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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
Right	
Type	Departmental Bulletin Paper
Textversion	publisher

MULTICYSTIC MALIGNANT MESOTHELIOMA OF THE TUNICA VAGINALIS WITH AN UNUSUALLY INDOLENT CLINICAL COURSE

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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 evidence of neither distant nor lymph node metastases. The testicular tumor markers were within the normal limits. Inguinal orchiectomy was performed under the suspicion of a malignant tumor. The cystic tumor consisted of fibrocellular, microcystic and adenomatoid elements microscopically was diagnosed biphasic malignant mesothelioma of tunica vaginalis but no invasion into the testis, epididymis and also scrotum. The patient has been disease-free for 72 months and is being followed on an outpatient basis with no further adjuvant therapy.

(Acta Urol. Jpn. 50 : 511–513, 2004)

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 as 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



Fig. 1. Ultrasonography revealed a 9.5×9.0×9.0 cm multicystic mass in right scrotal sac.

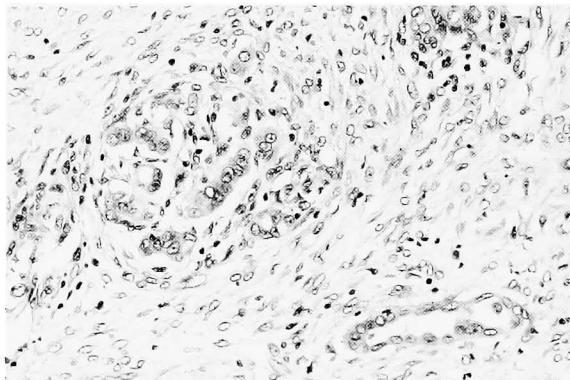


Fig. 2. Pathological findings showed the biphasic malignant mesothelioma of tunica vaginalis with fibrocellular, microcystic and adenomatoid elements (H & E, ×200 magnification).

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^{1,2)}, and none of them showed multicystic form of malignant mesothelioma originating in tunica vaginalis. Wagner et al.³⁾ 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 poor⁴⁾ 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 months¹⁾. Only 2 of the 110 cases perviously reported survived more than 5 years after radical orchidectomy^{1,2)}. 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.

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(Received on October 6, 2003)
(Accepted on March 15, 2004)

和文抄録

緩徐な経過を示した精巣鞘膜原発多嚢胞性悪性中皮腫

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緩徐な経過を示した精巣鞘膜原発多嚢胞性悪性中皮腫の1例を経験した。48歳の男性が1カ月来の右陰嚢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で右陰嚢内に多嚢胞性腫瘍を認めた。遠隔臓器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。悪性腫瘍を疑い高位精巣摘除術を施行した。多嚢胞性腫瘍は

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、陰嚢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)