

## Case Report

# Papillary Cystadenoma of the Epididymis

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**ABSTRACT**

Papillary cystadenoma of the epididymis is a rare benign neoplasm that arises from the efferent duct epithelium. It may present as a unilateral or bilateral epididymal mass and its association with von Hippel-Lindau syndrome is very high, particularly in bilateral lesions. A 24-year-old male presented with a right-sided scrotal mass of six months duration. Ultrasonography of the scrotum revealed a well-defined extra-testicular hyperechoic

mass, 2.5 cm in diameter in the region of the head of the right epididymis. Fine needle aspiration cytology diagnosis was suggestive of adenomatoid tumor but this was later confirmed histologically as papillary cystadenoma of the head of epididymis. There was no evidence of von Hippel-Lindau syndrome at the time of diagnosis and during two years of follow-up.

KEYWORDS: epididymis, papillary cystadenoma, von Hippel-Lindau syndrome

**INTRODUCTION**

Papillary cystadenomas (PC) of the epididymis are partially or completely cystic or solid lesions, between 1-3 cm in diameter and arise from the efferent duct epithelium of the head of epididymis<sup>[1]</sup>. The lesion is bilateral in 40% of cases<sup>[2]</sup> and 65% of the bilateral PC occur in patient with von Hippel-Lindau syndrome (vHLS)<sup>[3]</sup>. The lesion is usually benign, but malignant variant has also been reported<sup>[4]</sup>. A case of PC of the epididymis, not associated with vHLS is reported because of its rarity and the difficulties in differentiating it from other papillary lesions of the testis by fine needle aspiration cytology (FNAC) alone.

**CASE REPORT**

A 24-year-old single male presented with a painless scrotal mass of six months duration. He had no significant past medical history. On local examination, a non-tender nodule, about 2.0 cm in size was palpable on the head of the right epididymis. Results of routine biochemistry, hematological tests, urinalysis and X-ray chest were all normal. Ultrasonography (US) of the scrotum revealed a normal testis with homogenous granular echotexture. A well defined hyperechoic mass, 2.5 cm in diameter in the region of globus major of the right epididymis and close to the superior pole of the right testicle was detected. FNAC smears showed diffuse sheets of apparently monomorphic round to oval cells, spermatozoa and eosinophilic proteinaceous material in the background. No inflammatory cells were detected. The possibility of

adenomatoid tumor was considered. However, the presence of large numbers of spermatozoa in the aspirate was against the possibility of any neoplastic process and hence biopsy was advised. The nodule was excised and sent for histopathology examination.

**Gross Finding**

The tissue measured 1.5 cm in greatest dimension, was well circumscribed, grayish-brown and there was no visible cyst on the cut surface.

**Microscopic Findings**

Sections revealed a strikingly papillary neoplasm that appeared very similar to thyroid tissue (Fig. 1). Thick hyalinized fibrous capsule separated large numbers of dilated efferent ducts both within and outside the neoplasm. Many of the ducts contained spermatozoa (accumulated due to obstruction of vasa efferens by tumor tissue), detached epithelial buds and eosinophilic colloid-like material that was PAS positive but Mayer's mucicarmine negative. The cystic structures contained papillary projections with fibro-vascular core and were lined by double layers of low cuboidal to flattened epithelium (Fig. 2). The cytoplasm showed fine granularity and the nuclei were bland in appearance. In the outermost area, part of the dilated efferent ducts discerned small fibro-vascular stalks covered by flat epithelium and projecting into the luminal spaces at the vicinity of frank papillary lesion, suggesting transition from normal efferent ducts to neoplasm. The overall

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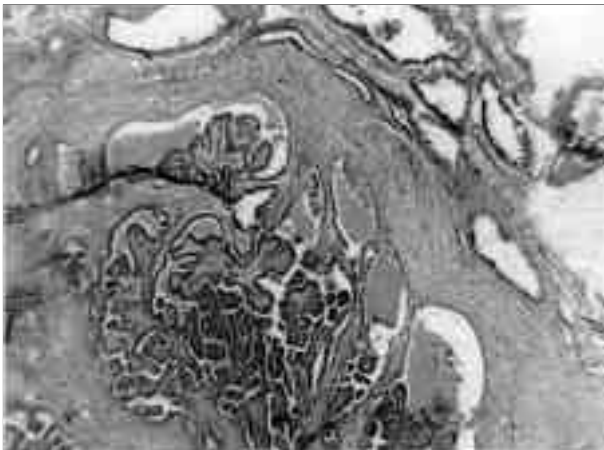


Fig. 1. Photomicrograph of PC of the epididymis showing striking papillary formations and thick capsule with underlying dilated ducts containing colloid-like material (H&E x 40)

histology findings were fairly classical of papillary cystadenoma of the epididymis.

#### Follow Up

Post-operatively, the patient was subjected to ultrasonography of the whole abdomen, intravenous pyelography, fundoscopy and X-ray skull. These did not show any features of vHLS. Two years after the operation, the patient is in good health and doing well.

#### DISCUSSION

Ever since the first case of PC of epididymis was described precisely by Sherrick<sup>[5]</sup> in 1956, about 40 cases have been reported in the world literature and the largest series studied so far comprised of 18 cases<sup>[6]</sup>. It is a rare benign neoplasm, the mean age of the patients being 36 years and the incidence of its association with vHLS is about 40%<sup>[2]</sup>. The epididymis may be unilaterally or bilaterally affected and 65% of the bilateral PC occurs in patients with vHLS<sup>[3]</sup>. Cases of PC in families have also been reported<sup>[1]</sup>. The cell morphology may be clear looking at times and is usually positive for keratin and carcinoembryonic antigen<sup>[7]</sup>. Since there is a close relationship between vHLS and renal cell carcinoma (RCC), patients with vHLS and PC of clear cell morphology, metastatic RCC in the epididymis needs to be excluded by lectin histochemistry, as the PC cells are soybean agglutinin positive<sup>[8]</sup>. Other papillary lesions in the paratesticular or testicular location include mesothelioma and papillary serous tumor of low malignant potential (PSTLMP). FNAC smears are akin to be similar in all three conditions. In histopathology however, the very lack of cellular atypia, stratifications and mitotic figures clearly favors the diagnosis of PC. Moreover, psammoma bodies and detached epithelial buds are prominent

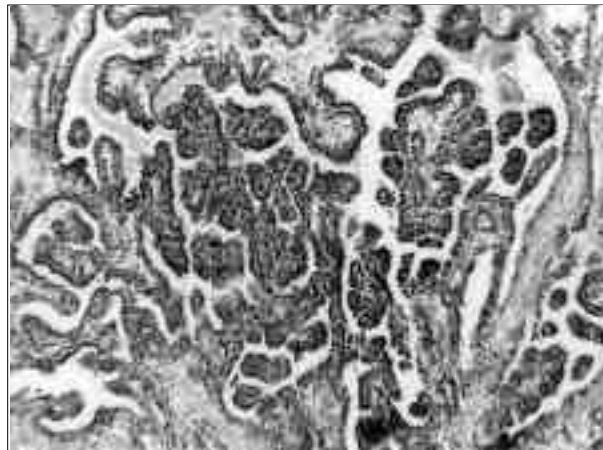


Fig. 2. Photomicrograph showing papillary structures lined by double layers of cuboidal cells with light eosinophilic cytoplasm (H&E x 100)

in paratesticular PSTLMP than mesothelioma. In our case, dilated efferent ducts with spermatozoa inside, refute any consideration of PSTLMP or mesothelioma.

FNAC of the testis has been used in the investigation of male infertility. However, for testicular tumors, whether it carries any risk of tumor spread has not been satisfactorily investigated. Delayed diagnosis of malignancy due to conservative treatment of a supposed inflammatory or benign lesion may be more harmful than the hypothetical risk of tumor spread by FNAC<sup>[9]</sup>. Nowadays, US plays an important role in evaluation of scrotal masses because its accuracy is 98 to 100% in differentiating intratesticular and extratesticular pathology<sup>[10]</sup>. This distinction is important because most of the extratesticular masses are benign, but majority of the intratesticular lesions are malignant<sup>[11]</sup>. Incision biopsy of testicular mass is contraindicated in patients with clinical suspicion of malignancy and US confirmed intratesticular lesion with characteristic echotexture. However, percutaneous FNAC can still be carried out in these cases with caution and proper planning so that immediate surgery including excision of the needle track can follow, if the result is positive for malignancy<sup>[12]</sup>.

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