

MALIGNANT MESOTHELIOMA OF THE TUNICA VAGINALIS TESTIS: A CASE REPORT AND LITERATURE REVIEW

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Malignant mesothelioma of the tunica vaginalis testis is a rare but often fatal malignancy. Here, we report one patient with locally advanced disease who has a history of asbestos exposure. We review the literature concerning current management strategies of the disease. Radical surgery plus adjuvant radiotherapy seems to provide the best results.

Key Words: asbestos, malignant mesothelioma, tunica vaginalis
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Malignant mesothelioma is a very rare [1] but often fatal type of testicular malignancy that originates from mesenchymal tissue [1–4]. It usually presents as an incidental finding at the time of hydrocele surgery [4], particularly in patients with prior exposure to asbestos [5–7]. Here, we describe a Taiwanese patient with malignant mesothelioma of the tunica vaginalis, together with a review of the literature.

CASE PRESENTATION

A 67-year-old man, who was previously diagnosed with infected right hydrocele in 2004 and had long-standing bilateral hydrocele for 30 years, visited our clinics with presentation of painless enlargement of the right scrotum for 2 months. Grossly, the tumor had a size of 8 × 10 × 15 cm and occupied the whole right scrotum up to the right external inguinal canal. Ultrasonography revealed left hydrocele and heterogeneous right testicular tumor. The subsequent abdominal

and pelvic computerized tomography (CT) demonstrated a localized heterogeneous right scrotal tumor without evidence of local lymphadenopathy (Figure 1). Elevated serum β-HCG (27.5 mIU/mL) was detected. Surgical exploration through scrotal incision was performed. The stony hard lesion with purulent discharge had destroyed the right testicle and had invaded the right external inguinal canal. Local resection was done because of chronic inflammation of thickening tunica vaginalis, as confirmed by frozen section examination. Pus culture grew enterococcus species.

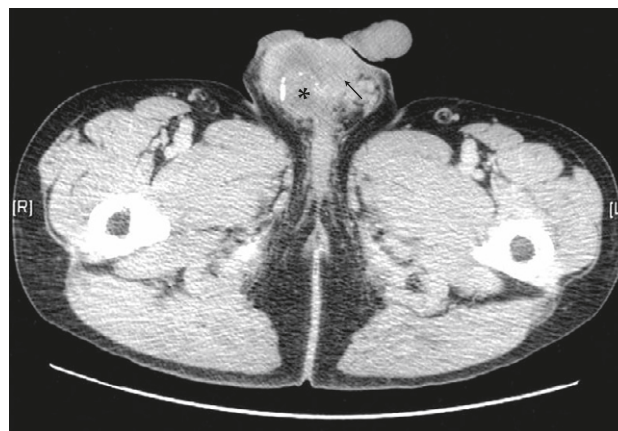


Figure 1. Computed tomography of the testis shows one heterogeneous tumor (arrow) involving the scrotal wall and invading the right testicle (asterisk).



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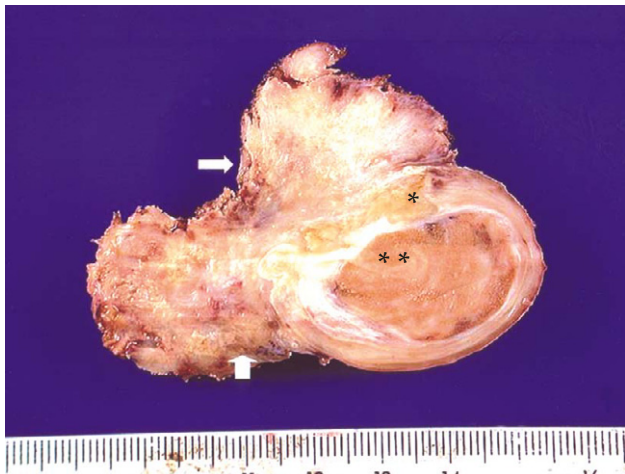


Figure 2. Gross inspection of the bivalve specimen. Note the tumor (arrows) arising from the tunica vaginalis (asterisk) and invading the testicular parenchyma (double asterisks).

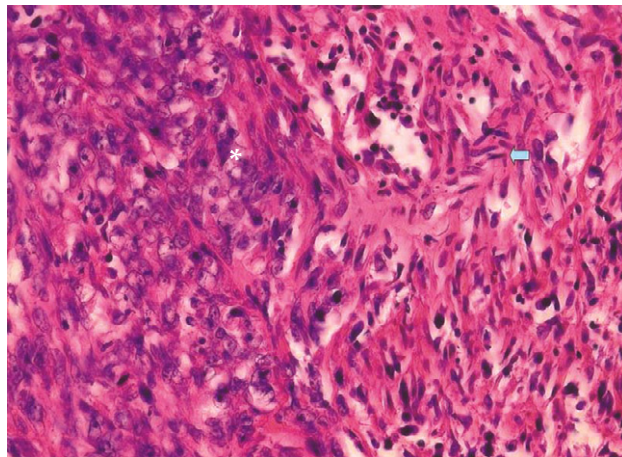


Figure 3. Histology of malignant mesothelioma of the tunica vaginalis testis in a biphasic pattern (hematoxylin & eosin, 400 \times). The epithelial (asterisk) and sarcomatous (arrow) types exist concomitantly.

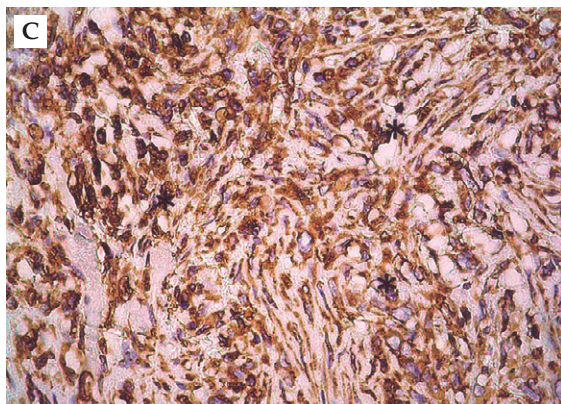
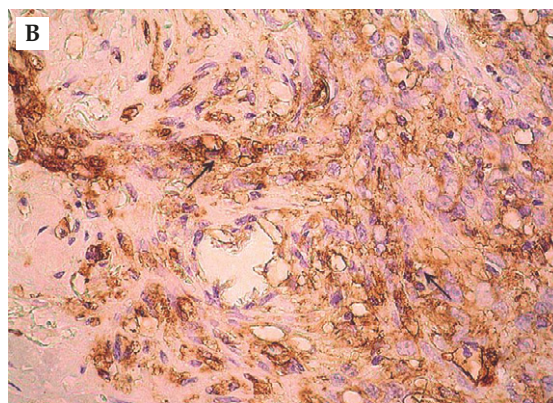
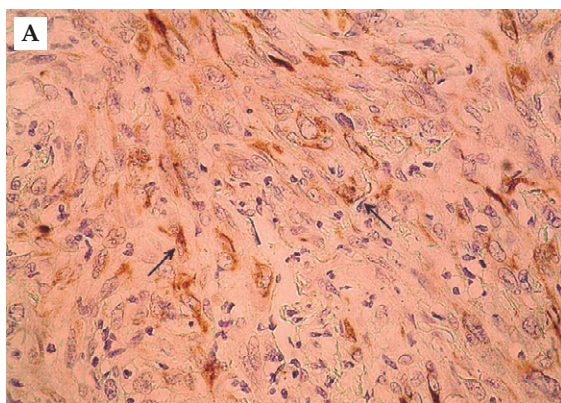


Figure 4. Immunohistochemistry of malignant mesothelioma of the tunica vaginalis testis: (A) CK(++); (B) EMA(++); (C) vimentin (++) (LSAB, 400 \times).

However, pathology demonstrated malignant mesothelioma arising from the tunica vaginalis of the testis (Figure 2). Histopathology was characterized by a biphasic pattern with admixed epithelial and spindle cell proliferation (Figure 3). Immunohistochemical staining showed CK (++) (Figure 4A) and EMA (++)

(Figure 4B) positivity in epithelial nests, while the spindle cell was positive for vimentin (++) (Figure 4C) and negative for desmin (-). Because of poor wound healing, wide resection was performed to achieve an adequate margin 1 week postoperatively. His postoperative course was smooth and the wound

healed well. No local recurrence has been noticed at regular clinic follow-ups for 7 months.

Retrospectively, the patient had short-term occupational exposure to asbestos 40 years ago.

DISCUSSION

Since the first case of malignant mesothelioma at the tunica vaginalis of testis was described by Barbera and Rubino in 1957 [1], approximately 100 cases have been reported. Malignant mesothelioma of pleura, pericardium, and peritoneum are uncommon and malignant mesothelioma originating in the tunica vaginalis is extremely rare. In the United States the incidence of malignant mesothelioma is about 11 cases per million people per year [2]. About 68–85% of the mesotheliomas arise in the pleura, 9.1–24.1% in the peritoneum, and only 0.3–5% in the tunica vaginalis testis [2–4,8]. In patients with malignant mesothelioma at the tunica vaginalis of the testis, more than two-thirds of the cases were patients older than 45 years of age with a median age of 60 years [4], although malignant mesothelioma has been reported in a 10-year-old child [9]. Patients usually present with hydrocele (56.3%) and sometimes a testicular tumor (32.8%) [4]. Although the diagnosis of malignant mesothelioma of the tunica vaginalis is seldom observed preoperatively, it should be considered in any patient presenting with scrotal pathology with a history of exposure to asbestos [5–7]. Direct contact with asbestos (34.2–41%) [4,10] or a familial occupational history can significantly increase the risk of developing a malignant mesothelioma by a factor of 10 [11–13]. In addition to asbestos, potassium bromate in drinking water was shown to increase the risk of developing malignant mesothelioma of the tunica vaginalis testis in an animal model [14] and long-standing hydrocele was also considered to be a risk factor of malignant mesothelioma of the tunica vaginalis, as proposed by Gurdal and Erol [15]. The localization of the tumor is presented with equal incidence in both testicles [4], and the presence of bilateral malignant mesothelioma in the tunica vaginalis is very rare, with only three cases presented before the present case [16–18].

According to different histopathologic traits, malignant mesothelioma has been subclassified into three types: epithelial type, the most frequently seen (60.8–75%) in the peritoneal cavity and tunica vaginalis; the biphasic type, as reported in this case, occurring in the

serosa membrane (25–37.3%); and the mesenchymal or sarcomatous type, which is found in the pleural cavity (1.9%) [3,9]. Malignant mesothelioma has an expansive and infiltrative growth pattern and nearly 40% of the patients presented initially presented with local invasion, and the two most frequently involved sites were subtunical connective tissue (25.8%) and testicular parenchyma (19.4%). Metastasis occurs early via the lymphatic system to inguinal, para-aortic or supraclavicular nodes, and was reported in 14.9–31% of cases [4,19]. In 11 patients with primary metastatic disease in the study by Plas et al, retroperitoneal lymph nodes were most involved in five cases, followed by inguinal nodes in three and iliac nodes in two. In cases showing disease progression, the common sites of metastasis were lymph nodes (13.8%), lung (9.7%) and liver (4.2%) [4]. In the study by Plas et al, tumor recurrence was more than 60% within 2 years and over 90% within 5 years [4]. Radical primary resection may decrease the rate of tumor recurrence.

Univariate analysis to assess the prognostic parameters revealed a significantly better correlation in patients of younger age. However, in a multivariate Cox's regression model, there was no statistically significant result [3]. The mean disease-specific survival for patients with or without systemic treatment was 26 and 36 months, respectively [20]. Although some cases were responsive to radiotherapy or chemotherapy [21–24], adjuvant treatments such as chemotherapy or immunotherapy have limited effects in advanced disease [4]. Radiotherapy appeared to be more effective than chemotherapy and as good as the combination of chemotherapy and radiotherapy in the patients with metastatic disease [4]. The DNA hypomethylating agent 5-aza-2'-deoxycytidine may be effective based on the induction and upregulation of the expression of cancer/testis antigen [25].

Malignant mesothelioma of tunica vaginalis is a rare but often fatal malignancy. It should be considered a differential diagnosis of inguinoscrotal mass, particularly in patients with exposure to asbestos. Despite aggressive surgical procedures or systematic adjuvant therapies, the prognosis remains poor.

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睪丸白膜鞘惡性間質細胞瘤： 病例報告與文獻回顧

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睪丸白膜鞘惡性間質細胞瘤是一種少見但致命的惡性腫瘤。我們報告一位有石棉接觸史且局部大範圍侵犯的睪丸白膜鞘惡性間質細胞瘤病人，並且回顧關於目前治療方針的相關文獻，現今仍以廣泛切除加上放射線治療為主。

關鍵詞：石棉，惡性間質細胞瘤，睪丸白膜鞘
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