Oncocytic Papillary Cystadenoma with Tyrosine-rich Crystalloids in the Parotid: Case Report with Cytology and Histology Correlation and Literature Review

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Abstract

Oncocytic papillary cystadenomas are rare benign salivary gland neoplasms. Minor salivary glands are frequent localization for this neoplasm. We report a case of oncocytic papillary cystadenoma of the parotid gland, occurring in a 53-year-old male as a slowly growing, well defined cystic nodule of 1.5 cm in diameter. Fine needle aspiration (FNA) was performed by using 25-gauge needle by the (cyto) pathologist. Few small sheets of epithelial and oncocytic degenerative cells with extensive granular debris on the background were observed on FNA smears. Few tabular or petal form, non-birefringent crystalloids were noted in the background of scanty cellular specimens composed predominantly of oncocytic cells. On histopathology, unilocular cystic lesion with papillary structures lined by oncocytic cells and tyrosine-rich crystalloids within the lumen were identified. In conclusion, oncocytic papillary cystadenoma is uncommon benign tumor in the parotid gland and can be accompanied by tyrosine-rich crystalloids. It is useful to be familiar with the type of the crystalloid as they may help in differentiating benign and malignant lesions. To the best of authors’ knowledge, this is the second reported case of oncocytic papillary cystadenoma with crystalloids with cyto-histo correlation.

Keywords: Papillary cystadenoma; Oncocytic cells; Parotid gland; Tyrosine-rich crystalloid; Fine needle aspiration cytology

Introduction

Cystadenoma is an uncommon benign epithelial neoplasm of salivary glands [1]. The majority of these tumors are located in minor salivary glands and about 45% of cases arise in the parotid gland [1]. Histologically, they are composed of cystic spaces lined by single layer of epithelial cells. Most cases are multicellular and 20% of cystadenomas are unilocular. Extensive oncocytic metaplasia of epithelial layer in addition to the crystalloids within the cyst-lumen are rare; to the best of our knowledge, five cases of oncocytic papillary cystadenoma with crystalloids have been reported [2-6]. Fine needle aspiration (FNA) was applied to seven cases of several parotid lesions with crystalloids reported in the literature however only one previous case of oncocytic papillary cystadenoma with crystalloids has included FNA in diagnostic work-up [3,7-9]. We report a case of parotid oncocytic papillary cystadenoma with crystalloids with a review of the literature. To the best of our knowledge, the case presented here represents the second reported case of oncocytic papillary cystadenoma with crystalloids with cyto-histo correlation.

Case report

A 53-year-old male was admitted to the Department of Oral and Maxillofacial Surgery with a swelling in the right parotid region since 5 years associated with slight pain. Ultrasonography displayed a homogeneously hypoechoic lesion of size 1.5 cm with posterior acoustic enhancement, consistent with a benign cystic mass in the superficial lobe of the right parotid gland. By ultrasound and physical examination, the cervical lymph nodes were without any changes. FNA was performed by the (cyto) pathologist using 25-gauge needle and 20 mL syringe attached to the syringe-holder. Little amount of viscous-quality fluid was obtained by aspiration, not suitable for centrifugation or filtration. Two direct smears were prepared by the (cyto) pathologist; one smear was air-dried for May-Grünwald-Giemsa (MGG) staining while the other one was alcohol-fixed for Papanicolaou (PAP) method. No residual mass was detected to continue the aspiration. Few small sheets of epithelial and oncocytic degenerative cells, and foamy macrophages with extensive granular debris on the background were observed in hypocellular smears. Few tabular-form crystalloids with blunt ends, staining dark blue by MGG and faintly by PAP stainings were noted in the background of scanty cellular specimens composed predominantly of oncocytic cells and they were not discernible under polarized-light (Figure 1).

Cytologic material was reported as benign cytology, being compatible with “oncocytic cystic lesion of the salivary gland” and microscopic data concerning the crystalloids were noted. The patient underwent a right superficial parotidectomy. In gross examination, a cystic lesion measuring 15×12×7 mm was detected within the parotid gland. The external surface of the lesion was smooth and it was soft in consistency. On sectioning, the cut surface revealed an unilocular cyst with relatively smooth inner walls. The tissue was fixed in neutral buffered formalin and embedded in paraffin to be stained with hematoxylin and eosin (H&E).
Subsequent histopathological examination showed unilocular cystic lesion with papillary structures lined by oncocytic cells and some crystalloids within the luminal space of the lesion. The microscopic and staining features of the crystalloids were evaluated as being compatible with tyrosine-rich crystalloids. There was a thick connective tissue layer partially surrounding the cystic cavity. The epithelial lining cells displayed the characteristic oncocytic features such as single round nucleus with conspicuous central nucleolus and abundant eosinophilic granular cytoplasm (Figure 2). The tabular-form, non-birefringent crystalloids stained pale with a yellow hue on H&E slides (Figure 3). 

Discussion

The frequency of cystadenoma is between 4.2-4.7% out of benign salivary gland tumours [1]. There is a female predominance and the mean age of the patients is about 57 years [1].

The oncocytic variant of cystadenoma is characterized by a single or double layer of oncocyes lining the cyst walls, resembling Warthin’s tumor without the lymphoid stroma. Clinically, cystadenoma presents as a slow growing, painless cystic mass, usually smaller than 1 cm in diameter with the first impression of a mucocele. It can be detected by routine radiological methods easily and accordingly, sonography has been observed as the most applied method in diagnostic process of the reported cases.

Histopathologically, there are mucinous and papillary types of cystadenoma. Papillary cystadenoma are well-defined, unilocular or multilocular cysts, with intraluminal papillary projections [7,10]. The lining epithelial cells are cuboidal or columnar, and usually two layers in thickness. Oncocytic differentiation occurs rare and can be extensive [11]. Cases exhibiting with apocrine and focal squamous metaplasia and ciliated pseudostratified columnar epithelium have also been reported [9,12,13].

FNA material of papillary cystadenoma is usually adequate for assessment. In the aspiration cytology; papillary structures, sheets of benign epithelial/oncocytic cells, histiocytes/macrophages with granular debris on the background could be seen [3,7-9]. In the literature, there was not any case of papillary cystadenoma reported as false positive by FNA. However it should always be kept in mind that any cellular elements may be diluted in a considerable specimen volume and this emphasizes the need for careful aspiration for cystic lesions and for repeating the aspiration for the residual masses of the salivary gland [14].

Most cases of salivary glands can be approached by FNA with growing confidence by clinicians and (cyto) pathologists. Several institutions indicate that the results obtained when (cyto) pathologists both perform and interpret aspirations are better than when these
functions are split between a clinician and a pathologist who has not seen the patient [14,15].

There are quite a few cases of oncocytic papillary cystadenoma with intraluminal crystalloids reported in the English literature. Crystals in different shapes and nature may associate oncocytic type of salivary gland neoplasms. Only in 5 cases of cystadenoma; crystalloids (2 tyrosine-rich, 1 intraluminal and 2 non-tyrosine) have been reported; the location of the lesions were major salivary glands (4 lesions in parotid and 1 lesion in submandibular). Oncocytic metaplasia was seen in all of the cases with crystalloids [2-6].

Several types of crystalline structures such as amylase, tyrosine, collagenous, oxalate and intraluminal crystalloids have been illustrated in neoplastic and nonneoplastic lesions of salivary gland [5,6,15-17].

On the other hand, Bezic et al. [6] focused on crystalloids arising in oncocytic and papillary lesions may pose difficulties in differential diagnosis. It is useful to be familiar with the type of crystalloid as they may help in differentiating benign and malignant lesions.

References


