

CASE REPORT

PLEOMORPHIC ADENOMA OF MINOR SALIVARY GLANDS

Mohammad Naeem Khan, Syed Shahmeer Raza, Syed Aizaz Hussain Zaidi, Ihtisham Ul Haq, Amer Kamal Hussain, Muhammad Daniyal Nadeem, Khayyam Farid

Khyber Medical College, Peshawar-Pakistan

Pleomorphic adenoma is the most common benign tumour of salivary glands which is known for its wide pleomorphic architecture. It accounts for 45–75% of all salivary gland neoplasm. It can involve major as well as minor salivary glands. Among minor salivary glands (5–10% of cases) the palate lip, nasal cavity, pharynx, larynx and trachea are the most common sites. Diagnosis is made with biopsy along with histopathology. Wide excision with biopsy and removal of underlying extension of tumour is the treatment of choice. Sixty years old farmer presented with painless swelling in the upper lip for the last 8 years. History revealed recurrent mass in the midline of upper lip with no other complaints. He was operated 3 times for this complaint in the past. Belonging to poor socioeconomic status no biopsy records were found. On examination 3×4 cm hard and mobile mass was found. Lymph nodes of head and neck and parotid gland revealed no enlargement. Surgery by wide excision was planned. After baseline investigation surgery was done and the mass sent for histopathology. Biopsy reports showed pleomorphic adenoma on unusual site. Dissection of salivary gland tumour is important as they have propensity to metastasize. Wide local excision along with biopsy is the method of choice. Proper surgical techniques are required to avoid recurrence.

Keywords: Pleomorphic Adenoma, Minor Salivary Glands; Recurrent Lip mass

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INTRODUCTION

Pleomorphic adenoma is the most common benign tumour of salivary glands. As the name suggests this tumour have mixed histology and consists of 3 components: An epithelial, a myoepithelial and a stromal (mesenchymal) component. It is also known as benign mixed tumour which describes its pleomorphic appearance on light microscopy having mixed origin from epithelial and myoepithelial elements. Epidemiologically, it is the most common tumour in both children and adults accounting for 45–75% of all salivary gland neoplasm.¹ The annual incidence is approximately 2–3.5 cases per 100,000 population. It occurs in individuals of all ages. However, these are most common in the third to sixth decades; the mean age at presentation is between 43 and 46 years.² Forty percent of them are male, 60% female. These tumours can occur in major as well as minor salivary glands. Among the major salivary glands, the tail of the superficial lobe of the parotid salivary gland is the most common site and accounts 70–80% of cases, the submandibular salivary gland accounts 10% and sublingual gland 1%.³ As far as minor salivary glands (5–10% of cases) are concerned the palate (specifically, the junction of the soft and hard palates) and the lip, nasal cavity, pharynx, larynx and trachea are the most common sites for pleomorphic adenoma. The treatment of choice is

surgical removal with wide local excision along with periosteum and involved bone. The risk of the PA becoming malignant is about 6%. The aetiology of pleomorphic adenoma is unknown, however, radiation exposure (after 20 years) and simian virus (SV40) may play an important role.⁴ Clonal chromosome abnormalities with aberrations involving 8q12 and 12q15 have been reported.

CASE REPORT

A 60 years old farmer consulted the ENT out patients department (OPD) of Khyber Teaching Hospital (KTH) with the chief complaint of recurrent painless swelling in the upper lip for the last 8 years. Careful history and examination revealed that the swelling started as a small mass in the midline of upper lip that has gradually increased in size. There was no associated pain, fever, dysphagia, dyspnoea, hoarseness, difficulty in chewing, and epistaxis. He had three previous admissions during which he was operated via excision of the mass for the indication of cosmetic disfigurement of the face. There was no history of Hypertension, Diabetes, smoking and chewing tobacco. He was not on medication or diet restriction at the time of admission. His family history was positive for hypertension but negative for tumours of head and neck. Recurrences, admissions and surgeries had greatly affected his life socially as well as economically.

On examination an alert old man with a visible swelling in the midline of upper lip was found. On palpation mass was 3×4 cm. Mass was hard and mobile. Palpation of the lymph nodes of head and neck and parotid area revealed no swelling. Ear, nose and throat examination was normal revealing no signs of epistaxis, tonsillar enlargement, visible mass or any other lesion. Rest of the systemic examination was normal. As far as investigations are concerned no records of X-ray, C.T scan, MRI or biopsy were found.

Surgery with wide local excision and removal of periosteum and underlying bone was planned. Base line investigations including Hepatitis B and C screening, Serum Urea and electrolytes, blood glucose levels and clotting profile was done. Anaesthesia consultation was taken. Patient was prepared for surgery and possible risks were discussed. Site of incision identified and midline vertical incision was given. Skin and underlying tissue was separated. Mass excised along with surrounding skin and sent for biopsy. Skin sutured. Enucleation was not done to avoid spillage and chance of recurrence. Mass was confined to upper lip alone and no superficial and deep structures were involved. (Figure-1)



Figure-1: Patient Post-operative



Figure-2: Excised mass

DISCUSSION

Tumours occurring in small salivary glands accounts for 22% of all cases reported.⁵ The chances of malignant potential are greatly increased with smaller glands involvement. Pleomorphic adenoma usually presents as a slow-growing, painless mass, which may be present for many years. Symptoms and signs depend on the location.⁵ Our patient presented with recurrent mass that was present in the mid line of upper lip. Patients with minor salivary gland can present with other symptoms such as dysphagia, dyspnoea, hoarseness, difficulty in chewing, and epistaxis. In this case no such complaint was reported by the patient. The diagnosis of pleomorphic adenoma is based on complete and thorough history, physical examination, cytology and histopathology. An incisional biopsy along with histopathology is necessary to determine the proper management regimen, treatment and to differentiate benign from malignant tumour.⁶

CT scan and MRI can help in determining the location, size of tumour and extension to the surrounding areas specially bone. Apart from conventional methods of diagnosis newer technique like Immunohistochemistry (IHC) can be used to help differentiate pleomorphic adenoma from other tumours. The IHC stains that have proved useful are Keratin Cam 5.2 and EMA P-63 Calponin, maspin, S-100 HHF-35 Muscle-specific actin Glial fibrillary acidic protein BMP and Aggrecan.⁷ Despite three prior surgeries no biopsy was done due to poor socioeconomic status of the patient. After excision the mass was sent for biopsy that showed mixed salivary gland tumour (Figure-2, 3).

Pleomorphic adenomas of the minor glands have little propensity for recurrence (a recurrence rate of 2 to 44%, but mainly of the parotid gland. Lack of proper surgical technique was reported to be the main cause of failure. Surgical problems that lead to recurrence are pseudopodia, capsular penetration and tumour rupture. In our patient inappropriate surgery was found to be the main reason.

Dissection of salivary gland tumour should always be carried out as they have propensity to become malignant. Wide local excision with negative margins along with biopsy and histopathology should be the first line treatment option. Appropriate surgical technique should always be used to avoid recurrence.

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Address for Correspondence:

Muhammad Naeem Khan, Community Medicine Department, Khyber Medical College, Peshawar-Pakistan

Cell: +92 300 590 1841

Email: eaglebook@hotmail.com