

Unravelling ectopic tumour pleomorphic adenoma of cheek - A Case Report

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Abstract

Pleomorphic adenoma is a benign tumor most commonly occurring in parotid gland. The tumour was originally believed to be “mixed” neoplastic cells derived from both duct epithelial cells and myoepithelial cells. However, it is now known to be a neoplasm of purely ectodermal cells. It affects most of the major as well as minor salivary glands. Palate is the most commonly affected minor salivary gland amongst other site followed by lips, cheeks, gingiva, floor of the mouth, and tongue. Pleomorphic Adenoma of buccal minor salivary glands is a very rare occurrence both in adults and children. Pleomorphic adenomas can grow into extensive sizes if

left untreated and hence need to be diagnosed early. The reputation of the pleomorphic adenomas for recurrence is due to great surgical difficulties of complete removal of tumour. Pleomorphic adenomas have an unusually strong tendency to seed and recur in the incision scar if incompletely excised. In this report, a case of Pleomorphic adenoma of minor salivary gland cheek in a patient is presented which was treated by wide local surgical excision with adequate margins under general anesthesia, and the patient was followed for 1-year post operatively with no recurrence.

Keywords: Pleomorphic adenoma, minor salivary gland tumour, cheek, buccal mucosa

Introduction

Pleomorphic adenoma (PA) accounts for about 75% of parotid tumours, but a lower proportion of intraoral salivary gland tumours. Salivary gland tumors are uncommon and constitute 2%–6.5% of all the head and neck neoplasms. Tumors of the minor salivary glands account for <25% of the total salivary neoplasms^[1].

The term Pleomorphic adenoma (PA) was coined by Willis. PA is defined by World Health Organization in 1972 as “a circumscribed tumour characterized by its pleomorphic or mixed appearance clearly recognizable epithelial tissue being intermingled with tissue of mucoid, myxoid or chondroid appearance”^[2]. Microscopically pleomorphic adenoma has a highly variable appearance, hence the name pleomorphic. It is characterized by mixed proliferation of polygonal epithelial and spindle-shaped myoepithelial cells in a variable stroma matrix of mucoid, myxoid, cartilaginous or hyaline origin. The tumor lacks the true capsule and is surrounded by a fibrous pseudo capsule of variable thickness.^[3] PA is a clinically well-defined, slow-growing, asymptomatic lesion with hard consistency and varied diameters. The majority of intraoral PA is solid or rubbery and is found in the submucosa. Although ulcerations are seen in certain cases, the mucosal lining stays intact^[4]. In this report, we emphasize on making differential diagnosis of buccal space lesions to rule out rare pathologies like pleomorphic adenoma of cheek which accounts for as an uncommon site of occurrence with 5.5% incidence^[5]. Although PA arising from the minor salivary glands rarely affects the buccal space, the condition should still be considered in the differential diagnosis of buccal mucosa masses.^[5] This report presents a case of PA of buccal minor salivary gland in an adult patient which was successfully treated by wide local surgical excision.

Case Report

A 22-year-old male patient reported to our Department of Oral and Maxillofacial surgery PCDS & RC, Bhopal with complain of painless swelling over right cheek region since 1 year. (Fig: 1)



Figure 1: Pre-operative picture of patient with swelling present over right cheek region.

Patient had no significant dental history and no past history of use of any medication for continuous use, alcoholism, smoking or history of head and neck radiation. On Extraoral examination a swelling of 3 × 2 cm over the buccal mucosa on the right side of the face was detected extending superior-inferiorly from ala-tragus line to the right lower border of mandible. Antero-posteriorly it was extending 1 cm from the right angle of mouth up to 1 cm short of right pterygomandibular raphe region. It was mildly uncomfortable and freely mobile over the underlying structures and firm in consistency, nonreducible, and unattached to the underlying structures on palpation. The lesion could easily be felt between the cheek mucosa and the skin with bimanual palpation. The color and texture of the cheek mucosa were both normal on inspection. The patient had a normal mouth opening. The lymph nodes examination was normal, with no enlargement. All the necessary blood investigations were done; on ultrasonography of right cheek, there was well-defined, oval shape solid appearing lesion showing isohypoechoic echotexture, measuring 2.8 x 1.9 cm in size

noted in deep subcutaneous region of right cheek region. On color Doppler the lesion appeared to be non-vascular. Radiological examination showed no abnormality in the panoramic radiograph.

Fine-needle aspiration cytology was performed which was suggestive of chronically infected sub mucosal cyst, nonmalignant cells. Since the lesion was small in size, excision of the lesion was planned. After the informed surgical consent from patient, we planned surgery with the knowledge that the lesion was mostly benign. The surgical operation was under general anesthesia under aseptic conditions for wide local excision of the mass. A horizontal incision was given over the prominence of the swelling parallel to the Stenson's duct, submucosal blunt dissection was carried out, and tumor mass was excised and sent for histopathological evaluation. (FIG:2) The surgical wound closure was done using 3-0 vicryl. (FIG:3A) On histopathological examination, specimen(FIG:3B) showed well encapsulated mass lesion, comprising of nests and trabeculae and sheath of epithelial and myoepithelial cells in the chondromyxoid stroma (FIG:4). No evidence of malignancy was seen suggestive of benign PA of minor salivary glands of right buccal mucosa. The patient was discharged after the procedure. He was stable and oriented. There was no postoperative complication. The patient is undergoing clinical follow-up for one year with no changes indicative of recurrence and with a satisfactory healing. (FIG: 5).



Figure 3: Transoperative images.(A) Suture after excision; and (B) Measurement of the surgical specimen.

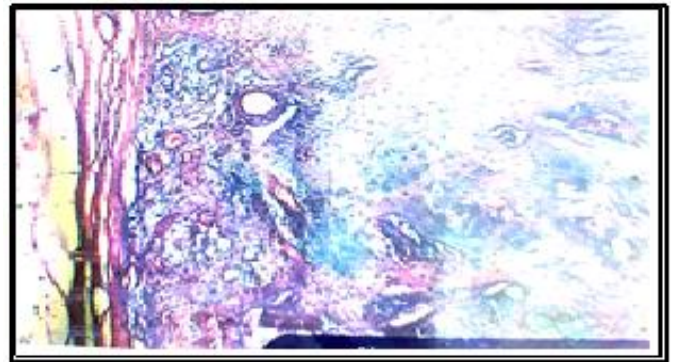


Figure 4: Histopathology image of pleomorphic adenoma



Figure 5: Post-operative image of patient after 6 months of surgery.

Discussion

This case describes the diagnosis and treatment of a pleomorphic adenoma in the buccal mucosa. This is an unusual case since this lesion most commonly affects the parotid gland (70% of cases) ^[6] and when minor salivary glands are involved (5–25% of cases) ^[6], usually the lip and palate are the regions of highest occurrences. Pleomorphic adenomas mostly occur in the young- and



Figure 2: Intraoperative exposure of the tumor mass.

middle-aged adults, between 30 and 60 years. The literature reports suggest female predilection. Location of occurrence is predominantly in the parotid gland occurring in the superficial lobe and presenting as a swelling on the ramus of the mandible in front of the ear^[7]. Differential diagnosis for pleomorphic adenoma includes Warthin tumor, parotid nodal metastasis, facial nerve schwannomas, myoepitheliomas, mucoepidermoid and adenoid cystic carcinoma, and a large variety of other neoplasms nonspecific to salivary glands. Histopathology remains the gold standard in differentiating them all.^[3] PA ectopically exists even in the soft tissue of the neck, lymphonodes, tongue, mandible, hypophysis, mastoid bone, thyroid, parathyroid, subcutaneous layer of the nose, and skin of the external auditory canal^[8]. The size, extent of lesions and the bone involvement can be assessed by imaging like CT scan, MRI and ultrasonography which are useful tools during the pre-operative phase.^[9] Incisional biopsy of PA is contraindicated because can predispose to a recurrence. The preferred diagnostic modality is Fine needle aspiration cytology. According to Willis and colleagues, there are three hypotheses to explain this kind of heterotopias: abnormal differentiation of the local tissues (heteroplasia), abnormal persistence and development of vestigial structures and dislocation of portion of a deficient rudiment during mass movement and development. It can happen due to metaplasia, neoplastic degeneration of ectopic salivary gland tissue or to the implantation after surgical excision of the salivary gland tumor^[10]. The risk of recurrence of PA is generally associated with inadequate surgical procedure, which could have been spillage of tumor or tumor capsule. Recurrent PA occurs as multiple, separate nodules. Surgical risks involved are pseudopodia, capsular penetration, and tumor rupture^[11]. The patient in

question has been followed-up for approximately one year and has not shown any evidence of recurrence. Long term follow-up after excision of the lesion must be performed for a minimum of five years.^[12]

Conclusion

Pleomorphic Adenoma of cheek is not a common lesion compared to that of lesion of parotid gland. A meticulous differential diagnosis of such lesion is of utmost importance. The gold standard treatment for pleomorphic adenoma cheek is surgical excision and thorough follow up as highlighted by this case report. Although the lesion is benign but reported recurrence rate is 7-15%.^[11]

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