Papillary cystadenoma of right testis: Case report and literature review

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ABSTRACT

Testicular cystadenoma is ranked the second commonest benign neoplasm. Other benign epididymal neoplasms include adenomatoid tumor (most common), leiomyoma, serous (nonpapillary) cystadenoma, cavernous hemangioma, and melanotic neuroectodermal tumor. Adenocarcinoma, mesothelioma, and metastatic renal cell carcinoma are malignant tumors that can affect the epididymis. A 24-year-old male undergraduate with a 3-month history of mildly tender right testicular swelling histologically diagnosed as papillary cystadenoma is presented. This case is presented from our locality as the first of its kind; and because it can be a possible manifestation of other diseases like von Hippel-Lindau (VHL) disease.

Key Words: Cystadenoma, Testes

1. INTRODUCTION

Tumours of the epididymis comprise about 5% of intrascrotal tumours. Papillary cystadenoma (PCE) of the testis is second only to adenomatoid tumour in this category.[1–4] It is an epithelial tumour believed to develop in the efferent ductules.[2–4] Sherrick was the first to describe it in a 21-year-old in 1956.[5] The history however dates as far back as 1921, when Brandt found an “epididymal cyst” in an autopsy of a patient for which von Hippel had previously reported retinal lesions.[6] Papillary cystadenoma can occur sporadically or as a manifestation of VHL disease (about 2/3 have VHL disease).[7–9] Tsuda et al. have described a familial pattern.[10] PCE shows a strong histologic resemblance to metastatic renal cell carcinoma.[7–9] Macroscopic lesions arise in 50% of VHL male patients and it is believed that microscopic lesions will be present in all male VHL patients.[9] The walls are usually studded with nodules of epithelial cells arranged in a globular or papillary configuration. Treatment is surgery with good prognosis. A few cases of late transformation into cystadenocarcinoma have been reported.[11]

2. CASE REPORT

A 24-year-old male with a 3-month history of mildly tender right testicular swelling. No history of trauma, abdominal pains or swelling. No history of impaired vision, breathing, hearing or gait disorders suggestive of VHL disease.

Examination revealed a young man in apparent general good health with vital signs within normal range. Both testicles were about 15 ml volume and the left epididymis as well as both vasa deferentia were essentially normal. There was a 1 cm firm oval nodule over the caput of the right epi-
didymis. There were bilateral varicoceles but no regional lymphadenopathy.

Testicular ultrasound scans revealed slight probe tenderness and a bulky right epididymis measuring 13.7 mm × 15.6 mm with a nodular hypoechoic area measuring 11.9 mm × 13.9 mm in size. It also showed marginal increase in blood flow on colour doppler.

Blood picture revealed packed cell volume of 42%, lymphocytes of 44% and serum prolactin of 13.4 ng/ml (3.6-16.3). Seminal fluid analysis was essentially normal. Urinalysis revealed no abnormal findings.

He underwent excision biopsy and histology showed tissues composed of dilated ducts with cystic spaces and papillary projections lined by a single layer of columnar cells, having vacuolated cytoplasm. The stroma was densely fibrous with focal areas of hyalinization and patchy chronic inflammation (see Figures 1 & 2). No evidence of malignancy was seen. The patient was requested to obtain abdomino-pelvic ultrasound and brain CT-scans for follow up.

![Figure 1. Papillary cystadenoma of the testis (H&E ×40)](image1)

3. DISCUSSION

Papillary cystadenoma is common in post-pubertal males 16-65 years (mean age 36).\(^7\) Unilateral cases are commoner and bilateral cases occur especially in patients that have been diagnosed with VHL disease before.\(^12\) In a study of 8 patients with unilateral PCE, none was found to develop VHL disease at a follow-up period of between 18 months and 15 years.\(^13\) In recent studies however, PCE or its precursors have been found in all the epididymii of patients having VHL disease that were examined.\(^14,15\)

Two-thirds of patients with PCE have an increased probability of developing benign cysts of the kidneys and pancreas, as well as tumours of the CNS, adrenals, and reproductive adnexial organs.\(^9\) This risk is higher in bilateral cases.\(^11\)

PCE is most commonly located at the head of the epididymis\(^7\) (as in our own case), although Gaffey et al. have described a solitary case in the broad ligament associated with VHL disease.\(^16\) It often presents as a painless, slowly growing scrotal swelling.\(^12\) Typically PCE is less than 5 cm in size.\(^8,17\)

Histologic characteristics are cysts filled with prominent intracystic papillary projections.\(^12\) These papillae contain fibrovascular cores lined by single layer of bland cuboidal or columnar epithelium.\(^12\) Cytoplasmic clearing is an obvious finding almost in all reported cases.\(^12\)

Metastatic renal cell carcinoma, a strong differential diagnosis of PCE, may co-exist with it in patients with VHL\(^7\) possibly due to the mesos nephric tissue origin of both renal tubules and the efferent ductules of the epididymis. The early recognition and management of this tumour may help prevent possible late malignant transformation and also pave way for early identification of more pathologic entities like VHL and its associated lesions.
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