A Case of Intratesticular Endometrioid Papillary Cystadenocarcinoma

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We report a case of intratesticular endometrioid papillary cystadenocarcinoma. A 73-year-old man was admitted for a painless right scrotal swelling. Ultrasonography and computed tomography revealed a large cystic mass in the right testis. Right scrotum puncture revealed xanthochromic fluid with negative cytology. Three months later, follow-up computed tomography showed enlargement of the cystic mass. Right high orchiectomy was performed because a testicular malignancy was suspected. The pathological diagnosis was endometrioid papillary cystadenocarcinoma, and the cells were strongly positive for the estrogen and progesterone receptors. Testicular neoplasms resembling common ovarian-type epithelial tumors are very rare. This is the first report of endometrioid papillary cystadenocarcinoma of the testis.

Key words: testicular tumor – endometrioid carcinoma – estrogen receptor – progesterone receptor

INTRODUCTION

Endometrioid papillary cystadenocarcinoma of the testis is a rare neoplasm that is practically indistinguishable from that found in the ovary (1). The histology of ovarian-type epithelial tumors of the testis has been classified into the following types: borderline microinvasive serous tumor and serous carcinoma, intra- and para- testicular mucinous tumor (with different grades of malignancy), and endometrioid adenocarcinoma and Brenner (transitional cell) tumor (2). The histogenetic of ovarian-type epithelial tumors may be associated with remnants of müllerian tissue or müllerian metaplasia of the tunica vaginalis in a testis or paratestis (3). Here we present a case of endometrioid papillary cystadenocarcinoma of the testis.

CASE REPORT

A 73-year-old man was admitted for a painless right scrotal swelling that persisted for a year and rapidly increased in size over the past month, causing discomfort in the right groin. Physical examination revealed swelling of the right hemiscrotum and a normal testis on the left. Ultrasonography showed a heterogeneous cystic lesion with a maximum diameter of 5 cm in the right testis. Serum human chorionic gonadotropin (hCG) and alpha-fetoprotein (AFP) levels were within normal limits. Computed tomography (CT) demonstrated a cystic mass in the right testis without invasion to adjacent structures or distant metastasis (Fig. 1). Right scrotum puncture revealed xanthochromic fluid with negative cytology. These clinical findings enabled careful monitoring of the patient without treatments. However, a follow-up CT carried out 3 months later demonstrated enlargement of the right cystic mass. Right high orchiectomy was performed because a malignant testicular tumor was suspected.

Pathological examination of the surgical specimen revealed a 9 cm multilocular cystic lesion in the testis containing viscous fluid. The lining surface showed several grayish-white papillary excrescences (Fig. 2). The epididymis and spermatic cord appeared normal. Microscopically, the cyst wall was composed of dense fibrous tissue lined by a partially ciliated cuboidal and columnar epithelium, and intracystic excrescences composed of fibrovascular villous structures lined by stratified columnar epithelium with formation of micropapillary tufts (Fig. 3). The cells showed mild nuclear pleomorphism but no increased mitotic activity. Detached clusters of epithelial cells were present in the cyst space. The lining...
tumor cells were strongly positive for estrogen receptor (ER), progesterone receptor (PgR) and placental alkaline phosphatase (Fig. 4A and B). Immunohistochemical staining of other testicular tumor markers such as carcinoembryonic antigen (CEA), AFP and hCG were negative. The surrounding compressed testicular tissue as well as the epididymis and spermatic cord were unremarkable. Consequently, this case was diagnosed as endometrioid papillary cystadenocarcinoma.

The postoperative period was uneventful, and no recurrence has been observed after 3 years of follow-up.

**DISCUSSION**

Testicular and paratesticular tumors resembling ovarian neoplasm of the common epithelial type are rare and not well described in the medical literature. The first series of cases was reported in 1986 by Young and Scully, in which 3 original cases and 11 reviewed previous cases were documented (4). Serous-type tumors are the most predominant among common ovarian-type epithelial tumors arising in the testis, and other types, including Brenner and mucinous tumors, reportedly occur (5). Common ovarian-type testicular tumors are usually benign or of borderline malignancy. Endometrioid carcinoma of the testis is extremely rare and has not yet been reported.

Although the mean age of patients with common ovarian-type testicular tumors is 56 years (14–77 years) (6), patients who developed the disease in childhood have also been reported. The most common clinical presentation is an indolent testicular mass, which cannot be clinically and completely distinguished from other neoplasms. The tumors are usually treated by orchiectomy alone and have a good clinical course without recurrence or metastasis (7). The light microscopic features are identical to their ovarian counterpart, showing a papillary pattern with mild atypia and microstromal invasion (8). In addition, the present case shows a dense fibroblastic stroma resembling an ovarian type (8).

Recently, testicular and paratesticular tumors of common ovarian-type epithelial tumors have been studied by immunohistochemistry profiling, electron microscopy and DNA ploidy analysis. Immunohistochemical analysis of endometrioid adenocarcinoma has proved to be identical to that of the ovarian counterpart with positivity to cytokeratin, vimentin and absence of CEA. In addition, expressions of ER and PgR are generally positive in endometrioid adenocarcinoma but absent in serous carcinoma (9). In the present case, immunohistochemical findings, positive staining of ER and PgR, and negative staining of CEA strongly support the diagnosis of endometrioid carcinoma.

The origin of common ovarian-type epithelial tumors in the testis and paratesticular tissue remains unclear. Several
possibilities have been suggested in previous literature. The most reasonable origin is remnants of müllerian ducts persisting in the male appendix, testis or extratesticular scrotal contents such as the epididymis, connective tissue between the testis and epididymis and spermatic cord. Another possibility is müllerian metaplasia of the tunica vaginalis. Intratesticular tumors may develop from mesothelial inclusions or represent monodermal teratoma (10). In the present case, dense stroma resembling ovarian-type cortical stroma was present in the tumor, further supporting a mesothelial origin of these tumors. A testicular endometrioid adenocarcinoma has a very peculiar microscopic finding. The rarity of the histopathology in this location may lead to difficulty in diagnosis, and the tumor may be confused with other more aggressive tumors such as mesothelioma and metastatic carcinoma (11). The histological distinction between mesotheliomas and ovarian-type epithelial tumors may be difficult, but it can be facilitated by the use of immunohistochemistry. Calretinin, thrombomodulin and keratin 5/6 were the best positive mesothelioma markers, and MOC-31, B72.3, Ber-EP4, CA19-9 and leu-M1 (CD15) were the best negative mesothelioma markers for differentiating mesotheliomas from ovarian-type epithelial tumors (12).

Conflict of interest statement
None declared.

References