Synchronous Ipsilateral Parotid Tumors with Cytologic–Histologic Correlation

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Abstract Synchronous ipsilateral tumor formation within a major salivary gland is a very rare event. In this case, a 54-year-old female tobacco smoker presented with a slowly enlarging left parotid gland. Computed tomography of the neck demonstrated a solid mass superficial to a cystic mass in the deep lobe of the gland. Ultrasound-guided fine needle aspiration yielded oncocytic cells, lymphoid cells, and granular debris along with rare cohesive groups of basaloid cells. Parotidectomy was performed, and the resected gland was found to contain two adjacent but distinct masses. One mass, a predominantly solid, well-circumscribed lesion composed of ribbons of double-layered oncocytic cells and a lymphoid stroma with germinal center formation, was a Warthin tumor. The other mass, a predominantly cystic lesion composed of cords and nests of basaloid cells with associated deposits of basement membrane-like material, was a basal cell adenoma of the membranous type. To our knowledge, this is the first reported case of synchronous Warthin tumor and basal cell adenoma of the parotid gland with cytologic–histologic correlation attributable to each tumor.

Keywords Warthin tumor · Basal cell adenoma · Salivary gland · Parotid gland · Head and neck pathology · Cytopathology

Introduction

Warthin tumor (papillary cystadenoma lymphomatosum) is the second most common benign salivary gland tumor that presents most often in the parotid gland [1]. It occurs more commonly in males than in females (2.2:1) [2]. Diagnosis of the tumor by fine needle aspiration is often possible and may be used as a guide to operative management [3]. Cytologically, the defining features of this entity include cohesive sheets of oncocytes, a lymphocytic background, and cystic debris [1]. Similarly, a lymphoid stroma surrounded by a double layer of oncocytes is seen histopathologically.

Basal cell adenoma is another benign salivary gland tumor that may also be diagnosed cytologically with high accuracy [4]. It comprises about 2 % of all benign salivary gland tumors [5]. Basal cell adenoma occurs predominantly in major salivary glands and more commonly in females than in males (2:1) [2]. Cytologically, sheets of small basaloid cells containing oval nuclei with fine chromatin that are usually devoid of prominent nucleoli are seen [6, 7]. However, histopathologic confirmation of basal cell adenoma is necessary in order to exclude potential benign and malignant mimickers, including basal cell adenocarcinoma, canalicular adenoma, cellular pleomorphic adenoma, and the solid variant of adenoid cystic carcinoma [8]. Histologically, this tumor type is composed of nests of uniform basaloid cells, often displaying peripheral palisading, separated by a collagenous stroma [5, 8]. The membranous variant is further distinguished by the presence of prominent, hyalinized basement membrane-like material [5]. While most basal cell adenomas are well-circumscribed and show low recurrence after excision, the membranous type is often unencapsulated and multifocal, making the histopathologic distinction important for further patient management [5].

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Most cases of synchronous unilateral major salivary gland tumors that have been described in the literature have appeared in the parotid gland. While Warthin tumor has been described as a common tumor presenting as one of two synchronous unilateral tumors, pleomorphic adenoma in conjunction with Warthin tumor has been described as the most frequent synchronous unilateral neoplastic combination [9]. Although the membranous type of basal cell adenoma can occur as a multifocal lesion within the parotid gland, it may present as one of two synchronous parotid tumors of different types [9]. We herein describe a case of ipsilateral synchronous Warthin tumor and basal cell adenoma with relevant radiologic, cytopathologic, and histopathologic findings.

Case Report

A 54-year-old female tobacco smoker presented to our institution with a slowly enlarging left parotid gland. Her past medical history beyond her smoking was noncontributory. Computed tomography of the neck demonstrated a solid mass superficial to a cystic mass in the deep lobe of the gland (Fig. 1). The two lesions were targeted separately for sampling. Ultrasound-guided fine needle aspiration of the solid component demonstrated oncocytes (Fig. 2), lymphoid cells, and granular debris, while the aspirate of the cystic component yielded predominantly blood elements with rare cohesive groups of basaloid cells (Fig. 3).

Fig. 1 Computed tomography showing solid (arrowhead) and cystic (arrow) masses in the left parotid gland

Fig. 2 Cluster of oncocytic cells aspirated from the solid mass (Papanicolaou, ×400)

The prospective diagnosis based on cytology was Warthin tumor. Due to the paucity of basaloid cells present, however, an additional diagnosis of basal cell adenoma was not issued with the final cytopathology report. Based on the imaging and cytopathologic findings, total parotidectomy was performed with dissection and preservation of the facial nerve.

The resected gland was found to contain two adjacent but distinct masses (Figs. 4, 5). On gross examination, the firmer mass measured $6.0 \times 3.8 \times 2.3$ cm, while the brown fluid-containing mass measured $4.0 \times 3.9 \times 3.0$ cm. The firmer mass, a predominantly solid, well-circumscribed lesion composed of ribbons of double-layered oncocytic cells and a lymphoid stroma with germinal center formation, was a Warthin tumor (Fig. 6). The other mass, a predominantly cystic lesion composed of cords and nests of basaloid

Fig. 3 Cluster of basaloid cells aspirated from the cystic mass (Papanicolaou, ×400)

Fig. 4 Adjacent solid and cystic masses in sections of the resected parotid gland

Fig. 5 Parotid gland tissue with adjacent solid and cystic masses (hematoxylin and eosin, $\times 40$)

Fig. 6 Warthin tumor with ribbons of oncocytic cells and associated lymphoid stroma demonstrating occasional germinal center formation (hematoxylin and eosin, $\times 200$)

cells with associated deposits of basement membrane-like material, was a basal cell adenoma (Fig. 7). Periodic acid-Schiff staining highlighted the basement membrane-like material associated with the adenoma (Fig. 8). A diagnosis of synchronous Warthin tumor and basal cell adenoma of the membranous type was made. Resection tissue edges were negative for lesional involvement.

Discussion

The incidence of synchronous parotid tumors is greater in males than in females according to published reports [9–11]. In this case, the patient is a female. Her history of smoking could likely have been a predisposing factor in Warthin tumor development [12, 13], since a history of tobacco use was noted in 88 % of men and 89 % of women who developed this type of tumor [2]. On the other hand, the development of other salivary gland neoplasms, including basal cell adenoma, has not been linked to a history of smoking [5].

Gnepp and colleagues reported 25 cases of synchronous unilateral major salivary gland tumors, one of which included a 77-year-old man with Warthin tumor and basal cell adenoma [9]. The majority of the other synchronous lesions described by the authors included Warthin tumor and pleomorphic adenoma, the former being the most common to develop synchronously or metachronously in bilateral major salivary glands [9]. Goh and Cheah also described a case of synchronous parotid basal cell adenoma and adenolymphoma in an 83-year-old man [10]. Cytopathologic examination results were not reported; however, the histopathology revealed a Warthin tumor and a basal cell adenoma of trabecular type [10].

This case is unique in that all three diagnostic modalities indicated a dimorphic lesion. Computed tomography of the neck revealed solid and cystic areas that proved to be Warthin tumor and basal cell adenoma, respectively, via

Fig. 7 Basal cell adenoma with cords and nests of basaloid tumor cells (hematoxylin and eosin, $\times 400$)

Fig. 8 Basal cell adenoma containing membrane-like material flanking cords and nests of tumor cells (periodic acid-Schiff, $\times 200$)

cytopathologic and histopathologic examinations. Along with a few radiologic studies [11, 14], the present case also demonstrates the importance of imaging in identifying and carefully sampling potential synchronous parotid lesions. The cytopathologic and histopathologic identifications of the two tumor types and the extent of lesional involvement then has further management (extent of surgical resection) and prognostic (recurrence) implications.

In this case, the cytopathologic diagnosis of Warthin tumor was essential in guiding the extent of surgical resection. Histopathologic confirmation of concurrent basal cell adenoma did not alter the course of treatment, as both lesions were completely excised along with the parotid gland. The subclassification of the adenoma into membranous type, however, may be important prognostically and warrant further monitoring for the possible development of additional lesions [5].

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