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## Pleomorphic rhabdomyosarcoma of the spermatic cord and a secondary hydrocele testis: A case report

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### Abstract

#### BACKGROUND

Pleomorphic rhabdomyosarcoma (RMS) of the spermatic cord is a group of rare neoplasms, and a secondary hydrocele testis occasionally occurs. The misdiagnosis of paratesticular mass may lead to a therapeutic delay.

#### CASE SUMMARY

A 79-year-old man presented to our clinic complaining of a 1-mo history of painless scrotal swelling. Physical examination revealed approximately a 15 cm × 10 cm × 5 cm inguinal mass with limited mobility. Contrast-enhanced magnetic resonance imaging showed a hydrocele testis, several enlarged inguinal lymph nodes, and a heterogeneously enhanced lesion with a relatively well-defined margin in the left inguinal region. Due to the imaging findings, he was diagnosed with pleomorphic RMS and received a wide resection of the mass, an inguinal incision with a high section of the left spermatic cord, and a left radical orchiectomy. He experienced local relapse 1 mo postoperatively and received radiotherapy and anlotinib hydrochloride-based immunotherapy as adjuvant therapy. The patient died 3 mo after the surgery.

#### CONCLUSION

The optimal interventions for advanced-stage pleomorphic RMS patients should be investigated by more preclinical studies and clinical trials. Physicians need to be aware of the occurrence of pleomorphic RMS in unusual locations, especially when accompanied by a hydrocele testis.

**Key words:** Pleomorphic rhabdomyosarcoma; Spermatic cord; Secondary hydrocele testis; Case report

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**Core tip:** Pleomorphic rhabdomyosarcoma of the spermatic cord is rare and develops commonly in adults, which is still clinically challenging for definitive diagnosis and differentiation from other pathologies, especially when accompanied by a secondary hydrocele testis. The present case report as well as state-of-the-art literature review highlights the importance of comprehensive appreciation of laboratory, radiographic, and pathologic findings for the ultimate differential diagnosis of spermatic cord rhabdomyosarcoma.

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## INTRODUCTION

Rhabdomyosarcoma (RMS) is a highly malignant sarcoma originating from immature striated muscle, which mostly occurs in children<sup>[1,2]</sup>. Paratesticular RMS accounts for about 7% of all the rhabdomyosarcomas<sup>[3]</sup>. More than 200 cases of paratesticular RMS have been reported, including tumors of the testis, epididymis, and spermatic cord<sup>[2-7]</sup>. Although the therapeutic outcome of paratesticular RMS has been remarkably improved, it still depends on the tumor grade, distant disease, and tumor cell histology<sup>[4,5]</sup>. The 5-year overall survival rate of confined paratesticular RMS after combined therapeutic arms including surgery, radiotherapy, and chemotherapy was 94.6%<sup>[6]</sup>.

Pleomorphic RMS of the spermatic cord is one of the rarest histological types of paratesticular RMS and develops commonly in adults. Currently, only case reports with limited patient numbers and expert opinion are available for therapeutic instruction. The diagnosis of pleomorphic RMS of the spermatic cord is based on histological and specific immunohistochemistry features of the tumors. The imagological diagnosis of pleomorphic RMS of the spermatic cord is still challenging if the mass is not confined to paratesticular tissue, and is especially misled by a concomitant secondary hydrocele.

We herein report a case of pleomorphic RMS of the spermatic cord with a secondary hydrocele testis, treated by radical orchiectomy, high ligation of the spermatic cord, radiotherapy, and immunotherapy. An up-to-date literature review of pleomorphic RMS of the spermatic cord was also performed.

## CASE PRESENTATION

### Chief complaints

A 79-year-old man presented to our clinic and complained of a 1-mo history of painless scrotal swelling.

### History of present illness

A previous computed tomography (CT) scan at a local hospital identified a left 3 cm × 2 cm × 3 cm inguinal mass. The patient did not receive any treatment before he visited our clinic. His sleep and diet were not disturbed in the past 3 mo.

### History of past illness

The patient was in good health in the past and only received an appendectomy 40 years ago.

### Personal and family history

The patient reported no perceptible personal conditions related to the present clinical manifestation. Family history was also not noticeable.

### Physical examination upon admission

Physical examination revealed approximately a left 15 cm × 10 cm × 5 cm inguinal mass with limited mobility (Figure 1). The scrotum was irreducible, the left testicle was not palpable, and transillumination test was positive.





**Figure 1** Physical examination. A left scrotal mass measuring 15 cm was noted.

### Laboratory examinations

Laboratory tests showed elevated serum  $\beta$ -human chorionic gonadotropin ( $\beta$ -HCG; 240.70 mIU/mL) and squamous cell carcinoma antigen (3.20 ng/mL). Contrast-enhanced magnetic resonance imaging (MRI) showed a hydrocele testis, several enlarged inguinal lymph nodes, and a heterogeneously enhanced lesion (15 cm  $\times$  10 cm  $\times$  6 cm) with a relatively well-defined margin in the left inguinal region (Figure 2A and B).

## FINAL DIAGNOSIS

The final diagnosis of the presented case was pleomorphic RMS of the spermatic cord and a secondary hydrocele testis.

## TREATMENT

The patient received a wide resection of the mass, an inguinal incision with a high section of the left spermatic cord, and a left radical orchiectomy. Intraoperatively, the encapsulated tumor was found across the spermatic cord and extending into the scrotum, but without invasion to the testis or epididymis (Figure 2C). Pathological analysis identified that the mass was composed of sheets of giant atypical and pleomorphic cells with abundant eosinophilic cytoplasm, which led to a diagnosis of pleomorphic RMS (Figure 3A). Immunohistochemical examination showed a strong positivity for vimentin and Ki-67 (approximately 40%), as well as a weak positivity for desmin, smooth muscle actin (SMA), and myogenin (Figure 3B-F). According to the Intergroup Rhabdomyosarcoma Study Group staging classification, the tumor was in clinical group IIb and clinically staged as T2bN1M0.

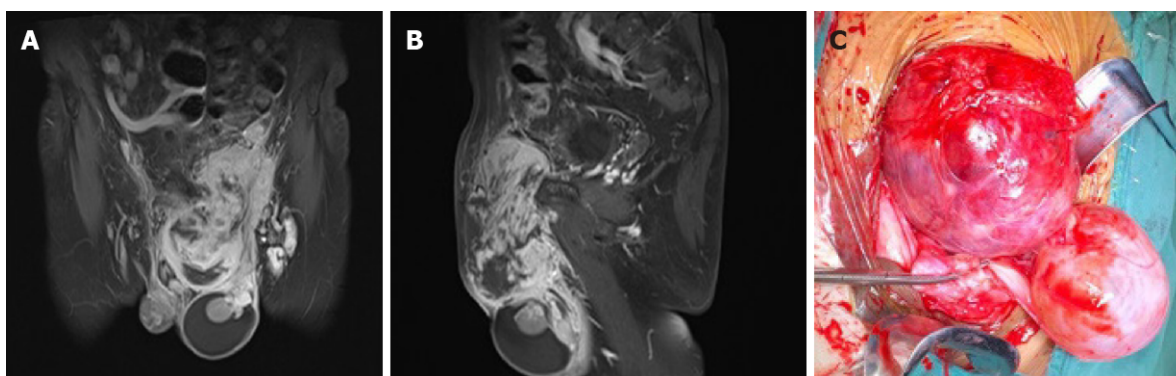
## OUTCOME AND FOLLOW-UP

The patient experienced local relapse 1 mo postoperatively. With a Karnofsky performance score of 40, the patient received combined radiotherapy and anlotinib hydrochloride-based immunotherapy without additional chemotherapies. Regretfully, the patient died from multiple organ failure 3 mo postoperatively.

## DISCUSSION

Spermatic cord RMS originates from mesenchymal components of the spermatic cord, with a poor prognosis<sup>[8]</sup>. It was commonly reported in old men<sup>[7]</sup>, but exceptional cases involved male teenagers<sup>[3]</sup>. Pleomorphic RMS of the spermatic cord is the rarest type of RMS (about 7% of all RMS and about 6% of all paratesticular tumours) with a high metastasis rate and remains a significant challenge for clinical diagnosis and





**Figure 2** Intraoperative finding. A and B: Coronal (A) and sagittal (B) views of contrast-enhanced magnetic resonance imaging showing a hydrocele testis and a heterogeneously enhanced lesion with a relatively well-defined margin; C: Intraoperative finding of the spermatic cord tumor and a secondary hydrocele testis.

treatment<sup>[8,9]</sup>.

MRI may provide significant information of tumor outline and adjacent bordering. The classic MRI feature of pleomorphic RMS of the spermatic cord is a paratesticular heterogeneously enhanced mass with or without compressing the testis. A paratesticular mass may be diagnosed empirically with hernia<sup>[10]</sup>, or hydrocele<sup>[11]</sup>, which potentially leads to a therapeutic delay. An ultrasound scan may echographically distinguish them. Tumors involving the scrotum might be masqueraded as reproductive system tumors. A secondary hydrocele testis occasionally accompanies the spermatic cord mass, which was only reported in less than ten cases since 2007 (Table 1)<sup>[10-17]</sup>. To the best of our knowledge, hydrocele testis secondary to pleomorphic RMS of the spermatic cord was not reported previously. Microscopically, the pleomorphic RMS was featured as a pleomorphic architecture of atypical/round rhabdomyoblasts and spindle cells<sup>[7]</sup>. Its determinant diagnosis is largely based on the positive immunostaining for several highly sensitive and specific markers, such as vimentin, desmin, myoglobin, myosin, and SMA<sup>[7,18]</sup>. Serum  $\beta$ -HCG generally tends to be normal in RMS patients, however, elevated  $\beta$ -HCG in our case should be highlighted as a potential diagnostic pitfall and raises the differential diagnosis from other germ cell tumors<sup>[19]</sup>.

Most of therapeutic information on pleomorphic RMS was available from some individual institutions<sup>[2-4,7]</sup>. A wide resection of tumor with a radical inguinal orchiectomy, followed by adjuvant therapy (*e.g.*, chemotherapy or radiotherapy) was the gold standard therapeutic arm for patients with a localized tumor. This is due to the fact that the tumor commonly recurs locally and invades the pelvic cavity through internal inguinal ring. However, the actual prognosis is dependent on the histology and metastatic stage of the tumors<sup>[7]</sup>. Lack of pain in the mass contributed sometimes to therapeutic delays. A wide resection of tumors with radical inguinal orchiectomy followed by VAC (vincristine, actinomycin D, and cyclophosphamide) based chemotherapy is the gold standard treatment for localized tumors<sup>[20]</sup>. The radiotherapy was recommended for controlling tumor local recurrence and remote metastasis<sup>[20,21]</sup>. The combination of these therapeutic arms may increase the overall survival of RMS towards about 70%-90%<sup>[4,22]</sup>. A poor prognosis cannot be avoided for advanced tumors. Since chemotherapy was not recommended for cases with a low Karnofsky performance score ( $< 70$ ), the present patient received only combined radiotherapy and immunotherapy after the surgery<sup>[23]</sup>. Emerging immunotherapy based on molecular mechanism of RMS is believed to improve the long-term survival of these patients<sup>[24]</sup>. However, more preclinical studies and clinical trials are warranted to identify the optimal interventions for patients in advanced stages of this disease.

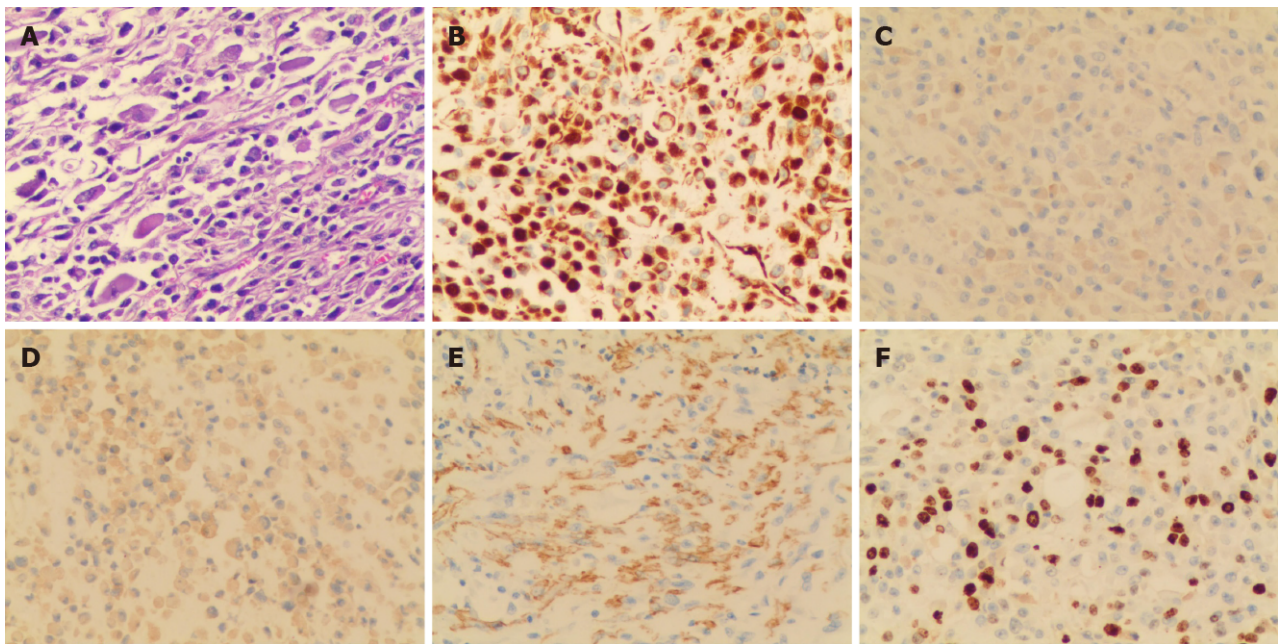
## CONCLUSION

Only scarce reports of paratesticular RMS have been reported, mainly in the form of single neoplasia without a secondary hydrocele testis. Although multi-disciplinary approach including surgery, radiotherapy, and chemotherapy/immunotherapy is performed to maximize survival, an early diagnosis should also be confirmed. Physicians need to be aware of the occurrence of pleomorphic RMS in unusual locations, especially when accompanied by a hydrocele testis.

**Table 1** Spermatic cord mass with a secondary hydrocele (review of the literature from 2007 to 2019)

Ref.	Yr	Patient's age (yr)	Side	Histologic diagnosis	Follow-up (mo after surgery)	Outcome
Li <i>et al</i> <sup>[12]</sup>	2007	71	Right	Lymphangioma	3	ANED
Lim <i>et al</i> <sup>[11]</sup>	2007	6	Right	Sparganosis	NS	NS
Domsa <i>et al</i> <sup>[13]</sup>	2008	75	-	Mixed liposarcoma	NS	NS
Chang <i>et al</i> <sup>[14]</sup>	2009	38	Right	Klatskin tumor	5	ANED
Uehara <i>et al</i> <sup>[15]</sup>	2010	59	Left	Liposarcoma	NS	NS
Park <i>et al</i> <sup>[16]</sup>	2011	65	Left	Mesothelioma	6	Died
Soda <i>et al</i> <sup>[17]</sup>	2012	67	Bilateral	Mesothelioma	3	Died
Londeree <i>et al</i> <sup>[10]</sup>	2014	70	Right	Liposarcoma	18	ANED

NS: Not stated; ANED: Alive with no evidence of disease.



**Figure 3** Histopathological and immunohistochemical analyses. A: Formalin-fixed paraffin-embedded tumor tissue was stained with hematoxylin and eosin (100 ×); B: Vimentin (200 ×, cytoplasmic and membranous staining); C: Desmin (200 ×, cytoplasmic staining); D: Myogenin (200 ×, nuclear staining); E: Smooth muscle actin (200 ×, cytoplasmic and membranous staining); F: Ki-67 (200 ×, approximately 40% positivity).

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