CASE REPORT

Pleomorphic adenoma of major and minor salivary glands: Report of 5 cases

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ABSTRACT:

Salivary gland tumors constitute approximately 3% of all head and neck tumors. Pleomorphic adenoma is the most common tumor of the salivary glands. Pleomorphic adenomas constitute 60% of all salivary gland tumors. It's more commonly seen in parotid, with less than 10% in submandibular, sublingual and minor salivary glands. Here, we report a series of 5 cases of pleomorphic adenoma.

Key words: Pleomorphic adenoma, salivary glands, diagnosis, tumour

Introduction

Pleomorphic adenoma is a benign tumour arising from cells of salivary gland tissue. It's also known as mixed tumour because of presence of epithelial and mesenchymal elements. 63% of pleomorphic adenoma occurs in parotid gland,[1, 2, 5,6,10] followed by 38% in minor salivary glands and 10% in submandibular glands.[10] Since asymptomatic patients visit dentist in later stage, diagnosis will be an incidental finding. The parotid gland and the palate are the most commonly affected sites.[2, 3, 8] Pleomorphic adenomas are generally slow growing and painless tumour.[4] Cases of rapid growth after a long period of quiescence have been reported and malignant transformation occasionally occurs.[4, 8]

Pleomorphic adenoma commonly presents in middle age women.[2, 3,5,7,9] Although it occurs most commonly in the major salivary glands, it may also occur in the minor salivary glands and extra-salivary tissue. It is usually encapsulated when it arises in the major salivary glands but not in the minor salivary glands.

Epithelial cells give rise to ductal structures and mesenchymal elements give rise to myxoid, hyaline, cartilaginous and osseous changes .Based on the presence of amount of different epithelial and mesenchymal elements and anatomical site of occurrence the tumour will show diversity in clinical, radiological and histopathological presentation.[10] Here we are describing 5 cases of pleomorphic adenoma of major and minor salivary gland reported in our institution over a period of 2 years.

Case Series

Case 1

A 42 years, old male patient reported to outpatient department with a chief complaint of slow growing asymptomatic swelling (Fig 1a, 1b) in the right side of face with 3 years evolution. Examination revealed a well defined solitary swelling in the region of angle of mouth and elevating the ear lobe measuring about 5 x 4 cm covered with the normal skin and was firm in consistency. Fine needle aspiration cytology was performed which was suggestive of pleomorphic adenoma of right parotid gland. The patient was referred to oral surgery for the surgical removal of tumour. Diagnosis was confirmed after the histopathological study of excised tissue.

Case 2

A male patient aged 60 years came to our hospital with a chief complaint of swelling on the right side of neck of duration 12 years. It was asymptomatic, slow growing and reached the present size (Fig 2). Extra oral examination revealed a well defined swelling of size approximately 4 x 6 cm in the right submandibular region extending into adjacent areas. Swelling was firm in consistency and was not fixed to underlying structures. Ultrasonography and FNAC were done which was suggestive of pleomorphic adenoma of right submandibular gland. Tumour was excised under general anesthesia in the dept of oral surgery and specimen was sent for histopathological examination which confirmed our diagnosis.

Case 3

Another wise healthy 55 years old male was referred to our outpatient clinic with a compliant of large mass in the palate (Fig 3) since 3 years which was affecting his speech. Swelling was slow growing and attained present size. On intra oral examination a well defined swelling was noticed in the mid palatal region extending from rugae region to soft palate measuring approximately 7x5 cm. Swelling was with normal overlying mucosa and was firm in consistency. FNAC was done which was suggestive of pleomorphic adenoma of minor salivary gland. Tumour excision and biopsy of specimen confirmed the diagnosis.

Case 4

A female patient of 66 years came to our institution with a complaint of swelling in upper right back teeth region of 1 month duration. Swelling was small, gradually growing and reached size approximately 3x3 cm (Fig 4). Surface of swelling was normal without any surface changes. Swelling was firm in consistency and nontender on palpation. FNAC was done which was suggestive of pleomorphic adenoma of minor salivary gland. Surgical excision and biopsy confirmed our diagnosis

Case 5

A male patient of 32 years reported to our college with a chief complaint of swelling in upper right back teeth region of 1 month duration. Swelling was small and gradually growing and reached size of about 3x3 cm (Fig 5, 6). Surface of swelling was normal without any surface changes. Swelling was firm in consistency and non tender on palpation. FNAC was suggestive pleomorphic adenoma of minor salivary gland. Surgical excision and biopsy confirmed the diagnosis.

Discussion

Pleomorphic adenoma is an insidious tumor that can reach into great proportion if not treated[4]. Four of five salivary gland tumors of the parotid gland and more than half the tumors of submandibular gland and the palate are pleomorphic adenomas.[8] Pleomorphic adenoma occurs less commonly outside salivary gland tissue and may arise from any glandular tissue with myoepithelial cells.[10] It can occur in the lacrimal glands, external auditory canal, skin, breast tissue, and vulva, and accounts for half of lacrimal gland tumours; the other half are malignant.[10] Late presentation is common; the majority of patients reported here had tumours in excess of 3 cm in diameter at the time of our diagnosis. This could be attributed to asymptomatic nature of tumour.

Diagnosis can be made using various techniques available. A radiograph doesn't show any bony changes hence it can be used for ruling out other type of lesions from pleomorphic adenoma. Fine needle aspiration cytology, with or without ultrasound guidance, is often used in investigations of superficial salivary gland masses, and provides high diagnostic accuracy.[6, 12] FNAC is economic and easy to perform and required minimum instruments. Differentiation from adenoid cystic carcinoma and low grade adenocarcinoma may be difficult with fine needle aspiration alone.[13]

Radiologically it is difficult to distinguish pleomorphic adenoma from its variants, myoepithelioma and basal cell adenoma.[14] Imaging techniques ultrasound, magnetic resonance (MRI), or computed tomography for diagnosis of salivary gland tumour depends on the site & size of the tumor.[6,15] Superficially placed tumours can be diagnosed easily using ultrasonography where as tumours placed deep inside structures can be diagnosed by MRI. Because pleomorphic adenoma is commonly located in the superficial lobe of parotid gland; high resolution ultrasound is suited for differentiating it from commonly found small reactive nodes within the parotid gland. On ultrasound it appears as a hypoechoic, homogenous well circumscribed mass with posterior acoustic enhancement.[10] It also helps to guide fine-needle aspiration (FNA) for cytological diagnosis. Adenoma arising from accessory parotid tissue lying anterior to main body of the gland or along the main duct clinically presents as cheek lump or buccomassetric mass that can be elevated with ultrasound. MRI is the method of choice for deep lobe parotid tumour which clinically presents as a preauricular or oropharyngeal mass.[15] It must be differentiated radiologically from adjacent deep neck spaces (Parapharyngeal and masticator space). Table 1.

Surgical excision is the treatment of choice for pleomorphic adenoma. Malignant transformation may take place in long standing untreated tumors and in recurrent tumors.[2, 4, 7] Recurrence rate is 1 to 5 %.[2,16] Histologically, tumours that have an irregular border with "tongues" of tumour growing into surrounding tissue are associated with a high risk of local recurrence after excision.[18] Even though it is histologically benign tumour, in some cases it may invade local blood vessels in the absence of any other features associated with malignancy.[19] Metastasis from cases of "benign" pleomorphic adenoma to lymph nodes, bone, skin, liver and lung have been reported, but the metastatic deposits do not show any malignant histological features.[16] There is no recognized feature for the primary tumour to predict its metastatic tendency. Carcinoma ex-pleomorphic adenoma is a malignant neoplasm arising from a coexisting or previously excised pleomorphic adenoma.[2,3,5,10] The carcinomatous element arises from the epithelial component of the benign tumour. On imaging, it may look similar to a pleomorphic adenoma, or may show infiltrative margins, necrotic areas, and regional lymph node metastases. For diagnosis histological evidence is needed that shows carcinoma ex-pleomorphic adenoma is arising from a pre-existing benign pleomorphic adenoma.

Conclusion

Pleomorphic adenomas may present in a variety of ways. As Oral physician we need to be aware of its diverse presentation as sound knowledge of this tumour will help in its early diagnosis and treatment thus prevent or reduce the mortality associated with the tumour.

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TABLE 1

Differential diagnosis of Pleomorphic adenoma of salivary gland in different locations[10, 17].

Pleomorphic adenoma of parotid gland	Pleomorphic adenoma of submandibular gland	Pleomorphic adenoma of minor salivary gland
Benign reactive lymphoid hyperplasia,Neuroma of facial nerve,Warthins tumour, Squamous cell carcinoma, Other malignancies	Abnormal lymphadenopathy, Low grade malignant tumour	Lymphoma,Paraganglionomas, Low grade mucoepidermoid carcinoma, Squamous cell carcinoma

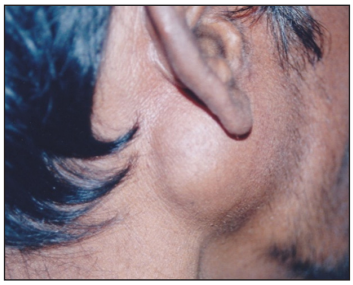

Figure 1a Extra oral swelling present on right parotid region causing elevation of ear lobe

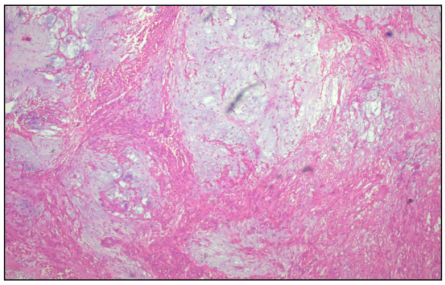

Figure 1b Photomicrograph shows myxoid area with epithelial cells arranged in sheets with interconnecting strands

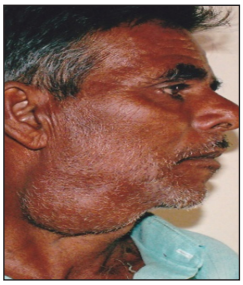

Figure 2 Extra oral swelling measuring 5 x 7cm present on right submandibular region

Figure 3 Intraoral swelling measuring 5 x 7 cm present on palate

Figure 4 Intra oral swelling measuring about 3 x 3 cm present on right side of palate

Figure 5 Intraoral swelling measuring about 3 x 3 cm present on right side of palate

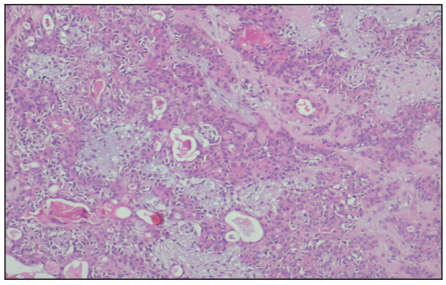

Figure 6 Photomicrograph shows clumps of epithelial cells, myxoid areas and duct like structures, blood vessels