Clinical data (age, sex, and site) were obtained from the laboratory archives derived from the information provided on the histopathology request forms. All the biopsy specimens were fixed in 10% formalin-saline, then processed into paraffin embedded sections and stained with hematoxylin and eosin (H and E). Special stains (e.g., for mucin) were occasionally employed. All the histopathology slides were reviewed by the authors and classified according to the World Health Organization (WHO) histological typing of tumors 2005. Re-staining of the slides and fresh sections of tissue blocks were performed whenever required. Data collected were analyzed and deductions observed were then discussed. Ethical approval was taken for this study from institutional ethical committee.
Results

A total of 104 specimens of salivary gland tumors were received during the study period. The number of cases were slightly higher in females with 54 cases (51.92%) compared to males with 50 cases (48.08%). The male to female ratio was 1:1.08. Histological diagnosis included 26 cases (25%) of non-neoplastic lesions and 78 cases (75%) of neoplastic lesions. Among the 78 neoplastic cases, benign tumors with 56 cases (71.79%) outnumbered the malignant ones with 22 cases (28.21%). Pleomorphic adenoma was the commonest benign tumor with 43 cases (76.79%) followed by Warthin’s tumor with eight cases (14.29%). Mucoepidermoid carcinoma was the most common lesion among the malignant group with seven cases (31.82%) followed by acinic cell carcinoma with five cases (18.18%). Among the non-neoplastic lesions, chronic sialadenitis was the most common lesion.

Benign tumors were more common in males than females with M:F ratio of 1.04:1. Pleomorphic adenoma (76.79%) was the commonest benign lesion with M:F ratio of 1.36:1 followed by Warthin’s tumor (14.29%) with M:F ratio of 3:1 [Table 1]. Malignant tumors were common in females with a M:F ratio of 1:3.14. Mucoepidermoid carcinoma (31.82%) was the commonest tumor with M:F ratio of 0.75:1, acinic cell carcinoma (22.73%) which affected only females followed by adenoid cystic carcinoma (18.18%) with M:F ratio of 0.33:1 and carcinoma ex pleomorphic adenoma (9.09%) with M:F ratio of 1:1. BCAC, lymphoepithelioma like carcinoma, well differentiated squamous cell carcinoma and metastatic undifferentiated carcinoma were seen in one case each.

Table 1: Frequency of benign tumours and their sex correlation

| Benign tumour                  | No. | %     | M:F     |
|--------------------------------|-----|-------|---------|
| Pleomorphic adenoma            | 43  | 76.79 | 1.36:1  |
| Warthin tumour                 | 8   | 14.29 | 3:1     |
| Myoepithelioma                 | 3   | 5.36  | 0.5:1   |
| Adenoma                        | 1   | 1.79  | 1:1     |
| Benign lymphoproliferative lesion | 1   | 1.79  | 1:1     |
| Total                          | 56  | 100   | 1.04:1  |

Table 2: Frequency malignant tumours and their sex correlation

| Malignancy                     | Total | %     | M:F     |
|--------------------------------|-------|-------|---------|
| Mucoepidermoid carcinoma       | 7     | 31.82 | 0.75:1  |
| Acinic cell carcinoma           | 5     | 22.73 | All females |
| Adenoid cystic carcinoma       | 4     | 18.18 | 0.33:1  |
| Carcinoma ex pleomorphic adenoma | 2    | 9.09  | 1:1     |
| Basal cell adenoma carcinoma    | 1     | 4.55  | All males |
| Lymphoepithelioma like carcinoma | 1    | 4.55  | All males |
| Well differentiated squamous cell carcinoma | 1 | 4.55 | All females |
| Metastatic undifferentiated     | 1     | 4.55  | All females |
| Total                          | 22    | 100   | 1:3.14  |

Sialadenitis (84.62%) was the commonest lesion with M:F ratio of 1.2:1 followed by cyst (11.54%) with M:F ratio of 2:1 [Table 2].

Age ranged from 5 years to 78 years. Tumors were rare below the age of 20 and only seven cases were detected. The most common incidence of tumors was among the age group 41-50 years with 23 cases detected, followed by the age group 21-30 years with 17 cases. Non neoplastic conditions were common in the age group of 21-30 years with 10 cases [Table 3].

Parotid (56.65%) was the commonest gland involved followed by submandibular gland (31.73%). Both benign and malignant tumors were common in parotid with 50 cases, followed by submandibular gland with 22 cases [Table 4]. Among benign tumors, parotid (66.07%) was the commonest gland involved followed by submandibular gland (28.57%). Pleomorphic adenoma was the commonest benign tumor affecting parotid, submandibular and minor salivary gland. Warthin tumor, myoepithelioma and benign lymphoproliferative lesion affected only the parotid gland [Table 5].

Parotid (59.09%) was the commonest gland involved by malignancy followed by submandibular (27.27%) and minor salivary gland (13.64%). Mucoepidermoid carcinoma was common in parotid whereas acinic cell carcinoma was common in submandibular gland. Adenoid cystic carcinoma involved both parotid and submandibular gland equally (two cases each). All cases of carcinoma ex pleomorphic adenoma affected only the parotid. Basal cell carcinoma, lymphoepithelial like carcinoma, well differentiated carcinoma, metastatic undifferentiated...

Table 3: Relative frequency and age distribution of various salivary gland lesions

| Age     | Total | %     | Non neoplastic | Benign | Malignant |
|---------|-------|-------|----------------|--------|----------|
| ≤20     | 09    | 8.65  | 02             | 7.69   | 05       | 8.93     | 02       | 9.09     |
| 21-30   | 27    | 25.96 | 10             | 38.46  | 14       | 25.0     | 03       | 13.64    |
| 31-40   | 14    | 13.46 | 04             | 15.39  | 08       | 14.29    | 02       | 9.09     |
| 41-50   | 30    | 28.85 | 07             | 26.92  | 16       | 28.57    | 07       | 31.82    |
| 51-60   | 13    | 12.5  | 02             | 7.69   | 07       | 12.5     | 04       | 18.18    |
| >60     | 11    | 10.58 | 01             | 3.85   | 06       | 10.72    | 04       | 18.18    |
| Total   | 104   | 100   | -              | 56     | -        | 22       | -        | -        |

Table 4: Distribution of primary sites involved

| Site               | Total | %     | Nonneoplastic | Neoplastic |
|--------------------|-------|-------|---------------|------------|
| Parotid            | 61    | 58.65 | 11             | 18.03      | 37        | 60.65    | 13        | 21.32    |
| Submandibular      | 33    | 31.73 | 11             | 33.33      | 16        | 48.48    | 06        | 18.18    |
| Sub-lingual         | 01    | 0.96  | 01             | 100        | -         | -        | -         | -        |
| Minor              | 09    | 8.66  | 03             | 33.33      | 03        | 33.33    | 03        | 33.33    |
| Total              | 104   | 100   | -              | 56         | -        | 22       | -        | -        |

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carcinoma involved only the parotid gland (one case each) [Table 6].

Sialadenitis (84.62%) was the commonest non-neoplastic lesion. Sialadenitis was common in submandibular gland followed by parotid. Salivary gland cyst and mucocele were the other conditions detected in our study.

**Discussion**

Salivary gland tumors are relatively uncommon constituting about 3%-10% of all head and neck neoplasms.\(^6,7\) In the present study, we found a predominance of neoplastic (75%) over non neoplastic lesions (25%) which was also highlighted by Ma’aita et al.\(^8\) Among the 78 neoplastic lesions, benign tumors predominated over the malignant ones similar to a series of 124 cases by Vargas et al.\(^9\) in a Brazilian population. Studies by Satko et al.\(^10\) in Slovakian population and Ahmad et al.\(^11\) in Kashmir were also comparable to our study. They also showed the highest incidence of benign tumors in 3\(^{rd}\) to 4\(^{th}\) decade and malignant tumors in 4\(^{th}\) to 5\(^{th}\) decades of life consistent with our study. The mean age for benign and malignant tumors was 40.2 years and 46.86 years respectively in our cases. However, the age profile in African studies was much younger than the Western Literatures.\(^12\)

According to the variable “gender”, most of the studies reviewed show a female predominance similar to our findings.\(^13\) Male to female ratio for benign, malignant and non-neoplastic lesions were 1.04:1, 1:3.14, and 1.73:1 respectively in contrast to more male predominance of salivary gland tumors as described by Frade Gonzalez et al.\(^6\) However, we found a slight male predominance in benign lesions. Studies citing a male predominance among malignant lesions are also reported in contrast to our study.\(^17\) This may be due to the regional differences or the size of the samples used in each study.

The vast majority of our salivary gland lesions occurred in major salivary glands (91.34%), especially, in the parotid gland (58.65%) followed by submandibular gland (31.73%). Among benign tumors, parotid (66.07%) was the commonest gland affected followed by submandibular gland (28.57%). In malignant tumors, again the parotid (59.09%) was the gland most affected followed by submandibular (27.27%) and minor salivary gland (13.64%). Lopes et al.\(^19\) reported a higher prevalence of malignant tumors in minor salivary glands. In our study, we noted three cases each of benign and malignant tumors of minor salivary glands of the total six cases suggesting equal incidence as described by Vargas et al.\(^9\)

In non-neoplastic lesions, parotid and submandibular gland were equally involved (42.3%) followed by minor salivary glands (11.6%). No case of salivary gland tumor was noted in the sublingual gland in our study. This result demonstrates the low prevalence of sublingual tumors, as has been reported by Eveson et al.,\(^16\) who observed 1 sublingual tumor to 100 parotid tumors.

Pleomorphic adenoma (76.79%) was the commonest lesion in benign tumors as observed by Li et al.\(^16\) Warthin tumor (14.29%) was the 2\(^{nd}\) commonest benign tumors as quoted by Chung et al.\(^20\) Out of 43 cases of pleomorphic adenomas, 25 involved the parotid, 16 cases involved the submandibular gland and two cases involved the minor salivary glands. All eight cases of Warthin tumor involved only the parotid gland, which is similar to what has been reported by Eveson et al.\(^21\)

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**Table 5: Benign tumours and site correlation**

| Benign     | Parotid | Submandibular | Sub-lingual | Minor |
|------------|---------|---------------|-------------|-------|
| Pleomorphic adenoma | 25      | -             | 16          | -     |
| Warthin tumour       | 8       | -             | -           | -     |
| Myoepithelioma       | 3       | -             | -           | -     |
| Adenoma              | -       | -             | -           | -     |
| Benign lymphoproliferative | 1 | -             | -           | -     |
| Total                | 37      | 66.07%        | 28.57%      | 5.4%  |

**Table 6: Malignant tumours and site correlation**

| Malignancy                  | Parotid | Submandibular | Sublingual | Minor |
|-----------------------------|---------|---------------|------------|-------|
| Mucoepidermoid carcinoma    | 4       | 1             | -          | 2     |
| Acinic cell carcinoma       | 1       | 3             | -          | 1     |
| Adenoid cystic carcinoma    | 2       | 2             | -          | -     |
| Carcinoma ex pleomorphic adenoma | 2 | -             | -          | -     |
| Basal cell adenoma carcinoma| 1       | -             | -          | -     |
| Lymphoepithelioma like carcinoma | 1 | -             | -          | -     |
| Well differentiated squamous carcinoma | 1 | -             | -          | -     |
| Metastatic undifferentiated carcinoma | 1 | -             | -          | -     |
| Total                       | 13      | 59.09%        | 27.27%     | 13.64%|
Out of 22 cases of malignant lesions, mucoepidermoid carcinoma affected seven cases, acinic cell carcinoma in five cases and adenoid cystic carcinoma in four cases. Pinkston et al. in their series noted mucoepidermoid carcinomas to be more common in parotids, similar to our study (four out of seven cases). [22] Primary squamous cell carcinoma of salivary gland is rare. Batsakis et al. reported the true incidence to be 0.3% to 1.5%. [21] We found only one case of squamous cell carcinoma in the parotid gland in our series.

Among 26 cases of non-neoplastic lesions, salialdenitis was noted in 22 cases followed by benign cyst in three cases. Sialadenitis commonly involved submandibular gland followed by parotid and minor salivary gland.

To conclude, neoplastic lesions of the salivary gland are not common. The principal site for salivary gland tumors in Manipur is the parotid gland and pleomorphic adenoma outnumbered all other tumors. Larger studies to determine the prevalence is the need of the hour.

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