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Case Report

Canalicular adenoma of parotid gland: A case report and review of literature

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ABSTRACT

Canalicular adenoma is a rare benign salivary gland neoplasm affecting predominantly the minor salivary gland. We present a case of a 55 year female who presented with a mucosal nodule in the left cheek and was diagnosed as Canalicular adenoma based on the morphology and immunohistochemistry findings. Our case is rare in that, it is a case of Canalicular adenoma affecting the major salivary gland – Parotid gland, and is the 7^{th} case reported in literature to the best of our knowledge.

1. Introduction

Canalicular adenoma (CA) is a benign salivary gland neoplasm first described in 1942 by McFarland as a variant of monomorphic basal cell adenoma.[1] In 1991, CA was identified as a separate entity in the World Health Organization classification. CA constitutes 1-3% of the salivary gland neoplasms[2] with the most common site of occurrence being the upper lip, followed by buccal mucosa, lower lip and soft palate. Rarely, affects the major salivary gland (Parotid gland) and esophagus. CA occurs in the fourth to seventh decade of life, usually after 50 years of age with a slight female predilection (Male: Female – 1:1.8).[3–7] CA is composed of monomorphous epithelial ductal cells arranged in anastomosing cords within the cell-poor vascular stroma.

Here, we report a case of Canalicular adenoma of the Parotid gland in a 55-year female.

2. Case Presentation

A 55-year female presented with a painless swelling in the left cheek for 3 months. The lesion was slow growing and had no associated symptoms. On examination, a nodule was noted in the parotid region which was mobile, non-tender and measuring 1cm in size. The patient underwent superficial parotidectomy.

Grossly, the nodule was well-circumscribed and soild pale yellow to tan in colour. No cystic areas or necrosis was seen. Microscopically, the lesion was encapsulated and composed of single layered monotonous cells arranged in anastomosing cords, canaliculi, cords and tubules. Focally, papillary projections into the cystic spaces noted. These cells are cuboidal to coulumnar with round to ovoid uniform nuclei with focal pseudostratification, coarse chromatin and scant to moderate cytoplasm. Stroma is edematous with mild lymphoplasmacytic infiltration. Variable sized cystic spaces are seen filled with mucinous and pale eosinophilic material. Mitosis was sparse. Surrounding compressed uninvolved salivary gland tissue was also seen.

On immunohistochemistry, these cells were immunoreactive for S100 and was negative for p63, confirming the lack of myoepithelial layer. Based on the clinical findings, morphology and immunohistochemistry, a diagnosis of Canalicular adenoma was made.

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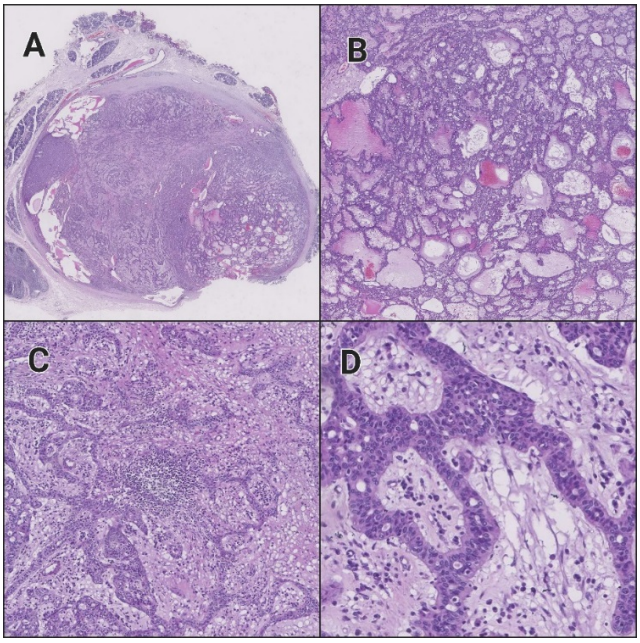

Fig. 1: H&E stain: **A:** 10x – Shows a well circumscribed encapsulated neoplasm of salivary gland. **B**&C: 20x – shows anastomosing cords, strands and tubules of neoplastic cells with cystic spaces filled with pale eosinophilic to myxoid material. **D:** 40x – Shows a single layered epithelium with pseudostratification with lack of myoepithelial layer.

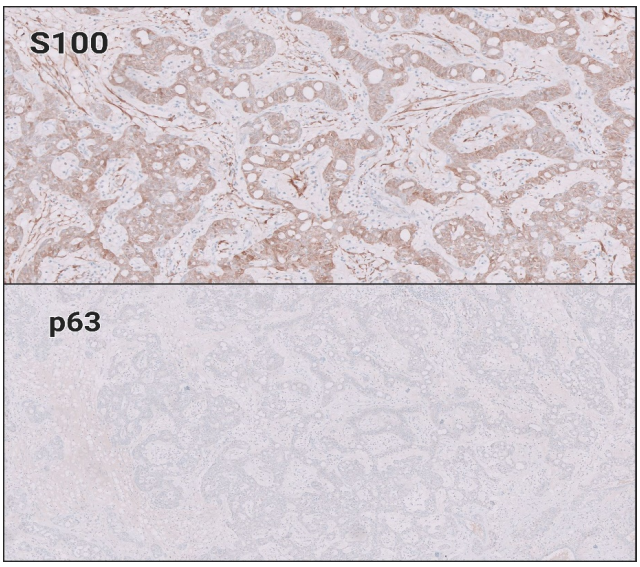

Fig. 2: IHC stain 40x: Show a moderate to strong cytoplasmic S100 positive and a negative p63 immunostain indicating the lack of myoepithelial layer.

3. Discussion

Salivary gland neoplasms represent around 3% of all head and neck tumors and Canalicular adenoma constitutes 1-3% of the salivary gland neoplasms. Most commonly occurs in the minor salivary gland and is very rare in the major salivary gland.[2,3] Based on the literature review, our case is the 7^{th} reported case of Canalicular adenoma of the parotid (Table 1). The peak incidence of CA is in the fourth to seventh decade of life. Clinically, CA presents as an asymptomatic mucosal nodule that is slow growing. Grossly, CA is a well-circumscribed tumor and often encapsulated. It can be either unifocal or multifocal. Microscopically presents as strands or ribbons or anastomosing cords with cystic spaces in between the strands. These tumors lack an outer myoepithelial layer and can infiltrate the capsule and show extracapsular tumor islands.[8]

Table 1:

S.No	Case report	Year
1.	Rossiell et al[9]	2002
2.	Philpott et al[10]	2005
3.	Liess et al[11]	2006
4.	Butler et al[12]	2009
5.	Kim D H et al[13]	2017
6.	Su V et al[8]	2023
7.	Current report	2023

The most common differential diagnosis based on clinical and morphological findings are pleomorphic adenoma (HMGA2-WIF1 fusion pleomorphic adenoma), basal cell adenoma, polymorphous low-grade adenocarcinoma and adenoid cystic carcinoma. Immunohistochemistry diagnosis is of utmost importance in differentiating CA from other tumors. Very few cases have diagnosed CA with the aid of immunohistochemistry. The most useful markers are S100 which shows strong to moderate diffuse cytoplasmic positivity and p63 immunostain which fails to highlight the myoepithelial layer, depicting the absence of the myoepithelial layer. Other useful markers are AE1/AE3, CK19, Pancytokeratin, CK7, CK8, CK13, CK15, CD117 and BCL2. Nuclear SOX10, CAM5.2 and cytoplasmic p16 are also noted. GFAP shows a focal distinctive linear immunoreactive pattern among cells in proximity to the connective tissue interface. The stroma of CA stains for Alcian blue and PAS stain. No relevant fusions or genetic signatures identified till date.[14]

CA has a favorable prognosis upon complete resection of the tumor. Persistence due to mutifocality can be indistinguishable from recurrence. The recurrence rates are 3-5% after the surgery.

4. Conclusion

Though CA is a rare entity, it is important to consider this type of tumor to be a differential before diagnosis of pleomorphic adenoma, basal cell adenoma and especially adenoid cystic tumor. Immunohistochemistry plays a major role in helping to differentiate CA from other entities. As the prognosis of CA is good, an accurate diagnosis aids in a better therapeutic management.

5. Conflicts of interest

There are no conflicts of interest.

6. Source of Funding

None.

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