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Editorial

The Interpolated Wen-Keratocystoma - Salivary Gland

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Keratocystoma emerges as an exceptionally discerned, benign tumour incriminating salivary glands. Initially scripted by Seifert as choristoma and subsequently by Nagao, tumefaction is configured of squamous epithelial cell layered multi-cystic spaces intermingled with focal areas of solid squamous epithelial cell nests.

An extremely exceptional neoplasm of salivary gland, keratocystoma may arise with paediatric population or adults. Tumefaction is devoid of cytological or nuclear atypia, focal necrosis and lymphatic or vascular invasion.

Keratocystoma is posited to emerge from ducts of salivary glands demonstrating squamous metaplasia [1,2].

Commonly, keratocystoma incriminates major salivary glands as the parotid gland. A benign neoplasm of salivary glands, keratocystoma is devoid of lobular architecture [2,3].

Upon microscopy, the tumefaction configures innumerable multi-cystic spaces. Cystic articulations are layered with squamous epithelial cells and appear commingled with foci of solid nests of epithelial cells. Characteristically, multi-cystic spaces layered by stratified squamous epithelium are permeated with keratotic lamellae [2,3].

Pathognomonic morphological features are comprised of a bland layer of stratified squamous epithelium devoid of granular cell layer impregnated within and coating multi-cystic structures, commingled with sharply segregated, solid islands and nests of squamous epithelial cells.

Layering stratified squamous epithelium lacks a distinct granular cell zone and exhibits foci of parakeratotic or orthokeratotic keratinization. Extraneous squamous epithelial cell layer enunciates bud-like protrusions. Extruded keratin engenders focal, chronic inflammatory cell reaction inculcated with foreign body giant cells [4,5].

Neoplastic cells are pervaded with abundant eosinophilic cytoplasm and bland, uniform nuclei. Mucous cells are absent. Tumour cells are devoid of cellular or nuclear atypia.

Tumefaction exhibits an expansive pattern of neoplastic progression.

Foci of tumour necrosis, tumour cell infiltration and vascular invasion, lymphatic invasion or perineurial invasion appear absent. Occasional and normal mitotic figures may be discerned. Tumour cell nests are encompassed by collagenous stroma [4,5].

Keratocystoma appears immune reactive to cytokeratin, AE1/AE3, CK14 or CK17. Focal immune reactivity to CK13 or CK19 is encountered. The extraneous, basal cell layer appears immune reactive to Ki67. Multi-cystic structures and cellular nests are enmeshed within a substance immune reactive to type IV collagen.

Keratocystoma is immune non reactive to alpha smooth muscle actin (α-SMA), S100 protein, CK8 or CK18 [6,7].

Figure 1: Keratocystoma depicting multi-cystic structures lined by parakeratotic and orthokeratotic stratified squamous epithelium and imbued with keratotic lamella. Solid nests and aggregates of squamous epithelial cells are intermixed with multi-cystic structures [8].

Figure 2: Keratocystoma delineating multi-cystic articulations layered by orthokeratotic and parakeratotic stratified squamous epithelium and permeated with keratotic lamellae. Solid nests and aggregates of squamous epithelial cells appear commingled with multi-cystic configurations [9].

Benign Epithelial Tumours	Malignant Epithelial Tumours
Pleomorphic adenoma	Mucoepidermoid carcinoma
Basal cell adenoma	Adenoid cystic carcinoma
Warthin tumour	Acinic cell carcinoma
Oncocytoma	Secretory carcinoma
Salivary gland myoepithelioma	Micro-secretory adenocarcinoma
Canalicular adenoma	Polymorphous adenocarcinoma
Cystadenoma of salivary gland	Hyalinising clear cell carcinoma
Ductal papilloma	Basal cell adenocarcinoma
Sialadenoma papilliferum	Intra-ductal carcinoma
Lymphadenoma	Salivary duct carcinoma
Sebaceous adenoma	Myoepithelial carcinoma
Intercalated duct adenoma and hyperplasia	Epithelial-myoepithelial carcinoma
Striated duct adenoma	Mucinous adenocarcinoma
Sclerosing polycystic adenoma	Sclerosing micro-cystic adenocarcinoma
Keratocystoma	Carcinoma ex pleomorphic adenoma
Sialolipoma (mesenchymal tumour of salivary gland)	Carcinosarcoma of salivary glands
	Sebaceous adenocarcinoma
	Lympho-epithelial carcinoma
	Squamous cell carcinoma
	Sialoblastoma
	Salivary carcinoma (NOS) and emerging entities

Table 1: WHO classification of salivary gland tumors (5th edition) [2].

Keratocystoma of salivary gland requires segregation from neoplasms such as epidermal inclusion cyst, dermoid cyst, mucoepidermoid carcinoma, necrotizing sialometaplasia, squamous metaplasia occurring within various disorders of salivary glands, primary or metastatic squamous cell carcinoma or metaplastic Warthin tumour.

Keratocystoma can be appropriately managed with comprehensive surgical extermination of the neoplasm [6,7].

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8. Image 1 Courtesy: Science direct.

9. Image 2 Courtesy: Nature.com.