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Mesothelioma in Situ of the Spermatic Cord Arising from a Patent Processus Vaginalis: A Case Report

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Word counts: Abstract, 90 words; Main text, 598 words

Number of pages and figures: 10, 3

Key words: atypical mesothelial cells; mesothelioma in situ; radical orchiectomy; spermatic cord tumor

ABSTRACT

Mesothelioma is an aggressive tumor originating from mesothelial cells. Mesothelioma of the spermatic cord is a very rare disease, and the most common presentation of this disease is that of aggressive mesothelioma with no description of mesothelioma in situ. We report an extremely rare case of mesothelioma in situ of the spermatic cord arising from a patent processus vaginalis. To our best knowledge, this is the first report of this finding. The identification of a patent processus vaginalis and investigation of single-layered atypical mesothelial cells led to the final diagnosis.

Keywords: atypical mesothelial cells; mesothelioma in situ; radical orchiectomy; spermatic cord tumor

INTRODUCTION

Mesothelioma is an aggressive tumor originating from mesothelial cells [1]. Mesothelioma of the spermatic cord is a rare disease, with only 9 cases reported in detail in the English literature and none describing mesothelioma in situ [2-10]. We report an extremely rare case mesothelioma in situ of the spermatic cord arising from a patent processus vaginalis.

CASE REPORT

An 82-year-old man complaining of a gradually enlarging swollen mass in his right inguinal area for the past three months was referred to our hospital. The right spermatic cord mass was hard, smooth and immobile. Chest and abdominal computed tomography showed right spermatic cord swelling and no other significant findings (Fig. 1). We diagnosed the patient as having right spermatic cord tumor, for which he underwent right radical orchiectomy. A spermatic cord tumor was found adhering to the surrounding tissue, and no obvious residual tumor existed intraoperatively.

Macroscopically, the spermatic cord was enlarged at 7.0 × 4.0 cm and had a smooth and capsulated surface. The cut surface was creamy white, solid, thickened and poorly circumscribed (**Fig. 2A, B**). Microscopically, the tumor was a mixture of tubulopapillary, trabecular and solid structures (**Fig. 2C, D**). Immunohistochemically, the tumor cells were positive for calretinin, Wilms' tumor 1, podoplanin, glucose transporter type 1 (GLUT-1) and

epithelial membrane antigen (EMA), negative for carcinoembryonic antigen, MOC-31 and Ber-EP4, and showed loss of BRCA1-associated protein (BAP1). We diagnosed the spermatic cord tumor as epithelial mesothelioma.

Interestingly, a patent processus vaginalis was found lined by a single layer of mesothelial cells from the proximal end to the tunica vaginalis (**Fig. 3A**). A single layer of atypical mesothelial cells with enlarged nuclei lined the ipsilateral side of the mesothelioma and flattened normal mesothelial cells lined the cavity on the opposite side (**Fig. 3B**). Immunohistochemically, the atypical mesothelial cells were positive for GLUT-1 and EMA, negative for desmin, and showed loss of BAP1, whereas the normal mesothelial cells showed an opposite pattern (**Fig. 3C-D**), indicating that the single-layered atypical mesothelial cells were mesothelioma in situ. Finally, we diagnosed mesothelioma in situ of the spermatic cord arising from a patent processus vaginalis. A second surgery and adjuvant therapy were not performed because there was no obvious tumor elsewhere. He was alive without signs of disease at 2 years after surgery.

DISCUSSION

Mesothelioma of the spermatic cord is a rare entity; only 9 cases have been reported so far in the English literature (**Table 1**) [2-10]. The patients most commonly presented with an inguinal mass, and all 9 patients underwent surgery. Histological findings showed no descriptions of

mesothelioma in situ. Mesothelioma arising from the peritoneum may have invaded the spermatic cord in Cases 4 and 6. Our patient being alive without signs of disease at 2 years after surgery excluded the possibility of a peritoneal origin.

BAP1 expression is detected by immunohistochemistry, which is useful for distinguishing benign and malignant mesothelial proliferations [11]. BAP1 expression is detected in the nuclei of benign mesothelial cells, whereas BAP1 loss is detected in the nuclei of mesothelioma cells [12].

Recently, Churg et al. reported two cases of mesothelioma in situ. They proposed that mesothelioma in situ be defined as well-confirmed in situ lesions and be related to genomic events such as BAP1 loss [13]. The single-layered atypical mesothelial cells in our patient were an in situ lesion and showed loss of BAP1. Thus, they were diagnosed as mesothelioma in situ of the spermatic cord arising from a patent processus vaginalis.

CONFLICT OF INTEREST

None.

FUNDING

This work did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

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FIGURE LEGEND

Figure 1. Abdominal CT showing the right spermatic cord swelling.

Figure 2. (**A**) Macroscopic findings of the spermatic cord. The spermatic cord was enlarged (arrowheads). (**B**) The cut surface showed a solid and thickened spermatic cord (arrowheads) and no involvement of the testis and tunica vaginalis (arrows). (**C**) Epithelioid mesothelioma with a papillary pattern of growth. (**D**) Epithelioid mesothelioma with trabecular and solid patterns of growth. (**C-D**): Hematoxylin and eosin stain.

Figure 3. (A) Cross-sections of the specimen of the right spermatic cord. (a) Mesothelioma of the spermatic cord (red line), (b) patent processus vaginalis (yellow line) and (c) vas deferens (green point). Patent processus vaginalis stained with hematoxylin and eosin (**B**), desmin (**C**) and BAP-1 (**D**). (**B-D**) Mesothelioma is present on the right side of the images. Cells on the right lining are mesothelioma in situ (arrowhead). Cells on the left lining are normal mesothelial cells (arrow). ※ = patent processus vaginalis.

Table 1

Table 1. Clinical and pathologic features of primary mesothelioma.

Case	Age (Y)	Asbestos exposure	Side	Symptom	Tumor metastasis	Operation	Histologic type	Postoperative recurrence	Follow-up		Ref
1	63	NA	Lt	Mass in the inguinal region	NA	NA	Biphasic	NA	NA		2
2	40	NA	Lt	High near groin	External iliac lymph node	RO, RALD	NA	No	NED	18 Y	3
3	57	NA	Rt	Inguinal hernia	No	SR	Epithelioid	Local at 12 M Periaortic, iliac lymph nodes at 34 M	DOD	42 M	4
4	46	No	Rt	Mass in the inguinal region	No	RA	Biphasic	NA	DOD	8 M	5
5	52	NA	Rt	Scrotal enlargement	No	RA	Epithelioid	Local at 9 M Iliac lymph nodes at 30 M	AWD	30 M	6
6	65	Yes	Lt	Mass in the inguinal region	No	RA	Biphasic	Local, peritoneal at 3 M	DOD	6 M	7
7	45	No	Lt	Mass in the inguinal region	No	SR	Epithelioid	No	NED	6 M	8
8	80	Yes	Rt	Mass in the inguinal region	No	RA	Biphasic	No	NED	12 M	9
9	45	No	Rt	Swelling in the inguinal region	No	RA	Biphasic	NA	NA		10
Our case	82	No	Rt	Swelling in the inguinal region	No	RA	Epithelioid	No	NED	2 Y	

Abbreviations: NA, not available; Lt, left; Rt, right; RO, radical orchiectomy; RALD, retroperitoneal and para-aortic lymph node dissection; SR, spermatic cord resection; NED, no evidence of disease; DOD, dead of disease; AWD, alive with disease; Y, years; M, months; Ref, reference.

Fig. 1

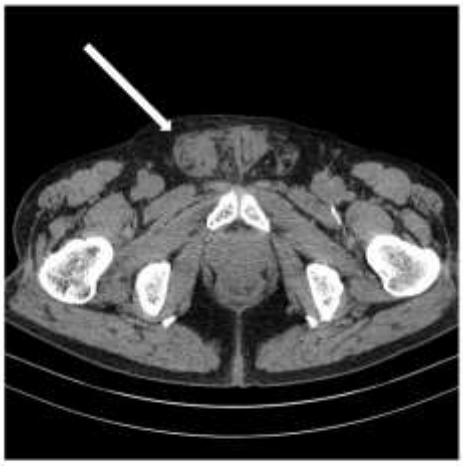
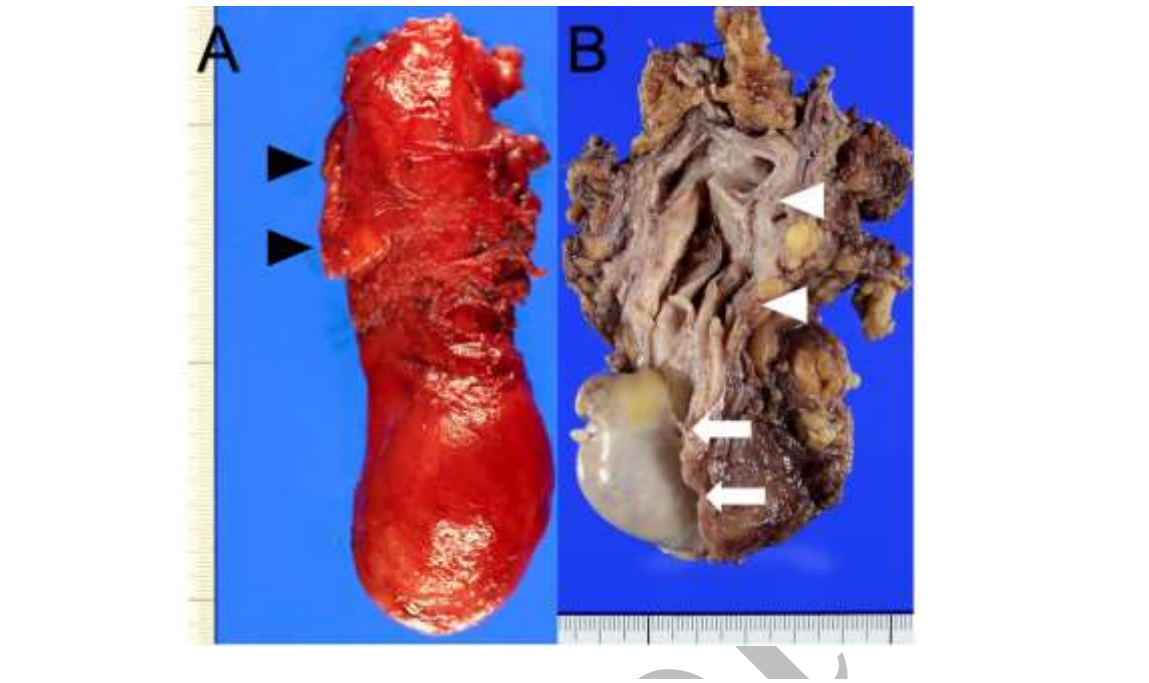


Fig. 2



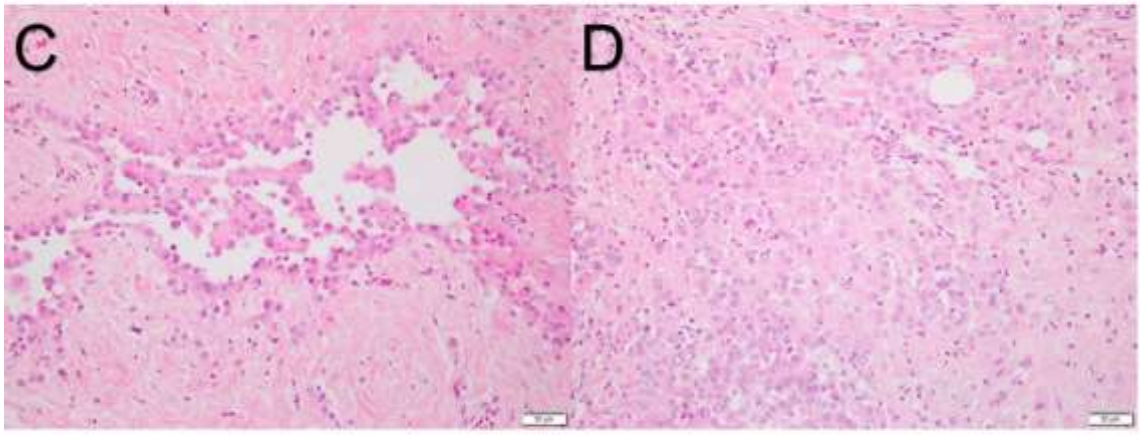


Fig. 3