



Original article

Retrospective Clinicopathological Study of 33 Cases of Pleomorphic Salivary Adenoma Diagnosed in Benghazi

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ABSTRACT

Background: Pleomorphic salivary adenoma (PSA) is the most frequently found neoplastic tumor in major and minor salivary glands. It has a high recurrence rate and there is a possibility for benign PSA to transform into malignant form. No study on clinical and histopathological features of pleomorphic adenoma in Libya was found. occurrence of benign PSA is more common than PSA with malignant foci.

Aim: The present study aimed to retrospectively analyze the clinical and histopathological features of cases diagnosed as pleomorphic adenoma and compare our findings to those of other studies.

Materials and Methods: In this retrospective study, the pleomorphic salivary adenoma cases diagnosed between 1994-2019 in the Department of Oral Pathology, Dental School, University of Benghazi, Libya were retrieved. The diagnosis was confirmed by reevaluation of hematoxylin and eosin-stained slides. Descriptive statistics were used to describe the clinical data of the patient's demographics and clinical features of the tumor.

Results: It was found that among 33 diagnosed cases of PSA, 11 cases were males and 22 cases were females. The most common age range was 10-29 years (21 cases). The most common intra-oral site was the palate (15 cases) followed by buccal mucosa (6 cases) and submandibular glands and parotid glands were most commonly affected among major salivary glands. Benign pleomorphic adenoma (24 cases) was most common, myoepithelium was reported in 2 cases and malignant foci were reported in 7 cases.

Conclusion: This study revealed that Females were more affected than males. The most common location of pleomorphic adenoma was the palate. The myoepithelioma is a rare.

Keywords: *Pleomorphic Salivary Adenoma, Retrospective, Clinicopathological, Libya.*

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INTRODUCTION

Pleomorphic adenoma, the most common salivary gland tumor, constituting up to two-thirds of all salivary gland tumors. It is also referred to as benign mixed tumors, because of its twofold origin from both epithelial and myoepithelial elements.^{1,2,3} It has a natural history of slow growth over a long period of time. The cause of pleomorphic adenoma is not well understood. However, the incidence of this tumor has been rising over the past 15-20 years, particularly in relation to radiation exposure. One study proposes that the oncogenic simian virus (SV40) might be involved in the onset or

progression of pleomorphic adenoma. Additionally, previous head and neck irradiation is considered a risk factor for the development of these tumors.⁴ It makes considerable interest for its relatively high recurrence rate (4% and 45%)^{5,8} and its potential for malignant transformation (2% to 24%).^{9,10}

Pleomorphic adenoma is diagnosed by its histopathological features. It consists of two kinds of cells, i.e., epithelial and myoepithelial cells and contains a variable stroma that can be of the hyaline, myxoid, chondroid or fibrous type. The epithelial cells are organized into various formations such as nests, chains, sheets or duct-like structures within these different types of stroma.^{7,10-13} No study on clinicopathological features of pleomorphic adenoma in Libya was found in the medical literature. The aim of the present study therefore was to analyze retrospectively the clinicopathological features of pleomorphic salivary adenoma such as age and gender of the patient, site of

the tumor, and its histopathological subtypes in Libyan patients and to compare our findings with the results of similar studies from other parts of the world.

MATERIALS AND METHODS

In this retrospective study, the pleomorphic salivary adenoma cases were diagnosed between 1994-2019 in the Department of Oral Pathology, Faculty of Dentistry, University of Benghazi. The diagnosis was confirmed by reevaluation of hematoxylin and eosin-stained slides, the pleomorphic adenomas of either major or minor salivary glands were predominantly well-circumscribed and were defined by epithelial cells organized in nests, sheets, and duct-like structures. The cell types included either epithelial or myoepithelial cells, with the myoepithelial cells displaying a plasmacytoid or spindle-shaped morphology. The stroma was either fibrous or myxo-chondroid. (Figure 1).

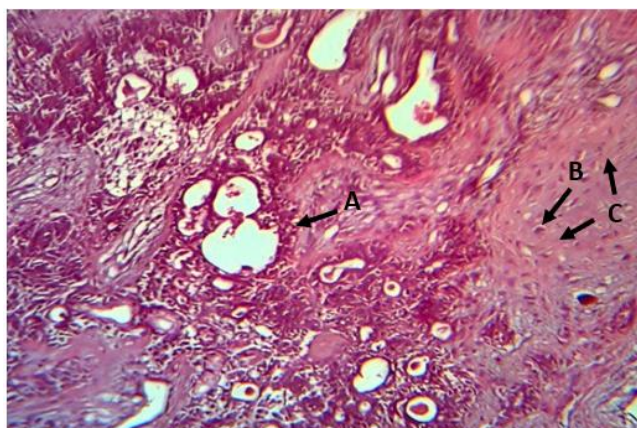


Figure 1: A photomicrograph of H&E stained section of pleomorphic adenoma showing neoplastic epithelial cells arranged in duct-like structure (A) and islands and also showing myoepithelial cells (B) surrounded by myxochondromatous stroma (C) (Magnification x 100).

Descriptive statistics were employed to outline patient demographics, including age at tumor diagnosis and gender, as well as clinical features of the tumor, such as site, recurrences, and histopathological types.

RESULTS

This study comprises 33 cases of pleomorphic adenoma with 11 cases occurring in men, and 22 cases occurring in women (M:F=1:2). Pleomorphic salivary adenoma occurred in patients over a wide age range (8-70years), most commonly in patients age group range from 10-29 years (Table 1). The mean patient age at the time of diagnosis was 28.5 years.

Table 1. Distribution of PSA according to age and gender

Age	Male	Female	Total
0-9	0	2	2
10-19	4	9	13
20-29	4	4	8
30-39	1	2	3
40-49	1	1	2
50-59	1	1	2
60-69	0	2	2
70-79	0	1	1
Total	11	22	33

Intra-orally, 15 cases occurred in the palate, followed by buccal mucosa (6 cases). Extra-orally, parotid glands (5 cases) are most commonly affected among major salivary glands followed by submandibular glands (3 cases) (table 2). The clinical symptoms and signs at diagnosis include swelling (32 cases), and pain, (one case), (Table 3).

Table 2. Distribution of PSA according to site

	Site	Number of cases
Major	Parotid	5
	Submandibular	3
	Sublingual	0
Minor	Palatal	15
	Buccal	6
	Labial	3
	Tongue	0
	Floor of mouth	1
	Total	33

Histologically, among the 33 cases of PSA, 2 cases (6.1%) were of myoepithelioma subtype and 7 cases (21.2%) showed malignant foci (table 4).

Table 3. Presenting symptoms of PSA

Symptom	Number of cases
Swelling	32
Pain and tenderness	1
Total	33

Table 4. Histological subtypes of PSA

	PSA	Myoepithe lioma	PSA with malignant foci	Total
Number of cases	24	2	7	33

DISCUSSION:

A pleomorphic salivary adenoma is the most common neoplasm arising from salivary gland tissue, accounting for about 81.2% of all salivary tumors.¹⁴ Clinically, PSA usually presents as a slow-growing, painless mass and has a relatively long course.¹⁵ To the authors' best of knowledge, despite the small number of cases, this is the first study to describe the PSA in a Libyan population. In this study several interesting findings were reported, some of them were in agreement with previous studies conducted elsewhere and other findings were surprisingly different. In the present study, females (22 cases) were more affected by PSA than males (11 cases), This finding is similar to the results of other studies such as that of Irani et al.¹⁶

In the present study, PSA is more prevalent in 10-29 years of age group; this finding is contrary to many studies which found PSA common in the 4th-5th decades of life.¹⁶ It is unclear why but this could have something to do with environmental risk factors specific to our country. However, further research is needed to understand why younger individuals are more likely to be affected by PSA.

Another surprising finding in the present study was that the palatal salivary tissue is the most common site for occurrence of PSA {15 cases representing (45.5%)}. In many studies, the parotid gland was reported to be the most common site for pleomorphic adenomas, followed by the submandibular gland.^{14,17}

It is difficult to explain this finding and further research is required to understand this phenomenon.

The present study has some limitations which should be considered before making conclusions. First, the study is retrospective in nature and hence the authors have little influence over the data collection process. For example, no additional information was collected about the risk factors and hence limited the ability to explain results. Given the constraints of the available data and the small sample size, our findings should be interpreted with caution. Nevertheless, the study provided new insights into the distribution of PSA among Libyans and can serve as a baseline for future studies.

CONCLUSIONS:

In conclusion, our study has found that females were more affected than males. The most common location of pleomorphic adenoma was the palate. The myoepithelioma subtype is a rare occurrence and benign PSA is more common than PSA with malignant foci.

REFERENCES

1. Mc Loughlin L, Gillanders SL, Smith S, Young O. The role of adjuvant radiotherapy in management of recurrent pleomorphic adenoma of the parotid gland: a systematic review. *Eur Arch Otorhinolaryngol.* 2019 Feb;276(2):283-295.
2. Lee JH, Kang HJ, Yoo CW, Park WS, Ryu JS, Jung YS, Choi SW, Park JY, Han N. PLAG1, SOX10, and Myb Expression in Benign and Malignant Salivary Gland Neoplasms. *J Pathol Transl Med.* 2019 Jan;53(1):23-30.
3. Meshram GG, Kaur N, Hura KS. Pediatric pleomorphic adenoma of the parotid: Case report, review of literature and novel therapeutic targets. *Children (Basel).* 2018 Sep 18;5(9):e172
4. Gündüz AK, Yeşiltaş YS, Shields CL. Overview of benign and malignant lacrimal gland tumors. *Curr Opin Ophthalmol.* 2018 Sep;29(5):458-468.
5. Renehan A, Gleave EN, McGurk M. An analysis of the treatment of 114 patients with recurrent pleomorphic adenomas of the parotid gland. *Am J Surg.* 1996;172:710-14.
6. Donovan DT, Conley JJ. Capsular significance in parotid tumor surgery: reality and myths of lateral lobectomy. *Laryngoscope.* 1984;94:324-9.
7. Naeim F, Forsberg MI, Waisman J, et al. Mixed tumors of the salivary glands: growth pattern and recurrence. *Arch Pathol Lab Med.* 1976;100:271-5.
8. Krolls SO, Boyers RC. Mixed tumors of salivary glands: long-term follow-up. *Cancer.* 1972;30:276-81.
9. Gunn A, Parrott NR. Parotid tumours: a review of parotid tumour surgery in the Northern Regional Health Authority of the United Kingdom 1978–1982. *Br J Surg.* 1988;75:1144-6
10. Phillips PP, Olsen KD. Recurrent pleomorphic adenoma of the parotid gland: report of 126 cases and a review of the literature. *Ann Otol Rhinol Laryngol.* 1995;104:100-4.
11. Dardick I. Salivary gland tumor pathology: text and color atlas. New York, Tokyo: Igaku-Shoin Medical Publishers; 1996

12. Ellis GL, Auclair PL. Tumors of salivary glands: atlas of tumor pathology. Washington DC: Armed Forces Institute of Pathology; 1996.
13. Neville BW, Damm DD, Allen CM, Bouquot JE. Oral and maxillofacial pathology. Philadelphia: W.B. Saunders; 2002.
14. Sharma S, Mehendiratta M, Chaudhary N, Gupta V, Kohli M, Arora A. Squamous metaplasia in pleomorphic adenoma: a diagnostic and prognostic enigma. *J Pathol Transl Med*. 2018;52(6):411-5.
15. Lotufo MA, Junior CA, Mattos JP, Franca CM. Pleomorphic adenoma of the upper lip in a child. *J Oral Sci*. 2008;50:225-8.
16. Irani S, Dehghan A, Kalvandi Z. Correlation of clinical and histopathological features of salivary pleomorphic adenoma. *J Dent Shiraz Univ Med Sci*. 2023;24(4):404-9.
17. Friedrich RE, Li L, Knop J, Giese M, Schmelzle R. Pleomorphic adenoma of the salivary glands: analysis of 94 patients. *Anticancer Res*. 2005;25(3A):1703-5.