Cystic and florid squamous metaplasia in pleomorphic adenoma of palate – A diagnostic dilemma

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ABSTRACT

Pleomorphic adenoma is a benign tumor of salivary gland. Controversy regarding the origin of various mesenchymal components exists and myoepithelial cell has a key role. Metaplastic process is triggered by minor trauma and probable etiology for this change is ischemia. It is diagnostically challenging, because diagnostic pit falls in the presence of mucinous and squamous metaplasia. Report a case of pleomorphic adenoma in the palate of a 35 years old Indian female. Biopsy revealed epithelial component with extensive squamous metaplasia and cystic degeneration. Here we discuss the role of myoepithelial cells in the pathogenesis of pleomorphic adenoma and diagnostic pit falls.

Key words: Myoepithelial cell, pleomorphic adenoma, squamous metaplasia

INTRODUCTION

Pleomorphic adenoma is a benign epithelial neoplasm of salivary gland, histopathologically characterized by a great diversity of morphological aspects.¹ Intercalated duct and myoepithelial cells determine the pleomorphic structure of the tumor. Controversy regarding the origin of various mesenchymal components pleomorphic adenoma, it is now well accepted that myoepithelial differentiation is predominant in these tumors and responsible for their morphologic diversity.² Myoepithelial cell has a key role in the genesis of these tumors, being capable of dedifferentiation, metaplasia and transdifferentiation.³ This case illustrates the difficulty of making a correct diagnosis in the initial tissue specimen and adding a note on diagnostic pitfalls of this pathological entity.

CASE REPORT

A 35 years female presented with swelling in the palate since 2 years, but the increase in the size is observed in last 3 months. Physical examination revealed a well defined growth on the hard palate extending from incisive foramen up to the middle of hard palate. Irregular in shape, soft in consistency and size of the tumor is 50 × 50 mm measuring antero-posteriorly and mediolaterally (Figure 1). Pleomorphic adenoma was the provisional diagnosis. Computed tomography scan reveals soft tissue tumor mass with complete destruction of turbinates (Figure 2). Incisional biopsy was performed that revealed clusters of slightly atypical squamous cells with nuclei and moderate amount of eosinophilic cytoplasm with evidence keratin pearl formation. These squamous cells were seen lining the cystic spaces. The connective tissue component was very minimal. Possibility of pleomorphic adenoma and low grade mucoepidermoid carcinoma or well differentiated squamous cell carcinoma was considered. Considering the differential diagnosis, the lesion was surgically excised. Exicisional specimen revealed squamous cells with extensive squamous metaplasia and keratin pearl formation (Figure 3). Multiple cystic spaces of varying sizes were lined by squamous cells (Figure 4). There were no mucin containing cells and the absence of malignant features ruled the mucoepidermoid carcinoma and well differentiated squamous cell carcinoma. The stroma was fibrous, with lipometaplasia and lymphocytic infiltration is evident. Minimal myxoid stroma was observed. The diagnosis of...
pleomorphic adenoma with extensive squamous metaplasia and cystic degeneration was made. Patient responded well with treatment and post operative palatal obturator was given covering the exposed area (Figure 5).

**DISCUSSION**

Salivary gland neoplasia is unique encompassing perhaps the most complex histopathology of any organ system. The occurrence of pleomorphic adenoma in parotid, submandibular and intra oral salivary gland is 66‑77%, 45‑60% and 19‑53% respectively.¹

Two types of cells determine the pleomorphic structure of the tumor, an inner row of the epithelial cells and an outer layer of myoepithelial cells. The great diversity of cytological differentiation present in pleomorphic adenoma can be explained by metaplastic potential of the neoplastic myoepithelial cells.²

Myoepithelial cells are the result of the differentiation of the pluripotent cells from the salivary ducts during the 10th week of embryo life but in adult life they may proceed from the reserve cells of the intercalated and striated salivary ducts. These myoepithelial cells have the ability to transdifferentiate into cells of the mesenchymal line. The modified myoepithelial cells derived from the neoplastic myoepithelial cells as a result of metaplasia, differentiation and transdifferentiation processes.²³

Squamous metaplasia is uncommon and incidental microscopic finding in benign salivary gland tumors. But squamous metaplasia has been noted in non neoplastic entities like chronic sialadenitis, necrotizing sialometaplasia and lymphoepithelial cyst.⁴ This change is commonly associated with repair following infarction and necrosis of the salivary glands. The metaplastic process might occur spontaneously or be triggered by minor trauma. The most probable etiology for this change is ischemia. This etiology is supported experimentally by the induction of squamous metaplasia in rat salivary glands by arterial ligation.⁴ The varying degree of squamous metaplasia is because of rapidity and ease of the switch in genetic programming of cytokeratin filaments induced by ischemia in salivary glands.⁵

25% of pleomorphic adenoma shows some degree of squamous metaplasia and this process appears to arise largely in relation to modified myoepithelial cells. The squamous differentiation in mucoepidermoid carcinoma is either by the luminal epithelial or modified myoepithelial cells.⁶⁻⁸

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**Figure 1:** Palatal swelling involving the whole palate with intact surface mucosa

**Figure 2:** Computed tomography scan reveals soft tissue tumor mass with complete destruction of turbinates

**Figure 3:** Epidermoid differentiation with keratinization occurring in sheet like areas of tumor cells (H & E stain ×50), Inset shows squamous cells with keratin form of pearl. (H & E, ×100)
The marked ability of acinar units in rat salivary gland to undergo squamous metaplasia lends further support to this premise and provides new evidence for histogenic pathways in human salivary gland tumors.7

Squamous metaplastic changes seen in benign salivary glands might lead to difficulties or misdiagnosis through FNAB and intraoral frozen section simulating malignant neoplasm probably high grade mucoepidermoid carcinoma and squamous cell carcinoma. The probable reasons are the squamous cells often reveal nuclear atypia and the scanty mesenchymal component in pleomorphic adenoma. This may have direct effect on preoperative and intraoperative surgical planning in surgery of the tumors.9,10

CONCLUSION

In conclusion, the feature of squamous metaplasia raises the possibility of carcinoma or carcinomatous even though these are related to ischemia as proven in the experiments. Awareness of these changes is important to prevent over interpretation of benign salivary tumors while excluding other malignancies.

REFERENCES


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