

Pleomorphic adenoma of upper lip: An unusual presentation

Gowri Balakrishnana¹, A. Shanmugam², Josna Thankachan³, Dr Jeevanandam⁴,
Karthik Shunmugavelu⁵, Evangeline Cynthia⁶, RN Mugundan⁷

Mercy Multispeciality Dental Centre, Chrompet, Chennai, Tamilnadu, India, Malar Fortis hospital, Vadapalani, Chennai, India, Dentistry/Oral and Maxillofacial Pathology, Mercy Multispeciality Dental Centre, Chrompet, Chennai, Tamilnadu, India, Mahatma Gandhi Post graduate Institute of dental sciences, Puducherry, India

Πλειόμορφο αδένωμα του άνω χείλους: μια ασυνήθιστη εντόπιση

Gowri Balakrishnana¹, A. Shanmugam², Josna Thankachan³, Dr Jeevanandam⁴,
Karthik Shunmugavelu⁵, Evangeline Cynthia⁶, RN Mugundan⁷

Mercy Multispeciality Dental Centre, Chrompet, Chennai, Tamilnadu, India, Malar Fortis hospital, Vadapalani, Chennai, India, Dentistry/Oral and Maxillofacial Pathology, Mercy Multispeciality Dental Centre, Chrompet, Chennai, Tamilnadu, India, Mahatma Gandhi Post graduate Institute of dental sciences, Puducherry, India

Case Report
Αναφορά περιστατικού

SUMMARY: Pleomorphic adenoma is the most common benign salivary gland tumor. Pleomorphic adenoma of lip is a rare presentation, only 10 % of pleomorphic adenoma occurs in minor salivary gland. The present report describes about the case of 55 year old male patient with asymptomatic oval shaped, solitary and lobulated swelling in upper lip. Complete excision of the lesion was done. The Histopathological examination revealed Pleomorphic adenoma. The patient was followed up for 6 months. No evidence of recurrence was seen.

KEY WORDS: Salivary gland tumor, lip, lobulated swelling.

ΠΕΡΙΛΗΨΗ: Το πλειόμορφο αδένωμα είναι ο πιο κοινός καλοήθης όγκος των σιελογόνων αδένων. Το πλειόμορφο αδένωμα του χείλους είναι μια λιγότερο συχνή εντόπιση, μόνο το 10% των πλειομόρφων αδενωμάτων εμφανίζεται σε ελάσσονες σιελογόνους αδένες. Η παρούσα αναφορά περιγράφει την περίπτωση ενός άνδρα 55 ετών με ασυμπτωματικό περιγεγραμμένη, μονήρη και λοβωτή διόγκωση στο άνω χείλος. Έγινε πλήρης εκτομή της βλάβης. Η ιστοπαθολογική εξέταση έδειξε πλειόμορφο αδένωμα. Ο ασθενής παρακολούθηθηκε για 6 μήνες. Δεν παρατηρήθηκαν στοιχεία υποτροπής.

ΛΕΞΕΙΣ ΚΛΕΙΔΙΑ: Όγκος σιελογόνων αδένων, χείλος, λοβωτό οίδημα.

¹ Consultant – Dentistry
² MDS (OMFS), FIOO (Fellow in Oral Oncology).
Consultant-Oral and Maxillofacial Surgeon
³ Consultant – Dentistry/Oral and Maxillofacial Pathology
⁴ MDS (Prosthodontics) Professor
⁵ BDS, MSC (London), MDS OMFP, MFDS RCS England, MCIP, FIBMS (USA), MASID (Australia) Consultant – Dentistry/Oral and Maxillofacial Pathology
⁶ Consultant – General Pathology
⁷ BDS MDS OMFP, Consultant – Dentistry/Oral and Maxillofacial Pathology

INTRODUCTION

Pleomorphic adenoma the most common salivary neoplasm accounts for more than 70% of benign epithelial tumors. These well-circumscribed tumors are derived from a mixture of myoepithelial and ductal elements and are found in both the major and minor salivary glands with most occurring in the parotid gland and minor salivary glands of palate (1). It occurs in all ages but most commonly occurs in 2nd to 3rd decade of life and occurs most commonly in female than in male (2). The clinical presentation of pleomorphic adenoma is asymptomatic and slow growing mass. Small tumors are smooth, mobile, firm lumps but larger tumors tend to become lobulated and may attenuate the overlying skin or mucosa (3).

CASE REPORT

A 55 year old male patient came with a chief complaint of painless swelling in the upper lip region, which was present for the past 2 years and swelling gradually got increased to the present size. The patient had no pain. The patient's medical history revealed that the patient is diabetic for the past 6 years and taking medication for the same. Patient had no history of injury on the site of swelling.

Intraoral examination reveals 4 × 3 cm swelling present on the left upper lip which was well defined, oval shaped, solitary, lobulated extending superior-inferiorly from the left anterior labial sulcus to left upper vermilion border of the lip and medio-laterally 0.5cm from the labial frenum to the left maxillary second premolar region (Fig. 1). On palpation it was tender free, firm in consistency and freely movable. Extra-orally no abnormality detected. FNAC was done and it was suggestive of Pleomorphic adenoma.

Complete excision of the lesion was done and it was delimited by bony walls without any communication to the maxillary antrum. It was sent to histopathological examination. The excised lesion was oval, multinodular and encapsulated, measuring 3.5 cm in diameter (Fig. 2). The histopathological features revealed plasmacytoid myoepithelial cells which were in multiple strands and clusters with eosinophilic cytoplasm and the nuclei was round to irregular in shape which was hyperchromatic. Many parts of the specimen revealed prominent ductal differentiation. Based on the histopathological features it was diagnosed as pleomorphic adenoma. The patient was followed up for 6 months. No evidence of recurrence was seen.

DISCUSSION

Pleomorphic adenomas are the foremost common benign tumor arising within the both major and minor

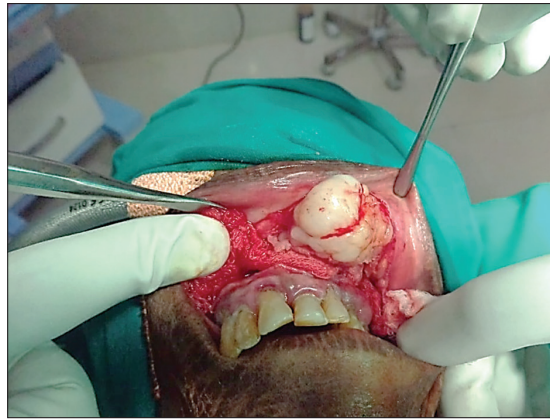


Fig. 1: Swelling in the upper lip



Fig. 2: Excised lesion measuring 3.5 × 2cm

salivary gland. Parotid gland accounts for about 80 % of the tumor arising in the major salivary gland. Pleomorphic adenoma makes up approximately 40% of intra-oral minor salivary gland tumors and palate is the most common site to be involved.² It occurs in all most all ages, but it peaks in fourth and fifth decade of life. Pleomorphic adenoma are usually painless and in most of the cases they are well encapsulated, rarely with pain, ulceration and bleeding of overlaying mucosa (3). Minor salivary gland pleomorphic adenoma presents as soft or firm mass, with most having a nodular, exophytic component. Pleomorphic adenoma most commonly occurs in upper lip than in lower lip. The lower lip lesion are usually considered as malignant (4). The list of case reports reviewed are shown in the Table-I.

Differentiation between benign and malignant tumors isn't possible without histopathology. Pleomorphic adenomas demonstrate a wide variety of histological features and growth patterns. Osseous and adipose metaplasia are very rare to be found (5). The tumors with numerous myoepithelial cells of plasmacytoid type are more common in Pleomorphic adenomas of the minor salivary glands (6). The presence of capsule in Pleomorphic adenoma is frequently seen. It is usually seen as thick and dense fibrous tissue, which may be complete or discontinued (7).

Differential diagnosis of Pleomorphic adenoma in the minor salivary gland are canicular adenoma and benign

Table 1

Case reports reviewed on pleomorphic adenoma of minor salivary gland

S.no	Year	Authors	Age/sex	Size range	Duration	Location	Investigation	CT findings	Provisional diagnosis	Treatment	Follow up
1	2012	Aggarwal A et al., ¹³	55/M	4.5x3 cm	9 years	Minor salivary gland of buccal mucosa near the superficial lobe of parotid gland	CT scan and MRI	A large well defined encapsulated homogeneously hypodense soft tissue swelling of size 3.3 cm (ML) X 5.6 cm (AP) X 4.4 cm (SI) in the minor salivary gland of buccal mucosa near the superficial lobe of parotid gland	Benign pleomorphic adenoma of minor salivary gland	Complete surgical excision	–
2	2016	Khan MN et al., ¹⁴	60/M	3x4 cm	8 years	Midline of upper lip	–	–	–	Mass excised along with surrounding skin	–
3	2020	Kazikdas KC et al., ¹²	20/M	4x3 cm	2 years	Superior to the vermillion border of the upper lip	Ct scan and Fine-needle aspiration biopsy	Demonstrated a 30 mm x 28 mm enhancing mass in the upper lip region with no invasion to the surrounding tissues	Benign mixed salivary gland tumour	Complete surgical excision	No recurrence or facial asymmetry was observed after 1 year of follow-up.
4	2020	PITSCH DW et al., ¹⁵	50/F	50mm	–	Posterior region of the palate	Incisional biopsy	–	Pleomorphic adenoma	Complete surgical excision	1-year follow-up with no signs of relapse.
5	2020	Shah BA et al., ¹⁶	68/F	2x2.5 cm	2 years	Upper lip	Fine-needle aspiration cytology	–	Pleomorphic adenoma	Complete surgical excision	Patient is on follow-up without any signs of recurrence.
6	2020	Sireesha K et al., ¹⁷	62/F	1.0x1.5 cm	1 year	Right side of the upper lip	–	–	Oral lipoma	Complete surgical excision	Patient was followed for 6 months with no evidence of recurrence
7	2020	Radhika T et al., ¹⁸	34/F	2x2.5 cm	2 months	Right Palate	CT scan IHC	A soft-tissue density measuring 2.3 cm x 1.5 cm x 1.7 cm on the right roof of the oral cavity, in the right lateral aspect of the hard palate with no definite extension to retro-molar regions, nasal cavity or pterygo-palatine fossa.	Minor salivary gland pathology of the right palate	Complete surgical excision	–



Fig. 3: Post-operative

and malignant mesenchymal tumors such as neurofibroma, lipoma and rhabdomyosarcoma (8). The myoepithelioma and basal cell adenomas are in first list of preference in differential diagnosis. Canalicular adenoma lacks chondroid or myxoid matrix, distinguishing it from Pleomorphic adenoma (9). Histologically, canalicular adenomas differ from pleomorphic adenoma by showing tumor cell beading and intraluminal squamous balls or morules (8). In myoepithelioma the ductal structures were rare and myoepithelial cells will be dominant. Basal cell adenoma has no capsule and more common in palatal region (9).

Complete surgical excision is the treatment of choice. To prevent recurrence complete resection with adequate margin of normal surrounding tissue (10). However, margin status, age at diagnosis, and tumor location were all associated with risk of recurrence (11). Recurrence of minor salivary gland tumors is uncommon but increased by intraoperative tumor spillage and inadequate surgical excision (12).

CONCLUSION

The pleomorphic adenoma occurring in the lip is very rare, hence clinical examination and histopathological study is required for proper diagnosis. Complete excision is important together with adequate tissue margins. Recurrence of pleomorphic adenoma in minor salivary gland is rare after adequate surgical excision.

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Address:

Dr. Karthik Shunmugavelu

Mercy Multispecialty Dental Centre

8/27, Parvathipuram 1st Street, Thiruneermalai Main Road, Chrompet, Chennai – 600034, Tamilnadu, India.

Tel: +91 9789885622/9840023697

e-mail: drkarthiks1981@gmail.com