Inverted Ductal Papilloma of the Salivary Gland

This is a case consult of slides stated to be from an excision of a buccal mucosa mass in a 58-year-old-man. The specimen was described as a 3 cm diameter roughly oval tan-gray tissue with a 2 x 1.5 cm mucosal ellipse on the surface that has a central ulcerated punctum. Cut section showed an underlying 1.7 cm diameter roughly oval well-circumscribed mass with a granular tan surface. Histological sections show a papillary lesion with an orifice on the mucosal surface and with epithelial nests invaginating into the underlying lamina propria in a non-infiltrative pattern. (Figure 1) The lesion is composed of papillary epithelial fronds with cleft-like spaces between the fronds. (Figure 2) The papillary fronds are lined by non-keratinizing basaloid stratified squamous cells with a superficial layer of columnar glandular cells along with mucous goblet cells interspersed among the squamous cells. (Figure 3) All the cellular components are devoid of cytologic atypia and mitoses. Based on these microscopic features we signed the case out as inverted ductal papilloma (IDP).

**Figure 1.** Papillary lesion with an orifice on the mucosal surface (single arrow) and with epithelial nests invaginating (“inverted”) into the underlying lamina propria (double arrow). Note the adjacent squamous epithelium of the oral cavity mucosa (asterisk) (Hematoxylin-Eosin, 40X magnification).
Ductal papillomas are uncommon benign epithelial tumors with a papillary configuration that originate from the excretory ductal system of salivary gland acini. The World Health Organization recognizes two sub-types depending on the growth pattern: an intraductal papilloma (IP) and an IDP. An IDP usually presents as an asymptomatic submucosal nodule, measuring about 1.5 centimeters in diameter, and most commonly involving the buccal mucosa, followed by the lips, palate, and floor of the mouth. Histological sections typically show an unencapsulated though well-circumscribed epithelial proliferation with a papillary configuration on the luminal surface, and a nodular, endophytic or invaginating ("inverted") configuration at its interface with the underlying lamina propria. Both the papillary and the invaginating areas are composed of basaloid, non-keratinizing stratified squamous epithelium that are often covered with a cuboidal or columnar ductal cell layer. Scattered among these are mucous goblet cells which can form microcysts. There is an overall morphological similarity to the sinonasal inverted papilloma. A relationship to trauma has been proposed. Association with Human Papilloma Virus (HPV) has also been reported. Others, however, have not been able to demonstrate this association.

Differential diagnoses primarily include IP - which is differentiated from IDP architecturally by being a unicystic intraluminal papillary proliferation within a dilated excretory duct - and sialadenoma papilliferum - which is predominantly polypoid and pedunculated with a verrucoid surface rather than a submucosal nodule, and an over-all morphologic similarity to the cutaneous tumor syringocystadenoma papilliferum. An important differential diagnosis that has to be ruled out is mucoepidermoid carcinoma (MECA) because of the presence of both squamous and mucin-secreting cells. MECA is distinguished by poor circumscription, and an infiltrative solid-cystic growth pattern.

IDP is benign and non-recurrent. Unlike the nasal tumor, there has been no report of malignant transformation. Complete surgical excision is considered curative. Reporting these cases is encouraged to further our knowledge of the entity and elucidate a potential association with HPV.

REFERENCES