

Case Report

Papillary cystadenofibroma of epididymis: a case report

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Received August 7, 2011; accepted August 9, 2011; Epub August 10, 2011; published August 15, 2011

Abstract: We present the first reported case of papillary cystadenofibroma of the epididymis. The tumor occurred in a 46-year-old man. The mass was 3.7 cm and included a hemorrhagic fluid-filled cyst. Microscopically, stromal-filled papillae were lined by low cuboidal to columnar epithelium. Epithelial cells were reactive for cytokeratin 7, cytokeratins AE1/3, and focally in the apical cytoplasm for CD10. Focal CD10 reactivity was also noted in the stroma. The lesion was negative for alpha-fetoprotein. These findings ruled out other lesions, including metastatic renal cell carcinoma.

Keywords: Papillary, cystadenofibroma, epididymis

Case history

We present the case of a 46-year-old man who had a 1-2 month complaint of right testicle enlargement. An ultrasound revealed a 3.9-cm intratesticular mass thought to represent a complex cyst with possible solid areas. The patient's serum alpha-fetoprotein was within normal range at 2.9, and his beta-hCG was undetectable. A right radical orchiectomy was performed, revealing a large cyst that contained altered blood and some excrescences near the hilum of the cyst, measuring up to 0.7 cm, that were suspicious for tumor.

Results

Microscopy revealed a cyst cavity, containing papillary structures supported by dense spindle cell stroma (Figure 1). This was lined by cuboidal cells, some of which were ciliated (Figure 2) and were focally crowded but lacked cytologic atypia and mitotic activity. The cyst lining was in continuity with a rim of residual epididymis. The adjacent testicular parenchyma that was sampled was normal. Epithelial reactivity was seen with cytokeratin 7 (Figure 3) and cytokeratins AE1/3. CD10 showed focally moderate reactivity in the stromal cells as well as rare staining of apical cytoplasm of the epithelial cells (Figure 4). There was no reactivity for alpha-

fetoprotein, ruling out a papillary yolk sac tumor. Outside consultative opinions confirmed the diagnosis.

Discussion

Papillary cystadenofibroma of the testis or epididymis has not been reported in the Medline literature. About 70 cases of papillary cystadenoma of the epididymis have been reported [1]. These occurred in men from ages 16 to 81; about 40% were bilateral, and about two-thirds occurred in conjunction with von Hippel-Lindau syndrome [2, 3] with lesions of the cerebellum, cerebrum, spinal cord, retina, pancreas or urinary bladder. Unlike in the epididymis, cystadenofibroma of the ovary [4] and fallopian tube [5], are fairly well-documented, and are thought to be of paramesonephric origin [5].

In this case, other cystic lesions of the testis or epididymis could be excluded. The differential diagnosis included tumors of mesothelial origin, which should not have ciliated cells or ovarian type stroma. There was focal atypia and crowding of nuclei, but these features comprised less than 10% of the tumor and were insufficient to diagnose a lesion of borderline malignant potential. Dermoid or epidermoid cysts would have characteristic squamous epithelium. Teratoma would have other heterologous glandular

Papillary cystadenoma of epididymis

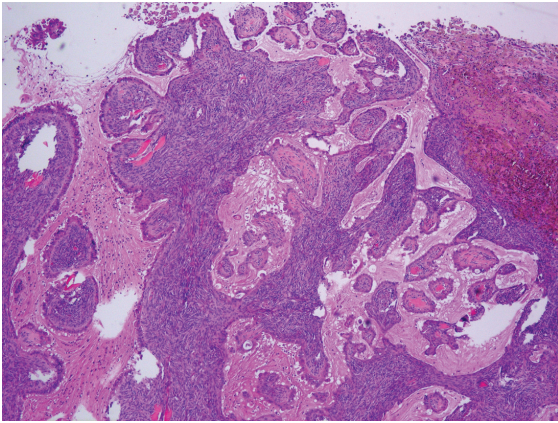


Figure 1. The tumor consists of a cystic space with papillary excrescences and hemorrhagic fluid (5x objective).

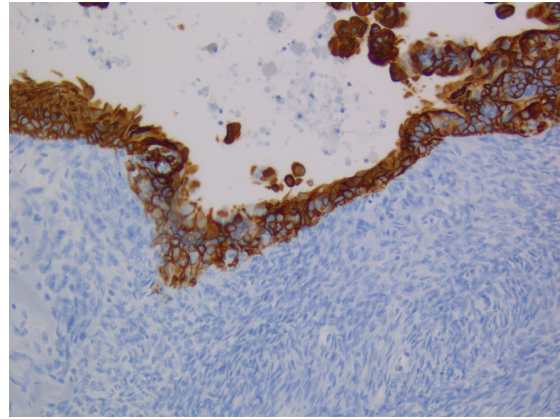


Figure 3. Epithelial reactivity for cytokeratin 7 (40x objective).

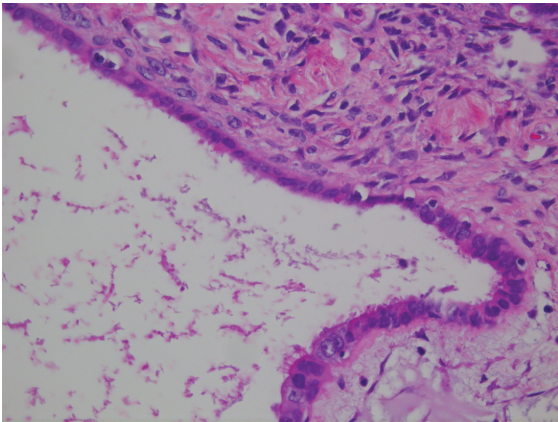


Figure 2. At high power, there are dilated spaces lined by papillae with stromal cores and a single to double layer of cuboid epithelium. Some cells (bottom) show cilia (40x objective).

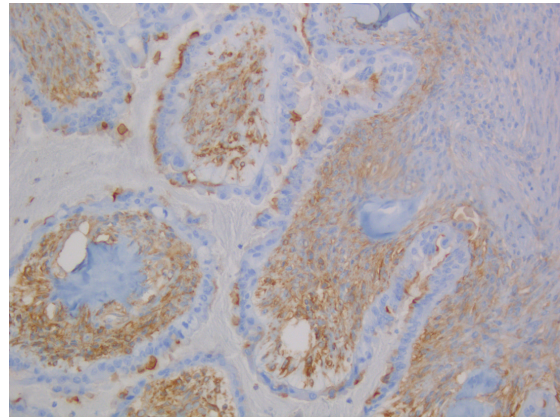


Figure 4. CD10 shows stromal and focal epithelial apical reactivity (40x objective).

and stromal elements. Finally, cystic trophoblastic tumor, a variant of choriocarcinoma, would be lined by atypical trophoblast cells.

Metastatic clear cell renal cell carcinoma is ruled out by the strong reactivity for CK7, and only focal apical reactivity for CD10. CD10 has been applied to this differential diagnosis in other cases of papillary cystadenoma of the epididymis. The cystadenomas showed no CD10 reactivity in 2 case reports [6,7] and reactivity in only one of five cases reported in another report [8]. The current case is unique because of its prominent fibrous component,

consistent with a cystadenofibroma. The CD10 immunostain was focally reactive in the stroma, as well as rarely in the apical cytoplasm of the epithelium as reported previously [8]. The significance of the stromal CD10 staining is not clear, although CD10 stromal staining has been reported in endometriosis [9] and in a minority of cases of breast fibroadenoma and phyllodes tumors [10].

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