International Surgery Journal
Gajbhiye AS et al. Int Surg J. 2018 Feb;5(2):657-661
http://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902

DOI: http://dx.doi.org/10.18203/2349-2902.isj20180371

Original Research Article

Pleomorphic adenoma of submandibular gland: not so common occurrence

Ashok S. Gajbhiye, Mukteshwar N. Deshmukh, Parag K. Jaipuriya*, Kishor B. Jeughale, Abhinav A. Kumar, Abhijeet P. Ghajbhare, Ankur A. Agrawal

Department of General Surgery, Indira Gandhi Government Medical College, Nagpur, Maharashtra, India

Received: 12 December 2017
Accepted: 03 January 2018

***Correspondence:**
Dr. Parag Jaipuriya,
E-mail: paragjaipuriya@gmail.com

ABSTRACT

Background: The salivary glands tumour are relatively rare, yet they represent a wide variety of both benign and malignant histologic subtypes. Several studies have been conducted on the tumors of the parotid and minor salivary glands, but very few reports in the literature have focused on submandibular gland tumors as they are rare and are usually grouped with other salivary glands. Approximately 70% of the salivary gland tumors affect parotid gland with the submandibular gland being affected in 5-10% of the cases, sublingual gland in 1% and minor glands in 5-15% of the cases. So, author describe a series of 50 patients of pleomorphic adenoma affecting submandibular gland.
Methods: The prospective study was conducted at Department of General Surgery, Indira Gandhi Government Medical College, Nagpur between January 2008 to December 2017.
Results: Out of the 50 patients, 10 were male and 40 were female. Thus, male to female ratio is 1:4. Most of the 42 (84%) patients presented with painless swelling in submandibular region. Clinically the tumour has the texture of cartilage and has an irregular and bosselated surface observed in all the patients. The size of tumour was 3 to 6 cm in 32 (64%) patients observed in the present study. Fine needle aspiration cytology was performed in all the patients. All patients were managed by surgical excision. Recurrence occurred in a single patient who was managed with radical excision including a neck dissection with postoperative radiotherapy.
Conclusions: Submandibular gland pleomorphic adenoma being common benign tumor, occurring commonly between the 3rd and 5th decade of life and presenting as slow growing asymptomatic swelling with female preponderance. The precise surgical excision along with whole submandibular salivary gland and postoperative radical radiotherapy required for recurrence as the accepted mode of management. However, further long-term studies involving submandibular gland have to be carried out to know the pattern of tumor recurrence.

Keywords: Excision, Pleomorphic adenoma, Submandibular gland, Salivary gland tumors

INTRODUCTION

Salivary gland tumors are uncommon and comprise only 1-4% of head-face-neck tumors. Majority of the salivary gland tumors affect parotid gland with more than 70% of the cases. Several studies have been conducted on the tumors of the parotid and minor salivary glands, but very few reports in the literature have focused on submandibular gland tumors as they are rare and are usually grouped with other salivary glands. Submandibular gland is affected in 5-10% of the cases with pleomorphic adenoma being the most common tumor.[1]

The most frequent neoplasms in the submandibular glands are: pleomorphic adenoma (36%), adenoid cystic

carcinoma (25%), mucoepidermoid carcinoma (12%) and malignant mixed tumor (10%). Clinical reports indicate that benign neoplasms are characterized by mostly painless enlargement of the submandibular triangle.[2] Becerril-Ramírez et al, in their 10-years study found a total of 22 cases of submandibular gland neoplasms, in which 19 cases (86%) were benign and 3 cases (14%) were malignant.[3] Munir and Bradley reviewed series of the pleomorphic adenoma affecting submandibular gland over a period of 16 years and reported that all patients presented with clinically visible and palpable mass of submandibular fossa among which 84% of cases were asymptomatic and 16% presented with pain.[4] Rapidis et al, analyzed clinicopathologic features of 23 patients with submandibular gland tumors, in which nine were benign and 14 were malignant tumors. They found that pleomorphic adenoma was the most frequent benign tumor and manifest a mild course of disease.[5]

Adeyemo et al reviewed a total of 36 patients submandibular salivary gland tumors over a 17 years, out of which 17 cases were benign and 19 cases were malignant. pleomorphic adenoma (36.1%) was the most frequent tumor, followed by adenoid cystic carcinoma (11.1%), anaplastic carcinoma (11.1%) and malignant lymphoma (11.1%).

Authors found progressive painless swelling (80.6%) was the most common mode of presentation and cases which presented with painful mass (11.1%) and ulceration (8.3%) were malignant.[6] In a Brazilian population, de Oliveira et al. found that the salivary gland tumors affect females more often, with a male: female ratio of 1:1.5. This ratio is reported as 1:1.6 in benign tumors and 1:1.5 in malignant tumors. The mean age for benign tumors was 43 years and for malignant tumors was 55 years.[7]

Alves et al, reviewed clinicopathological and immunohistochemical features of 60 cases of pleomorphic adenoma in Brazil and found that pleomorphic adenoma occurs commonly between 3 and 5 decades of life and 37/60 (62%) of them were women. Tumor sizes varied from 1 to 10cm. Only one patient experienced local recurrence, 3 years after treatment.[8] Fine needle aspiration findings provide evidence for a pre-operative diagnosis that is 70-80% accurate and also helps to differentiate between tumor and inflammatory conditions or enlarged lymph nodes. The final pathologic diagnosis is always established based on findings from surgical excision. The treatment of choice for submandibular gland is pleomorphic adenoma total submandibular gland excision along with tumor.[9]

Recurrence rate of submandibular gland tumors are less than parotid gland since entire gland is excised. Injury to the marginal mandibular nerve is the most common complication leading to temporary or permanent paralysis due to the stretching or compression of the nerve. Temporary paralysis may resolve spontaneously within a period of 3 months.[10] So, author describe a series of 50 patients of pleomorphic adenoma affecting submandibular gland.

METHODS

A prospective study was conducted in the department of surgery in a medical college and hospital in central India over a period from January 2008 to December 2017. A total of 50 patients of pleomorphic adenoma of submandibular gland who suspected clinically and confirm on histopathological examination were included in this study.

Figure 1: Pleomorphic adenoma rt. s/b gland.

Figure 2: Submandibular duct.

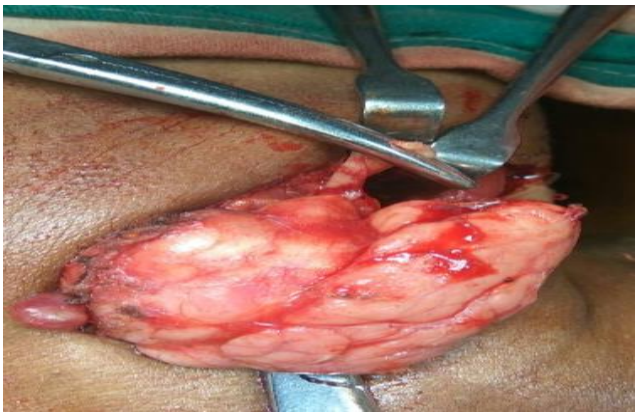

Figure 3: Submandibular gland excision.

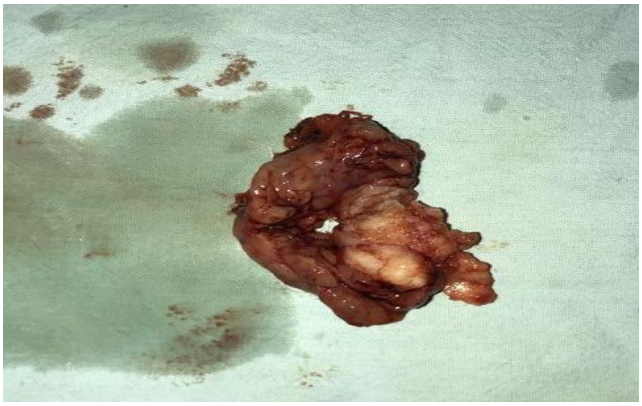

Figure 4: Pleomorphic adenoma seen in excised specimen.

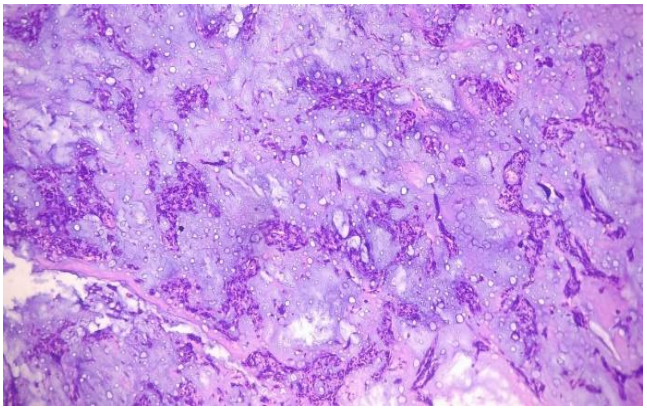

Figure 5: Histopathological slide of PA of submandibular gland.

Adenoid cystic carcinoma, mucoepidermoid carcinoma, anaplastic carcinoma, malignant lymphoma and malignant mixed tumor of submandibular salivary gland were excluded from the study. A detailed clinical history and examination of submandibular gland lump was done. Patients were subjected to fine needle aspiration cytology to confirm the diagnosis. Computed tomography and MRI were advice for exact localization of tumour. Postoperative complications and histopathologic results were evaluated prospectively. Patients who underwent surgery were followed up for any recurrence postoperatively. The profile photograph of pleomorphic adenoma of submandibular gland (Figure 1,2,3,4).

RESULTS

In the time period between 2008 and 2017, 50 patients were presented in the institute with clinical, radiological or histopathological diagnosis of pleomorphic adenoma of submandibular salivary gland. All of them were operated and confirmed on the histological examination. Majority of patients were in the age group 41-50 years (Table 1). Mean age of presentation was 37 years ranging from 12 years to 57 years. No patients were seen below ten years and above sixty years in the present study.

Table 1: Age wise distribution.

Age(in years)	No. of patients 50(100%)
11- 20	4(8%)
21- 30	10(20%)
31- 40	13(26%)
41- 50	15(30%)
50-60	8(16%)
Total	50(100%)

Out of the 50 patients, 10 were male and 40 were female. Thus, male to female ratio is 1:4 (Table 2).

Table 2: Sex distribution.

Sex	No. of patients 50 (100%)
Male	10(20%)
Female	40(80%)
M:F ratio	1:4

Maximum number of 30(60%) patients presented within five months. 30%,6%, and 4% patients presented between 5 to 10, 11 to 15 and more than 15 months respectively (Table 3).

Table 3: Duration of presentation.

Duration(in months)	No of patients 50 (100%)
<5 months	30(60 %)
5 -10 months	15(30%)
11-15 months	3(6%)
>15 months	2(4%)

Swelling was painless in 84% of patients while painful swelling observed in 7% of patients in submandibular region. Swelling with ulceration of skin seen in a single patient (Table 4). Right side was the comments site of swelling observed in 82% of the patients in the present study. Clinically the tumour has the texture of cartilage and has an irregular and bosselated surface observed in all the patients.

Table 4: Clinical presentations.

Clinical features	No of patient 50 (100%)
Painless swelling in submandibular region	42(84%)
Painful swelling in submandibular region	7(14%)
Swelling with ulceration in submandibular region	1(2%)

Table 5: Size of the tumor.

Size (cm)	No of patients 50 (100%)
<3cm	13(26%)
3- 6cm	32(64%)
>6cm	5(10%)

Out of 50 patients, 64 % of patients had swelling size in range of 3-6 cm. 13 patient had less than 3 cms and 5 patients had swelling size more than 6 cms (Table 5).

In the present study, fine needle aspiration cytology was positive in 42 patients (84%). Gold standard was histopathology after surgical excision of the mass. All patients underwent surgical excision of the mass along with whole submandibular gland. Patients were followed up and seen one recurrence after two years postoperatively. Recurrence patient was managed by radial surgical excision with radiotherapy. Complications like neuropraxia/paresis developed in marginal mandibular branch of facial nerve in 5 (10%) patients within first 24 hours postoperatively. It was completely resolved within 6 months. In addition, hematoma was developed in operative site of one patent and it was managed conservatively. Postoperative wound infection was seen in 3 (6%) patient.

DISCUSSION

Salivary gland tumors comprise only 1-4% of head and neck tumors and most commonly affect parotid gland. Very few reports in the literature have focused on submandibular gland neoplasms as they are rare and are usually grouped with other salivary glands.[1] The most frequent neoplasms in the submandibular glands are: pleomorphic adenoma (36%), adenoid cystic carcinoma (25%), mucoepidermoid carcinoma (12%) and malignant mixed tumor (10%). Clinical reports indicate that benign neoplasms are characterized by mostly painless enlargement of the submandibular triangle.[2] Becerril-Ramírez et al, in their 10-years study found a total of 22 cases of submandibular gland neoplasms, in which 19 cases (86%) were benign and 3 cases (14%) were malignant. The most common benign neoplasm was PA which accounted for 18 out of 19 cases. The mean age of occurrence of PA was 39.8 years with female to male ratio of 3.5:1.[3] In present study also, the mean age was 40.2 years with male to female ratio of 1:4 which showed female preponderance.

Munir and Bradley reviewed series of the pleomorphic adenoma affecting submandibular gland over a period of 16 years from 1988 to 2004. A total of 32 cases of submandibular gland PA were treated between the period among which 22 out of 32 (69%) cases were female and the mean age of occurrence of PA was 54 years. All patients presented with clinically visible and palpable mass of submandibular fossa among which 84% of cases were asymptomatic and 16% presented with pain.[4] In present study also most of the patients presented with painless submandibular swelling 42 (84%) cases and 7 (14%) with painful swelling. Clinically the tumour has the texture of cartilage and has an irregular and bosselated surface observed in all the patients.

Rapidis et al, analyzed clinicopathologic features of 23 patients with submandibular gland tumors, in which nine were benign and 14 were malignant tumors. They found that PA was the most frequent benign tumor and manifest a mild course of disease.[5] Submandibular salivary gland tumors over a 17 years period from 1990 to 2006. A total of 36 patients with submandibular gland tumors were reviewed among which 17 cases were benign and 19 cases were malignant. PA (36.1%) was the most frequent tumor, followed by adenoid cystic carcinoma (11.1%), anaplastic carcinoma (11.1%) and malignant lymphoma (11.1%). Authors found progressive painless swelling (80.6%) was the most common mode of presentation and cases which presented with painful mass (11.1%) and ulceration (8.3%) were malignant.[6] In present study also most of the patients presented with painless submandibular swelling 42 (84%) cases and 7(14%) cases with painful swelling.

In a Brazilian population, de Oliveira et al, found that the salivary gland tumors affect females more often, with a male: female ratio of 1:1.5. This ratio is reported as 1:1.6 in benign tumors and 1:1.5 in malignant tumors. The mean age for benign tumors was 43 years and for malignant tumors was 55 years.[7]

Alves et al, reviewed clinicopathological and immunohistochemical features of 60 cases of PA in Brazil and found that PA occurred commonly between 3 and 5 decades of life and 37/60 (62%) of them were women. Tumor sizes varied from 1 to 10cm. Only one patient experienced local recurrence, 3 years after treatment.[8] In the present study also most of the cases occurred between 3 to 5th decades with tumour size mostly between 3-6 cm in 32 cases (64%).

Fine needle aspiration findings provide evidence for a pre-operative diagnosis that is 70-80% accurate and also helps to differentiate between tumor and inflammatory conditions or enlarged lymph nodes. The final pathologic diagnosis is always established based on findings from surgical excision. The treatment of choice for submandibular gland PA is total submandibular gland excision along with tumor.[9] In present study also FNAC was diagnostic in 42 (84%) cases with treatment of choice being surgical excision.

Recurrence rate of submandibular gland tumors are less than parotid gland since entire gland is excised. Injury to the marginal mandibular nerve is the most common complication leading to temporary or permanent paralysis due to the stretching or compression of the nerve. Temporary paralysis may resolve spontaneously within a period of 3 months.[10]

In the present study, complications like neuropraxia/paresis developed in marginal mandibular branch of facial nerve in 5 (10%) patients within first 24 hours postoperatively and while it completely resolved within 3- 6 month. Other complications which developed were hematoma (2%) and surgical site infection (6%) cases.

CONCLUSION

Pleomorphic adenoma is most common benign tumor affecting submandibular gland, occurring commonly between the 3rd and 5th decade of life and presenting as slow growing asymptomatic swelling with female preponderance. The precise surgical excision along with whole submandibular salivary gland and postoperative radical radiotherapy required for recurrence as the accepted mode of management. However, further long-term studies involving submandibular gland have to be carried out to know the pattern of tumor recurrence.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

REFERENCES

1. Spiro RH. Salivary neoplasms: Overview of a 35-year experience with 2,807 patients. Head Neck Surg. 1986;8:177-84.
2. Hanna E, Lee S, Fan C, Suen J. Benign neoplasms of the salivary glands. In: Cummings C, Flint P, Harker L, Haughey B, Richardson M, Robbins T. editors. Cummings Otolaryngology Head and Neck Surgery. Philadelphia: Mosby; 2005:1348-1377.
3. Becerril-Ramírez PB, Bravo-Escobar GA, Prado-Calleros HM, Castillo-Ventura BB, Pombo-Nava A. Histology of submandibular gland tumours, 10 years' experience. Acta Otorrinolaringol Esp. 2011;62:432-5.
4. Munir N, Bradley PJ. Pleomorphic adenoma of the submandibular gland: An evolving change in practice following review of a personal case series. Eur Arch Otorhinolaryngol. 2007;264:1447-52.
5. Rapidis AD, Stavrianos S, Lagogiannis G, Faratzis G. Tumors of the submandibular gland: Clinicopathologic analysis of 23 patients. J Oral Maxillofac Surg. 2004;62:1203-8.
6. Adeyemo WL, Ajayi OF, Anunobi CC, Ogunlewe MO, Ladeinde AL, Omitola OG, et al. Tumours of the submandibular salivary gland: A clinicopathologic review of cases over a 17-year period. West Indian Med J. 2009;58:388-91.
7. de Oliveira FA, Duarte EC, Taveira CT, Máximo AA, de Aquino EC, Alencar Rde C, et al. Salivary gland tumor: A review of 599 cases in a Brazilian population. Head Neck Pathol. 2009;3:271-5.
8. Alves FA, Perez DE, Almeida OP, Lopes MA, Kowalski LP. Pleomorphic adenoma of the submandibular gland: Clinicopathological and immunohistochemical features of 60 cases in Brazil. Arch Otolaryngol Head Neck Surg. 2002;128:1400-3.
9. Eveson JW, Cawson RA. Salivary gland tumours. A review of 2410 cases with particular reference to histological types, site, age and sex distribution. J Pathol. 1985;146:51-8.
10. Hong KH, Yang YS. Surgical results of the intraoral removal of the submandibular gland otolaryngol Head Neck Surg. 2008;139:530-4.

Cite this article as: Gajbhiye AS, Deshmukh MN, Jaipuriya PK, Jeughale KB, Kumar AA, Ghajbhare AP, et al. Pleomorphic adenoma of submandibular gland: not so common occurrence. Int Surg J 2018;5:657-61.