Paratesticular Papillary Serous Tumor of Low Malignant Potential – A Case Report –

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Paratesticular papillary serous tumors have been rarely reported, and they often resemble ovarian serous tumors of borderline malignancy. We experienced a case of papillary serous tumor in the left paratestis of a 39-year-old man. This is the second case reported in the Korean literature. The tumor, which was found incidentally during an operation for a hydrocele, was composed of papillary structures lined by cuboidal to columnar epithelial cells that displayed low cytologic atypia and frequent psammoma bodies.

Key Words: Papillary serous tumor; Paratestis

In 1974, Herschman and Ross¹ first described a case of serous cystadenoma within the testis, and the tumor closely resembled its ovarian counterpart. Paratesticular papillary serous tumors are assumed to be derived from müllerian metaplasia of the mesothelium that comes from the tunica vaginalis or from a müllerian remnant in the testis. We report here on a case of paratesticular papillary serous tumor.

CASE REPORT

A 39-year-old man visited Chungnam National University Hospital for a painless scrotal swelling. Scrotal sonography revealed some fluid collection in the left scrotum. The patient then went through a left hydrocelectomy procedure. Approximately 100 mL of yellowish fluid was present in the tunica vaginalis, and multiple yellowish plaque-like lesions were also found in the left testis and epididymis. Some of the lesions were sent off to the laboratory for biopsy. Our initial diagnosis was consistent with papillary serous tumor of low malignant potential. A left radical orchiectomy was performed 17 days after biopsy.

On gross examination, we observed multiple, firm, gritty, plaque-like lesions that were attached onto the tunica vaginalis and epididymis (Fig. 1). On microscopic examination, the lesions were partially cystic with numerous intracystic blunt papillae. These papillae had wide fibrovascular cores and they were lined by cuboidal to columnar epithelial cells that displayed minimal to mild cytologic atypia with focal stratification (Fig. 2). There were one or two mitotic figures per 10 high power fields. There was no evidence of stromal invasion. Psammoma bodies were diffusely scattered, and a few foreign body giant cells, focal hyalinization and nonspecific inflammatory cell infiltration were also observed. The tumor cells were reactive for cytokeratin (CK) 7, and they were negative for CK20, estrogen receptor, progesterone receptor, CD15, carcinoembrionic antigen (CEA), and calretinin (Fig. 3).

DISCUSSION

Testicular and paratesticular neoplasms that resembe ovarian serous tumors have been rarely described; there are only approximately twenty such cases reported in the English literature. ¹⁻⁷ This case is only the second case reported in the Korean literature. The first reported case was a serous papillary cystadenoma arising in the paratesticular tissue of a 12-year-old boy. The microscopic findings in that case were intraluminal papillae that were lined by both ciliated and nonciliated secretory cells, and a similar cellular morphology was described in the salpinx. Electron microscopy also revealed ciliated columnar epithelial cells, but immunohistochemical study was not performed in this case.

Our 39-year-old patient was presented with a painless scrotal swelling. On physical examination, there were multiple tiny nodules observed on the left scrotum. The light microscopic findings of this lesion resembled that of the ovarian serous tumors

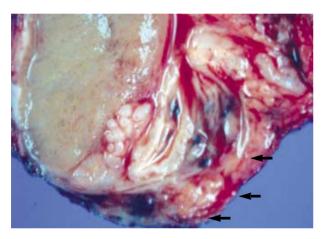


Fig. 1. Multiple, firm, gritty, plaque-like lesions (arrows) are attached on the tunica vaginalis and the epididymis.

of borderline malignancy. Multiple psammoma bodies were seen and both the paratesticular tissue and the tunica albuginea were involved with the tumor. We could not find any ciliated cells on light microscopy. Electron microscopy was not performed. The immunohistochemical findings were similar to those findings of ovarian serous tumors. The tumor cells were reactive for CK7, and the tumor cells were negative for CK20, CEA and calretinin.

A differential diagnosis for papillary serous tumor in paratesticular region includes malignant mesothelioma, adenocarcinoma of the rete testis, epididymal adenocarcinoma and metastatic carcinoma. Malignant mesothelioma shows either an epithelial, sarcomatoid or biphasic pattern. The epithelial mesotheliomas

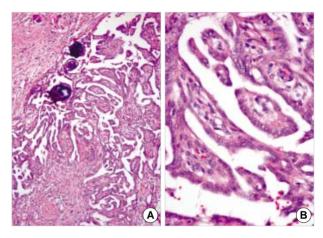


Fig. 2. (A) Low power light microscopic view shows numerous, intracystic, blunt, papillary structures with a few psammoma bodies. (B) High power view shows cuboidal to columnar lining cells having minimal to mild cytologic atypia and focal stratification.

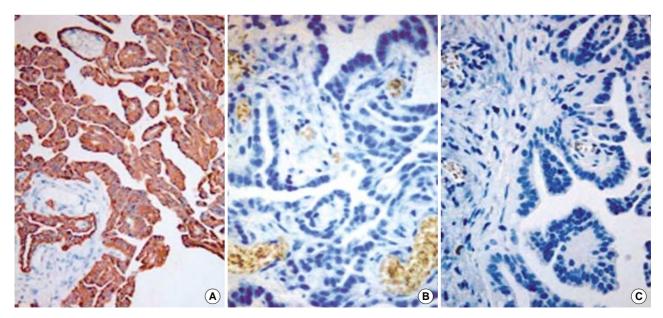


Fig. 3. (A) Tumor cells show strong positivity for cytokeratin 7. (B & C) The tumor cells are negative for cytokeratin 20 (B) and calretinin (C).

may be papillary, glandular or solid. Psammoma bodies may be seen, but they are not generally very numerous. A high mitotic rate, significant nuclear atypia, prominent nucleoli and invasion of adjacent structures are also seen. Mucin stains (periodic acid Schiff and mucicarmine) and immunostainings for CEA and factor VIII are negative, but stainings for pan-CK, CK 5/6, epithelial membrane antigen (EMA) and calretinin are positive. ⁹ This case was not reactive for calretinin. Adenocarcinoma of the rete testis requires the following criteria for diagnosis: 1) the absence of histologically similar extratesticular tumor, 2) the tumor centered in testicular hilum, 3) a morphology incompatible with any other type, 4) the presence of a transition from unaffected rete testis to tumor, and 5) a predominantly solid growth pattern.¹⁰ This tumor is usually reactive for CK, CEA and EMA.¹¹ However, our case was not reactive for CEA. Epididymal adenocarcinoma shows invasive tubules, papillae, and sheets of cells with clear to eosinophilic cytoplasm.¹² Our case did not show any clear cells. Metastatic tumors generally occur in patients having a known clinical history, and these cancers are positive for CEA. Our patient did not have any suspected cancerous condition previously, and the tumor cells were not reactive for CEA.

Testicular and paratesticular tumors of the müllerian type seem to arise from müllerian metaplasia of the peritoneal lining of the tunica vaginalis, appendix testis or müllerian remnants between the testis and the spermatic cord.^{2,9} Although a variety of müllerian epithelial subtypes in this location have been described by far, the most common subtype is the serous tumor. To date, any serous "borderline" tumor of the paratesticular region in the literature was not associated with recurrence or metastasis after resection.³ Only single case of serous cystadenoma of borderline malignancy has been reported to display focal transition into invasive cancer.⁴

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