Case Report

# Papillary Cystadenoma of the Epididymis

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## Abstract

Papillary cystadenoma of epididymis (PCE) is a benign neoplasm involving epididymis and frequently associated with Von Hippel-Lindau (VHL) disease. It is characterized by asymptomatic swelling, which is well circumscribed with papillary fronds on cut section. On microscopic examination, numerous cystic spaces with papillary infoldings are seen. These infoldings are covered with cuboidal to columnar epithelium. It is important to excise and do histopathological examination to rule out diagnosis of serous cystadenocarcinoma of paratestis, which mimic in symptomatology to papillary cystadenoma of epididymis but has poor prognosis. At the same time, regular ultrasonography done to rule out renal cell carcinoma as this entity is associated with VHL disease.

*Keywords:* Papillary; Von Hippel-Lindau Disease; Epididymis; Cystadenoma

## Introduction

Benign epididymal neoplasms take the origin from various structures leading to wide variety of tumors, which includes leiomyoma, serous (nonpapillary) cystadenoma, cavernous hemangioma, and melanotic neuroectodermal tumor, whereas malignant epididymal tumors include adenocarcinoma, mesothelioma, and metastatic clear cell renal cell carcinoma (CCRCC).[1]

PCE is the second most common neoplasm of the epididymis after adenomatoid tumor.[2] It can occur sporadically or in association with VHL disease. In fact, there is a strong association with VHLD, wherein more than one-third of papillary cystadenomas of the epididymis (PCE) reported in the literature have occurred in patients with VHLD, particularly when bilateral tumors are present.[1, 3]

First described by Sherrick[4] in 1956, since then, approximately 70 cases are reported and published in literature.[1, 2, 4] Usually asymptomatic swelling of scrotum, many a times associated with pain and hydrocele also, which can mimic in symptomatology to metastatic CCRCC, clear cell papillary RCC, or some low grade mesothelial proliferation. Hence, excision biopsy with histopathological examination is necessary for timely management and to rule out association with VHL.

## Case Report

A 49-year-old male presented with hydrocele associated with scrotal pain since 2 months. In family, patient's father had history of renal cell carcinoma (RCC), and he expired. Jaboulay's procedure was done and the tissue sent for HPE. On gross examination, a grey-white, tiny, cystic soft tissue piece measuring $0.5 \times 0.4 \times 0.1$ cm was received. Microscopically, sections show small cyst lined by double layered cuboidal to columnar epithelium with focal papillae formation. The cells have abundant eosinophilic cytoplasm with blend features. No significant mitosis, necrosis, and pleomorphism seen. Diagnosis of papillary cystadenoma of epididymis was given.

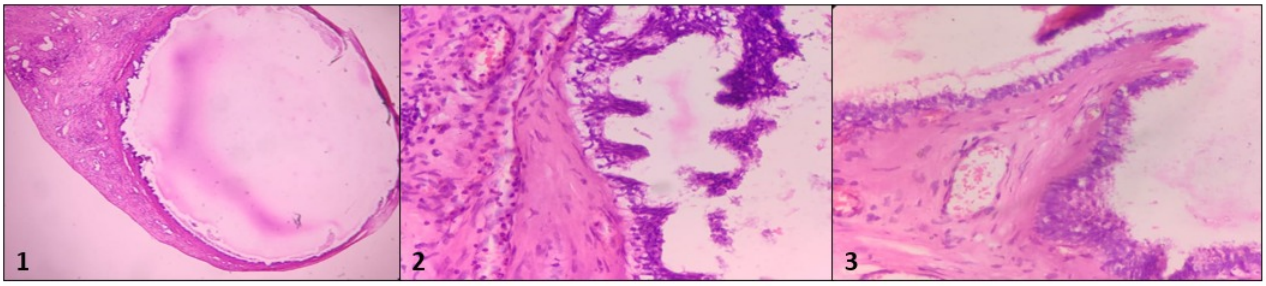


**Figure 1:** A: Cyst lined by cuboidal epithelium with focal papillae formation. (H & E stain, 40x) B: Papillae with stratification of epithelium. (H & E stain, 100x) C: Shows columnar epithelium. (H & E stain, 400x)

## Discussion

Papillary cystadenoma of the epididymis (PCE) is common benign lesion of male genital tract involving epididymis.[1] Approx 33% cases are associated with von Hippel-Lindau Disease (VHLD), a genetic condition that predisposes individuals to various tumors like as: clear cell RCC, hemangioblastomas, pheochromocytoma, pancreatic serous cystadenoma, etc. The average age of involvement is 35 years.[1]

In the studies of Lopez et al.,[3] Iczkowksi et al.,[5] and Shektai et al.[6] age of involvement between 30 to 50 years, which is in concordance with our case. Similarly, these case reports showed the unilateral scrotal swelling, as in our case.

The importance of this entity is to screen the patient for other lesions associated with VHL. In our case, the father of the patient expired due to RCC. Hence, thorough clinical evaluation is required for an occult renal cell carcinoma and other tumor in such cases.

## Conclusion

Although PCE is common lesion, yet rare in literature. Association with VHL and rare cases of transformation to carcinoma makes its diagnosis important.

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