

Blister and Flux-Keratocystoma Salivary Gland

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Keratocystoma emerges as an exceptionally discerned, benign neoplasm of salivary glands. Initially scripted by Seifert and Nagao, tumour is preponderantly composed of multicystic spaces layered by squamous epithelial cells commingled with focal areas of solid epithelial cell nests. Neoplasm is extremely exceptional within paediatric population or adults. Commonly implicating the parotid gland, neoplasm may be engendered from salivary ducts subjected to squamous metaplasia [1,2].

Upon microscopy, a benign tumour devoid of lobular architecture is observed. Tumour depicts an expansive pattern of neoplastic evolution [2,3]. Tumefaction is comprised of multicystic spaces layered by squamous epithelial cells along with admixed, focal aggregates of solid epithelial cell nests. Layering squamous epithelium depicts foci of parakeratotic and orthokeratotic keratinization and appears devoid of a granular cell layer. Extraneous cellular layer depicts bud-like protrusions [3,4]. Tumour cells are impregnated with abundant, eosinophilic cytoplasm and bland, uniform nuclei. Circumscribing stroma appears collagenous. Foci of foreign body giant cell reaction may appear in concurrence with keratin deposits. Mitotic figures are occasional [3,4]. Focal necrosis, cellular atypia, tumour invasion into encompassing stroma or angio-lymphatic spaces or perineurial invasion appears absent. Mucous cells appear absent [4,5].

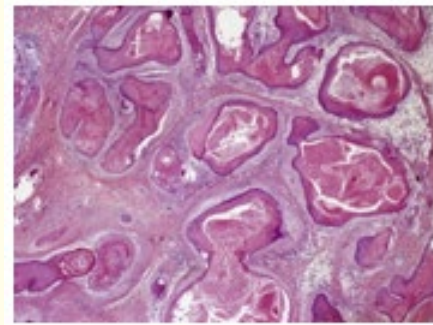


Figure 1: Keratocystoma demonstrating multiple cystic spaces layered by stratified squamous epithelium admixed with solid foci of epithelial cell nests surrounded by a collagenous stroma [7].

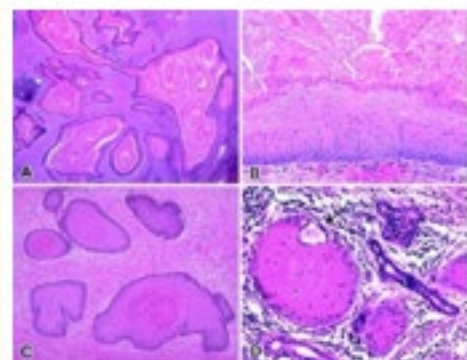


Figure 2: Keratocystoma delineating multiple cystic spaces lined by squamous epithelium with commingled solid epithelial cell nests. Circumscribing stroma is collagenous [8].

TNM staging of salivary gland neoplasms as per American Joint Committee on Cancer (AJCC) 8th edition.

Primary tumour

- TX: Primary tumour cannot be assessed
- T0: No evidence of primary tumour
- Tis: Carcinoma in situ
- T1: Tumour \leq 2-centimetre magnitude with absent extra-parenchymal extension
- T2: Tumour $>$ 2 cm but \leq 4 cm with absent extra-parenchymal extension
- T3: Tumour $>$ 4 cm or tumour along with extra-parenchymal extension
- T4a: Tumour of variable magnitude with invasion into cutis, mandible, auditory canal or facial nerve
- T4b: Tumour of variable magnitude with invasion into base of skull or pterygoid plates or tumour encasement of carotid artery

Extra-parenchymal extension comprises of clinical or macroscopic evidence of neoplastic invasion of soft tissues. Discernment of singular microscopic evidence does not constitute as extra-parenchymal neoplastic extension for tumour classification.

Regional lymph nodes

- NX: Regional lymph nodes cannot be assessed
- N0: No regional lymph node metastasis
- N1: Metastasis into a singular ipsilateral lymph node \leq 3 centimetre magnitude with absent extra-nodal extension
- N2a: Metastasis into singular ipsilateral lymph node \leq 3 centimetre magnitude along with extra-nodal extension or magnitude $>$ 3 centimetres and \leq 6 centimetres with absent extra-nodal extension
- N2b: Metastasis in multiple ipsilateral lymph nodes \leq 6 centimetre magnitude with absent extra-nodal extension
- N2c: Metastasis into bilateral or contralateral lymph nodes \leq 6 centimetre with absent extra-nodal extension
- N3a: Metastasis into a singular lymph node $>$ 6 centimetres with absent extra-nodal extension

- N3b: Extra-nodal extension into a singular ipsilateral lymph node $>$ 3 centimetre or singular contralateral lymph node or multiple lymph nodes with metastases with discernible extra-nodal extension

Extra-nodal extension (ENE) is designated as tumour confined to lymph node with extension through the lymph node capsule into circumscribing connective tissue along with or absence of associated stromal reaction.

Distant metastasis

- M0: Distant metastasis absent
- M1: Distant metastasis present [3,4]

Keratocystoma of the salivary gland appears immune reactive to cytokeratin AE1/AE3, CK14 or CK17 with focal immune reactivity to CK13 and CK19. Ki67 immunostaining appears confined to extraneous basal layer [4,5]. Cysts or cellular nests appear immune reactive to collagen type IV. Tumour cells appear immune non reactive to α smooth muscle actin (α SMA), S100 protein, CK8 and CK18 [4,5]. Keratocystoma of the salivary gland requires segregation from neoplasms as epidermal cyst, dermoid cyst, mucoepidermoid carcinoma, necrotizing sialometaplasia, squamous cell carcinoma or squamous metaplasia occurring within diverse disorders of salivary gland [5,6]. Keratocystoma of the salivary gland can be appropriately subjected to surgical extermination of the neoplasm. Surgical manoeuvres as enucleation remain non beneficial on account of enhanced possible localized tumour reoccurrence [5,6].

Lesions confined to superficial lobe of parotid gland may be managed with superficial parotidectomy. Total parotidectomy with preservation of facial nerve may be beneficially employed for treating enlarged superficial tumours or neoplasms confined to deep seated lobe of parotid gland [5,6].

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7. Image 1 Courtesy: Science direct
8. Image 2 Courtesy: Nature.com