Title: Multicystic malignant mesothelioma of the tunica vaginalis with an unusually indolent clinical course

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Citation: 泌尿器科紀要 (2004), 50(7): 511-513

Issue Date: 2004-07

URL: http://hdl.handle.net/2433/113402

Type: Departmental Bulletin Paper

Publisher: Kyoto University
We report an extremely rare case of a multicystic malignant mesothelioma in the tunica vaginalis with an unusually indolent clinical course. A 48-year-old man presented with a one-month history of painless swelling of right scrotal contents. Ultrasonography and computed tomography (CT) revealed a multicystic mass in the right scrotal sac with no invasion into the testis, epididymis or scrotum. The patient has been disease-free for 72 months and is being followed on an outpatient basis with no further adjuvant therapy.

**Key words:** Multicystic malignant mesothelioma, Tunica vaginalis testis, Indolent clinical course

**INTRODUCTION**

Multicystic malignant mesothelioma of the tunica vaginalis is an extremely rare form of scrotal tumor. The aggressive biological behavior of malignant mesothelioma results in a poor prognosis. In this case report we describe a case in a patient remaining well 6 years after diagnosis and radical orchiectomy.

**CASE REPORT**

A 48-year-old man presented with a one-month history of painless swelling of right scrotal contents. Ultrasonography (US) (Fig. 1) and computed tomography (CT) revealed a 9.5×9.0×9.0 cm, 250 g, multicystic mass in the right scrotal sac with no evidence of distant and lymph node metastases. The testicular tumor markers (human chorionic gonadotropin-β and alpha fetoprotein) and carcino-embryonic antigen (CEA) were within normal limits. Inguinal orchiectomy was intraoperatively decided under the suspicion of malignant tumor. Macroscopic appearance showed marked multicystic degeneration of tunica vaginalis, but the testis and epididymis were not affected. The tumor consisted of fibrocellular, microcystic and adenomatoid elements microscopically was diagnosed biphasic malignant mesothelioma of tunica vaginalis (Fig. 2). Although the tumor was negative for periodic acid schiff, and also negative for CEA, and myelomonocytic antigen (Leu-M1), cytokeratin and vimentin were immunohistochemically detected especially in the epithelial component. There were no tumor cells in the testis, epididymis or scrotum. Therefore, no further adjuvant therapy was performed on the patient. The patient has been...
disease-free for 72 months and is being followed on an outpatient basis.

**DISCUSSION**

Although the origin of tunica vaginalis is embryologically the same as that of pleura and peritoneum, malignant mesothelioma of the tunica vaginalis is relatively rare. Multicystic malignant mesothelioma of the tunica vaginalis is an extremely rare form of scrotal tumor. Previously, approximately 110 cases of malignant mesothelioma originating in the tunica vaginalis have been reported, and none of them showed multicystic form of malignant mesothelioma originating in tunica vaginalis. Wagner et al. advocated that malignant mesothelioma seems to be related to a history of exposure to asbestos. However, in our case there was no history of exposure to asbestos. It is difficult to diagnose the disease preoperatively, and most cases are preoperatively diagnosed as a hydrocele or a suspected testicular tumor. In our case, a multicystic mass detected by US and CT in the right scrotal sac was preoperatively diagnosed as a suspected testicular tumor. The inguinal orchiectomy is the standard treatment for malignant mesothelioma of the tunica vaginalis. Effective treatment by radiation therapy or immunotherapy with lymphokine-activated killer T cells and interleukin 2 has been reported, but the prognosis was poor. Malignant mesothelioma is classified histologically into three groups: the epithelial type, the sarcomatous type, and the biphasic type. However, the prognosis is not affected by histological pattern. Previously, Plas et al. reported that the median survival period was 24 months with a range of 2–64 months. Only 2 of the 110 cases previously reported survived more than 5 years after radical orchidectomy. In this case, the patient showed an unusually indolent course with a disease-free period of 72 months.

Although a long-term observation will be necessary to evaluate the prognosis, this case suggested that radical inguinal orchiectomy might contribute to a better prognosis.

**REFERENCES**


(Received on October 6, 2003)
緩徐な経過を示した精巢鞘膜原発多囊胞性悪性中皮腫

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緩徐な経過を示した精巢鞘膜原発多囊胞性悪性中皮腫の1例を経験した。48歳の男性が1カ月前より右陰囊の無痛性腫脹を主訴に当科受診した。超音波検査およびCTで右陰囊内に多囊胞性腫瘍を認めた。組織検査およびリンパ節転移は認めなかった。また精巣腫瘍マーカーはいずれも正常範囲内であった。悪性腫瘍を疑い精巣摘出術を施行した。多囊胞性腫瘍はfibrocellular, microcystic, adenomatoidな成分で構成され、精巣鞘膜原発のbiphasic malignant mesotheliomaと病理学的に診断された。精巣、精巣上体、陰囊への浸潤は認めなかった。術後補助療法なしで、術後72カ月経過した現在も再発転移は認めていない。

（泌尿紀要 50: 511-513, 2004）