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Case Report

CYSTIC SQUAMOUS METAPLASIA OF PLEOMORPHIC ADENOMA IN PALATE: A RARE CASE REPORT

Sahni P and Arun Kumar K.S.*

Department of Oral Pathology and Microbiology, Narsinhbhai Patel Dental College and Hospital, Visnagar, Gujarat State, India.

ABSTRACT

Pleomorphic adenoma (PA) is the common benign salivary gland neoplasm characterised by neoplastic proliferation of parenchymal cells along with components having malignant potentiality. Metaplastic process, triggered by minor trauma and ischemia might be the probable etiological cause. PA with extensive squamous metaplasia is unusual, moreover it can signify a difficulty in the histopathological diagnosis. A case report of pleomorphic adenoma of 45year- old- female has been discussed with histopathological features showing squamous metaplasia of minor salivary glands in the palate.

Key words: Pleomorphic adenoma, Neoplastic proliferation, Squamous metaplasia.

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INTRODUCTION

PA is the most common benign mixed neoplasm of salivary gland with more predilection for females. It has a wide range of occurrence with mean age of 43.6years and peak incidence between fourth and fifth decade of life [1]. It most commonly occurs in major and minor salivary glands (50%). Among the major salivary glands, parotid is the most affected gland and minor salivary glands in palate is the most common site of occurrence which accounts for 42.8-68.8% followed by upper lip (10.1%) and cheek (5.5%) [2]. Clinically PA presents as slow growing, painless, sessile and firm mass occasionally with ulcerated surface [3]. Histologic diversity although being the hallmark of PA sometimes it may be misinterpreted [4, 5]. The pleomorphic structure of the tumor is determined by the myoepithelial cells and intercalated ducts.

Corresponding Author

Arun Kumar KS Department of Oral Pathology and Microbiology, Narsinhbhai Patel Dental College and Hospital, Visnagar, Gujarat State, India. Email: drarun7027@gmail.com

Myoepithelial cells arise from ectoderm and located between the luminal cells and basal lamina in normal salivary gland. It is now well accepted that myoepithelial differentiation is predominant in these tumors and responsible for the morphological diversity. These cells play a vital role in the genesis of tumor being capable of dedifferentiation, metaplasia and transdifferentiation [6]. The histological patterns may vary considerably among the different parts of the same tumor. Metaplastic changes occur in the epithelium and stromal components with variations in their proportion between epithelial and chondromyxoidstroma which can pose a problem in diagnosing [7].Squamous metaplasia is a rare and incidental histological finding in which squamous epithelium is seen. Ischemia is the most probable cause of squamous metaplasia, the evidence for which is experimentally supported by induction of squamous metaplasia in rat salivary glands by arterial ligation [7, 8]. Local surgical excision is the treatment of choice. If it is incompletely exicised some residual which were left will

lead to its reccurence [9]. The presented case is of PA showing cystic squamous metaplasia.

CASE REPORT

A 45-years old female patient presented with a growth on the left side of the palate since 10years. Patient was relatively asymptomatic before 10years. The growth was initially peanut in size and gradually increased to the present size. The growth was well defined, cauliflower shaped, soft in consistency, pedunculated measuring about 1x1cm in size [Fig. 1]. There was no evidence of surgical history, trauma or infection. A provisional diagnosis of squamous papilloma was made. The lesion was excised and sent for histopathological diagnosis. The excised specimen was creamish yellow in color, with entrapped blood, firm in consistency, measuring about 1.8X 1.6X1.2 Cm in size [Fig.2].

Haematoxylin and Eosin (H&E) stained sections showed superficial parakeratinized stratified squamous epithelium. Stroma was comprised of well encapsulated neoplastic lesional areas. Neoplastic cells were plump spindle to polygonal in shape and were arranged in the form of islands, nests and sheets. Numerous enlarged ducts like structures were lined by cuboidal cells containing pale eosinophilic material. Areas of hyalinization were noticed with squamous metaplasia in most part of the section (Fig. 3). There were numerous keratin filled cystic spaces lined by squamous epithelium (Fig. 4). Deeper areas were comprised of salivary gland tissue, ductal structures and fat cells.

The histopathological features were suggestive of cystic keratinizing variant of pleomorphic adenoma. The patient was followed up for 8 months with uneventful healing and no recurrence was observed.

Table 1. Differential diagnosis of pleomorphic adenoma with squamous metaplasia

Tumor	Microscopic features		
Mucoepidermoid carcinoma	- Cords, sheets, clusters of mucous, squamous, intermediate and clear cells; low to high		
	grade		
SCC in salivary glands	-neoplastic squamous cells, metaplastic in origin, with varying degrees of nuclear atypia		
	-cysts lined by papillary proliferations of bilayeredoncocytic epithelium, inner columnar		
Warthin's tumor	and outer triangular or fusiform basaloid cells		
	-focal areas of squamous metaplasia and mucous cell proplasia		
Epithelial myoepithelial	- Both epithelial and myoepithelial components seen		
carcinoma	- Most tumor cells have myoepithelial features with clear cytoplasm or naked nuclei		
	- Focally, there are ducts or tubules with an outer rim of myoepithelial cells and inner,		
	dark ductal cells with scant eosinophilic cytoplasm and round, bland nuclei		

Fig 1. Intraoral photograph showing growth in the left
side of the palate.Fig 2. Photograph showing excised growth from the left
side of the palate





Fig 3. Photomicrograph 10x Haematoxylin & Eosin (H&E) stained section showing Stroma comprised of well encapsulated neoplastic lesional areas. Neoplastic cells were plump spindle to polygonal in shape, arranged in the form of islands, nests and sheets



Fig 4. Photomicrograph 40x Haematoxylin & Eosin (H&E) stained section showing keratin filled cystic spaces lined by squamous epithelium.



DISCUSSION

Pleomorphic adenoma was coined by Willis and it is the most common tumor of salivary glands which accounts for 53-77% if parotid tumors, 44-68% of submandibular tumors and 33-43% of minor salivary gland tumors among which palate is the most common. Women are affected most frequently in their fourth decade of life [10]. The presented case is of a 45-year- old female patient with a growth on the left side of the palate since 10years. PA with squamous metaplasia is rare and it can imply a potential difficulty in histopathological diagnosis [1]. Microscopically, PA consists of cells with epithelial and mesenchymal differentiation. The morphology of this tumor is highly variable as the result of interplay between these elements [2]. Exuberant squamous metaplasia of pleomorphic adenoma is uncommon and is difficult to diagnose. Focal squamous metaplasia in PA can be related to ischemia and may be found in about 25% of the PA [1, 3]. This case revealed squamous proliferation resulting in multiple squamous epithelium-lined cysts with keratotic lamellae and few solid squamous cell islands showing keratin pearls. Seifert, Donath and Jautzke (1999), described that parotid gland sometimes resembles hair follicle also presents multiple cystic spaces limited with keratotic lamellae. It also exhibits solid squamous cell islands, keratinized masses outside the cysts with multinucleated giant cells and focal calcification [14]. Nago et al (2002) [13] described two cases histologically identical to the choristoma previously reported, both of the parotid gland, but the authors proposed a new designation for the lesions: keratocystoma, since they believed that the choristoma described by Seifert, Donath and Jautzke (1999) is a peculiar variety of salivary gland tumor [14]. Squamous metaplasia is a finding in most of the salivary gland neoplasms which had exposed to preoperative FNAC [3]. In the present case as there is no history of trauma, extensive squamous metaplasia remains unexplained.

The differential diagnosis of the pleomorphic adenoma with squamous metaplasia and keratin cysts includes, mucoepidermoid carcinoma, squamous cell carcinoma in salivary glands, and Warthin's tumor, Epithelialmyoepithelial carcinoma [Table 1] [2, 10, 11].

Vaz Goulart MC *et al*, in their study mentioned that immunohistochemical analysis helped to identify nature of tumor cells in which the luminal cells were identified by low-molecular-weight cytokeratins (CK's), including CK7, CK8 and CK19, as well as CEA (carcinoembryonic antigen) and EMA (epithelial membrane antigen) by showing immune positivity and abluminal cells showed heterogeneous positivity for myoepithelial cell markers such as p63, high-molecularweight CKs, S-100, vimentin, SMA (smooth muscle actin), MSA (muscle specific action) and GFAP (glial fibrillary acidic protein). The solid sheets, nests and cords presented immune reactivity either for CK7 and CK19, suggesting a luminal cell phenotype, or for vimentin, GFAP and p63 [1].

Many theories were put forward by various authors for histogenesis of salivary gland neoplasms which helps in classifying and differentiating lesions for establishing early diagnosis. Among them are Basal reserve cell theory, Pluripotent unicellular reserve cell theory, Semi-pleuripotent bicellular reserve cell theory: (Advanced by Eversole 1971 and further refined and 161 developed by Batasakis*et al.*) and multicellular theory. For

better diagnosis and management, histogenetic concepts are correlated with cytoarchitectural features and profiles of these lesions [12].

The complete surgical excision of the lesion and follow up should be done in order to prevent recurrence and malignant transformation [1, 9]. Follow up of the present case was done for 8 months, without any recurrence.

CONCLUSION

PA even though is benign salivary gland neoplasm, when presents with features of squamous metaplasia, keratin and cystic spaces, leads to misinterpretation and pose difficulty in diagnosis. As it has features similar to malignancies like MEC and SCC, it is necessary to observe caution while reporting characteristic morphological features of PA and rule out mitosis and necrosis to delineate it from malignancy. This helps in accuracy in the treatment and avoids unnecessary aggressive treatment.

STATEMENT OF HUMAN AND ANIMAL RIGHTS

All procedures performed in human participant were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. This article does not contain any studies with animals performed by any of the authors.

CONSENT

Informed consent of the patient was obtained.

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CONFLICT OF INTEREST Nil

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